UNDERSTANDING SELF-RESTRAINT AND SELF-INJURIOUS BEHAVIOUR IN CHILDREN WITH AUTISM AND/OR INTELLECTUAL DISABILITY

By

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ABSTRACT

Background: Self-restraint, the self-initiated restriction of one's own body movements, is observed alongside self-injurious behaviour in populations with neurodevelopmental conditions. Self-restraint behaviours are understood through mechanisms of operant learning, however a purely operant account cannot adequately account for the presence of *both* self-injury and self-restraint. To bridge this theoretical gap, two complementary hypotheses have been proposed: the compulsivity and inhibitory control accounts. Thus far, these accounts are limited by a paucity of research, lack of focus on self-restraint measurement and reliance on indirect measures of inhibitory control. Therefore, this thesis aimed to describe self-restraint in high-risk groups, and test prevailing theoretical accounts of this behaviour across different parameters of self-restraint.

Method: A comprehensive systematic review of the existing literature provided meta-analytic estimates of the prevalence of self-restraint, self-restraint topographies, and quantified the size of association between self-restraint and putative correlates, including self-injury. A large-scale cross-syndrome survey examined transdiagnostic correlates of self-restraint, including compulsivity and anxiety, using a dimensional measurement approach. The presence and intensity of self-restraint displayed by autistic children with intellectual disability was assessed using naturalistic observational techniques. Finally, the feasibility of a novel battery of direct executive function tasks was assessed, and tasks were administered alongside indirect measures to examine the compulsivity and inhibitory control accounts.

Results: The pooled prevalence estimate of self-restraint in autistic individuals and/or those with intellectual disability was 39%, and self-restraint and self-injury shared a significant

moderate association. Compulsivity and anxiety were unique predictors of self-restraint across neurodevelopmental conditions, over-and-above confounding variables. Autistic children with intellectual disability displayed a high prevalence of self-restraint, with intensity of self-restraint varying both between and within topographies. The direct executive function tasks were deemed developmentally-appropriate for autistic children with varying degrees of intellectual disability. Indirect measures of executive function more closely associated with self-restraint than direct measures, with self-injury severity moderating the relation between poor executive function and greater self-restraint intensity.

Conclusions: The prevalence of self-restraint within populations who self-injure was consistently high, however the presence of 'multi-parameter' self-restraint was lower. Younger age, greater autism characteristics and lower adaptive functioning were important demographic factors associated with presence, topographies and intensity of self-restraint. Findings of impulsivity uniquely predicting self-restraint at high levels of self-injury, and of compulsivity and anxiety predicting self-restraint after accounting for covariates, allowed for proposal of a novel, integrated compulsivity/inhibitory control account. This suggests that 'impulsive' self-injury increases anxiety/arousal, leading to the emergence of habitual 'compulsive' self-restraint. Future research is required to examine this integrated theory.

DEDICATION

This thesis is dedicated to my Grandma and Grandpa. All that I do is for you.

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CHAPTER ONE

SELF-RESTRAINT AND SELF-INJURIOUS BEHAVIOUR: AN INTRODUCTION

1.1 Introduction

The focus of this thesis is on self-restraint, an under-researched behaviour that commonly occurs alongside self-injurious behaviour (SIB) in autistic individuals and/or individuals with intellectual disability (ID). The overarching aim of this thesis was to examine mechanisms that drive self-restraint, including SIB, and reconsider traditional accounts of operant learning to delineate putative relations with impulsivity, compulsivity and broader components of executive functioning (EF). This Chapter provides an overview of the existing literature examining self-restraint, current gaps in understanding, and rationale for the studies undertaken within this thesis. Self-restraint is described, approaches to measurement summarised, and prevailing theories discussed. Finally, an overview of each Chapter is provided.

1.2 Self-injurious behaviour

Self-injurious behaviour (SIB) refers to non-accidental behaviours that are initiated by and towards an individual, causing physical change, such as tooth marks or reddening skin, or damage, such as biting, scratching or head hitting (Oliver, McClintock, et al., 2003). The term 'SIB' was proposed by Tate and Baroff (1966) to describe behaviour that directs physical injury towards the self, as an alternative to terminology that suggests aggressive or destructive

intent, such as 'auto-aggression' and 'self-destructive behaviour'. The fact that self-injury[1] does not involve suicidal intent (Cooper et al., 2009), is constrained by operant reinforcement contingencies (section 1.4.7.1) and is primarily displayed by people with ID differentiates SIB from self-harm behaviours observed in neurotypical and autistic populations without ID (Swannell et al., 2014), which are conceptualised to occur voluntarily in response to a mental health need.

1.2.1 Consequences of self-injurious behaviour

SIB poses serious risks to physical health and wellbeing, particularly when it is repetitive, persistent and severe in nature. For example, chronic head hitting is highly prevalent among populations with autism and/or ID (Summers et al., 2017), and involves repetitive, forceful blows to localised areas of the head (Lewis & Bodfish, 1998; Symons, 2011). The strength of self-injurious head hitting has been equated to that of boxing punches (Newell et al., 2002), capable of causing permanent neurological damage, and in extreme cases, premature death (Nissen & Haveman, 1997). The negative impacts of SIB are far-reaching and unconstrained to physical damage; engaging in self-injury interferes with daily living and psychosocial development (Vandewalle & Melia, 2021), and increases risk of psychiatric hospitalisation (Mandell, 2008), intrusive restraint use (Emerson, 1990), breakdown in family care (Hastings, 2002) and use of anti-convulsant medication (Taylor et al., 2011). Therefore, SIB is considered pervasive, interfering with physical, psychological and adaptive functioning.

SIB generally emerges in early childhood and persists over-time. In a longitudinal study involving autistic individuals, Laverty et al. (2020) identified stability of self-injury over 10

[1] 'Self-injury' and 'self-injurious behaviour (SIB)' are used interchangeably throughout this thesis.

years, with baseline impulsivity, stereotyped behaviour and adaptive functioning predicting SIB stability. Taylor et al. (2011) found SIB persisted for 84% of participants over 18 years. There is suggestion that the risk of SIB emergence decreases after the age of 50 years (Davies & Oliver, 2014). Therefore, this thesis examines SIB and self-restraint displayed by individuals with neurodevelopmental conditions during childhood, a high-risk developmental period.

1.3 Neurodevelopmental conditions

'Neurodevelopmental conditions'[2] were introduced in the fifth edition of The Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association [APA], 2013). This diagnostic category describes life-long conditions characterised by differences in brain development that lead to difficulties with social, academic, personal and/or occupational functioning. Conditions onset during early childhood, rarely have a single cause and commonly co-occur (Thapar et al., 2017). Conditions include intellectual conditions, including rare neurogenetic conditions associated with ID, communication disorders, such as speech-sound disorder, Autism Spectrum Disorder (ASD)[3], Attention-Deficit/Hyperactivity Disorder (ADHD), specific learning disorders, motor disorders and tic disorders.

The combined prevalence of neurodevelopmental conditions is challenging to identify, in part due to variation in estimates of individual conditions. For example, Ogundele and Morton

[2] A growing shift towards a social model of disability has highlighted the pathologizing connotations of the word 'disorder' (Fletcher-Watson & Happe, 2019). Therefore, the alternative term 'condition' is adopted throughout this thesis to refer to 'neurodevelopmental disorders'.

[3] The term 'autism' is considered less pathologizing than 'disorder', therefore will be used in this thesis to refer to the cluster of diagnoses that the DSM-5 recognises as 'Autism Spectrum Disorder' (APA, 2013).

(2022) reported the prevalence of autism ranges within the literature from 0.8 to 3.5%, and ID from 2.1 to 3.6%. Changing diagnostic criteria and terminology over time (e.g., 'neurodevelopmental disorders' and 'neurodisabilities') and methodological variance (e.g., questionnaire versus observational assessments) may drive variance in prevalence estimates (Francés et al., 2022). Despite this, neurodevelopmental conditions are considered relatively common. In a population-wide study, Fleming et al. (2020) reported that 35,873 participants, 4.7% of schoolchildren in Scotland from 2009 to 2013, had at least one neurodevelopmental condition, while 4789 children, or 0.6%, had two or more co-existing diagnoses. Thus, while the relative number of individuals with at least one neurodevelopmental condition is low compared to the general population, the cumulative number of children in the United Kingdom (UK) with conditions such as autism and ID is substantial.

Autism is characterised by difficulties with social interaction and communication, and restrictive or repetitive behaviours/interests (APA, 2013; section 2.2). Recent estimates suggest the median global prevalence of autism is one in 100 people (Zeidan et al., 2022). Francés et al. (2022) reported that 0.7 to 3% of children are autistic[4]. ID is classified according to intellectual ability and adaptive functioning skills, whereby the individual has an intellectual quotient (IQ) score below 70 and impairments in adaptive daily functioning activities, such as feeding, dressing and washing (APA, 2013, section 2.2). It is estimated that 10.37/1000 people have ID (Maulik et al., 2011). Fleming et al. (2020) found autism and ID were the most common neurodevelopmental conditions to co-occur, with 2251 school

[4] This thesis strives to adopt identity-first language when referring to autism where possible, in keeping with preferences expressed by some members of the autism community (Kenny et al., 2016). It is acknowledged that person-first language is sometimes adopted when discussing individuals with coexisting diagnoses for grammatical reasons (e.g., person with autism and/or ID).

children, or 0.3% of the population, exhibiting both diagnoses. Further, a recent meta-analysis found 33% of autistic individuals were estimated to also have an ID (Zeidan et al., 2022).

1.3.1 Prevalence of self-injury across neurodevelopmental conditions

SIB is more prevalent in autistic populations than typically developing individuals. For example, a recent meta-analysis involving 14,379 individuals estimated 42% of autistic individuals self-injure (Steenfeldt-Kristensen et al., 2020). This is higher than estimates of self-harm in typically developing children (8%; Moran et al., 2012). Prevalence rates of SIB for individuals with ID range from 5.3% (Ruddick et al., 2015) to 47.2% (Simó-Pinatella et al., 2017), while severe SIB (the most life-threatening behaviours; Baghdadli et al., 2003) presents in 4 to 5% of individuals with ID (Ruddick et al., 2015). Populations with rare genetic disorders associated with ID also show high prevalence of self-injury, including Cornelia de Lange Syndrome (CdLS; 64%; Eden et al., 2014), Fragile X (55%; Richards et al., 2012), Tuberous Sclerosis Complex (27%; Eden et al., 2014) and Down syndrome (18%; Richards et al., 2012). Overall, degree of ID is considered a reliable correlate of SIB (Oliver & Richards, 2015). Given the association between SIB and self-restraint (section 2.2.3), and the lack of prevalence data examining self-restraint across conditions (section 2.4.2), this thesis focuses on SIB and self-restraint displayed by individuals with a variety of neurodevelopmental conditions, including autistic individuals, individuals with ID and/or individuals with rare genetic conditions associated with autism and/or ID.

1.3.2 Neurodevelopmental continuum and the ‘transdiagnostic revolution’

The high prevalence and co-occurrence of autism and ID highlights the propensity for an individual to have more than one neurodevelopmental condition (Francés et al., 2022). For example, Hansen et al. (2018) reported 21.7% individuals with neurodevelopmental

conditions had at least two conditions. Saito et al. (2020) found 88.5% of autistic children had at least one additional neurodevelopmental condition, while 23% of autistic children reported two further conditions: ID (37%) and borderline intellectual functioning (21%). Altogether, the co-existence of multiple neurodevelopmental conditions supports the growing shift towards adopting a transdiagnostic approach to neurodevelopmental conditions, exploring attributes along a continuum rather than according to diagnosis (Finlay-Jones et al., 2019).

Traditionally, neurodevelopmental conditions are studied according to discrete diagnostic categories. Transdiagnostic approaches aim to transcend diagnostic boundaries, recognising the wide variety of experiences and characteristics displayed by individuals with the same diagnosis (Astle et al., 2022) as well as the shared attributes between diagnoses (Finlay-Jones et al., 2019). The neurodevelopmental continuum model suggests that meeting diagnostic criteria is less important in understanding behavioural outcomes than the presence of specific traits (section 3.2). This has implications for understanding and treating behaviours such as SIB in populations with 'sub-threshold' conditions, who may not meet diagnostic criteria or have access to a diagnosis, despite experiencing similar behavioural challenges (Astle et al., 2022). This is important given barriers to accessing diagnoses in the UK. For example, 86% of people (121,000 individuals) waited longer than the recommended three months for an autism diagnosis in December 2022 (NHS Digital, 2023). Therefore, to expand upon current understanding, Chapter 3 studied self-restraint across the neurodevelopmental continuum.

1.4 Self-restraint

Self-restraint refers to non-accidental behaviours that are initiated by an individual and directed towards the self to restrict one's own physical movement(s). Self-restraint has been

observed alongside pervasive self-injury in autistic individuals, individuals with ID and/or those with associated genetic syndromes. Most existing research focuses on understanding the functional role of self-restraint in attenuating SIB (section 1.4.7.2). However, consistent reports of self-restraint in the absence of SIB confound understanding (section 2.4.5). Therefore, while this thesis primarily tests hypotheses related to self-restraint in the presence of SIB, an additional focus is placed on understanding the theoretical and clinical significance of self-restraint in the absence of SIB.

1.4.1 Background of self-restraint and self-injury: Lesch-Nyhan syndrome

Around 20 years before the term 'self-restraint' was used in the context of self-injury (e.g., by Pace et al., 1986), reports of self-restraining behaviours shown by individuals with Lesch-Nyhan syndrome were published (Lesch & Nyhan, 1964). Lesch-Nyhan syndrome is a rare genetic disorder with a prevalence of 0.0005 cases per 100 people (D'Souza & Karmiloff-Smith, 2017), and is associated with ID. Severe SIB is a pervasive behavioural challenge for individuals with Lesch-Nyhan syndrome (McCarthy et al., 2011; Nyhan, 1976). Estimates suggest 45% of individuals obtain permanent lip or inner-cheek damage (Anderson & Ernst, 1994; Christie et al., 1982), with self-injury so extreme many children partially amputate fingers or the tongue (Nyhan, 1976). Thus, SIB is uniquely common and clinically damaging for individuals with Lesch-Nyhan syndrome.

The most common intervention for severe SIB in Lesch-Nyhan syndrome is imposed restraint (Olson & Houlihan, 2000; Wurtele et al., 1984), including the use of gloves, helmets, arm splints and arm or hands straps (McCarthy et al., 2011). Many individuals with Lesch-Nyhan syndrome require restraint devices 100% of the time (e.g., 18 out of 37 patients; Anderson &

Ernst, 1994). Interestingly, many individuals with Lesch-Nyhan syndrome show a strong preference for restraint and appear "terrified" when restraint is removed (Nyhan, 1976, p.239). Restraint removal is typically followed by immediate self-injury, such as hand biting (Nyhan, 1976), and/or attempts to physically restrict movement (self-restraint), for example by clasping hands behind the back (Wurtele et al., 1984), sitting on hands, or tucking hands under seatbelts (Christie et al., 1982). Self-restraint following removal of mechanical restraint has therefore been theorised to function as avoidance of SIB (Christie et al., 1982). A change in personality has also been reported in response to restraint removal. Nyhan (1976) described how a relaxed and co-operative restrained individual becomes agitated, screams, and desperately attempts to re-access restraint if removed. Consequently, a strong preference for restraint, and the emergence of self-restraint in the absence of mechanical restraints, has prompted commentary on the seemingly 'involuntary' displays of self-injury.

Silverman et al. (1984) described self-injury in Lesch-Nyhan syndrome as appearing to "belong to someone else" (p.545), whereby individuals self-restrain seemingly in attempt to regain behavioural control. Insightful evidence from an individual with Lesch-Nyhan syndrome was reported by Buzas et al. (1981):

> Curiously enough, Bonny [*sic*] believed that he could not control his mutilative behavior and as a result, he, himself, had asked the staff not to remove his restraints. He even dictated a memo to the unit director asking to have the attendants stop asking if he wanted his restraints off. During the periods when his restraints were removed, 10 minute baths and infrequent swim sessions, Bobby either sat on his hands or clutched them to his sides. (Buzas et al., 1981, p.50).

Altogether, early reports of 'uncontrollable' self-injury and a unique preference for restraint in Lesch-Nyan syndrome laid the groundwork for exploring the phenomenon of restraint-seeking. The term 'self-restraint' was later applied (e.g., Pace et al., 1986; Rojahn et al., 1978;

Silverman et al., 1984) to describe both self-initiated restraint (i.e., use of own body to restrict movement) and a preference for imposed restraint (i.e., being held by other people or use of mechanical devices). Self-restraint has since attracted academic interest due to its clinically significant consequences.

1.4.2 Consequences of self-restraint

Self-restraint is of interest for both practical and theoretical reasons. Practically, self-restraint has been associated with a range of deleterious consequences. Extreme and prolonged restraint can render individuals immobile (Isley et al., 1991) posing serious risks to physical health, including halted motor development and muscular atrophy (Smith et al., 1992). Ill-fitting restraint devices can lead to bruising or loss of circulation (Hagopian et al., 2015). Powers et al. (2007) described a 16-year-old girl with moderate ID and SIB who self-restrained by looping her fingers through arm splint straps for 97% of the observation time, cutting of blood circulation to her fingers and impacting motor, educational and adaptive abilities. Chronic self-restraint can also impact socialisation and reduce opportunities to engage in independent functional skills (Scheithauer et al., 2015), and quality of relationships with others (Christie et al., 1982). Hagopian et al. (2023) described self-restraint as "debilitating and self-limiting" (p.575) due to its impact on adaptive functioning. Preferred long-term restraint may also impact academic achievement by contributing towards school-withdrawal (Jennett et al., 2011). Altogether, seeking a state of restraint can impact physical and psychological wellbeing. Importantly, self-restraint is most often displayed by individuals who also show SIB (e.g., section 1.4.1), behaviours that have deleterious consequences for individuals and their carers (section 1.2.1). Hagopian et al. (2023) reported that individuals with automatically-reinforced SIB and self-restraint show high treatment resistance. Thus, the

concomitance of both behaviours poses significant challenges to clinical outcomes.

Theoretically, the presence of self-restraint in response to self-injury suggests that the operant learning model of self-injury, the prevailing explanatory model for SIB, cannot account fully account for behaviour (section 1.4.7.1). Despite the clinical significance of self-restraint, however, there is a lack of research focusing on self-restraint, and as a result the phenomenon is not well-understood.

There are several possible explanations for the paucity of self-restraint research. Firstly, purposeful self-restrictive practices are relatively common within the general population. For example, it is not unusual to sit on hands or cross arms to prevent fidgeting (Sturmey, 2015). The breadth of topographies and their use in populations without SIB mean it is challenging to distinguish seemingly 'typical' behaviours from the clinically significant behaviours. In contrast, the dearth of self-restraint research may be explained by the relative rarity of 'functional' self-restraint that controls SIB (section 1.4.7.2). For example, Kerth et al. (2009) stated that professionals were likely to encounter only a "handful" of individuals exhibiting both SIB and self-restraint throughout their entire careers (p.192). More recently, however, cross-sectional studies have identified a high (but variable; section 2.2.1) prevalence of concomitance, where up to 92% of individuals who self-injure also self-restrain (Oliver et al., 2003). Thus, rather than rarity of behaviour, the relative paucity of self-restraint research may be better explained by the highly complex relation it appears to share with SIB (Kerth et al., 2009).

1.4.3 Prevalence of self-restraint

The prevalence of self-restraint varies widely across studies (section 2.4.2). Fovel et al. (1989) found up to 2.9% of individuals with ID showed self-restraint, while Oliver and colleagues (2003) reported a prevalence of 76.1%. Variation is attributed to differences in the conceptual and practical definitions of self-restraint between studies, and methodological variance, such as study characteristics (e.g., sample size) and participant characteristics (e.g., age, sex[5], diagnosis; section 2.2.1). Such variance also influences prevalence estimates of self-restraint shown by those known to self-injure (7-92%; Iwata et al., 1994; Oliver et al., 2003; section 2.3.3). Given the potentially adverse consequences of self-injury and prolonged self-restraint, Chapter 2 aims to establish the meta-analytic prevalence estimate of self-restraint in populations at high-risk for SIB.

1.4.4 Definitions of self-restraint

Self-restraint is a challenge to define, partly due to the wide breadth of topographies that may be considered 'self-restraint', and differences in the interpretation of behaviours. Nevertheless, most, if not all, descriptions of self-restraint in the literature agree on two core definitions: self-initiation of behaviour and restriction of own body movements. Self-initiation refers to engaging in self-restraint without being asked or told to, and engagement in self-restraint without the help of another person (except to request imposed or physical restraint, section 1.4.5.1). Descriptions of individuals seeking, gesturing for and even begging for mechanical restraints (Seegmiller, 1980) indicate self-initiation. While the function of behaviour cannot be inferred from the presence of self-initiation alone, it is important to

[5] Sex, the biological construct, and gender, the social and cultural construct, are separable. For clarity, terminology adopted within this thesis reflects terminology presented to caregivers during data collection, which varies between Chapters. For simplicity, references to 'sex' and 'gender' are both considered to represent biological sex unless otherwise stated.

establish that self-restraint behaviours are voluntary, rather than involuntary. Other definitions of self-restraint only specify restriction of own body movement (e.g., Bruhl et al., 1982; Fovel et al., 1989; Isley et al., 1991; Jennett et al., 2011; Laverty et al., 2020), while others define self-restraint by topography (e.g., Iwata et al., 1994; Kerth et al., 2009; Lerman et al., 1994; Oliver et al., 2003). Rojahn (1986) defined self-restraint as stereotypy, suggesting behaviours are not driven by internal goals. Finally, purely topographical definitions of self-restraint make no suggestion of behavioural function. Therefore, there are several definitions of self-restraint observed in individuals with neurodevelopmental conditions that do not assign a functional relation with SIB.

Many researchers define self-restraint in relation to SIB. Some suggest that self-restraint functions to prevent, avoid or suppress SIB (Hyman et al., 2002; King, 1993; Laverty et al., 2020; Powell et al., 1996; Rooker et al., 2022; Silverman et al., 1984; Smith et al., 1992) while others have defined self-restraint as a behaviour that is physically incompatible with self-injury (Hagopian et al., 2015, 2017, 2020, 2023; Rooker et al., 2020). For example, Lovaas and Simmons (1969) described an individual who held cups in both hands so that they could not hit their head. Forman (2003) suggested self-restraint that is incompatible with SIB functions to avoid or suppress SIB. However other researchers have combined two or more parameters of self-restraint in their definition of the behaviour. For example, Schroeder and Luiselli (1992) defined self-restraint both topographically ("entangle their legs and arms in clothing or furniture or lie on their arms or legs", p.293) and functionally (i.e., behaviour functions to prevent SIB). Agreement on a single definition of self-restraint is complicated by the range of topographies of self-restraint. For example, Forman (2003) described 94 topographies of self-restraint shown by 55 individuals. Therefore, to understand the

phenomenon of self-restraint and interpret the existing literature, it is necessary to first define the parameters of self-restraint.

As noted above, self-restraint behaviours may occur for a variety of reasons. Oliver et al. (2003) proposed an important distinction between the presence of self-restraining behaviours (i.e., solely topographical definition) and the use of self-restraint to manage behaviour. Authors suggested self-restraint behaviours alone may be common and have no significant functional properties. Rather, it is the presence of additional parameters of behaviours observed alongside behaviours that indicate functional self-restraint. These parameters are now described in turn.

1.4.5 Parameters of self-restraint

1.4.5.1 Topography

Topography refers to the specific behaviour observed. Isley et al. (1991) recognised the breadth of topographies reported in the literature and grouped these into three categories of self-restraint. Each category, alongside an additional fourth category proposed by Oliver et al. (2003), is described.

The first category involves *restriction of body movements using clothing* that is worn, such as entwining hands inside one's shirt (Hardy et al., 1984), hiding hands under a coat (Korten et al., 1975), pushing hands into pockets (Rojahn et al., 1978), and trapping arms in the main body of clothing (e.g., t-shirt or jumper; Forman et al., 2002). Material that is not worn may also be used to restrict movement, such as pushing hands and arms into a rolled-up towel (Bruhl et al., 1982) and wrapping one's body in a sheet (May et al., 1981; Underwood et al.,

1989) or blanket (Bruhl et al., 1982).

The second category involves *using one's own body to limit movement*, for example sitting on hands (Ball et al., 1975), holding arms behind back (Myers & Deibert, 1971), using own body weight to pin legs or arms down (Silverman et al., 1984) and putting hands under thighs (Pace et al., 1986). The third category involves *holding or squeezing objects to restrict movements*, such as holding a cup (Lovaas & Simmons, 1969), a ball (Tarpley & Schroeder, 1979) or a pillow (Bruhl et al., 1982). Individuals may also cling to furniture (Callias et al., 1973) by locking arms tightly under arm rests of chairs (Rojahn et al., 1978) or tucking arms under tables (Underwood et al., 1989).

Oliver et al. (2003) identified a fourth category describing *a preference for imposed physical or mechanical restraint*. This includes gesturing for or requesting another person to physically restrain the body (May et al., 1981) or hands (Ball et al., 1975). For example, an individual may cling to people, climb into someone's lap (Tate & Baroff, 1966), hug people (Underwood et al., 1989) or hold another's hand with both hands (Derby et al., 1996). This fourth category also includes a preference for mechanical restraint devices (Saposnek & Watson Jr, 1974) such as leg restraint (Lovaas & Simmons, 1969), arm splints (Derby et al., 1996; Foxx & Dufrense, 1984; Oliver et al., 1998; Smith et al., 1992), helmets (Muttar, 1975), posey belts (Bruhl et al., 1982; Underwood et al., 1989) or straightjackets (Kinnell, 1984; Zehr & Theobald, 1978).

Regarding imposed restraint, there are reports in the literature of 'stimulus control' devices. This refers to the use of an object to control behaviour even if it does not restrict movement.

For example, a survey of individuals with Lesch-Nyhan syndrome revealed 46% of participants sought restraint in response to self-injurious biting that did not restrict hand movement, such as seeking lightweight gloves or bandages (Anderson & Ernst, 1994). Understanding of stimulus control has developed from case studies whereby restraints are systematically faded (i.e., the restrictive properties decreased). For example, Oliver et al. (1998) found that a non-restrictive arm splint (i.e., 100% flexion was possible) maintained reinforcing properties, such that while it was worn self-injury and self-restraint did not occur. Scheithauer et al. (2015) reported low levels of self-restraint while splints were worn, even when full flexion was possible. Such non-restrictive devices have been termed "symbolic restraints" (Pace et al., 1986, p.384), and examples include eyeglasses (Foxx & Dufrense, 1984) and wrist-bands (Pace et al., 1986). In short, the presence of symbolic restraints in some cases serves as discriminative stimuli for injury-free behaviour.

It is not uncommon for an individual to display multiple self-restraint topographies from different categories. For example, Smith et al. (1992) described a child who displayed at least six forms of self-restraint, including using clothing, objects and other people. Cross-sectional research has revealed similar findings: Forman (2003) administered the Self-Restraint Questionnaire (SRQ; Oliver et al., 2003), which measures 23 forms of self-restraint, to 63 individuals with ID who showed SIB. Findings indicated that individuals displayed an average of two behaviours, ranging from one to 16. Furthermore, the Self-Restraint Checklist (SRC; Powell et al., 1996) measures seven topographies (including an additional 'other' item). Powell et al. (1996) reported that while many participants, adults with ID and SIB, displayed one self-restraint topography (52%), the mean number of topographies displayed was 1.98, ranging from one to five behaviours. Finally, Hyman et al. (2002) reported 48% of

participants with CdLS displayed one topography. Interestingly despite the SRQ measuring over three times the number of topographies than the SRC, across both measures the average number of topographies displayed was around two. Therefore, there are many forms of self-restraint, and individuals with ID tend to display more than one topography. It is important to identify the number of self-restraint topographies an individual shows, as research has shown that different forms of self-restraint may be associated with different functions of behaviour, for example to prevent different topographies of SIB (Rojahn et al., 1978). Therefore, Chapter 3 describes the first study to measure self-restraint as a continuous variable representing the number of topographies displayed.

The parameter of topography is vital for describing phenomenon. It may not be adequate, however, to infer the function of behaviour based solely on a topographical definition of self-restraint. This is because some forms of self-restraint, such as sitting on hands or wrapping a blanket around oneself, are typical of all individuals, and are not exclusively exhibited by individuals who self-injure. However, a topography-only definition of self-restraint is not without value. For example, identification of incompatible topographies of self-restraint and self-injury may reveal important information about the putative function of behaviours. Therefore, topography is a fundamental parameter for identifying and describing self-restraint.

1.4.5.2 Intensity

Intensity refers both to how many times an individual self-restrains (i.e., frequency) and/or the amount of time an individual spends engaging in any given topography of self-restraint (i.e., duration). Intensity represents the pervasiveness of self-restraint. The combination of

frequency and duration reflects the conceptual overlap of the individual parameters and highlights the lack of precise data reporting frequency and duration of self-restraint.

Oliver (1991) provided detailed frequency data using the SRQ with individuals with ID who showed SIB. The behaviour 'pushes hands inside trousers' occurred 'half of the time' for 3% of participants. 'Entwines hands in the front part of clothes', 'lies on stomach holding hands behind back' and 'indicates desire for hands to be held' occurred 'most of the time' for 3% of participants. Finally, 'carries object around without manipulating, using or playing with it' occurred 'all of the time' for 8% of participants. Overall, while these data indicate the frequency of self-restraint, and potential for behaviours to become persistent (section 1.4.2), a focus on frequency alone may underrepresent the severity or impact of self-restraint. It is therefore useful to combine frequency and duration data. Forman et al. (2002) reported duration of self-restraint behaviours (e.g., trapping arms in clothing) observed in a 26-year-old man with Down syndrome and profound ID. Across the 16.5 hours of observations over three weeks, self-restraint occurred for 96% of the total observation time. Under conditions of experimental functional analysis (section 1.4.7.2), Scheithauer et al. (2015) reported a 12-year-old girl with Down syndrome showed self-restraint for 9.8% of the observation time and did not show self-restraint when wearing arm splits with any rigidity.

Case studies have repeatedly noted extreme amounts of time spent in restraint, but precise durations are scarcely reported. For example, Jennett et al. (2011) described a 17-year-old autistic female with a severe ID who gestured for 'homemade' restraints "approximately 23h a day" (p.1111). The restraint involved wearing long socks over hands and forearms, then attaching forearms together and securing behind the back using Velcro straps. Pace et al.

(1986) described an 18-year-old man with a profound ID held in place by rigid tubes that restricted arm movement "24 hours a day" (p.382), and O'Reilly et al. (2003) described a 27-year-old man with Down syndrome and profound ID who wore rigid arm sleeves "24 hours per day" (p.542). This was despite the individual having the physical ability to remove the sleeves himself, therefore this, coupled with the fact sleeve removal led to distress and SIB, suggested the devices had obtained reinforcing properties and therefore can be considered 'self-restraint'. Finally, Kinnell (1984) described a 17-year-old autistic female with severe ID who had an 'addiction' to wearing a straightjacket, which was worn "more or less round the clock" (p.78). Overall, there are many reports of individuals with ID seeking prolonged restraint. However, it is important to acknowledge the sample bias of case study reports and therefore existing data on self-restraint duration are not necessarily typical of all forms of self-restraint for all individuals who self-injure.

In summary, the parameter of intensity combines both frequency and duration to indicate the clinical significance of self-restraint. For example, an individual who seeks self-restraint (including mechanical restraint) for prolonged durations may be at higher risk of the deleterious consequences of self-restraint (section 1.4.2). However, there are limited data directly reporting intensity of different topographies of self-restraint (i.e., beyond mechanical restraints) in samples more representative of the broader population. This was therefore the primary aim of Chapter 5.

1.4.5.3 Preference for behaviour

Preference for behaviour refers to negative affect and increase in SIB when self-restraint is terminated, alongside positive affect when self-restraint is available. Preference for self-

restraint may be indicated by anxiety upon restraint removal. For example, an individual may appear fearful (Lovaas & Simmons, 1969; Nyhan, 1976), agitated (Foxx & Dufrense, 1984) and angry when self-restraint is unavailable (Kinnell, 1984). The individual may scream, appear pained (Nyhan, 1976) and cry without tears (Saposnek & Watson Jr, 1974). Restraint removal may also lead to refusal to eat or sleep (Kinnell, 1984; Lovaas & Simmons, 1969; Nyhan, 1976). Importantly, distress alleviates once restraint is available or reapplied.

The immediate occurrence of self-injury once self-restraint is terminated is another indication of preference for behaviour (e.g., Foxx & Dufrense, 1984). Examples include hand biting and tearing flesh (Nyhan, 1976) and intense slapping of the temples resulting in bruising and abrasions (Saposnek & Watson Jr, 1974). SIB may persist until restraint is reapplied.

Preference for self-restraint is also indicated by the enjoyment of engaging in self-restraint (Favell et al., 1981), and feelings of safety and contentedness while self-restraining (Bruhl et al., 1982). An individual may seek (Foxx & Dufrense, 1984), gesture for (Jennett et al., 2011) or look for restraint (Bruhl et al., 1982), and assist with the application of restraint (Bruhl et al., 1982; Foxx & Dufrense, 1984). Anderson and Ernst (1994) reported 87% of participants (*n*=33 individuals with Lesch-Nyhan syndrome) made decisions around accessing restraint, with 92% expressing a desire to be restrained. Finally, an individual may engage in alternative forms of self-restraint when preferred forms are unavailable. For example, Buzas et al. (1981) described an individual who sat on their hands when arm restraints were removed. Overall, the parameter of preference for behaviour (i.e., enjoyment of self-restraint, and anxiety and SIB upon restraint removal) may be indicative of self-restraint that functions to avoid, prevent or terminate SIB (section 1.4.7).

In conclusion, there are multiple parameters of self-restraint. Previous research suggests that the greater number of parameters observed provides stronger evidence of functional self-restraint (Forman, 2003; Oliver et al., 2003). For example, self-restraint topographies that occur at a high frequency with a long duration (i.e., intense self-restraint), that are self-initiated, preferred, and incompatible with SIB, or control behaviour through stimulus control contingencies, are most likely functioning to escape SIB. To look beyond the case study literature, it is important to measure parameters of self-restraint and test hypotheses of behaviour within larger samples of high-risk populations.

1.4.6 Existing measures of self-restraint: A taxonomy

It is important to examine the parameters assessed by different measures of self-restraint to make inferences about behaviour. There are four main measures of self-restraint: the SRC (section 3.3.4.2.5), the SRQ (section 4.3.4.2.2), the Self-Restraint Questionnaire 2 (SRQ2; Forman, 2003; section 4.3.4.2.3) and direct observation (section 5.3). Topographies measured by the SRC, such as 'positions self to restraint' (item three), and the SRQ, such as 'pushes hands into pockets' (item five), derive from the case study literature describing individuals who also showed self-injury, such as 'pinning legs or arms down using body weight' (Silverman et al., 1984) and 'tucking hands in pockets' (Rojahn et al., 1978). Table 1.1 summaries the parameters measured and scoring criteria of these tools adopted within this thesis. Figure 1.1 provides a diagrammatic representation of self-restraint taxonomy utilised throughout Chapters.

In summary, self-restraint measures vary in the number and types of parameters assessed, and restrictiveness of definition. Given such variation, it is important to examine the relation

between different assessments, and associations with putative correlates. Informant-report tools and observational methods have not yet been compared between and within samples, limiting understanding of the relative value of each measure. Therefore, this thesis used all described measures of self-restraint to compare associations with demographic, behavioural and cognitive variables. This allowed for testing of prevailing explanatory hypotheses of self-restraint based on different definitions and behavioural parameters.

Table 1. 1 Description of self-restraint measurement tools, parameters assessed and scoring criteria

Measure		**Parameter**		**Scoring**			**Chapter used**
Name	Brief description	#	Name	'Self-restraint' group	'No self-restraint' group	Total score	
SRC	Informant-report questionnaire. Checklist of seven behaviours, including free textbox to describe 'other'.	1	Topography	Answered 'yes' to at least one item	Answered 'no' to all items	**'Total topography score'** Sum number of topographies checked 'yes' (0-8). Higher scores represent greater number of topographies.	Chapter 3
Direct observation of behaviour	Records topography and intensity of self-restraint behaviours within a specified timeframe under certain conditions. Naturalistic observation or experimental functional analysis.	2 [a]	Observed topography and intensity	Displayed at least one form of self-restraint at least once during observation time	Did not display any self-restraint during observation time	'**Total observation intensity score'** Percentage of time spent engaging in self-restraint while visible (intensity proportion) transformed to a continuous score [b]. Higher scores represent higher intensity.	Chapter 5

Note. # = number of parameters; SRC = Self-Restraint Checklist.
[a] Parameters refer to those extracted in this thesis (Chapter 5). [b] See section 5.3.4 for full description.

Table 1.1 cont.

Measure		Parameter		Scoring			Chapter used
Name	Brief description	#	Name	'Self-restraint' group	'No self-restraint' group	Total score	
SRQ	Informant-report questionnaire. Measures the frequency of 23 listed behaviours ('never', 'some', 'half', 'most' or 'all of the time'), including one free textbox to elaborate on frequent behaviours, and one free textbox to describe 'other' forms of self-restraint.	2	Informant-report topography and intensity	Scored above zero ('some', 'half', 'most' or 'all of the time') on at least one item (1-23)	Scored zero ('never') on all items (1-23)	**'Total SRQ intensity score'** Sum scores of items 1 to 23 (each item scored from zero, 'never', to four, 'always'). Higher scores represent greater number of topographies *and* higher intensity of behaviour.	Chapters 5 and 6

Note. # = number of parameters; SRQ = Self-Restraint Questionnaire.

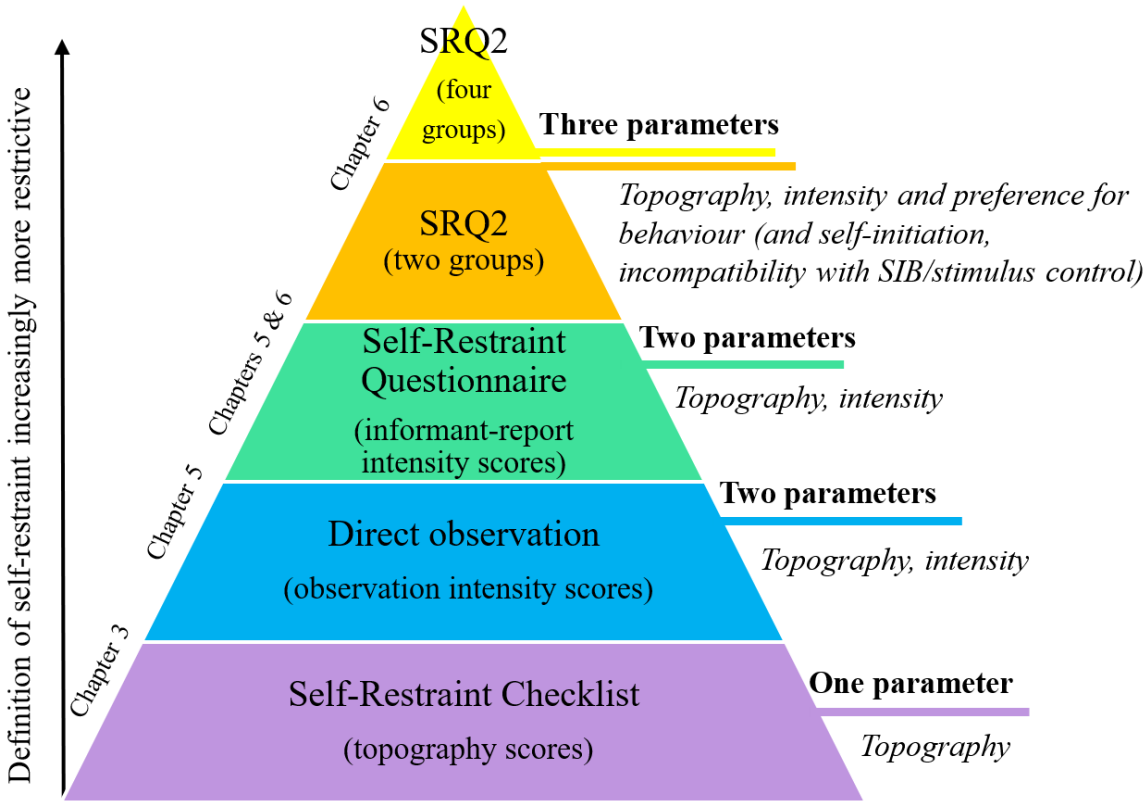
Table 1.1 cont.

Measure		Parameter		Scoring		Chapter used
Name	Brief description	#	Name	Groups	Total score	
SRQ2	Informant-report questionnaire. Measures additional parameters of behaviour reported in SRQ (six items for individuals who SIB, four items for those who do not SIB).	3	Topography [c], intensity [c] and preference for behaviour (and confirms self-initiation, incompatibility with SIB/ stimulus control [d])	**'Multi-parameter groups'** *If child self-injures* - Yes self-restraint/yes SIB (met criteria for all parameters) - No self-restraint/yes SIB (did not meet criteria for all parameters) *If child does not self-injure* - Yes self-restraint/no SIB (met all parameters except incompatibility with SIB) - No self-restraint/no SIB	N/A	Chapter 6

Note. # = number of parameters; SRQ2 = Self-Restraint Questionnaire 2.

[c] Preliminarily measured by SRQ. [d] Factors are not considered parameters, rather core descriptors of self-restraint.

Figure 1. 1 Diagrammatic representation of self-restraint measurement taxonomy



Note. SRQ2 = Self-Restraint Questionnaire 2.

1.4.7 Dominant theories of self-injury and self-restraint

1.4.7.1 Operant learning model: Self-injurious behaviour

Most theories of self-restraint build upon theoretical understanding of SIB, which is best understood through the Skinnerian theory of operant learning (Emerson, 1998). According to this model, SIB emerges as an adaptive learned response, functioning to access environmental and/or sensory rewards contingent upon triggering antecedents. Automatic sensory feedback may positively reinforce SIB, while removal of aversive sensations or pain can serve as negative reinforcement. Environmental antecedents such as low social attention or demanding tasks may 'trigger' SIB. Here, a 'mutual reinforcement cycle' (Oliver & Richards, 2015) may

form, whereby the individuals' SIB is reinforced by socially mediated environmental contingencies (e.g., caregiver attention or demand removal)[6], but the caregivers' socially mediated response is also, in turn, reinforced by the cessation of the SIB.

Despite operant theory dominating understanding of SIB maintenance, and the extensive supporting evidence (e.g., Iwata & Dozier, 2008; Matson & Lovullo, 2009), the operant model has limitations for understanding behaviour. This is evidenced by the intriguing phenomenon of individuals attempting to restrain their own body movements. It is paradoxical that an individual, who, according to the operant model, exhibits SIB as a learned response to access reinforcing consequences, would self-initiate a behaviour that terminates the self-injury preventing access to those reinforcing consequences. If SIB is purely a learned behaviour, it should be constrained by the environmental or sensory contingencies that it functions to obtain, and therefore should not require any external motoric inhibition (i.e., self-restraint). Thus, a purely operant account of SIB is theoretically incompatible with the "curious" phenomenon of self-restraint (Fisher & Iwata, 1996, p.93)

1.4.7.2 Operant learning models: Self-restraint

Many explanations for self-restraint suggest these behaviours are reinforced through operant learning. First described by Baroff and Tate (1968), the prevailing theory is that through negative reinforcement self-restraint is reinforced by avoidance or escape from SIB (Luiselli, 1993). This suggests that self-restraint emerges as a response to the pain or discomfort caused by self-injury, and it is the removal of this pain that negatively reinforces self-restraint for

[6] Crucially, the caregiver does not intend to reinforce behaviour, rather they intend to stop self-injury. For the caregiver, the removal of SIB following intervention negatively reinforces the intervening behaviour.

future occurrences of SIB (Figure 1.2). Over time this cycle may become an avoidance-motivated behaviour, whereby the individual seeks self-restraint in the presence of the antecedent before the SIB occurs, avoiding any experience of SIB-related discomfort altogether (Figure 1.3). This may explain surprisingly high reports of self-restraint in the absence of SIB (e.g., 66%, Laverty et al., 2020; section 2.4.2).

Figure 1. 2 Diagrammatic representation of self-restraint negatively reinforced by escape from self-injurious behaviour

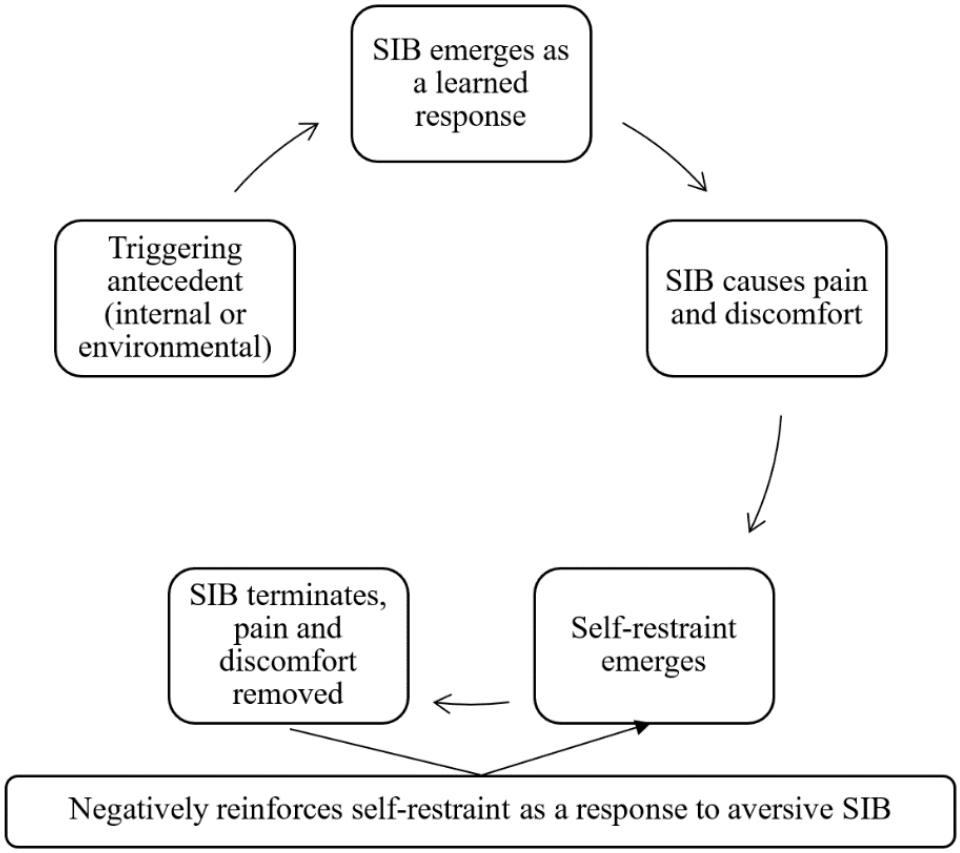
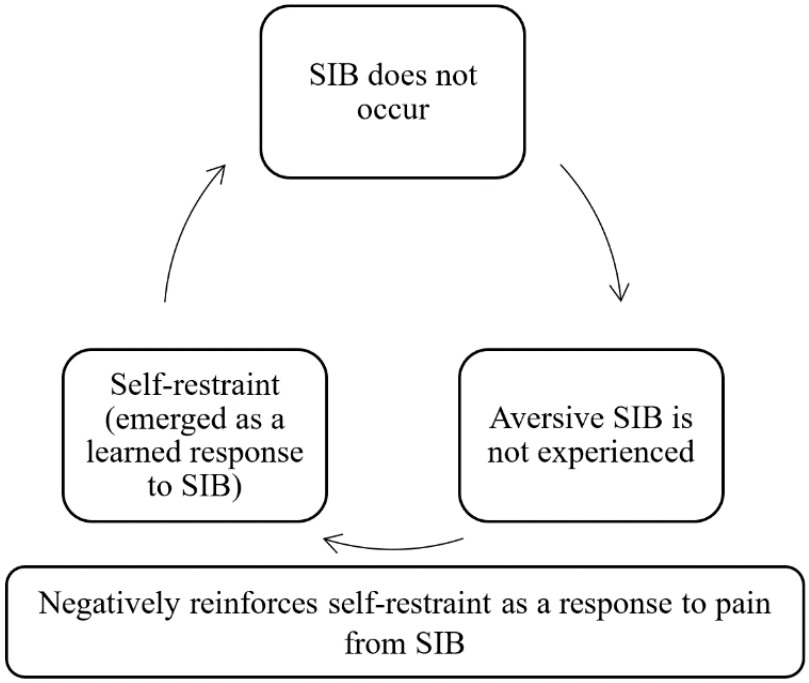


Figure 1. 3 Diagrammatic representation of self-restraint negatively reinforced by avoidance of self-injurious behaviour



Experimental functional analysis, a procedure that identifies operant functions of behaviours by systematically manipulating antecedents and consequences (Matson & Lovullo, 2009), supports this operant account. An inverse-relation between self-restraint and SIB has been established, whereby SIB (e.g., face hitting) decreases when self-restraint (e.g., entwining fingers in material) is available (Powers et al., 2007). The self-injury may function to escape task demands (e.g., Smith et al., 1992), access objects such as food (Kerth et al., 2009) or serve as automatic reinforcement (e.g., Hagopian et al., 2015; Powers et al., 2007; Rooker et al., 2020). This reveals a complex cycle of reinforcement between antecedents, SIB and self-restraint. The relation between SIB and self-restraint is examined using experimental functional analysis sub-conditions where self-restraint is available and unavailable. Within non-verbal males with profound ID, Smith et al. (1992) found results from two participants

that were consistent with negative reinforcement (i.e., low levels of SIB and high levels of self-restraint occurred in the self-restraint-available sub-condition, and high levels of SIB occurred during the self-restraint-unavailable sub-condition). Similarly, Kerth et al. (2009) reported that the self-injury of a 16-year-old autistic male with moderate-to-severe ID reduced by 52% from baseline when self-restraint (non-contingent access to a hooded sweatshirt) was available. However, existing experimental research typically employs single-case study designs to identify behavioural function, where the characteristics of the individual are largely unimportant and findings are not generalised to the wider population. Thus, this thesis aimed to further the field by employing population-based cross-sectional designs across cohorts of individuals with neurodevelopmental conditions.

Not all data support a theory of negative reinforcement, for example Smith et al. (1992) found one participant displayed higher levels of SIB than self-restraint across all conditions, and no clear function of behaviours could be ascertained. Thus, consideration of alternative or complementary operant models of behaviour is warranted. Schroeder and Luiselli (1992) proposed that self-injury and self-restraint are maintained by the same reinforcing properties (i.e., share the same functional class). This theory suggests that the function of self-restraint is not the avoidance of SIB, rather the function of self-restraint is similar to reinforcers of SIB (Pace et al., 1986). For example, self-restraint may be positively reinforced by caregiver attention (Derby et al., 1996; Figure 1.4) or negatively reinforced by task escape (Figure 1.5). Importantly, self-restraint may become the prevailing behavioural response over-and-above SIB as the same reinforcing contingencies are accessed while the aversive properties associated with SIB are avoided (Pace et al., 1986).

Figure 1. 4 Mutual reinforcement cycle of social positive reinforcement of self-injury and self-restraint

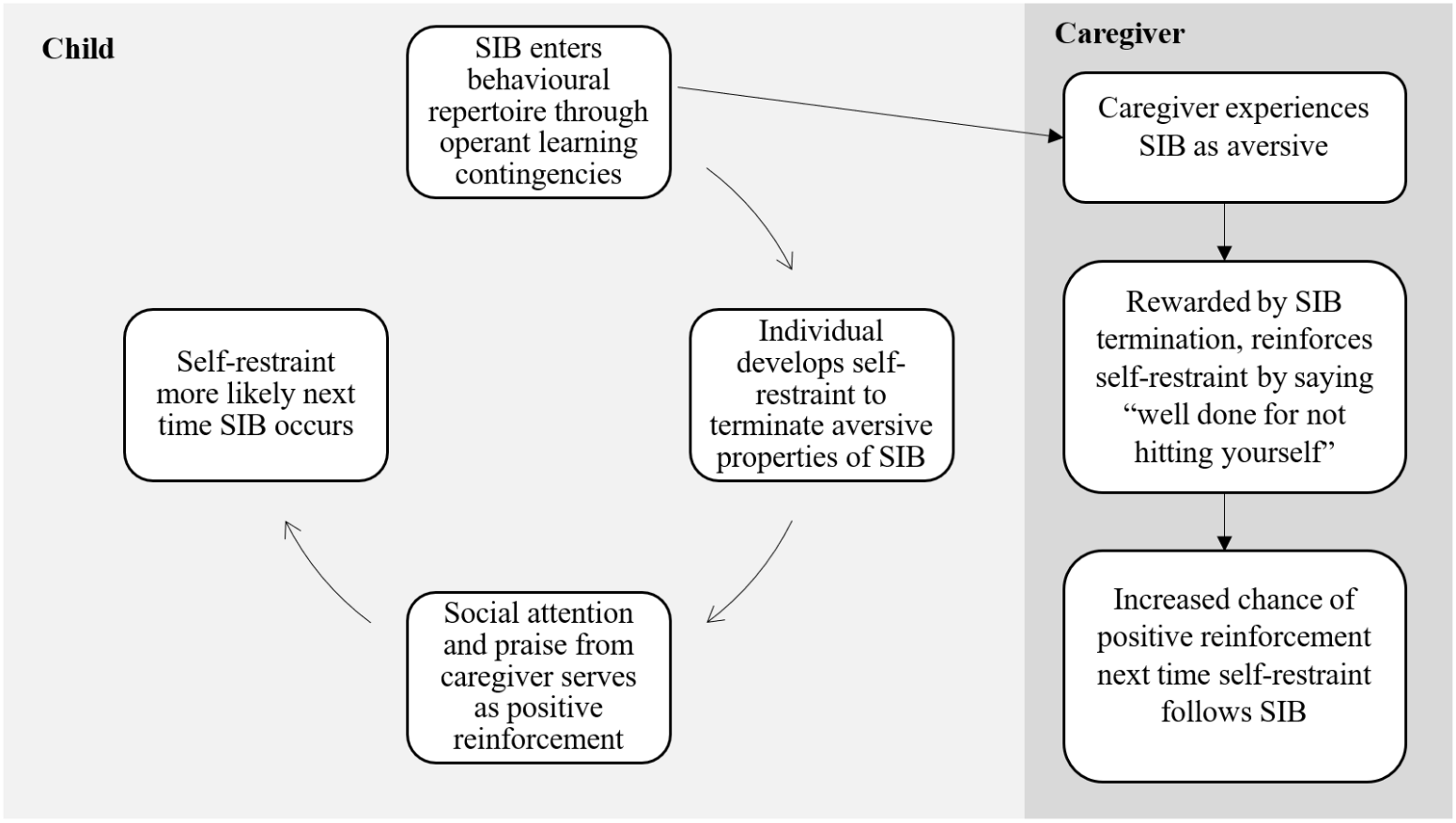
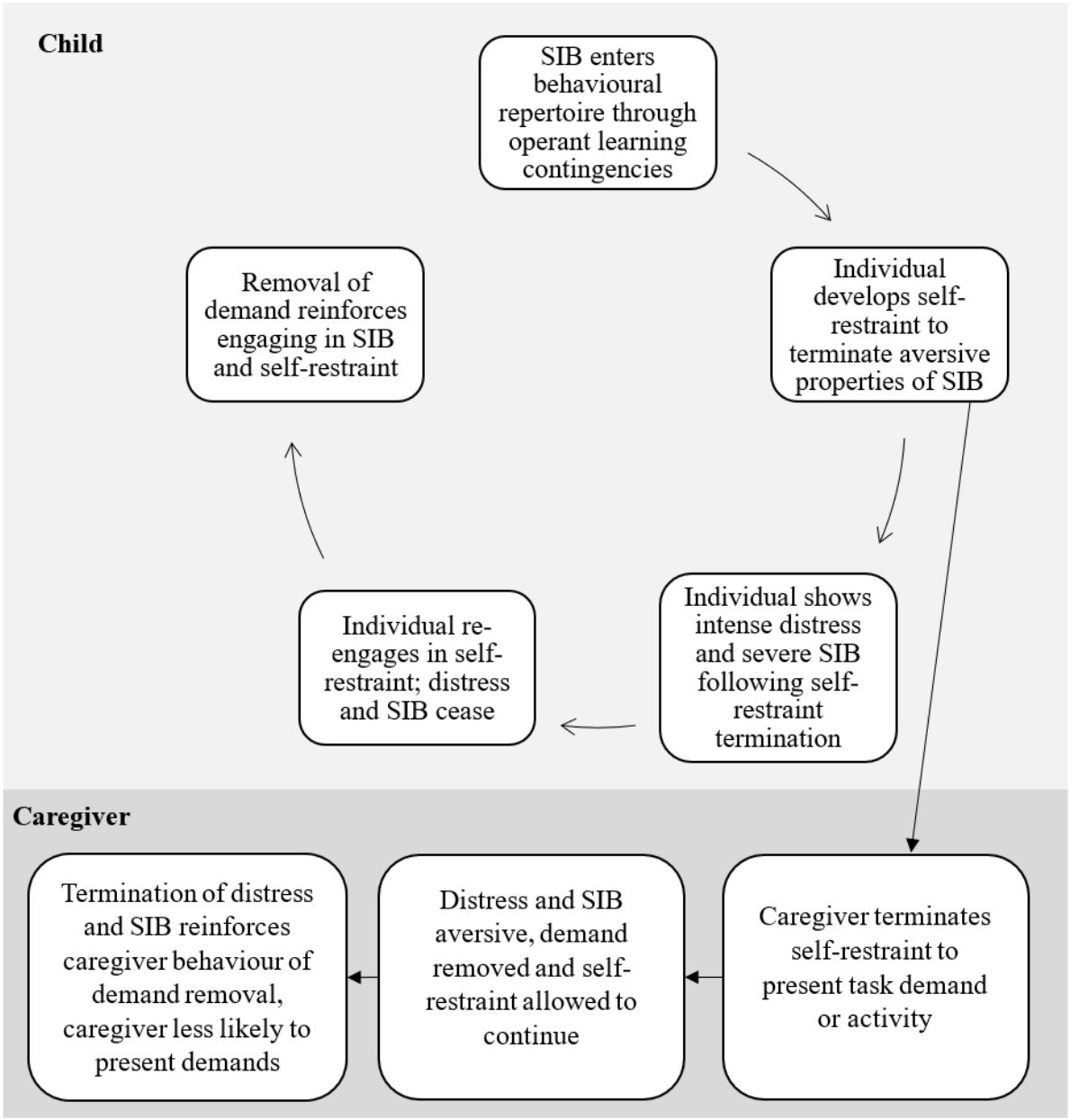


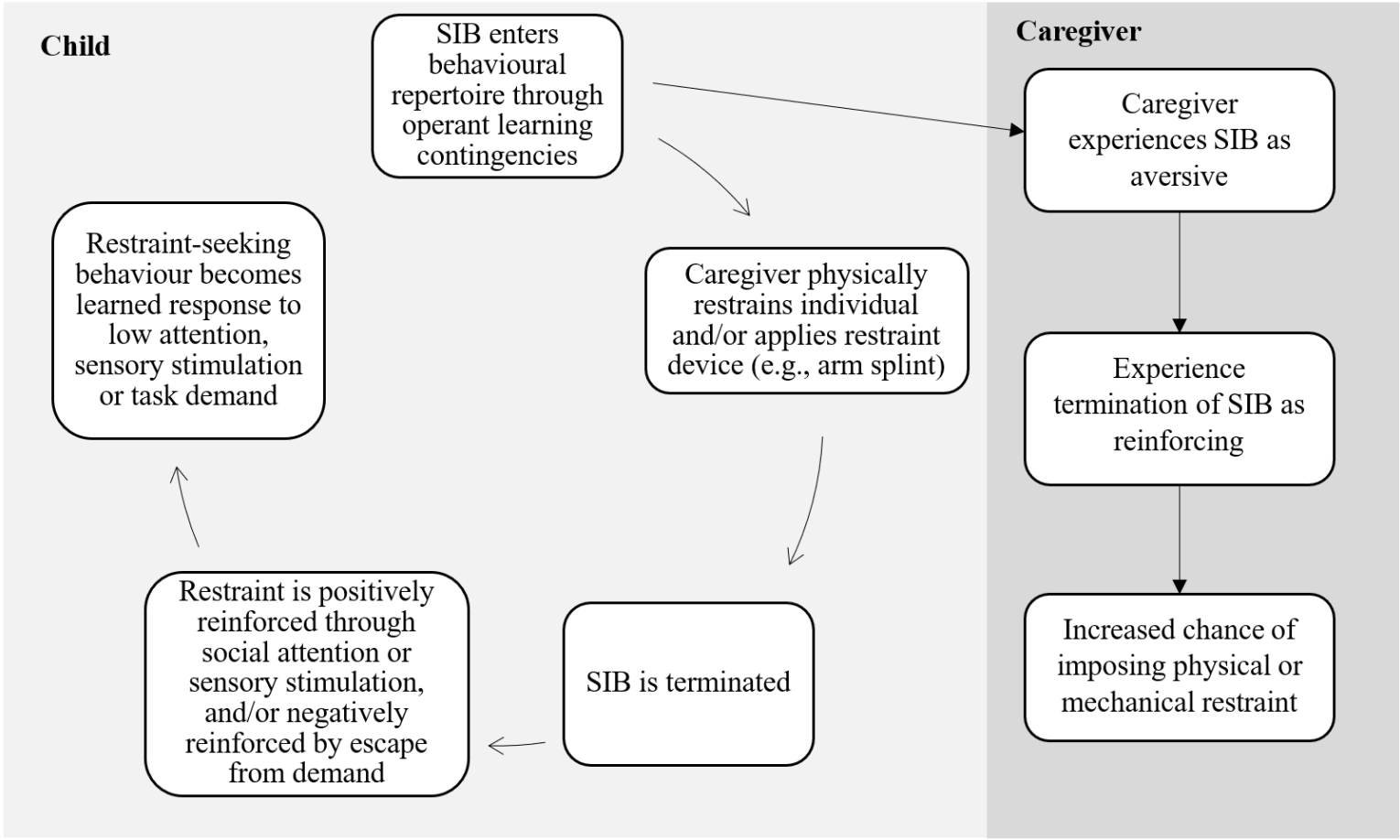
Figure 1. 5 Mutual reinforcement cycle of negative reinforcement of self-injury and self-restraint



In contrast, research has suggested that self-restraint and SIB are functionally independent behaviours, such that self-restraint does not function in relation to SIB. Early evidence from Peterson and Peterson (1968) suggested that the topography of 'wrapping oneself in a blanket' serves as discriminative reinforcement for rewards unrelated to SIB, such as warmth, sensory stimulation and comfort. Fisher and Iwata (1996) suggested it was unlikely that SIB and self-restraint would develop independent functions by chance alone, therefore it is perhaps more likely that both behaviours initially share a functional relationship and become functionally independent over time (e.g., Figure 1.6). For example, evidence of self-restraint continuing to occur after SIB has been eliminated through intervention (e.g., Rojahn et al., 1978) reveals how self-restraint may develop new functions that are unrelated to SIB. Alternatively, Favell et al. (1978, 1981) suggested that self-restraint, such as imposed physical or mechanical restraint, may initially begin as neutral stimuli introduced in response to SIB, however over time acquire reinforcing properties through positive reinforcement, such as social attention or sensory stimulation, or negative reinforcement such as escaping tasks or demands, that are unrelated to SIB.

In summary, operant models of self-restraint are highly regarded and theoretically supported. However, an overarching limitation of operant paradigms is the self-initiated restriction of movement that may risk losing access to the reinforcing consequence. In response to this theoretical conundrum, authors have speculated the role of behavioural control in driving SIB and self-restraint.

Figure 1. 6 Example of functional independence of SIB and self-restraint



1.4.7.3 Models of compulsivity and impulsivity

Early literature describing SIB and self-restraint noted the seemingly 'involuntary' nature of the behaviour for some individuals (Dizmang & Cheatham, 1970). Lovaas and Simmons (1969) described an eight-year-old male with severe ID who appeared frightened of his own SIB, who only settled once restrained. Christie et al. (1982) reported of a man with Lesch-Nyhan syndrome who would tell his hand to "be good" when restraints were briefly removed (p.298). Evidence of a 'subtype' of self-injurers who display automatically-reinforced self-injury and co-occurring self-restraint suggests self-restraint might not be constrained to environmental contingencies, turning attention towards the role of cognitive control in the relation between self-restraint and SIB (Hagopian et al., 2018). Initially, a compulsivity hypothesis of SIB was proposed to account for 'out of control' SIB (King, 1993; section 3.2), with broader deficits in inhibitory control (and other components of EF) driving behaviours (Oliver & Richards, 2015; Richards et al., 2017; section 6.2.3).

Impulsive behaviour[7] reflects the inability to stop the initiation of behaviour, leading to behaviours that appear premature, risky and occur without foresights (Robbins et al., 2012). Building upon this 'trait' is compulsivity, which reflects the inability to stop a behaviour once it is initiated (i.e., persistence or perseveration). Compulsive behaviours are typically repetitive, habitual and chronic in nature, and are driven by a lack of voluntary control over behaviour (Figee et al., 2016). Compulsive behaviours are not goal-driven, are inappropriate for the situation, and lead to negative outcomes (Robbins et al., 2012). Both compulsivity and impulsivity are associated with frontal-striatal brain circuitry, which is considered to drive

[7] A note on terminology: 'inhibitory control' refers to the cognitive process (i.e., executive function) and is used when referring to the hypothesis (e.g., hypothesis of impaired inhibitory control). 'Impulsivity' refers to the behavioural representation of a lack of inhibitory control (e.g., the putative correlate of impulsivity). Impulsivity and impulsive behaviour are synonymous. Inhibition refers to the ability to override an impulsive behaviour.

top-down control over behaviour (Hook et al., 2021), commonly co-occur, and are associated with long-term negative outcomes (Chamberlain et al., 2019). Both constructs are evidenced by motor disinhibition (Robbins et al., 2012). Drawing on the Obsessive-Compulsive Disorder (OCD) literature, King (1993) suggested self-restraint compensates for SIB that is compulsive, with some studies finding elevated rates of compulsivity in those who self-restrain (e.g., Powell et al., 1996). Understanding of the role of compulsivity in driving self-restraint across neurodevelopmental conditions is limited by a lack of empirical investigation and direct measures of behaviour (section 1.3.2). Therefore, Chapters 3 and 6 examine relations between indirect (i.e., informant-report) and direct (i.e., performance-based) markers of compulsivity with self-restraint in individuals with autism and/or ID.

Compelling reports of a preference for self-restraint (section 1.4.5.3) and evidence of anxiety "appear[ing] to stem from a fear of self-mutilation" (Wurtele et al., 1984, p.229) suggests individuals may be 'anxious' to seek a state of restraint due to 'compulsive' SIB (King, 1993). This theory stems from understanding of OCD, whereby compulsions are driven by an urge to reduce anxiety or distress (APA, 2013). Romanczyk et al. (1992) proposed SIB induces high arousal, which is lowered through self-initiated restraint. Self-restraint is thereby associated with the removal of high arousal, negatively reinforcing the behaviour that led to this 'soothing' self-restraint, which may be SIB. A 'vicious cycle' of self-injury, anxiety and self-restraint may thereby develop (section 3.2). Altogether, the role of anxiety/high arousal is implicated in a compulsive hypothesis of self-restraint and SIB, however the relation is complex, and the evidence-base is small, contradictory and theoretical in nature (section 3.2). A barrier to understanding is the challenge of measuring anxiety-related traits in individuals known to self-restrain, particularly those with severe ID, who may be unable to introspect and

self-report internal experiences. Thus, while there is a need to unpick the complex relation between compulsivity, anxiety, SIB and self-restraint in individuals with ID, this is methodologically challenging. This thesis, therefore, aimed to address this literature gap and clinical need by utilising a novel measure of behavioural anxiety designed for use with populations with severe/profound ID across the neurodevelopmental continuum (Chapter 3).

Inhibitory control is considered one of three core executive functions (EF), a set of top-down cognitive abilities that underpin the control of thought and behaviour (Diamond, 2013). Inhibition requires behavioural and emotional control to override temptations, predispositions, and habits. Inhibition is a basic component from which more complex EF skills, such as working memory and cognitive flexibility, build (Devine et al., 2019). A lack of inhibition is measured by 'impulsive' behaviour (Barkley, 1997). Links between executive dysfunction and autism are well established (e.g., Demetriou et al., 2018), while less research attention has focused on EF in individuals with ID, or individuals with co-occurring autism *and* ID, despite suggestion of inhibitory control difficulties in this group (e.g., Benallie et al., 2021; McClain et al., 2022; Spaniol & Danielsson, 2022; Trezise et al., 2014). This is largely due to methodological challenges of direct measurement of EF in these groups (section 4.2). Preliminary data have implicated deficits in inhibitory control as predicting self-restraint in autistic individuals (Richards et al., 2017), however understanding is limited by a lack of inclusion of individuals with ID (a high-risk group for self-restraint) and reliance on indirect measures of EF (section 6.2.3). Therefore, this thesis aims to extend existing work by creating developmentally-appropriate, direct measures of EF for autistic children with ID (Chapter 4) and investigate relations between EF, self-restraint and SIB (Chapter 6).

1.5 Conclusions and aims

The literature reviewed above described the phenomenon of self-restraint observed within populations who show SIB, including autistic individuals, individuals with ID and/or associated genetic syndromes. It is important to delineate the putative demographic and behavioural variables driving self-restraint due to the potentially deleterious consequences of self-restraint for adaptive and physical functioning. Understanding of self-restraint, however, is limited both by a paucity of empirical investigation, and a lack of nuance in measuring self-restraint and putative correlates, such as EF and compulsivity, in neurodevelopmental conditions. This thesis, therefore, aims to unpick the complex relation between self-restraint, self-injury and putative correlates by addressing limitations and gaps identified within the existing literature.

1.5.1 Overall thesis aims

This thesis capitalised on two large-scale studies to further understanding of self-restraint and its relation with self-injury. Across Chapters, the following aims were examined:

- Aim 1. Delineate the prevalence of self-restraint in populations known to self-injure
- Aim 2. Identify common topographies of self-restraint
- Aim 3. Describe demographic characteristics of participant groups that are associated with self-restraint
- Aim 4. Examine the relation between self-restraint and self-injury
- Aim 5. Test the compulsivity hypothesis of self-restraint and self-injury
- Aim 6. Test the inhibitory control hypothesis of self-restraint and self-injury
- Aim 7. Compare measurements of self-restraint according to different parameters

A brief overview and rational for each Chapter are provided.

1.5.2 Chapter 2

Self-restraint is observed across neurodevelopmental conditions, however prevalence estimates vary widely according to participant and study characteristics. Therefore, Chapter 2 presents a systematic synthesis of the existing cross-sectional literature, and undertakes a robust quality assessment of the literature. Prevalence data are meta-analysed to identify a pooled prevalence estimate of self-restraint, its topographies and relations with putative correlates, including SIB, compulsivity and impulsivity.

1.5.3 Chapter 3

Individuals with low adaptive functioning and greater autism characteristics are at high risk of displaying SIB, and self-restraint has been conceptualised as a compensatory behaviour in response to impulsive and/or compulsive self-injury. However, associations have not been examined in a large, heterogeneous sample of individuals with neurodevelopmental conditions using a continuous measure of self-restraint. Therefore, Chapter 3 describes the prevalence, correlates and predictors of self-restraint (measured according to topography) across the neurodevelopmental continuum (i.e., children and adults with genetic syndromes associated with autism and/or ID).

1.5.4 Chapter 4

Impaired inhibitory control is hypothesised to drive self-restraint in the context of self-injury. So far, this has only been assessed using indirect measures of EF, and there are very few available measures of EF that are suitable for autistic children with ID. Direct and indirect measures of EF arguably capture different constructs. Therefore, Chapter 4 provides an overview of a novel battery of direct play-based EF tasks used to assess impulsivity, working memory and cognitive flexibility in this population. The feasibility, validity and

developmental appropriateness of the remote tasks was assessed. Self-restraint was measured using a multi-parameter approach, and the underlying factor structure of the SRQ was assessed. Methods outlined in Chapter 4 form the basis of Chapters 5 and 6.

1.5.5 Chapter 5

Direct observations of self-restraint provide precise data around the frequency and duration (i.e., intensity) and topographies of self-restraint. Intense self-restraint impacts physical wellbeing and adaptive functioning. Thus far, self-restraint intensity has not been examined beyond informant-report or case study methods, nor using a continuous 'intensity' measure of self-restraint. This limits understanding of behavioural intensity across neurodevelopmental conditions. Therefore, Chapter 5 describes a naturalistic observation of the intensity of self-restraint topographies displayed by autistic children with co-occurring ID in the home environment. The clinical utility of observational methods was assessed through comparisons with informant-rated self-restraint.

1.5.6 Chapter 6

Self-restraint is considered to attenuate SIB, particularly when the self-injury is automatically-reinforced and driven by impulsivity and/or compulsivity. No study has yet attempted to unpick the complex relation between self-restraint, behaviours of concern and EF, despite clear implications for theoretical understanding. 'Multi-parameter' self-restraint accounts for key parameters of behaviour, providing a stringent 'functional' definition of behaviour. Therefore, Chapter 6 outlines the first study to compare multiple measurements of self-restraint with direct and indirect assessments of EF.

1.5.7 Chapter 7

This Chapter reviews key findings, presents a new integrated model of self-injury and self-restraint, and discusses methodological, theoretical and clinical contributions of this thesis to the field of self-restraint.

CHAPTER TWO

THE PREVALENCE AND CORRELATES OF SELF-RESTRAINT IN INDIVIDUALS WITH AUTISM AND/OR INTELLECTUAL DISABILITY: A SYSTEMATIC REVIEW AND META-ANALYSIS

2.1 Preface

Perhaps due to the apparent rarity of 'functional' self-restraint, empirical evidence identifying, describing and explaining self-restraint is characteristically small. However, chronic self-restraint poses risk to health and wellbeing, and the presence of self-initiated bodily restriction challenges operant understanding of self-injurious behaviour. The few existing cohort studies of self-restraint have revealed variable rates of prevalence, complicating understanding of the fundamental commonality of self-restraint within self-injurious populations. Therefore, Chapter 2 presents a systematic synthesis of the existing cross-sectional literature to establish pooled prevalence estimates of self-restraint and its topographies. To evaluate evidence for hypotheses of operant learning and/or hypotheses of impaired behavioural control, this Chapter examines the prevalence of self-restraint in the presence and absence of self-injury, and estimates the size of the relation between self-restraint and putative correlates, including self-injury, compulsivity and impulsivity.

2.2 Introduction

Self-restraint refers the self-initiated restriction of own body movements. Topographies include restricting movement using clothing, material, own body parts, objects, other people and mechanical devices (section 1.4.5.1). While the implementation of restraint by other people may not appear to be *self*-restraint, the distinction from reactive involuntary restraint is that the individual presents a desire for their movements to be restricted, possibly through verbalisations, actions, or serious distress upon restraint removal (Oliver et al., 1998). The overarching aim of this study is to synthesise the existing evidence describing self-restraint in autism and intellectual disability (ID) and identify the overall prevalence of self-restraint and its topographies.

Intentional self-restrictive practices are relatively common within the general population (Sturmey, 2020). However, self-restraint has been more commonly observed in individuals with neurodevelopmental conditions than in neurotypical peers, with high rates of self-restraint shown in autistic individuals and/or those with ID (Richards et al., 2017; Smith et al., 1992). The increased likelihood of self-restraint in these populations may be linked to the elevated prevalence of self-injurious behaviour (SIB) also observed within such groups (Adamek et al., 2011). SIB refers to pervasive behaviours that are initiated by and towards an individual, causing physical change or damage (section 1.2). Despite heterogeneity in diagnoses, autism and ID are both commonly associated with SIB.

Autism is a life-long neurodevelopmental condition characterised by differences in social communication and interaction, and restricted/repetitive behaviours. The most recent diagnostic criteria (DSM-5, American Psychiatric Association [APA], 2013; ICD-11, World

Health Organization [WHO], 2022) defined social differences as difficulties[8] with social-emotional reciprocity, such as turn-taking within conversations, non-verbal communication, such as eye-contact, and the development or understanding of relationships. Individuals show two or more restrictive/repetitive behaviours, such as repetitive, stereotypic movements, an insistence on sameness, restricted and focused interests, and sensory sensitivity (i.e., hyper- or hypo-reactivity to sensory input). Characteristics present either early in life or at the emergence of relevant social contexts, cause difficulties in day-to-day functioning, and are not better explained by other diagnoses, such as ID.

Autism commonly co-occurs with ID (section 1.3). ID is typically classified according to intellectual ability and adaptive functioning skills. The DSM-5 (APA, 2013) defines deficits in intellectual functioning as difficulties with planning and reasoning, and academic and experiential learning. Intelligent quotient (IQ) tests reveal such difficulties, and an IQ score of less than 70 (i.e., two standard deviations below the average IQ of 100) indicates ID. Alongside intellectual ability, the DSM-5 also describes difficulties with adaptive functioning. For example, an individual with ID may have difficulties with verbal or non-verbal communication, such as functional communication, social skills, such as following societal rules or customs, and daily living skills, such as dressing and feeding. Such difficulties must present within the developmental period (i.e., are not acquired later in life) and lead to additional support needs. Estimates suggest that up to 50% of autistic individuals self-injure,

[8] The term ‘difficulties’ refers to the experiences of neurodivergent individuals navigating societal systems tailored towards neurotypical individuals (Fletcher-Watson, 2022). This thesis recognises the negative connotations of using terms such as ‘difficulty’ and ‘deficit’ when referring to experiences of autistic individuals (Fletcher-Watson & Happe, 2019). However, in order to test theoretical accounts of behaviour and cognition, such terminology is necessary to make the direction of difference clear.

while prevalence rates in individuals with ID are approximately 12% (Oliver et al., 1987; Steenfeldt-Kristensen et al., 2020). Vulnerability to SIB increases when an individual has a co-occurring diagnosis of autism and ID (Matson & Rivet, 2009). Self-restraint is hypothesised to be a functional response that emerges to compensate for self-injury (Schroeder & Luiselli, 1992), suggesting that populations with autism, ID and co-occurring autism and ID represent high risk groups in which the prevalence of self-restraint and co-occurrence of self-restraint and SIB should be evaluated.

2.2.1 Prevalence of self-restraint

It is important to delineate the overall prevalence of self-restraint and identify those most at-risk because intense self-restraint has been associated with a range of deleterious consequences (section 1.4.2), including risks to physical health (Smith et al., 1992) and adaptive functioning (Scheithauer et al., 2015), with downstream effects on overall wellbeing. To consider theoretical implications derived from operant theory (section 1.4.7.2), self-restraint may be negatively reinforced by the termination of pain caused by SIB. This theory postulates an inverse association between self-restraint and SIB, where low rates of SIB are related to high rates of self-restraint. Conversely, when self-restraint becomes unavailable, SIB occurs as a "side effect" until self-restraint is re-introduced (Isley et al., 1991, p.90). Alternatively, self-restraint may maintain and increase SIB through positive reinforcement. Opportunities to self-restrain may provide internal reinforcement via sensory feedback or external reinforcement via environmental consequences, that positively reinforce the self-injury elicited in order to access self-restraint (Favell et al., 1978). Through such mechanisms of operant learning, self-restraint can acquire powerful stimulus control, and in extreme cases, self-restraint can become the dominant behavioural response (Baroff & Tate, 1968; Pace et

al., 1986; Rojahn et al., 1978) with individuals seeking a state of restraint for up to 24 hours a day (O'Reilly et al., 2003; section 1.4.5.2). Altogether, the negative consequences of SIB and contingent self-restraint can present serious threats to wellbeing and quality of life. Despite this, there is limited understanding of its prevalence in the most vulnerable groups. Therefore, the first aim of this study is to address this gap in understanding by pooling together extant data to estimate the prevalence of self-restraint.

Most studies exploring self-restraint have described prevalence estimates within populations with ID. Fovel et al. (1989) reported 2.5 to 2.9% of attendees at a school for ID showed self-restraint, while four out of 21 individuals with ID (19%) demonstrated a preference for restraint (Favell et al., 1981). Research in Cornelia de Lange syndrome (CdLS), a rare genetic syndrome associated with ID, identified a prevalence of 53.4% (Hyman et al., 2002), while Oliver et al. (2003) reported that 76.1% of a sample with ID showed self-restraint. More recent research, however, has suggested that self-restraint is not uniquely displayed by individuals with ID: Richards et al. (2017) found 40.9% of autistic children and 42.6% of autistic adults self-restrained. These data demonstrate a need to look beyond ID populations and examine prevalence of self-restraint across heterogeneous samples with and without autism. Prevalence statistics from individual studies provide useful information about expected occurrence of self-restraint. However, statistical synthesis of all existing prevalence data would provide clearer, more representative estimates of real-world occurrence. These data would lay the groundwork for future large-scale research aiming to understand the prevalence, intensity and correlates of self-restraint across neurodevelopmental conditions.

Existing prevalence data suggest self-restraint occurs in 2.5 to 76.1% of vulnerable groups.

This wide variation in estimates likely results from three main methodological sources. First, self-restraint is a challenge to conceptualise, with researchers employing differing definitions (section 1.4.4) and parameters (section 1.4.5). For example, Fovel et al. (1989) defined self-restraint according to body restriction, while Forman (2003) adopted a parameter-based approach. Second, the measurement format may under- or over-estimate self-restraint. Informant-report measures, such as the Self-Restraint Questionnaire (SRQ; Oliver et al., 2003) rely on caregiver interpretation of behavioural presentations over self-defined units of time, whereas observational studies define a timeframe within which self-restraint may occur (e.g., within 16.5 hours; Forman et al., 2002). Finally, differences in study (e.g., sample size) and participant characteristics (e.g., age, gender, diagnosis) can impact estimates. For example, Rojahn (1986) found a prevalence rate of 12% across 25,000 non-institutionalised individuals with ID, while Powell et al. (1996) studied a sample of 99 institutionalised adults with severe and profound ID and identified that 46% showed self-restraint. Overall, the prevalence of self-restraint remains contested due to these limiting factors. Therefore, the current study also aimed to examine whether participant and study characteristics influenced the pooled prevalence estimate of self-restraint.

2.2.2 Prevalence of self-restraint topographies

Existing studies present mixed findings about common topographies of self-restraint. Richards et al. (2017) and Hyman et al. (2002) both identified the most prevalent form of self-restraint to be ‘holding onto others and holding objects’ (24% and 43% of respective samples), while Powell et al. (1996) found participants were most likely to ‘hold or squeeze objects’ (50%). Oliver et al. (2003) reported use of materials to be common behaviours, such as ‘wrapping hands into material’ and ‘pulling sleeves over hands’ (both 57%), however the

most common topography was 'sitting on hands' (64%). Across these samples, 'choosing mechanical restraint' was consistently the least common topography (0%, Hyman et al., 2002; 8%, Powell et al., 1996; 0%, Richards et al., 2017). However, Oliver et al. (2003) indicated that participants in their sample had a high preference for imposed physical restraint (50% 'requested hands to be held' and 43% 'requested arms to be held'). Individual cohort studies are useful for describing self-restraint. However, estimating topographical prevalence over multiple samples provides more representative data regarding patterns of behaviour. Research has yet to synthesise these data, therefore the second aim of this study was to identify the prevalence of different topographies of self-restraint.

2.2.3 Correlates of self-restraint

The functions of self-restraint are widely debated within the literature. Research testing an account of negative reinforcement, whereby self-restraint functions to control aversive self-injury, has identified an association between self-restraint and SIB (Hyman et al., 2002), such that the presence of SIB predicts the presence of self-restraint (Richards et al., 2017). This research builds on earlier experimental evidence showing a significant decrease in self-restraint when SIB occurs (Forman et al., 2002; Kerth et al., 2009; Rojahn et al., 1978; Smith et al., 1992) and a decrease in self-restraint following SIB intervention (Silverman et al., 1984). Prevalence data also point to an association, with estimates of individuals showing both self-restraint and SIB ranging from 7 to 92% (Iwata et al., 1994; Oliver et al., 2003). Wide variation in these estimates can be attributed to distinctions in how self-restraint is conceptualised and measured as well as participant and study characteristics, and make it difficult to extrapolate a true prevalence estimate of concomitance. Furthermore, alternative theories suggest that SIB and self-restraint function to elicit similar (Pace et al., 1986), or

distinct environmental or sensory rewards (Peterson & Peterson, 1968). Such ideas challenge existing theoretical accounts by raising the possibility that, for some individuals, self-restraint is unrelated to SIB. Supporting data has reported that self-restraint occurs in the absence of self-injury within a group of autistic children (27%), autistic adults (28%) and individuals with ID (13%; Hyman et al., 2002; Richards et al., 2017). These contradictory data suggest a need to identify robust individual pooled estimates for the prevalence of self-restraint in both the presence and absence of self-injury.

Cohort studies have repeatedly demonstrated a strong statistical correlation between the presence of SIB and self-restraint (e.g., Hyman et al., 2002). This association appears unequivocal, with research suggesting that self-restraint co-occurs more frequently with SIB than other behaviours of concern including aggression and property destruction (Fisher & Iwata, 1996; Isley et al., 1991). Self-restraint and self-injury consistently share a positive linear relation. However, the size of association varies from small, r=.01 (Rojahn, 1986), to large, r=.47 (Oliver et al., 2003). To further quantify this relation, the final aim of this study was to estimate the overall strength of association between self-injury and self-restraint.

Identification of other behavioural correlates of self-restraint may help to establish the mechanisms underpinning behaviour. For example, qualitative data suggest that among autistic adults without ID, self-restraint can be employed to prevent negative restrictive/repetitive behaviours (Collis et al., 2022). Additionally, King (1993) theorised that self-restraint was related to compulsivity, suggesting that involuntary self-injury produced compensatory self-restraint behaviours (section 3.2), while Richards et al. (2017) reported that impulsivity/overactivity predicted self-restraint two-fold. Finally, Rojahn (1986) suggested

self-restraint was stereotypic in nature, with stereotypies described as repetitive movements unrelated to achieving goals. To evaluate these accounts, this study also aimed to identify overall effect sizes correlating self-restraint with other behaviours reported to occur in autistic individuals and/or those with ID.

In summary, this study had three aims: (1) calculate the pooled prevalence of self-restraint among people with autism and/or ID, and examine effects of study and participant characteristics on these pooled estimates, (2) calculate pooled prevalence estimates for each topography of self-restraint, and (3) examine the relation between self-restraint and putative correlates, including SIB, restrictive/repetitive behaviours, compulsivity, impulsivity/overactivity and stereotypy.

2.3 Methods

2.3.1 Search strategy

This study was preregistered on PROSPERO (https://tinyurl.com/self-restraint-meta) and was performed and reported in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Page et al., 2021). Systematic searching of electronic bibliographic databases aimed to identify both published and unpublished data (i.e., theses, preprints and conference abstracts). Search terms were generated through inspection of previous meta-analyses of self-injury (McClintock et al., 2003; Steenfeldt-Kristensen et al., 2020) and examination of relevant literature. All databases were systematically searched from their earliest available record to the date of the search (February 08, 2021). The full search strategy, including exact search terms, truncations and Boolean operators, is provided in Appendix 2.1. Beyond exclusion criteria outlined in Table 2.1, no further search restrictions were set on participant characteristics, such as sample age or gender.

The self-restraint literature is characterised by inconsistency in terminology and definitions (Forman, 2003). Three strategies were applied to ensure inclusion of all relevant papers through additional backward citation searches. First, reference lists were hand-searched to identify relevant papers not retrieved by database searching. References of both excluded papers, including reviews and qualitative studies, and eligible papers were searched. Any titles deemed relevant were subsequently screened at full text. Second, published titles of authors known to study self-restraint and SIB were searched within PubMed on April 16, 2021. Third, abstracts from major academic ID conferences Seattle Club (available digitally from 2001 to 2018) and Gatlinburg (2021 available only) were hand-searched via respective website browsers on April 20, 2021.

Table 2. 1 Eligibility criteria for screening records

	Inclusion criteria	Exclusion criteria
1	Study reported on self-restraint	Study did not report on self-restraint
2	Participants had an ID, and/or diagnosis of autism, and/or an identified genetic syndrome associated with ID and/or autism (e.g., Fragile X, Tuberous Sclerosis Complex)	Participants were not autistic, did not have an ID or did not have an associated genetic syndrome
3	Study yielded quantitative empirical data	Study reported qualitative data only
4	Paper reported in English, or an English translation was available	No English language translation was available
5	Cross-sectional or longitudinal study design	Case study or case series
6	Peer reviewed published articles or unpublished empirical articles, theses or dissertations	Reviews, opinion pieces, non-empirical studies
7[a]	Prevalence data for self-restraint were reported or obtained	Study reported on self-restraint, but prevalence data were unreported, uninterpretable or unavailable

Note. ID = intellectual disability.
[a] Additional criterion imposed at full text screening only.

2.3.2 Selection strategy

Once identified, papers were screened for eligibility (Table 2.1) using a standardised screening tool. First, titles were reviewed and ineligible papers excluded. Second, abstract and method sections of papers deemed relevant were reviewed for inclusion. If during these first two steps eligibility could not be deduced, the full text was examined. The final step of screening involved assessment of full texts of papers not yet excluded.

2.3.3 Data collection

Once eligible papers were identified, data were extracted. Extracted data included participant and study characteristics, measure and prevalence of self-restraint and its topographies, prevalence of self-restraint in the presence and absence of self-injury, effect sizes of the association between SIB and self-restraint, and effect sizes between self-restraint and other correlates. Papers were also screened to identify whether participants were known to self-injure (i.e., if the presence of SIB was included within the study's inclusion criteria). Attempts were made to contact authors to obtain further relevant data via email, social media, and in-person conversations. Further data were obtained for six samples (Appendix 2.2). In most cases, additional data sources provided information about the sample reported in the publication, such as the prevalence of self-restraint, the frequency of self-restraint topographies, and participant demographic information. However, in the case of Oliver et al. (2003), the additional data source (the author's doctoral thesis; Oliver, 1991) employed different inclusion criteria, and yielded a larger group size than the corresponding journal article. Use of data extracted from additional sources (i.e., describing some demographic information, and analysing the self-injury and self-restraint correlation statistic) is noted.

The first author undertook data screening and extraction. To evaluate inter-rater reliability, a second rater evaluated 25% (k[9]=73) of the 292 journal articles screened at full text. Papers included in the inter-rater reliability analysis were identified by assigning each reference with a numerical value and generating 73 numbers at random. Inter-rater reliability was assessed

[9] *k* refers to the number of samples, κ refers to Cohen's kappa.

using Cohen's kappa (κ). Initially there was moderate agreement[10] between reviewers, κ=.65. Following further discussion and clarification of eligibility criteria, disagreements were resolved resulting in perfect agreement, κ=1.00. Both raters then extracted prevalence data, including the prevalence of self-restraint and its topographies, for 100% (*k*=15) of the final sample, κ=1.00.

2.3.4 Quality review

A quality assessment framework (QAF) was created to capture information about potential threats to validity (Table 2.2). Based on previous meta-analyses (Mingins et al., 2020; Richards et al., 2015; Surtees et al., 2018), the QAF was designed to rate each individual study included within the final sample for bias. Studies were scored across four domains as required: sample identification, autism measurement, ID measurement and self-restraint measurement. Not all domains were applicable to each paper. For example, a study that recruited individuals with ID without co-occurring autism did not require an autism measure rating. Thus, mean quality ratings were calculated by summing individual scores and dividing by the maximum *possible* score (i.e., nine or 12). Across samples, the maximum possible mean rating was 1, and scores ranged from .22 to .67.

Two coders independently rated all (100%, *k*=15) of the included papers to estimate reliability of the QAF. Agreement was assessed using intraclass correlation coefficients (ICC). Results indicated good reliability, ICC=.88, $p<.001$[11]. Disagreements in ratings were resolved during

[10] Cohen's kappa interpreted according to McHugh (2012): .00-.20 = no agreement, .21-.39 = minimal, .40-.59 = weak, .60-.79 = moderate, .80-.90 = strong, >.90 = perfect agreement.

[11] Interpretation is according to interpretation outlined by Koo & Li (2016), where a value of less than .50 is considered poor, .50 to .75 is moderate, .75 to .90 is good, and greater than .90 is excellent reliability.

consensus meetings. Appendix 2.3 displays amendments to the QAF resulting from meetings, and Appendix 2.4 displays justification for quality ratings.

Table 2. 2 *Quality assessment framework*

Domain		**Quality rating**			
		Poor (0)	**Fair (1)**	**Good (2)**	**Excellent (3)**
A	Sample identification of an autistic and/or sample with ID	Unspecified	Single restricted or non-random sample (e.g., a specialist clinic or previous research study). Single regional sample (e.g., a regional parent support group).	Multiple (i.e., two or more) restricted or non-random samples (e.g., multi-region specialist clinics, multiple schools). National non-random sampling (e.g., national parent support group).	Random or total population sample (e.g., national/clinical/birth registry).
B	Measurement of autism and reliability/validity of measurement of autism	Unspecified	Recruitment from specialist school, service or similar (not confirmed in this study). Score above threshold on screening instrument (e.g., 15 on SCQ [a]). Clinician judgement against specified diagnostic criteria (e.g., DSM-V or ICD-10). Diagnosis of autism obtained from official records.	One diagnostic measure of autism (e.g., ADOS, ADI-R). Previous diagnosis of autism by multidisciplinary team using multiple assessment methods unconfirmed but specified in the present study.	Consensus from multiple assessments, including at least one diagnostic instrument, confirmed in this study (not in previous study or as part of assessment through clinic).

Note. ID = intellectual disability; SCQ = Social Communication Questionnaire; DSM-V = Diagnostic and Statistical Manual of Mental Disorders, fifth edition; ICD-10 = International Classification of Disease, tenth edition; ADOS = Autism Diagnostic Observation Schedule; ADI-R = The Autism Diagnostic Interview-Revised.

[a] According to recommended cut off (see section 3.3.4.1.1).

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Table 2.2 cont.

Domain		**Quality rating**			
		Poor (0)	**Fair (1)**	**Good (2)**	**Excellent (3)**
C	Measurement of ID and reliability/validity of ID measure	Unspecified	Recruited from specialist ID school, service or similar (not confirmed in this study). Diagnosis of ID obtained from official records. Diagnosis of ID included in study eligibility criteria. Syndrome group known to be associated with ID (e.g., CdLS).	Self/parent/teacher informant report using a well-validated measure of adaptive functioning (e.g., The Wessex Questionnaire).	Formal IQ test administered by researcher or clinician (e.g., Wechsler Intelligence Scale for Children, Mullen Scales of Early Learning, Stanford-Binet) *and/or* formal measure of adaptive functioning administered by researcher or clinician (e.g., The Vineland Adaptive Behavior Scales).
D	Measure of self-restraint	Unspecified (e.g., "a survey").	Clinical case records. Observational measure with unreported interrater reliability.	Self/parent/teacher report using a validated measure (e.g., SRC, SRQ) *or* observational measure reporting at least substantial ($\kappa \geq .70$) inter-rater reliability, conducted in this study.	Self/parent/teacher report using a validated measure (e.g., SRC, SRQ) *and* observational measure reporting at least substantial ($\kappa \geq .70$) inter-rater reliability, conducted in this study.

Note. CdLS = Cornelia de Lange Syndrome; IQ = Intelligence Quotient; κ = Cohen's kappa; SRC = Self-Restraint Checklist; SRQ = Self-Restraint Questionnaire.

2.3.5 Data synthesis

Raw summary statistics were extracted and entered into SPSS (v.27.0.1.0). Where possible, relevant but unreported data were obtained through contacting study authors, and/or conducting backwards calculations. For example, to meta-analyse correlates of self-restraint, all reported effect sizes were converted to Pearson's *r* (Borenstein et al., 2021). Estimates were then converted to Fisher's *Zr* to minimise the sampling distribution of Pearson's *r* and identify a less skewed and biased statistic (Corey et al., 1998; Lenhard & Lenhard, 2014). Where authors had not conducted analyses of the association but had reported prevalence figures for both self-restraint and the associated behaviour (e.g., self-injury), data were re-analysed to generate Fisher's *Zr*.

Meta-analyses were conducted in R (v.1.3.1093; R Core Team, 2021). Weighted prevalence values were generated using the inverse-variance method. Random effects models were adopted due to observation of high heterogeneity according to the Higgins I^2 statistic (i.e., I^2 figures over 30%; Higgins et al., 2003). Since the distribution of prevalence estimates were non-normal, a restricted maximum likelihood estimator was used to calculate between-study variance. This estimator is considered more robust than traditional DerSimonian-Laird estimates when used with non-normal distributions due to its restriction of likelihood estimates to control for underestimation (Cheung, 2013). Sensitivity analyses were then performed to identify studies bearing disproportionately high influence on the overall meta-analytic effect. These included visual inspection of Baujat scatter charts, and 'leave-one-out' omitting analyses whereby the weighted prevalence was re-calculated following removal of each study in turn (Baujat et al., 2002). An additional quality rated analysis was also conducted, adjusting study weights according to overall quality ratings according to the QAF.

Individual meta-analyses were conducted to identify the prevalence of self-restraint across all samples, the prevalence of self-restraint in individuals known to self-injure and the prevalence of self-restraint in individuals known to *not* self-injure. The prevalence of 31 topographies of self-restraint was extracted and meta-analysed.

Meta-regression examined the influence of participant characteristics on prevalence rates, including the influence of age, gender, presence of ID and presence of autism. Characteristics were examined where eight or more studies had reported data, including age (k=9) and presence of autism (k=8). While falling below the commonly-cited rule of ten studies per analysis (Higgins et al., 2011), such analyses were conducted in order to retain data and identify characteristics that may influence prevalence estimates. Results evaluating the influence of age and presence of autism on the prevalence of self-restraint should therefore be considered with caution. Due to insufficient data (i.e., data from <8 studies), the effect of ID severity and presence of health conditions on the prevalence of self-restraint could not be evaluated. Subgroup analyses were conducted to identify the influence of study characteristics (i.e., how self-restraint was measured) on the prevalence rates.

Meta-analyses of effect sizes of putative correlates were conducted when two or more papers reported data. An a priori power analysis using G*Power (v.3.1.9.7; Faul et al., 2009) identified a pooled sample size of 614 individuals was required to detect a small effect size, r=.10, with an alpha of .05 and power of .80. Interpretation of effect sizes follows those proposed by Gignac and Szodorai (2016), where r=.10, .20 and .30 represent respective small, moderate and large effects. These guidelines are suggested to be more realistic than those proposed by Cohen (1988), where r=.10, .30 and .50 represent respective small, moderate and

large effects, as a meta-analysis revealed only 3% of 708 correlations met threshold for a 'large' effect (Gignac & Szodorai, 2016).

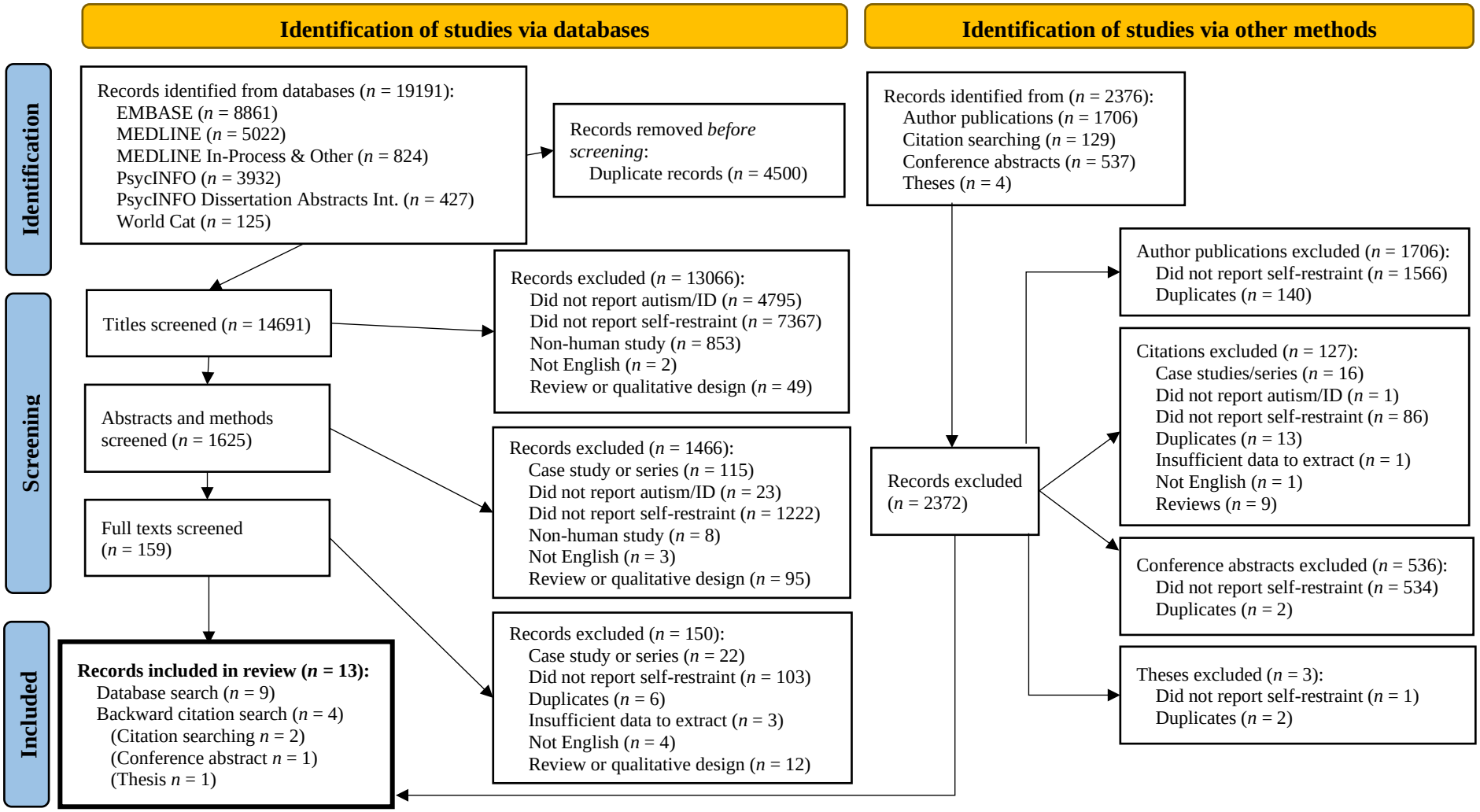
2.4 Results

2.4.1 Study selection

Initial database searches yielded a response of 19,191 papers (Figure 2.1). A further 2376 studies were identified through other methods. After duplicates were removed (*k*=4500) the remaining 17,067 papers were reviewed at different stages: 14,691 database papers were reviewed at title stage only, 3868 papers underwent title and abstract review (*k*=1625 database, *k*=1706 author publications, *k*=537 conference abstracts), and 292 papers were reviewed at full text (*k*=159 database, *k* = 129 citation searching, *k*=4 dissertations). In total, 17,054 papers were excluded (*k*=13,066 at title review, *k*=3708 at title and abstract review, *k*=280 at full text). This left a total of 13 eligible papers, of which two reported more than one sample. Thus, data were extracted from a total of 15 samples.

During full text screening, four studies (Breau et al., 2003; Nagy et al., 2019; Schroeder et al., 1982; Shapira et al., 2004) that met initial criteria for inclusion were excluded due to insufficient or uninterpretable prevalence data (Table 2.1). Authors were contacted in attempt to obtain these data prior to exclusion.

Figure 2. 1 PRISMA flow chart for all stages of study screening



2.4.2 Study characteristics

In total, 2309 participants, *M*=154, *Mdn*=67, *range*=18–686, were identified across 13 studies conducted between 1981 and 2020. Records included published journal articles (*k*=14) and two unpublished doctoral theses (Forman, 2003; Oliver, 1991). Studies were conducted in the United Kingdom (*k*=7), United States (*k*=7) and Germany (*k*=1). Further study and participant characteristics alongside outcome prevalence data are displayed in Table 2.3. A visual matrix of quality ratings is provided, colour-coded to represent QAF domain scores: red items reflect scores of zero (poor), orange scores of one (fair), yellow scores of two (good), green scores of three (excellent), and white items represent non-applicable domains.

Participant ages ranged from one to 72 years. Gender information was provided for 1541 participants (66.78% of the total sample), of whom 62.36% were male (*n*=961). Table 2.4 displays classifications of autism and ID used across the samples. Six samples included individuals with ID only (*n*=852 with severity of ID known), three samples included individuals who were autistic only (or had a similar diagnosis; *n*=491) and five samples included participants with autism and/or ID (*n*=197 participants). Furthermore, one study included a group with CdLS (*n*=88). A total of 11 genetic syndromes were identified within 124 individuals across 6 samples (Appendix 2.5).

The most common assessment of self-restraint was the Self-Restraint Checklist (SRC; Powell et al., 1996) survey, used across five samples. A total of four studies employed observational methods. The remaining survey instruments utilised are outlined in Table 2.3.

Table 2. 3 Quality criteria, study and sample characteristic and outcome data for studies reporting prevalence of self-restraint

Sample	**Quality rating**					**Study and sample characteristics**							**Outcome data**		
	Domain				Mean overall rating	*N*	SIB known (Y/N)	Location	Mean age (SD) *range*	*n* male (%)	*n* autistic (%)	*n* ID (%)	Measure of self-restraint	Prev. SR *n* (%)	Prev. SR + SIB *n* (%) [a]	Prev. SR without SIB *n* (%) [a]
	A	B	C	D												
Bruhl et al., 1982					.42	18	Y	USA	15.39 (5.73) *7 – 31*	11 (61.10)	4 (22.20)	18 (100.00)	Observation	10 (55.60)	10 (100.00)	0 (0.00)
Favell et al., 1981					.44	21	Y	USA	24.60 (8.39) *15 – 30*	2 (66.10)	N/A	3 (14.29)	Friedin Rating Scale	4 (19.05)	4 (100.00)	0 (0.00)
Forman, 2003					.67	63	Y	UK	30.90 (12.60) *6 – 64*	40 (63.50)	53 (84.13)	63 (100.00)	SRQ + SRQ2	41 (65.00)	41 (100.00)	0 (0.00)
Fovel et al., 1989					.22	789	N	USA	NR NR *24 – 72*[b]	16[b] (47.06)	N/A	34[b] (5.00)	Unspecified survey	34 (4.96)	28 (82.35)	6 (17.65)

Note. SIB = self-injurious behaviour; SR = self-restraint; Prev. = prevalence; N/A = not applicable; NR = not reported; SRQ = Self-Restraint Questionnaire; SRQ2 = Self-Restraint Questionnaire 2.

[a] Percentages calculated as a proportion of the total number of individuals who self-restrained (data were not all reported directly and so were calculated based on other information, e.g., number of individuals who self-restrained in the absence of SIB = the difference between total population who self-restrained and those who self-restrained and self-injured). [b] Demographic data were only reported for those who self-restrain, *n*=34.

Table 2.3 cont.

Sample	Quality rating				Study and sample characteristics						Outcome data		
	Domain A B C D	Mean overall rating	*N*	SIB known (Y/N)	Location	Mean age (SD) *range*	*n* male (%)	*n* autistic (%)	*n* ID (%)	Measure of self-restraint	Prev. SR *n* (%)	Prev. SR + SIB *n* (%)	Prev. SR without SIB *n* (%)
Hagopian et al., 2015		.50	52	Y	USA	NR NR *3 – 21*	37 (71.15)	38 (73.77)	45 (86.54)	Observation	8 (15.38)	8 (100.00)	0 (0.00)
Hyman et al., 2002		.67	88	N	UK	12.89 (8.02) *1 – 38*	42 (47.72)	N/A	N/A	SRC	47 (53.41)	36 (76.60)	11 (23.40)
Iwata et al., 1994		.56	152	Y	USA	NR NR *'1 - 10 to 50+'*	87 (57.24)	N/A	152 (100.00)	Observation	11 (7.24)	11 (100.00)	0 (0.00)
Laverty et al., 2020		.58	67	N	UK	24.58[c] (8.75)[c] *14 - 58*	54 (80.60)	67 (100.00)	N/A	SRC	29[c] (43.28)	10[c] (34.48)	19[c] (65.52)
Oliver et al., 2003 Devices		.42	41	Y	UK	21.70 (9.06) *'0 - <5 to 45+'*[d]	22 (53.66)	16[d] (29.63)	41 (100.00)	SRQ	24 (58.50)	24 (100.00)	0 (0.00)

Note. SIB = self-injurious behaviour; SR = self-restraint; Prev. = prevalence; NR = not reported; SRC = Self-Restraint Checklist; N/A = not applicable; SRQ = Self-Restraint Questionnaire.

[c] Data extracted from additional data source (see Appendix 2.2). [d] Data retrieved from additional data source with a larger total *N* of 54.

Table 2.3 cont.

Sample	**Quality rating**					**Study and sample characteristics**					**Outcome data**		
	Domain A B C D	Mean overall rating	*N*	SIB known (Y/N)	Location	Mean age (SD) *range*	*n* male (%)	*n* autistic (%)	*n* ID (%)	Measure of self-restraint	Prev. SR *n* (%)	Prev. SR + SIB *n* (%)	Prev. SR without SIB *n* (%)
Oliver et al., 2003 No devices		.44	47	Y	UK	18.10 (5.40) *NR*	22 (46.81)	N/A	47 (100.00)	SRQ	43 (91.50)	43 (100.00)	0 (0.00)
Powell et al., 1996		.44	99	Y	USA	NR	NR	N/A	99 (100.00)	SRC	46 (46.00)	46 (100.00)	0 (0.00)
Richards et al., 2017 Adults		.67	216	N	UK	34.10[c] (11.06)[c] *18 – 61*[c]	152 (70.37)	216 (100.00)	N/A	SRC	92 (42.60)	61 (66.30)	31 (33.70)
Richards et al., 2017 Children		.67	208	N	UK	13.72[c] (2.45)[c] *6 – 17*[c]	181 (87.02)	208 (100.00)	N/A	SRC	85 (40.90)	54 (63.53)	30 (35.29)
Rojahn, 1986		.56	528	Y	Germany	NR NR *3 – 62*	278 (52.95)	N/A	519 (98.30)	Unspecified survey	57 (10.80)	52[e] (86.67)	NR
Rooker et al., 2020		.25	23	Y	USA	NR NR *'5 - 12 to 18+'*	17 (73.91)	20 (87.00)	22 (95.65)	Observation	8 (34.78)	8 (100.00)	0 (0.00)

Note. SIB = self-injurious behaviour; SR = self-restraint; Prev. = prevalence; N/A = not applicable; SRQ = Self-Restraint Questionnaire; NR = not reported; SRC = Self-Restraint Checklist.

[c] Data extracted from additional data source (see Appendix 2.2). [e] Self-restraint and self-injury prevalence reported for subsample (*n*=431).

Table 2. 4 Sample classification of autism and/or intellectual disability

Sample	Classification of autism	Classification of ID
Bruhl et al. (1982)	"Autism-schizophrenia"	"Mental reta*dation"
Favell et al (1981)	-	"Reta*dation"
Forman (2003)	"Possible autism"	"Learning disability"
Fovel et al. (1989)	-	"Mental reta*dation"
Hagopian et al. (2017)	"ASD"	"Intellectual disability"
Hyman et al. (2002)	-	-
Iwata et al. (1994)	-	"Developmental disability", "Reta*dation"
Laverty et al. (2020)	"Autism"	-
Oliver et al. (2003) Devices	"Atypical autism" (*n* = 12), "Childhood disintegrative disorder" (*n* = 4)	"Mental reta*dation"
Oliver et al. (2003) No devices	-	"Mental reta*dation"
Powell et al. (1996)	-	"Mental reta*dation"
Richards et al. (2017) Adults	"Autism Spectrum Disorder"	-
Richards et al. (2017) Children	"Autism Spectrum Disorder"	-
Rojahn (1986)	-	"Mental reta*dation"
Rooker et al. (2020)	"Autism Spectrum Disorder"	"Intellectual disability"

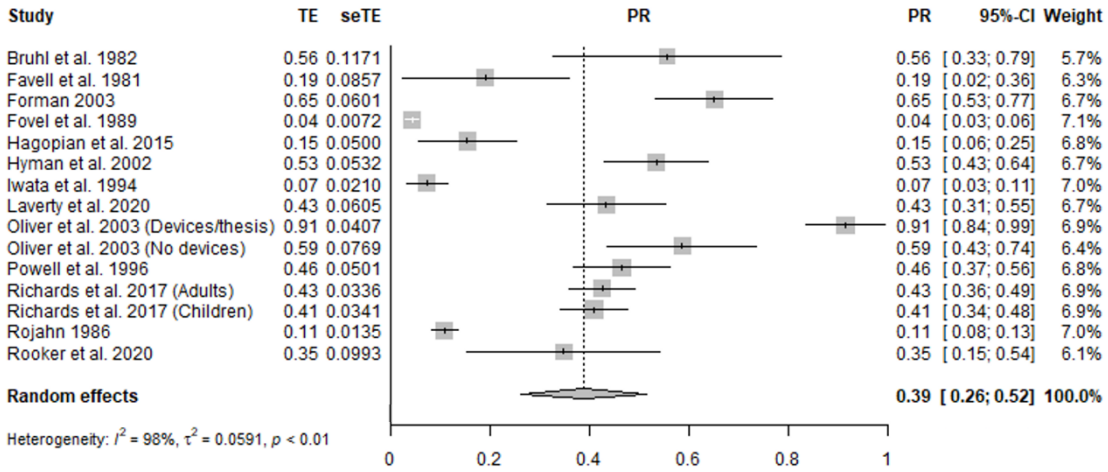
Note. Dash (-) indicates condition was not recruited or reported in study. Derogatory words are censored.

2.4.3 Prevalence of self-restraint

The inverse-variance weighted pooled prevalence estimate indicated that self-restraint occurred in 39%, z=6.02, p<.0001, 95%CI [26.25, 51.59], of participants with autism and/or ID (Figure 2.2). Heterogeneity between the prevalence rates was high, $tau^2 = 0.059$, Higgin's $I^2 = 99\%$, Q(14)=924.42, p<.0001, suggesting high variability between studies. When effects were weighted by quality ratings, the pooled prevalence estimate remained largely unchanged, 40%, z=5.99, p<.0001, 95%CI [27.05, 53.35] (Appendix 2.6). Analyses exploring study

heterogeneity and influence on the pooled prevalence estimates were conducted (Appendix 2.7). Results indicated no studies were bearing disproportionate influence, and thus no studies were removed from the prevalence analyses.

Figure 2. 2 Random effects forest plot for the overall pooled prevalence estimate for self-restraint



Note. Diamond represents mean effect, horizontal lines represent 95% confidence intervals, and the dotted vertical line represents the mean effect size.

2.4.3.1 Influence of participant characteristics on prevalence rates

The data allowed for exploration of the influence of several participant characteristics on the prevalence rates of self-restraint, including age, gender and presence of ID and autism (Table 2.5). Mean ages were extracted from nine (60%) studies, and the percentage of male participants was extracted from 14 (93%) studies. The percentage of participants reported to have an ID was extracted from 11 (73%) studies, and the percentage of participants known to be autistic was extracted from eight (53%) studies.

Table 2. 5 Influence of participant characteristics on self-restraint prevalence rates

Covariate	**Estimate**	**S.E.**	**Z**	***p***	**Confidence intervals**		***k***
					Lower 95%	**Upper 95%**	
Age (mean age)	.562	.232	2.424	**.015***	0.108	1.016	9
Gender (% male)	.421	.372	1.134	.257	-0.307	1.150	14
Presence of ID (%)	.052	.209	0.246	.805	-0.359	0.462	11
Presence of autism (%)	.809	.206	3.937	**<.001***	0.406	1.212	8

Note. S.E. = standard error. *k* = number of samples.
*Significant p-values in bold (p<.05).

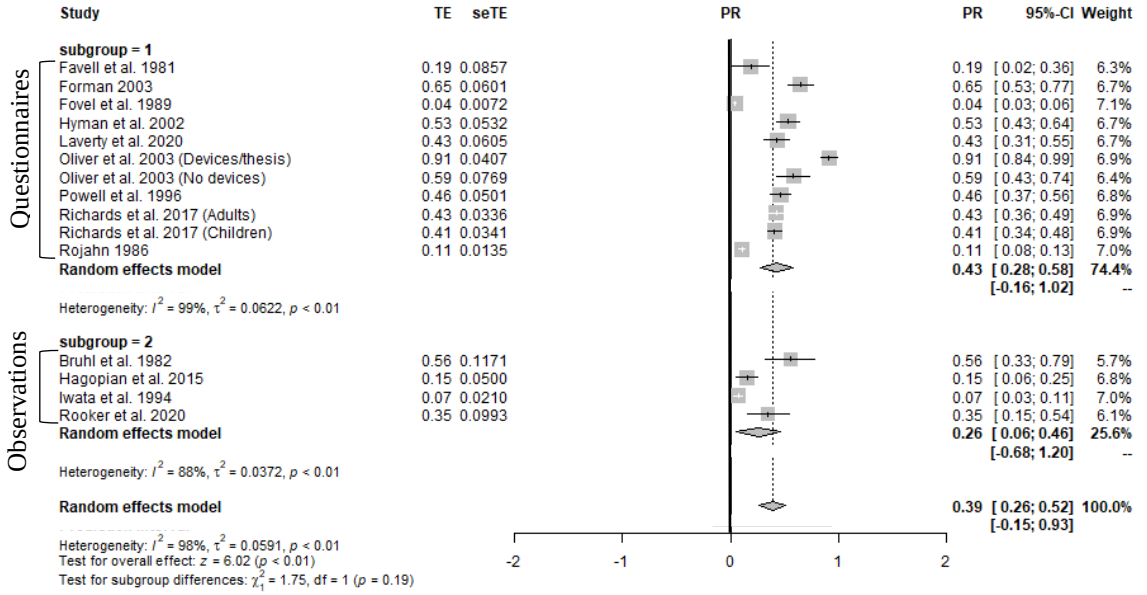
The meta-regression revealed that the association between the proportion of male participants and prevalence rates of self-restraint, and the proportion of participants with an ID and prevalence rates of self-restraint were non-significant. This suggests neither gender nor presence of ID influenced the prevalence of self-restraint. There was a significant association between mean age of participants and prevalence of self-restraint such that studies with the older participants had the highest levels of self-restraint. The presence of autism and prevalence of self-restraint were also significantly associated, such that as the percentage of individuals with autism in the sample increased, the prevalence of self-restraint increased.

2.4.3.2 Influence of study characteristics on prevalence rates

To examine the influence of measurement of self-restraint on the prevalence rates, subgroup analyses were conducted. Questionnaires were used by 11 samples, and four samples used direct observations. Figure 2.3 shows the forest plot of the random effects model splitting data into subgroups. Overall, there was no significant difference between the subgroups, χ^2=1.75,

p=.186. This suggested measurement of self-restraint did not significantly influence prevalence rates.

Figure 2. 3 Random effects forest plot for the subgroup analysis exploring measurement of self-restraint



2.4.4 Prevalence of self-restraint topographies

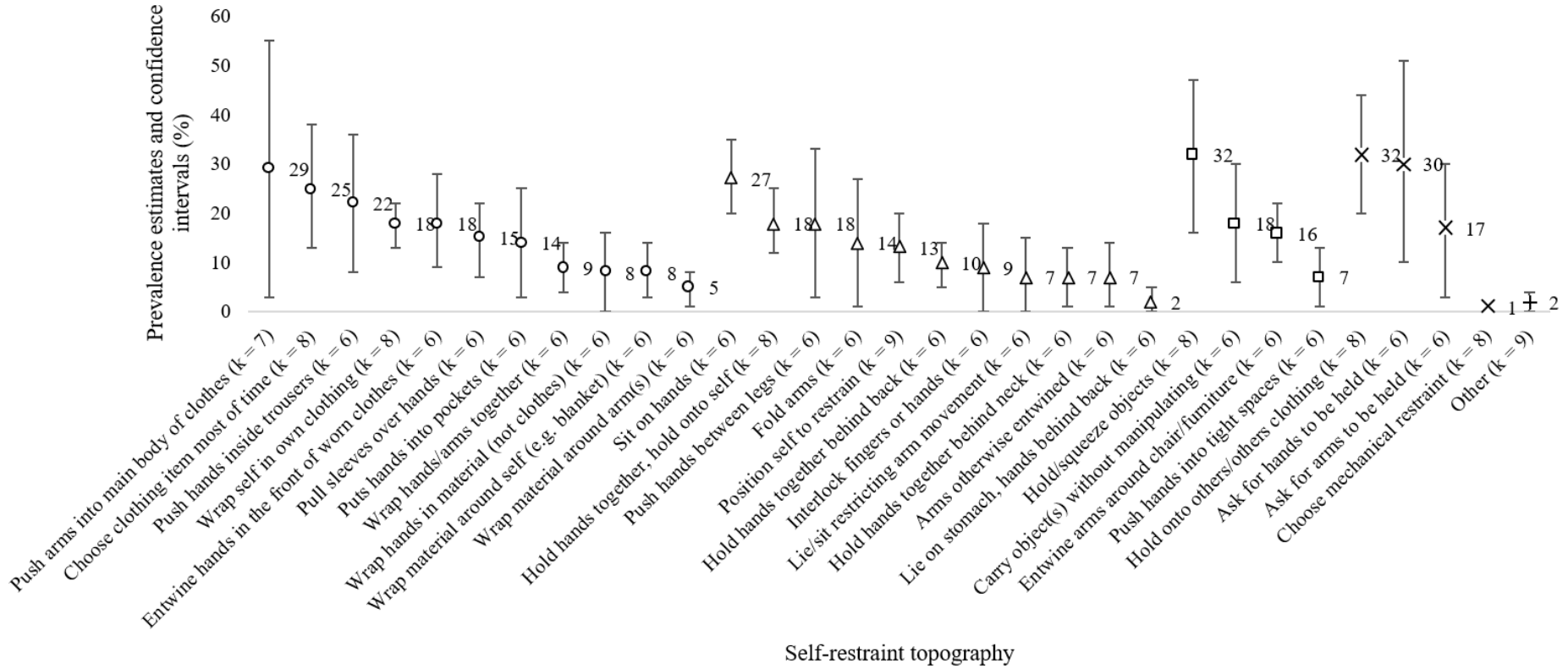
A total of 12 samples (*n*=1448) reported the prevalence of different topographies of self-restraint. Prevalence data for 31 topographies were extracted to generate individual pooled prevalence estimates. Topographies were based on items listed in the SRQ (23 items) and SRC (seven items). Where studies used observational techniques, topographies were assigned to the questionnaire item they most closely represent (Appendix 2.8).

A series of meta-analyses were conducted to identify and compare pooled prevalence estimates of the 31 topographies. Figure 2.4 represents topography estimates calculating

prevalence from individuals reported to show self-restraint. Pooled prevalence estimates for each topography calculated from the total number of participants (i.e., those with and without self-restraint) can be found in Appendix 2.9.

Within the pooled sample of individuals known to self-restrain, the most prevalent forms of self-restraint were 'holding or squeezing objects' and 'holding onto others or holding onto others' clothing' (both 32%). The least common topographies were 'chooses mechanical restraint' (1%) and 'other' (2%; Appendix 2.10 displays qualitative descriptions of "other" forms of self-restraint).

Figure 2. 4 Prevalence estimates and confidence intervals of each topography of self-restraint in those reported to self-restrain, grouped by the five overarching categories of self-restraint

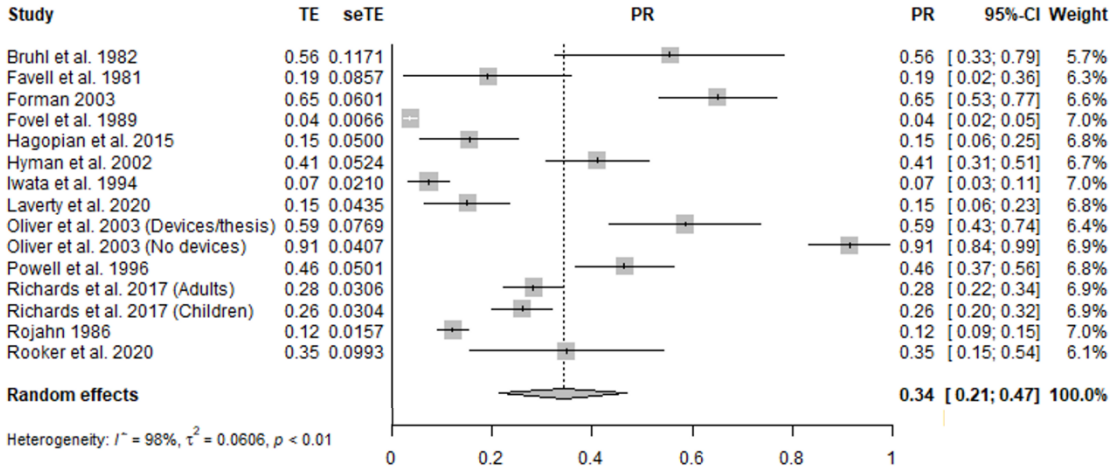


○ Estimates for topographies involving the use of clothing or materials
△ Estimates for topographies involving the use of body
□ Estimates for topographies involving the use of objects
× Estimates for topographies involving a preference for imposed restraint
+ Estimates representing other forms of self-restraint

2.4.5 Correlates of self-restraint

To examine the relation between self-restraint and SIB, a prevalence analysis only including participants known to self-injure (*k*=15) was conducted. Results indicated that self-restraint occurred in 34% of the pooled sample, *z*=5.23, *p*<.0001, 95%CI [21.36, 46.97] (Figure 2.5). Heterogeneity was high, $tau^2 = 0.061$, Higgin's $I^2 = 98\%$; Q(14)=799.77, *p*<.0001, requiring further inspection. Heterogeneity checks revealed no studies bore disproportionate influence on the estimate (Appendix 2.11). The random-effects model weighted by quality rating also indicated a prevalence of 34%, *z*=5.06, *p*<.0001, 95%CI [21.04, 47.63] (Appendix 2.12).

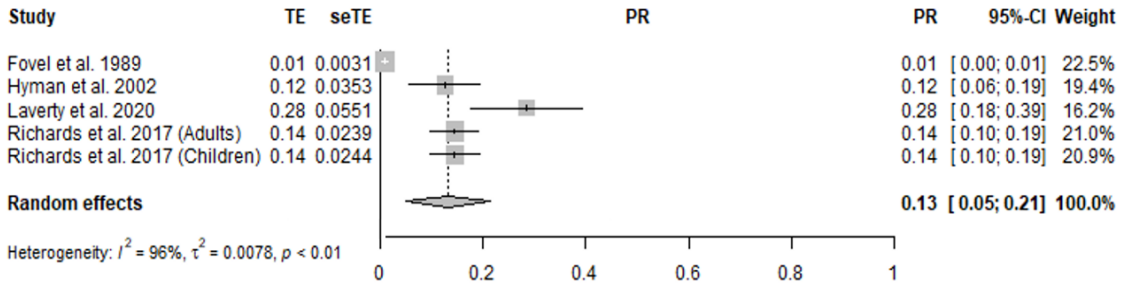
Figure 2. 5 Random effects forest plot for the prevalence of self-restraint among those who self-injure



An additional random-effects weighted analysis was conducted to identify the prevalence of self-restraint in the absence of self-injury (*k*=5). An estimate of 13% was identified, *z*=3.16, *p*=.0016, 95%CI [5.01, 21.43] (Figure 2.6). Heterogeneity was high, $tau^2 = 0.0078$, Higgin's $I^2 = 96\%$, Q(4)=96.20, *p*<.0001. The random-effects model weighted by quality rating showed a prevalence of 15%, *z*=3.42, *p*=.0006, 95%CI [6.45; 23.75] (Appendix 2.13). Exploration of

heterogeneity and influence identified no studies bore disproportionate influence on the estimate (Appendix 2.14).

Figure 2. 6 Random effects forest plot for the prevalence of self-restraint in the absence of self-injury



To further quantify the strength of the association between self-restraint and self-injury, as well as other related correlates of restricted/repetitive behaviours, compulsive behaviours, impulsivity/overactivity and stereotyped behaviours, effect sizes were meta-analysed. Table 2.6 reports the pooled effect sizes for each correlate, using the generic inverse variance method meta-analysing Fisher's z transformation of correlations. Random effects and quality-weighted forest plots for each correlation analysis can be found within Appendix 2.15-2.16. Results revealed a moderate positive correlation between self-restraint and SIB, while other correlations were non-significant.

Table 2. 6 Pooled correlation coefficients correlating behaviours with self-restraint

Correlate	**Pooled effect size**	**95% confidence intervals**	***p***	***k***	***N***
Self-injurious behaviour	.208 [a]	.141; .273	**<.001***	13	1576
Restricted/repetitive behaviours	.001	-.002; .004	.504	3	491 [b]
Compulsive behaviours	.003	-.005; .011	.444	2	187 [b]
Impulsivity/overactivity	.001	-.002; .004	.463	3	491 [b]
Stereotyped behaviours	.002	-.026; .049	.551	2	108 [b]

Note. [a] Data from one sample included within analysis (Oliver et al., 2003) were derived from the additional data source, with backwards calculations using a larger sample size (N=54) than the sample size included in the journal article (N=41). [b] Sample size too small to provide sufficient power according to a priori power analysis.
*Significant pooled correlation in bold (p<.05).

2.5 Discussion

This meta-analysis of 15 samples involving 2309 participants revealed three key findings. First, the overall pooled prevalence estimate of self-restraint was 39%. This prevalence rate was influenced by participant age and presence of autism, however participant gender, presence of ID and measurement of self-restraint did not significantly influence the prevalence rates. Second, the prevalence of 31 self-restraint topographies were meta-analysed, revealing the most common behaviours were 'holding or squeezing objects' and 'holding onto others or holding onto others' clothing' (both 32%), and 'choosing mechanical restraint' the least common (1%). Third, there was a moderate positive correlation between self-restraint and self-injury, r=.21, such that self-restraint occurred among 34% of participants who self-injured, while self-restraint occurred in 13% of participants who did not self-injure. Restricted/repetitive behaviours, compulsive behaviour, impulsivity/overactivity and stereotypy were not significantly correlated with self-restraint. The theoretical and clinical implications of these findings are now discussed.

Over one third of autistic individuals and/or individuals with ID included in the existing literature were reported to self-restrain. This represents a clinically significant proportion of individuals and provides an informative estimate extending previously heterogeneous figures. It is important to acknowledge that since the inclusion criteria of this study involved investigating self-restraint, it would be erroneous to conclude that 39% of *all* autistic individuals and/or those with ID self-restrain. A degree of sample bias is recognised, as many studies recruited participants based on displaying SIB. Therefore, the prevalence figures reported in this study reflect the pooled estimate of relevant samples, who were autistic and/or had ID, and/or showed SIB.

Self-restraint and self-injury co-occurred at a rate of 34% and shared a significant medium positive correlation, r=.21. To explore theoretical implications, the high prevalence of self-restraint among those who self-injure supports existing accounts that self-restraint and self-injury are bound together by paradigms of positive and negative reinforcement. For example, it is possible that self-restraint is a functional response to escape from aversive self-injury (Luiselli, 1993). The prevalence estimate of self-restraint in the absence of self-injury was, as expected, substantially lower (13%), deserving discussion. At face value, this suggests that for a small subgroup of individuals, self-injury and self-restraint are not related nor do they share a functional relationship. Perhaps for some, self-restraint provides sensory comfort unrelated to behavioural control, such as the warmth provided when wrapped up in a blanket (Peterson & Peterson, 1968) or the physical sensory feedback provided by contact behaviours. Alternatively, self-restraint in the absence of self-injury may be explained by the stimulus control hypothesis, suggesting that some restraining behaviours or devices ('symbolic restraints'; Pace et al., 1986) obtain reinforcing properties indicative of injury-free behaviour (section 1.4.5.1). Future research is needed to examine mechanisms underpinning self-restraint in individuals who do not SIB.

The high prevalence of self-restraint in some neurodevelopmental conditions and the association between autism and self-restraint has practical implications. Given the consequences of self-restraint to physical and emotional wellbeing (Scheithauer et al., 2015; Smith et al., 1992) it is suggested that practitioners in clinical settings consider self-restraint, and the possible extreme impact on quality of life. The presence of autism influenced the prevalence of self-restraint, such that as the percentage of autistic individuals in the sample increased, self-restraint prevalence also increased. This suggests autism may be a risk factor

for self-restraint, over and above ID. Individuals with autism presenting to clinical services for SIB should also be assessed for the presence and severity of self-restraint. Assessment using informant measures such as the SRC or SRQ, and/or behavioural observations, should occur both at initial formulation and routinely throughout care, as it is not yet understood how self-restraint may persist and impact wellbeing over the lifespan.

Participant age significantly influenced the prevalence rates of self-restraint. Previous literature has both supported and contradicted this finding; for example, Fovel et al. (1989) reported lower age was associated with self-restraint, however Oliver et al. (2003) and Richards et al. (2017) reported that age did not have an effect. The meta-regression suggested that as participant age increased, so did the prevalence of self-restraint. It is not possible to identify a particular age group most likely to self-restrain based on this analysis alone, however from observing the range of mean ages in the current studies (13 to 34 years), it is concluded that older participants were more likely to self-restrain.

Participant gender did not influence overall prevalence rates. Previous research has produced equivocal findings about the role of gender in showing self-restraint. For example, Oliver et al. (2003) found males were significantly more likely to show at least one form of self-restraint than females, however Hyman et al. (2002) revealed gender had no effect. Further research is needed to explore gender differences in self-restraint. Finally, whether authors employed questionnaire or observational methods to measure self-restraint did not influence prevalence rates. This may be reflective of the shared approach both methods take in noting topographies. Both approaches require informants to identify self-restraint in terms of topographies displayed by the individual, such as ‘sitting on hands’. This suggests both

approaches measure a similar construct. This implicates topography as a primary parameter of self-restraint (section 1.4.6) and points to a theoretical correlation between informant-report and observational methods of self-restraint. Future research employing both measures within the same sample is required to identify potential similar and disparate underlying constructs.

There were fewer than ten studies included in the meta-regressions assessing participant age and presence of autism. To examine the existing data, analyses were conducted on the respective nine and eight samples. The limited power of these analyses means that the results should be interpreted with caution. Importantly, the lack of participant data regarding individuals who showed self-restraint potentially limits understanding who may be at highest risk of behaviour. There were insufficient data to meta-analyse the impact of ID severity, characteristics of autism, and health characteristics such as poor vision, hearing problems, digestion problems and skin problems on self-restraint prevalence. Thus, it is imperative future research describes participant characteristics of those who self-restrain.

A total of 31 topographies of self-restraint were identified during data extraction. Individual meta-analyses of pooled prevalence estimates were conducted to identify common topographies within individuals known to self-restrain. Consistent with prior literature, 'holding/squeezing objects' and 'holding onto others, holding onto others' clothing' were the most common self-restraint behaviours (both 32%). 'Asking for hands to be held' was the second most common (30%), while 'choosing mechanical restraint' was the least common (1%). These data define self-restraint within autistic groups and/or individuals who have ID and indicate that some forms of imposed restraint are favoured. There was high variation in prevalence between categories of self-restraint (i.e., use of clothing/materials, body, devices

and preference for imposed restraint). For example, a preference for imposed restraint included the most and least common behaviours. This suggests conceptualising behaviours according to broad categories may not be useful. Further research is required to explore the underlying factor structure of the SRQ, which calculates categorical scores, to examine utility of categories.

Identification of common self-restraint behaviours has clinical implications. Identification of self-restraint, particularly within individuals known to self-injure, may help clinicians to understand the function and severity of self-injury. Self-restraint behaviours incompatible with SIB may suggest the restraint occurs to supress self-injury, providing information for behavioural management and intervention. Furthermore, given its rarity, showing a strong desire to access mechanical restraint may be an example of 'extreme' self-restraint. The presentation and removal of restraint during care and intervention must be carefully evaluated and considered. Given that a large majority of the population vulnerable to self-restraint and self-injury are often unable to report internal experiences, these findings shed light on how to recognise self-restraint in vulnerable groups.

The third and final aim of this study was to identify behaviours correlated with self-restraint. As discussed, self-injury was positively correlated with self-restraint, but restrictive/repetitive behaviours, compulsivity, impulsivity/overactivity and stereotypy were not significantly associated with self-restraint. It should be noted that few studies provided sufficient data for inclusion within the analyses, however all analyses included at least two samples (Valentine et al., 2010). Further research using large, well-powered samples is needed to test these associations further. Identification of correlates may further elucidate the mechanisms

underpinning self-restraint. For example, research exploring the role of executive functions on SIB has revealed that impulsive individuals are five times more likely to show severe SIB than non-impulsive individuals (Richards et al., 2017) suggesting self-restraint may emerge as a compensatory tool to regain control over the most severe, 'compulsive' self-injury (King, 1993). Supporting this, Richards et al. (2017) reported that the presence of SIB and impulsivity predicted the presence of self-restraint two-fold in autistic individuals. Research replicating this finding could pinpoint impaired inhibitory control as a cognitive mechanism underpinning self-restraint.

2.5.1 Limitations

Although this meta-analysis was conducted using robust search, extraction, and analytic methods, limitations are acknowledged. First, the field of self-restraint is characteristically small, and by excluding single-case designs the overall number of samples yielded for meta-analysis was low (k=15). This exclusion criterion was essential to avoid bias and sensitivity analyses revealed that no single study exerted undue influence on the overall effect sizes reported.

Second, a range of measures and definitions of self-restraint were employed across studies. The most common measure, the SRC, rated self-restraint according to topographies displayed over the lifetime. Consequently, analyses have identified estimates of individuals who have shown self-restraint across the lifetime, not necessarily individuals who showed self-restraint at the time of assessment. Arguments around the usefulness of characterising self-restraint across the lifetime and only by topography raise questions around the rigor of such measures (Forman, 2003; section 1.4.4). Future research should aim to define better the parameters and

critically evaluate the measurement of self-restraint.

Finally, the quality assessment revealed variation in study quality, with scores ranging from .22 to .67 (maximum = one). By observing quality ratings, it is revealed that most research exploring self-restraint relies on obtaining participant records through specialist services to identify autistic individuals and/or those with ID, rather than by employing direct diagnostic assessments. It must be acknowledged that diagnoses are broad, heterogeneous umbrella terms, with each individual presenting unique strengths, skills and differences. Additionally, transdiagnostic approaches value the severity of behaviour over categorical diagnoses (Astle et al., 2022). However, in order to test theoretical models and suggest implications for clinical practice, it is important researchers define participant characteristics that may be related to showing self-restraint. Therefore, where possible, future research should strive to use direct assessments of autism (e.g., ADOS, ADI-R), intellectual functioning (e.g., Wechsler Intelligence Scale for Children) and adaptive ability (e.g., the Vineland Adaptive Behavior Scale) to describe participant characteristics, whether diagnostically or on a continuum.

2.5.2 Conclusions

In summary, the pooled prevalence of self-restraint among individuals with autism and/or ID was estimated to be 39%. Self-restraint and self-injury were found to be positively correlated when accounting for all existing research, such that one third of those who self-injure also self-restrain. However, a prevalence figure of 13% for self-restraint in the absence of self-injury suggests a more complex relationship than that explained solely by models of positive and negative reinforcement. ‘Holding onto others or holding onto others’ clothing’ and ‘holding or squeezing objects’ were identified to be the most common topographies, while

'chooses mechanical restraint' was rare. Restrictive/repetitive behaviours, compulsivity, impulsivity/overactivity and stereotypy did not significantly associate with self-restraint, highlighting the need for larger-scale studies of the associations between self-restraint and putative correlates.

CHAPTER THREE

A CROSS-SYNDROME INVESTIGATION OF SELF-RESTRAINT: THE BEOND SURVEY

3.1 Preface

Chapter 2 revealed the prevalence estimate of self-restraint in autistic individuals and/or those with intellectual disability was 39%. Within this pooled sample, 34% of individuals who self-injured also showed self-restraint, with behaviours significantly associated, r=.21. Despite associations, a purely operant account of self-injury cannot account for the presence of self-restraint, raising challenges for this prevailing theory. An alternative explanation is that the self-injury is beyond the individual's control, turning attention to hypotheses of impaired inhibitory control and compulsivity. However, the theoretical mechanisms supporting models are not well-understood due to a lack of existing data, and the absence of consideration of additional correlates, as revealed in Chapter 2. Cross-syndrome studies have the potential to identify transdiagnostic markers of behaviour and inform theoretical understanding, however thus far, self-restraint has only been assessed categorically and not as a continuous, parameter-based variable. Therefore, this Chapter examines correlates and predictors of self-restraint in a large, heterogeneous sample of individuals with neurogenetic conditions to investigate operant learning and behavioural control accounts.

3.2 Introduction

Self-restraint is observed in populations where self-injurious behaviour (SIB) is common, including individuals with idiopathic autism[12] (Laverty et al., 2020; Richards et al., 2017), intellectual disability (ID) (Favell et al., 1981; Fovel et al., 1989; Rojahn, 1986), autism and co-occurring ID (Bruhl et al., 1982; Forman, 2003; Hagopian et al., 2015; Oliver et al., 2003; Rooker et al., 2020), and syndromic ID (i.e., Cornelia de Lange Syndrome; CdLS; Hyman et al., 2002). SIB severity, overactivity/impulsivity (Richards et al., 2017), compulsivity (Hyman et al., 2002) and anxiety (King, 1993) are putative correlates of self-restraint, supporting both operant (section 1.4.7.2) and impaired behavioural control accounts (section 1.4.7.3). However, current understanding of self-restraint displayed by individuals who self-injure is limited by two main factors. First, statistical power and generalisability of findings is limited by relatively small sample sizes (e.g., Oliver et al., 2003). Second, the putative correlates of self-restraint have not yet been assessed using a continuous measurement approach, with implications for identifying transdiagnostic risk markers of behaviour in high-risk groups. Therefore, the overarching aim of this study was to identify correlates and predictors of self-restraint in a large sample of individuals with a range of neurodevelopmental conditions.

The term 'neurodevelopmental conditions' refers to a heterogeneous constellation of conditions including autism, ID, and rare genetic syndromes associated with autism and/or ID (section 1.3). A neurodevelopmental continuum model indicates that there may be behaviours or traits that cross-cut traditional diagnoses, such that the same outcome may have multiple different causes (Morris-Rosendahl & Crocq, 2020). This model implies that it is not the

[12] 'Idiopathic' refers to cases where there is no known associated genetic cause for autism.

presence of a diagnosis per se (i.e., ability to fully meet criteria) that is important for understanding behavioural outcomes, rather the presence of specific traits that underpin behaviour. Most self-restraint research has involved individuals with autism and/or ID (section 2.4.2), with very few individuals with neurogenetic conditions included in self-restraint research (Appendix 2.5; although it is possible that a lack of reporting of diagnoses has led to under-identification). This is surprising given the high prevalence of SIB in neurogenetic populations (e.g., Arron et al., 2011; section 1.3.1). Identifying correlates of self-restraint in syndromes at elevated risk of SIB can provide insight into potential transdiagnostic risk markers of behaviour (Astle et al., 2022). Therefore, this study examined transdiagnostic correlates of self-restraint across 23 genetic syndromes associated with autism and/or ID.

A neurodevelopmental continuum model favours the use of continuous 'severity-based' variables over categorical data (Fletcher-Watson, 2022). Most cross-sectional research has measured self-restraint using the Self-Restraint Checklist (SRC; Powell et al., 1996; section 2.4.2), an informant-report questionnaire that explores differences between 'self-restraint' and 'no self-restraint' groups (section 1.4.6). Decisions around group classifications are arbitrary and rarely clearly reported. Given that self-restraint behaviours are not solely displayed by individuals who self-injure (section 1.4.2), and that self-restraint may function for reasons unrelated to SIB, including to access warmth, comfort and sleep (Peterson & Peterson, 1968), it is important to extend understanding by looking beyond comparison of group differences. A grouping approach, comparing those with and without self-restraint, does not account for the number of topographies an individual displays. Topography is an essential parameter of self-restraint (section 1.4.5.1), with individuals with ID reported to display up to 16 different

behaviours (Forman, 2003). Thus, measuring self-restraint according to a continuous 'topography score' not only accounts for the presence of self-restraint (replicating previous studies), but also accounts for multiple forms of self-restraint (section 1.4.6). No study has yet explored the relation between topography scores and correlates including SIB, compulsivity and impulsivity (discussed below). Therefore, this Chapter extended use of the SRC beyond traditional categorical scoring to identify links between variables and number of self-restraint topographies shown.

The estimated pooled prevalence of self-restraint in the existing literature for individuals with autism and/or ID is 39% (section 2.4.3). However, estimates vary between and within conditions. The prevalence of self-restraint in those with ID ranges from 11 to 92% (Iwata et al., 1994; Oliver et al., 2003; Rojahn, 1986), in individuals with idiopathic autism from 41 to 43% (Laverty et al., 2020; Richards et al., 2017) and in autistic individuals with ID from 15 to 65% (Forman, 2003; Hagopian et al., 2015). Hyman et al. (2002) conducted a cross-sectional study examining self-restraint in syndromic ID, reporting a prevalence of 53% in individuals with CdLS. Despite variation in estimates (section 2.2.1), these data suggest self-restraint is relatively common across conditions. It is important to delineate factors that might contribute towards the presence of self-restraint due to the potentially deleterious consequences of intense, preferred and persistent self-restraint (section 1.4.2). The meta-analysis in Chapter 2 suggested that older age and the presence of autism contributed towards showing self-restraint (section 2.4.3.1), while smaller-scale cross-sectional studies found male gender (Oliver et al., 2003) and lower ability relate to self-restraint behaviour (Richards et al., 2017). It is currently unclear whether such findings replicate in broader populations known to show self-injury. Thus, before testing theoretical accounts of behaviour, it is first vital to describe the profile

and person characteristics of individuals who self-restrain within a large, high-risk sample. Furthermore, growing recognition of selection bias against individuals with lower ability, characterised by exclusion of individuals with ID from autism research and limited reporting of ability where co-occurring ID is identified (Russell et al., 2019), indicates the importance of transparent reporting of participant characteristics (e.g., genetic syndrome, ability levels and autism characteristics). Therefore, the first aim of this study was to describe the prevalence, topographies and demographic characteristics associated with self-restraint in a well-characterised sample of individuals with autism and/or ID.

Questionnaire studies have revealed individuals who self-restrain display greater SIB severity (Richards et al., 2017), poorer impulsivity and overactivity (Laverty et al., 2020; Richards et al., 2017) and greater repetitive (Laverty et al., 2020) and compulsive behaviours (Hyman et al., 2002) in comparison to those who do not self-restrain. Such findings have contributed towards building theoretical models of behaviour (discussed below). Further, thus far self-restraint has been uniquely associated with SIB, and is not considered to be driven by or associated with other behaviours of concern (BoC)[13] such as aggression and property destruction (e.g., Forman, 2003). This suggestion is solely theoretical however, as associations between self-restraint and other BoC have not been examined empirically. Given the high co-occurrence of BoC (Emerson et al., 2001), it is important to extend theoretical and practical understanding by assessing the relation between self-restraint and broader BoC. While several variables are hypothesised to 'drive' self-restraint, unique relations between these variables and self-restraint (controlling for the presence of other correlates) have yet to

[13] Often referred to as 'challenging behaviours' within the literature. The alternative term 'behaviours of concern' has been used to refer to self-injury, aggression and property destruction to place emphasis on the concerning nature of the *behaviour*, and remove connotations that the individual who displays such behaviours are *themselves* 'challenging'.

be examined, complicating current understanding. Therefore, the second aim of this study was to examine correlations between self-restraint and behavioural variables, such as SIB, other BoC and anxiety, and cognitive variables, such as impulsivity and compulsivity.

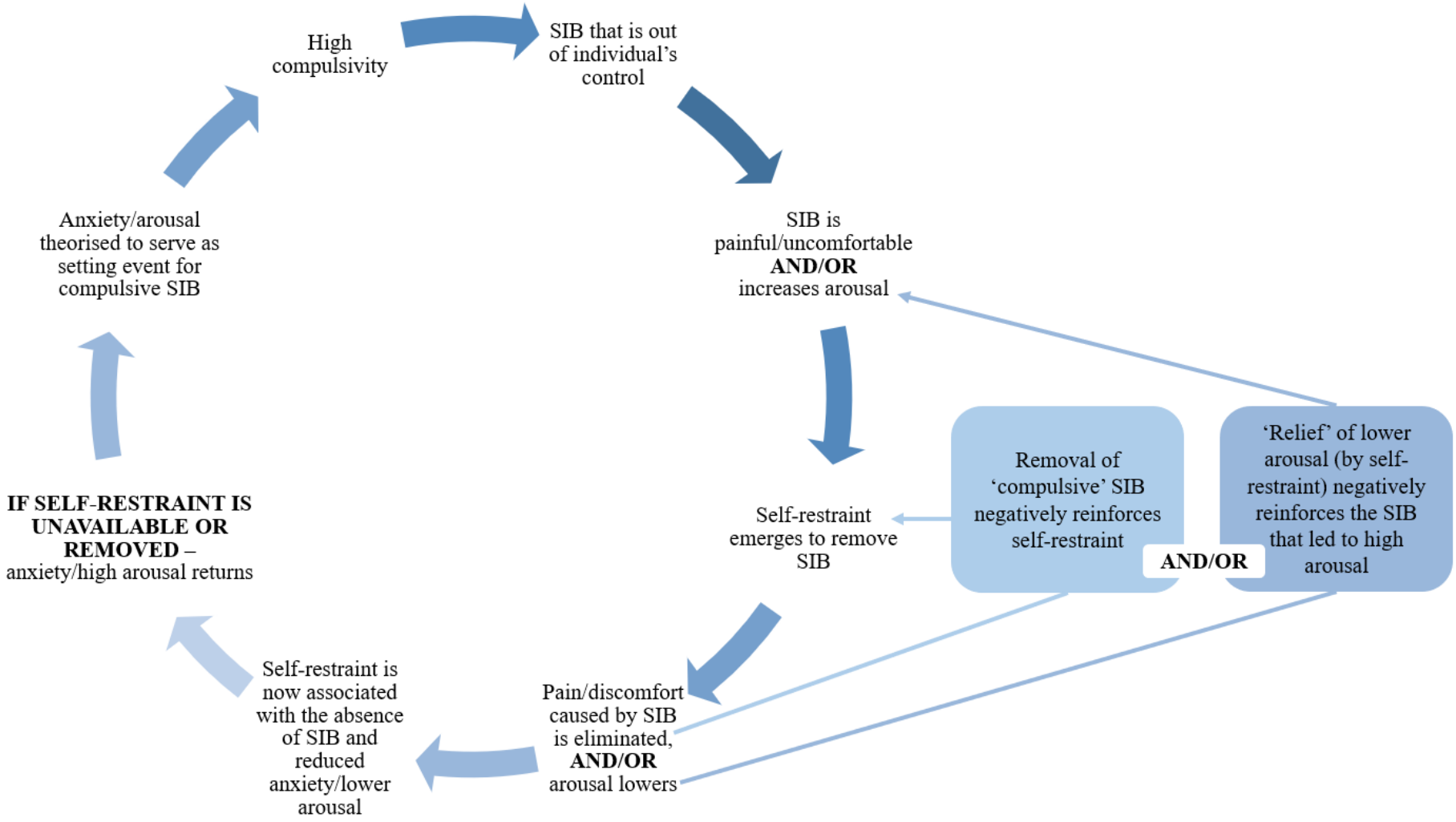
According to the operant learning account, self-restraint functions to remove the aversive properties of SIB (section 1.4.7.2). A positive moderate effect between self-restraint and SIB identified by meta-analysis supports this account (section 2.4.5). However, Smith et al. (1992) argued that the self-initiated termination of self-injury, which the operant model theorises to occur in order to access desirable contingences, indicates that SIB carries aversive properties that are not fully under the individual's control. Thus, the individual engages in self-restraint despite risking losing access to the reinforcing consequence of SIB (section 1.4.7.1). King (1993) conceptualised this 'loss' of behavioural control as compulsivity, evidenced by ritualised, habitual SIB that appears unconstrained by environmental contingencies (i.e., driven by automatic, internal reinforcers). The evidence-base examining compulsivity in individuals who self-restrain is small and equivocal: Hyman et al. (2002) identified a significant association between SIB and self-restraint for individuals with ID who displayed compulsive behaviours, but not for those who did not show compulsive behaviours. Similarly, Powell et al. (1996) found individuals with ID who showed both SIB and self-restraint were significantly more compulsive than those who only showed SIB. Interestingly, within a sample of autistic individuals without ID, Laverty et al. (2020) revealed no significant differences in compulsivity between individuals who did and did not self-restrain. Finally, using a multi-parameter definition of self-restraint (section 1.4.6), Forman (2003) found no significant differences in number of compulsions exhibited by individuals with ID who did and did not self-restrain. Thus, these conflicting data present a need to examine the role of

compulsivity across the neurodevelopmental continuum (i.e., individuals with autism and/or ID) to examine the compulsivity hypothesis.

The compulsivity account of SIB and self-restraint derives from anecdotal reports of individuals with Lesch-Nyhan syndrome, a genetic syndrome associated with ID, severe SIB and self-restraint (section 1.4.1), who appear fearful of their own inability to prevent SIB, and distress when self-restraint is unavailable (e.g., Nyhan, 1976). The desire to access self-restraint has been noted as 'anxiety' (Baumeister & Rollings, 1976). King (1993) suggested that compulsive (i.e., 'out of control') SIB is experienced as undesirable, painful and aversive by the individual. The cessation of SIB by self-restraint is therefore experienced as soothing and relaxing, negatively reinforcing self-restraint in the termination of 'compulsive' SIB. Romanczyk et al. (1992) further proposed that self-restraint functions to reduce high arousal, negatively reinforcing the behaviour that raises arousal and leads to self-restraint, which may be SIB. Thus, a vicious cycle of behaviour forms, whereby the individual engages in SIB to access the calming 'effect' of self-restraint. Crucially, both accounts indicate that once this cycle has formed, the removal of self-restraint leads to anxiety/high arousal[14], increasing the likelihood of compulsive SIB. King (1993) likened the individual with ID's access to self-restraint to a tightrope walker's access to a safety net. This complex relation is represented diagrammatically in Figure 3.1.

[14] Due to the conceptual overlap and lack of understanding and research into the role of anxiety and/or arousal in driving self-restraint and SIB, anxiety and arousal are at times grouped together when discussing theories.

Figure 3. 1 Relation between self-restraint, self-injury, compulsivity and anxiety/arousal



Limited research has examined the role of anxiety/arousal in self-restraint. Jennett et al. (2011) used a psychophysiological measure of anxiety with an autistic female with severe ID who self-injured and self-restrained. The individual's heart rate was at baseline when self-restraint was available but dramatically increased (e.g., by 60 beats per minute [bpm] within 30 seconds, maximum of 185 bpm) when self-restraint was terminated and SIB was initiated. Critically, when self-restraint was reinstated, the participant's heart rate returned to resting state and SIB ceased. This served as supporting evidence for King (1993)'s prediction of an inverse relation between SIB and self-restraint. Furthermore, Forman et al. (2002) findings of elevated SIB both before and *after* self-restraint did not support a purely operant account, leaving authors to theorise that the participant's anxiety to access self-restraint drove elevated SIB following self-restraint termination. Altogether, the relation between anxiety/arousal, compulsivity, SIB and self-restraint is complex, and perhaps as a result of this intricacy, there is a paucity of empirical evidence examining theories. However, better understanding of this relation has implications for intervention (particularly at a transdiagnostic level), implicating compulsivity and anxiety/arousal as areas to target.

One way to measure anxiety/arousal in individuals with ID, where cognitive anxiety is challenging to measure (section 1.4.7.3), is to examine behavioural anxiety. This refers to measurable, physical indicators of anxiety, including body movements, such as pacing, trembling, or gasping for breath, avoidance of people or situations or freezing in response to situations, and/or becoming disinterested in previously favoured activities. The measurement of such behaviours and their relation to self-restraint has not yet been studied. Therefore, to address this literature gap, the second aim of this study was to examine the compulsivity theory of SIB and self-restraint in individuals with autism and/or ID using a developmentally-

appropriate measure of anxiety.

In contrast to the compulsivity account, Oliver and Richards (2015) hypothesised that deficits in inhibition, a core executive function (EF; section 1.4.7.3) underpinned automatically-reinforced SIB, suggesting self-restraint emerges to compensate for 'impulsive' SIB. Whereas compulsivity refers to the perseveration of behaviour, impulsivity represents the inability to prevent the initiation of behaviour, resulting in acts that occur without foresight that may be harmful (Robbins et al., 2012; section 1.4.7.3). Supporting this account, greater impulsivity predicted self-restraint in autistic individuals (Richards et al., 2017; see section 6.2.3 for review). Overall, the theories of self-restraint described implicate SIB, impulsivity/compulsivity and anxiety/arousal as important drivers of self-restraint. Given the range of associated variables, a regression analysis approach is warranted to isolate the unique associations between variables and self-restraint, and thus inform theory. Therefore, the third aim of this study was to examine predictors of self-restraint, including SIB severity, impulsivity, compulsivity and behavioural anxiety, after controlling for covariates.

In summary, this study had three aims: (1) describe the prevalence, topographies and demographic correlates of self-restraint in neurogenetic syndromes associated with autism and/or ID, (2) examine correlations between behavioural variables and self-restraint, such as BoC and anxiety, and cognitive variables and self-restraint, such as compulsivity and impulsivity, and (3) examine whether SIB severity, impulsivity, compulsivity and behavioural anxiety predicted self-restraint after controlling for demographic covariates.

3.3 Method

3.3.1 Design and development

This cross-syndrome study capitalised on a wider study within the Cerebra Network for Neurodevelopmental Disorders. The Behavioural and Emotional Outcomes in Neurodevelopmental Disorders (BEOND) study was a collaboration across four research sites situated at Aston University, Surrey University, University of Birmingham and University of Warwick and measured a range of behavioural, cognitive and psychological outcomes for individuals with neurogenetic disorders and/or neurodevelopmental conditions. BEOND was designed to support individual research projects across the University sites. The pre-registered protocol for the BEOND study can be found on the Open Science Framework: https://osf.io/n89x7, DOI:10.17605/OSF.IO/N89X7.

3.3.2 Recruitment and screening

Participants were identified through three main routes. First, participants of previous studies were contacted through a database held at the University of Birmingham. Second, national syndrome-specific charities and parent support groups, such as Smith-Magenis Syndrome Foundation UK and Prader-Willi Syndrome Association UK, shared recruitment materials. Finally, the study was promoted using Cerebra Network social media.

Parents and caregivers[15] consented (Appendix 3.1) and completed questionnaire measures using an online survey platform. Informants reported on behalf of the individual they care for. Eligibility criteria included: ability to read and complete consent forms and survey measures in English; caring for an individual with a neurogenetic syndrome associated with ID and/or

[15] Henceforth ‘caregivers’.

autism (Appendix 3.2); and caring for an individual who was at least one year old (with no upper age restriction).

3.3.3 Participants

In total, 353 participants accessed the online survey. Of these, 272 (77%) caregivers completed the primary measure ('included' sample), while 81 (23%) caregivers provided demographic information but did not complete the SRC ('excluded' sample). Group comparisons revealed no significant differences in demographic characteristics between the included and excluded samples (Table 3.1) suggesting the SRC subsample was representative of the broader BEOND sample.

Table 3. 1 Demographic characteristics of the included and excluded BEOND study sample

Variable		**Sample**		**Mann-Whitney U/ χ^2**	***p***
		Included (*N*=272)	**Excluded (*N*=81)**		
Age	Mean, *SD* (range)	19.53, *13.78* (1-58)	16.28, *11.83* (2–47)	9519	.070
Sex	N female (%)	118 (43)	35 (43)	.003	.958
Verbal ability	N verbal (%)	192 (71)	61 (75)	.980	.322
ID	N ID (%)	256 (94)	78 (96)	.582 [a]	.329
Autism	N autism (%)	93 (34)	30 (37)	.223	.637

Note. [a] Fishers exact test used as crosstabs contained group <5

Participants had one of 23 neurogenetic syndromes associated with ID, of whom 192 participants (70.6%) were diagnosed by a clinical geneticist, 49 (18.0%) by a paediatrician, two (0.7%) by a General Practitioner, and 29 individuals (10.7%) by 'other' healthcare professionals (e.g., neurologist). In total, 256 participants (94%) were reported by caregivers

to have an ID and 93 (34%) were reported to be autistic. A total of 88 participants (32%) had a neurogenetic syndrome, autism and ID.

Table 3.2 displays participant demographic information. Information on service use was provided for 260 participants (96% of total sample), revealing 193 caregivers (74% of subsample) had accessed professional support or service(s) in the past two years for their child's physical, emotional and/or behavioural needs. Appendix 3.3 reports the prevalence of service use for BoC.

Table 3. 2 Demographic characteristics of included BEOND survey sample

Developmental variables	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Chronological age (years)	271		19.53 (16.00)	1 – 58 (13.78)
Sex (female/male/other [a])	272	118/153/1 (43/56/1)		
Mobility (walk unaided/unable)	272	232/40 (85/15)		
Genetic syndrome	272			
19p13		1 (0.4)		
8p23		1 (0.4)		
8p inv dup del		1 (0.4)		
Angelman		25 (9.2)		
CHAMP1[b]		1 (0.4)		
Coffin-Siris		9 (3.3)		
Cornelia de Lange		20 (7.4)		
Fragile X		40 (14.7)		
Jansen de Vries		9 (3.3)		
KBG		31 (11.4)		
Kleefstra		18 (6.6)		
Pallister Killian		1 (0.4)		
Potocki-Lupski		1 (0.4)		
Prader-Willi		12 (4.4)		
Rubinstein-Taybi		31 (11.4)		
SATB2-associated		10 (3.7)		

Note. [a] Other "no gender self-identification yet"; [b] 'Chromosome Alignment-Maintaining Phosphoprotein.

Table 3.2 cont.

Developmental variables	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Smith Magenis		23 (8.5)		
SWAN [c]		1 (0.4)		
SYNGAP1		1 (0.4)		
Tuberous Sclerosis Complex		20 (7.4)		
Wiedemann-Steiner		9 (3.3)		
Williams		1 (0.4)		
Wolf-Hirschhorn		6 (2.2)		
SCQ total scores	244		17.39 (18.00)	0 – 33 (6.60)
SCQ total verbal		158 (65)	16.91 (16.50)	0 – 33 (6.72)
SCQ total non-verbal		86 (35)	18.27 (19.00)	2 – 31 (6.33)
Autism/no autism [d]	244	155/89 (64/36)		
Intellectual disability	255			
Mild		30 (12)		
Moderate		83 (33)		
Severe		92 (36)		
Profound		21 (8)		
Other		5 (2)		
Unknown		24 (9)		
Wessex self-help score	271		6.25 (7.00)	3 – 9 (1.86)
Able [e]		69 (26)	8.58 (9.00)	8 – 9 (0.50)
Partly able [f]		112 (41)	6.62 (7.00)	6 – 7 (0.49)
Not able [g]		90 (33)	4.00 (4.00)	3 – 5 (0.75)

Note. SCQ = Social Communication Questionnaire. [c] 'Syndrome without a name'. [d] 'Autism' indicated by scoring 15 or above on SCQ (score 15 – 21 'ASD', 22 and above 'autism'). [e] Indicated by Wessex self-help score 8-9. [f] Indicated by Wessex self-help score 6-7. [g] Indicated by Wessex self-help score 3-5.

3.3.4 Measures

3.3.4.1 Characteristics and adaptive functioning

3.3.4.1.1 Social Communication Questionnaire (SCQ; Rutter et al., 2003).

This 40-item screening questionnaire (Appendix 3.4) is based on the Autism Diagnostic Interview–Revised (ADI-R; Lord et al., 1994) and was used to identify the presence of clinical autistic characteristics in this study. The SCQ is widely used due to its strong sensitivity and specificity, and concurrent validity with the ADI-R and Autism Diagnostic

Observation Schedule, Second Edition (Chandler et al., 2007). It is useful for identifying autism in individuals with lower intellectual ability (Snow & Lecavalier, 2008). The lifetime version produces three subscale scores: communication, social interaction and repetitive behaviour. Informants responded 'yes' or 'no' to items, where one point indicated autism-related behaviour, and zero points indicated absence of autism-related behaviour. Six additional items were administered for verbal children, with total possible scores ranging from 0-39, while non-verbal children scored between 0-33. A score of 15 or above is regarded as the clinical cut-off for autism (Berument et al., 1999), yielding a sensitivity value of .47 and specificity of .89 (Wiggins et al., 2007). Participants with scores of 14 or below were categorised as 'below threshold for autism'. Individuals with scores of 15 to 21 were categorised as 'Autism Spectrum Disorder' (ASD), representing presence of a subtype of autism, such as Pervasive Developmental Disorder, where the full clinical diagnosis of autism is not met (Chandler et al., 2007; Fombonne, 2003). Finally, individuals with scores of 22 or above were categorised as autistic. The ASD and autism groups are combined to create an overall 'autism' group (Chandler et al., 2007). The mean SCQ score, M=17.45, SD=6.62, was significantly higher than the clinical cut-off, $t(245)=5.812$, $p<.001$, 95% CI [1.62, 3.28]. Internal consistency was measured using McDonald's Omega (ω), and was acceptable, ω=.78 (section 3.3.6).

3.3.4.1.2 The Wessex Questionnaire (Kushlick et al., 1973).

The Wessex (Appendix 3.5) is a 16-item questionnaire that assesses adaptive functioning. Using a Likert scale, caregivers provide information across five subscales: continence, mobility, self-help skills, speech, and literacy. Higher scores represent higher ability. The self-help subscale was used a proxy measure of intellectual functioning, representing degree of adaptive behaviour (Kushlick et al., 1973; Oliver et al., 2012; Palmer & Jenkins, 1982). A

'self-help' score of 3-9 is generated by summing scores from questions regarding feeding, washing and dressing. Self-help scores of eight or nine group children as 'able', six or seven as 'partly able' and three to five as 'not able'[16] (Oliver et al., 2019). Psychometric analysis within a sample of children and adults with ID revealed moderate inter-rater reliability of Wessex subscales, mean κ=.62, and item-level reliability of κ=.54 (Kushlick et al., 1973; Palmer & Jenkins, 1982). Within the present sample, internal consistency of the self-help score was strong, ω=.87

3.3.4.2 Behaviour

3.3.4.2.1 Challenging Behaviour Questionnaire (CBQ; Hyman et al., 2002).

The CBQ (Appendix 3.6) determined the presence of self-injury, aggression and property destruction across the lifetime and within the previous month. Where behaviours were observed within the past month, information on topography and severity was collected, such as frequency of physical restraint to manage behaviour. The CBQ is suitable for use in verbal and non-verbal populations with ID and has demonstrated inter-rater reliability κ values ranging from .72 to .91 across subscales (Hyman et al., 2002). Scores on three items (rated from 0-4 or 1-5) assessing duration, severity and frequency of behaviours were summed to generate total severity scores for SIB, aggression and property destruction. Higher scores indicated more severe behaviour. Within the present sample, subscale internal consistency ranged from ω=.62, .64 to .67, respectively.

[16] For clarity and consistency with previous literature, original Wessex group labels have been used. It is important to note that labels relate to abilities measured by the questionnaire, and 'not able' does not indicate absence of all adaptive abilities.

3.3.4.2.2 Clinical Anxiety Screen for People with Intellectual Disability (ClASP-ID; Waite et al., 2019).

The ClASP-ID (Appendix 3.7) is a 34-item informant-report measure designed to assess behavioural anxiety in individuals with ID who may be unable to self-report anxiety. Caregivers rated the frequency of behaviours displayed in the previous month. Frequency of behaviour was measured on a seven-point Likert scale, ranging from 'almost never' to 'more than once a day'. A factor analysis revealed a four-factor structure: anxiety (minimum score = 14, maximum score = 98), pain (9–63), low mood (6–42) and consolability (4–28; Waite et al., 2019). Continuous subscale scores were generated, with higher scores indicating higher domain severity. Psychometric analyses, including identification of clinical cut-offs, are currently under investigation by the questionnaire authors (pre-registration: https://tinyurl.com/clasp-prereg). Within the present sample, there was strong internal consistency across all items, ω=.88, ranging from .88, .70, .80 to .57 across respective subscales.

3.3.4.2.3 Repetitive Behaviour Questionnaire (RBQ; Moss et al., 2009).

The RBQ (Appendix 3.8) is a 19-item questionnaire that measures frequency and severity of repetitive behaviours displayed in the last month. A five-point Likert scale measured behaviours such as 'object stereotypy' occurring 'never' to 'more than once a day'. Five subscales were produced: stereotyped behaviour (sum items 1-3), compulsive behaviour (sum items 4-7, 12, 16 and 18-19), insistence on sameness (sum items 15 and 17), restricted preferences (sum items 8, 10 and 13 for verbal participants only) and repetitive speech (sum items 9, 11 and 14 for verbal participants only), as well as a total score representing repetitive behaviour. In the present study, total scores were calculated by summing all items that did not

require verbal ability (i.e., excluding items 8, 9, 13 and 14). Higher scores represented higher frequency of repetitive behaviour. The RBQ is suitable for children and adults with ID, autistic individuals and individuals with and without verbal ability (Oliver et al., 2019). Psychometric analyses by Oliver et al. (2019) regarding 103 individuals with ID revealed variable item-level inter-rater reliability, .46 to .80, with 73% of coefficients above .60. Internal consistency across the five subscales was .74, .78, .50, .54 and .65 respectively. The RBQ has good concurrent and content validity with the repetitive behaviour subscale of the Autism Screening Questionnaire, r=.60, p<.001. Within the present study, internal consistency of subscales ranged from ω=.76, .79, .70, .51 to .75 respectively. Internal consistency across all items, excluding those relating to verbal ability, was strong, ω=.85.

3.3.4.2.4 The Activity Questionnaire (TAQ; Burbidge & Oliver, 2008).

This 18-item questionnaire (Appendix 3.9) assessed activity: six items assessed impulsivity (sum items 11-13 and 16-18), nine items assessed overactivity (sum items 1-9), and three items assessed impulsive speech (sum items 10, 14 and 15 for verbal participants only). The TAQ was developed for use in populations with ID, including individuals who are non-verbal and/or non-mobile. Developed in accordance with DSM-IV criteria for Attention Deficit/Hyperactivity Disorder (ADHD; APA, 1994) the TAQ yields robust inter-rater reliability for verbal and non-verbal participants, r=.74 and .78 respectively, and test-retest reliability for verbal and non-verbal participants, r=.88 and .94 respectively, across all three subscales among individuals with ID (Burbidge et al., 2010). Items are measured using a five-point Likert scale ranging from zero (‘never/almost never’) to four (‘always/almost all of the time’). Subscale scores and a total score were generated, with higher scores indicating higher

levels of activity. Within the present sample, subscale internal consistency was strong, ω=.90, .92 and .84 respectively. Internal consistency across all items was strong, ω=.93.

3.3.4.2.5 The Self-Restraint Checklist (SRC; Powell et al., 1996).

This eight-item checklist (Appendix 3.10) asked caregivers to indicate the presence or absence of seven topographies of self-restraint, such as 'wraps self in own clothing' and 'chooses mechanical restraint'. Informants indicated 'yes' (score of one) or 'no' (score of zero) to each item, with the eighth item asking caregivers to specify any 'other form of self-restraint'. Participants were grouped as showing self-restraint if caregivers indicated 'yes' to at least one item. A total 'SRC topography score' was calculated by summing the number of items, with possible scores ranging from zero to eight. Higher topography scores indicated a greater number of different topographies. The SRC was adapted for this study to ask about self-restraint observed in the last month. Powell and colleagues (1996) calculated interrater agreement for the SRC with adults with severe to profound learning disabilities. Agreement between two raters (i.e., familiar adults including teachers) was 91%. Within the present sample, internal consistency was acceptable, ω=.66.

3.3.5 Procedure

Questionnaire order in both online and paper surveys were counterbalanced in attempt to minimise order effects. Data were collected from November 2022 to August 2023. Health Research Authority, Health and Care Research Wales and Wales Research Ethics Committee provided ethical approval for this study (IRAS Project ID 299757; Appendix 3.11).

3.3.6 Data analysis

To examine the internal consistency of questionnaires, McDonald's Omega (McDonald, 1999) was calculated using 'the OMEGA macro' (Hayes & Coutts, 2020) in SPSS (v.29). Cronbach's alpha (α, Cronbach, 1951) is widely-used but relies on assumptions that are rarely met (Flora, 2020). For example, α relies on unidimensionality, such that each item measures the same construct, and assumes factor loadings across all items are equal ('essential tau-equivalence'; Dunn et al., 2014; Flora, 2020). As a result, α can easily be misused and misinterpreted (Hayes & Coutts, 2020). Researchers have proposed ω as suitable alternative (Ravinder & Saraswathi, 2020). Omega does not assume essential tau-equivalence, and is therefore considered a less biased (Flora, 2020) and more realistic coefficient (Watkins, 2017). Much like α, there is not universal consensus on the interpretation of ω coefficients (Watkins, 2017), however .65 to .80 has been suggested as acceptable reliability, with coefficients above .80 as strong evidence (Kalkbrenner, 2023).

Normality of data was inspected through Shapiro-Wilk and Kolmogorov-Smirnov tests. P-values of <.05 are typically considered non-normal, requiring the use of non-parametric tests. However, parametric t-tests and linear regression analyses are considered appropriate when using a relatively large sample size (i.e., >100; Lumley et al., 2002). Missing data were handled using pairwise deletion, whereby cases with missing values were excluded from respective analyses. In comparison to listwise deletion, where the entire participant case is deleted where at least one data point is missing, this approach preserves more data (Kang, 2013).

To examine aim one, the prevalence of self-restraint and concomitant self-injury were reported. Associations between self-restraint and SIB were examined using chi-square (χ^2). The prevalence of self-restraint topographies were reported, and SRC topography scores were compared across neurogenetic conditions. Relations between self-restraint and demographic variables, and behavioural and cognitive variables (aim two), were examined using Spearman's rho[17].

Multiple comparisons were controlled for using the Benjamini-Hochberg method (Benjamini & Hochberg, 1995). Methods to control for multiple comparisons aim to reduce the probability of committing a Type I error (false rejection of the null). A traditional approach is to control the family-wise error rate (FWER; the risk of committing at least one Type I error) using a Bonferroni correction. However, this approach is considered stringent, and increases the risk of committing a Type II error (false acceptance of the null; Kwong et al., 2002) and overlooking meaningful effects. Mechanisms related to severe BoC displayed by individuals with ID, an underrepresented population (Russell et al., 2019), may consequently be missed, with implications for theory and practice. Benjamini and Hochberg (1995) proposed a sequential modified Bonferroni correction controlling for the False Discovery Rate (FDR); the proportion of false positives among the total number of rejected hypotheses. This method is considered a less stringent and more powerful approach in comparison to traditional Bonferroni corrections (Haynes, 2013). The Benjamini-Hochberg method compares significance values against a critical value, accounting for the total number of statistical tests per analysis using a false discovery rate of 0.05.

[17] See section 2.3.5 for interpretation of r_s values.

To test aim three, examining predictors of self-restraint, a hierarchical multiple regression was conducted. An a priori power analysis was conducted using G*Power (v.3.1.9.7; Faul et al., 2009). Based on a medium effect size of .15, significance level of .05 and power of .95, a sample size of 160 participants was considered suitable for the regression. Continuous predictor variables were entered into five steps. In the first step, covariates age (in years) and ability (Wessex self-help scores) were entered. In the second, CBQ SIB severity scores were entered. In the third, TAQ impulsivity scores were entered, and RBQ compulsivity scores were entered into the fourth step. Finally, ClASP-ID anxiety subscale scores were added in the fifth step. The dependent variable was SRC topography score. Each predictor was continuous, had non-zero variance, and had a linear relationship with the dependent variable. Multicollinearity, the intercorrelation of predictor variables, was inspected using the Variance Inflation Factor (VIF) and tolerance statistics (i.e., reciprocal of the VIF, $1–R^2$; Kim, 2019). Tolerance statistics were above 0.2 and VIF statistics less than 10, suggesting the absence of multicollinearity (Kim, 2019; Menard, 1995; Myers, 1990). Visual inspection of the standardised residuals plot (standardised residuals, *ZRESID*, by standardised predicted values, *ZPRED*) suggested the assumption of independent errors had been met (i.e., residuals were independent of one another). Values fell within the limits of ±3.3 suggesting no significant outliers (Tabachnick & Fidell, 2001). Inspection of histogram and P-P plots suggested the assumption of normally-distributed errors had been met. Finally, inspection of the *F*-ratio significance value suggested that overall the model had a significant fit to the data ($p<.05$).

3.4 Results

3.4.1 The prevalence and profile of self-restraint in rare genetic syndromes

3.4.1.1 The prevalence and topographies of self-restraint

In total, 185 participants (68%) displayed self-restraint in the previous month (i.e., displayed at least one topography on the SRC). Behavioural characteristics are reported in Table 3.3. A total of 255 caregivers completed both the SRC and CBQ, measuring self-restraint and SIB respectively. There was a significant association between SIB and self-restraint, χ^2=8.897, *p*=.003, Phi (φ)=.187[18]. Of this subgroup, 92 participants (36%) had shown self-restraint and SIB in the past month, and 83 individuals (33%) had shown self-restraint without SIB. A total of 54 individuals (21%) showed neither self-injury nor self-restraint, and 26 participants (11%) showed self-injury without self-restraint.

All eight self-restraint topographies measured by the SRC were observed (Figure 3.2). The most common behaviours across the sample were 'holds or squeezes objects', shown by 117 participants (43%), and 'holds onto others or holds onto others clothing', shown by 97 individuals (36%). The least common behaviour was 'chooses mechanical restraint', displayed by seven participants (3%). 'Other' forms of self-restraint were displayed by 10 participants (4%), with examples including "wrap[s] himself in a sleeping bag-type blanket".

[18] Phi effect size is interpreted according to correlation coefficient interpretation (Ben-Shachar et al., 2023).

Table 3. 3 Behavioural characteristics of BEOND sample

Developmental variables	**Possible score range**	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Self-restraint - SRC		272			
Monthly self-restraint/no SR			185/87 (68/32)		
Total topography score	0 – 8			1.45 (1)	0 – 8 (1.46)
1 topography/2+ topographies			76/109 (41/59)		
Behaviours of concern – CBQ					
Monthly SIB/no current SIB		255	118/137 (46/54)		
Total SIB severity score	0 – 14	255		2.90 (0)	0 – 13 (3.57)
Monthly AGG/no current AGG		257	126/131 (49/51)		
Total AGG severity score	0 – 14	257		3.28 (0)	0 – 14 (3.82)
Monthly DoP/no current DoP		257	111/146 (43/57)		
Total DoP severity score	0 – 14	257		2.85 (0)	0 – 13 (3.68)
Activity – TAQ					
Impulsivity subscale	0 – 24	272		14.83 (16)	0 – 24 (7.11)
Overactivity subscale	0 – 36	272		13.90 (12)	0 – 36 (9.93)
Impulsive speech subscale	0 – 12	192		4.76 (4)	0 – 12 (3.74)
Total score	0 – 72	272		32.10 (31)	0 – 72 (17.15)

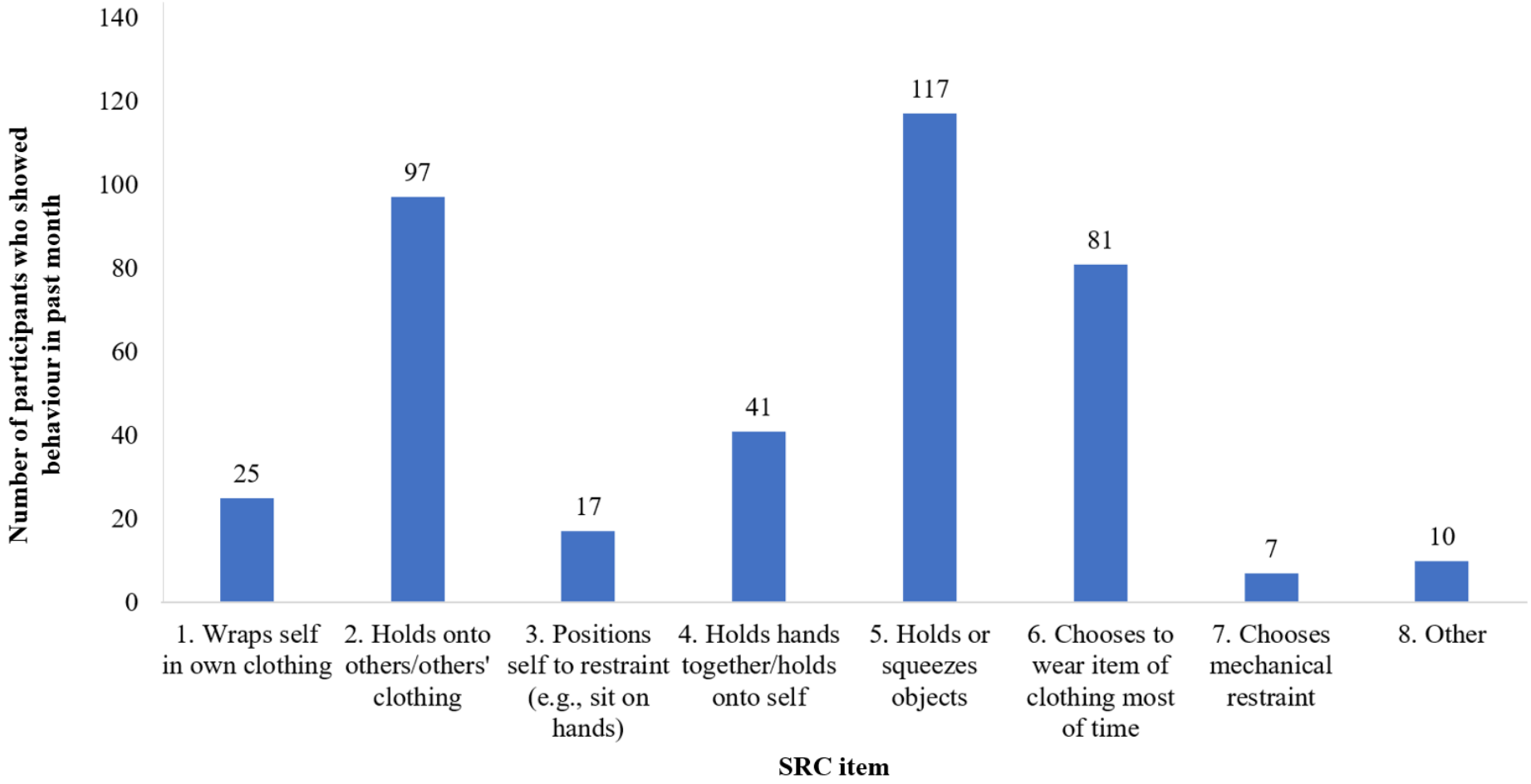
Note. SRC = Self-Restraint Checklist; SR = self-restraint; CBQ = Challenging Behaviour Questionnaire; SIB = self-injurious behaviour; AGG = aggression; DoP = destruction of property; TAQ = The Activity Questionnaire.

Table 3.3 cont.

Developmental variables	**Possible score range**	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Repetitive behaviour – RBQ					
Stereotyped behaviour subscale	0 – 12	272		6.06 (6)	0 – 12 (4.27)
Compulsive behaviour subscale	0 – 32	272		5.08 (3)	0 – 32 (6.31)
Insistence on sameness subscale	0 – 8	272		3.14 (3)	0 – 8 (2.82)
Restricted preferences subscale	0 – 12	192		5.43 (5)	0 – 12 (3.55)
Repetitive speech subscale	0 – 12	192		5.84 (5)	0 – 12 (3.97)
Total score	0 – 60	272		17.36 (15)	0 – 58 (12.21)
Anxiety – ClASP-ID		254			
Anxiety	14 – 98			36.89 (35)	14 – 96 (16.31)
Pain	9 – 63			15.82 (14)	9 – 58 (7.20)
Low mood	6 – 42			13.10 (11)	6 – 37 (6.95)
Consolability	4 – 28			20.26 (21)	4 – 28 (5.08)

Note. RBQ = Repetitive Behaviour Questionnaire; ClASP-ID = Clinical Anxiety Screen for People with Intellectual Disability.

Figure 3. 2 Prevalence of each self-restraint topography



Across the 23 syndrome groups, nine 'groups' contained one participant. Of these syndromes, individuals with 19p13, 8p inv dup del, CHAMP1, SWAN and Williams syndrome were reported to have shown self-restraint in the previous month (i.e., *n*=1, self-restraint prevalence = 100%). Individuals with 8p23, Pallister Killian, Potocki-Lupski and SYNGAP1 syndrome had not shown self-restraint (i.e., *n*=1, self-restraint prevalence = 0%). Of the remaining syndrome groups, the highest prevalence of self-restraint was displayed by Jansen de Vries and Wiedemann-Steiner syndromes (both 89%, *n*=8). The lowest prevalence of self-restraint was displayed by those with Wolf-Hirschhorn (50%, *n*=3). Table 3.4 displays self-restraint prevalence and topography scores across syndromes where groups contained more than one participant.

The highest mean topography scores were displayed by the individual with SWAN (4.00), and participants with Jansen de Vries (2.22) and Prader-Willi syndrome (2.17). The lowest mean topography scores[19] were displayed by those with Wolf-Hirschhorn (0.67) and Rubinstein-Taybi syndromes (1.00).

[19] Excluding the four syndromes where self-restraint was not displayed.

Table 3. 4 Self-restraint scores across participant groups where *N* >1

Group name	***N***	***n* self-restraint (%)**	**Self-restraint topography score**	
			Mean (median)	**Range (SD)**
Angelman	25	17 (68)	1.20 (1.0)	0 – 5 (1.23)
Coffin-Siris	9	6 (67)	1.78 (1.0)	0 – 5 (1.86)
Cornelia de Lange	20	13 (65)	1.40 (1.0)	0 – 4 (1.35)
Fragile X	40	30 (75)	1.63 (1.0)	0 – 7 (1.55)
Jansen de Vries	9	8 (89)	2.22 (2.0)	0 – 5 (1.64)
KBG	31	21 (68)	1.55 (1.0)	0 – 8 (1.79)
Kleefstra	18	10 (56)	1.22 (1.0)	0 – 4 (1.35)
Prader-Willi	12	9 (75)	2.17 (2.5)	0 – 7 (1.99)
Rubinstein-Taybi	31	19 (61)	1.00 (1.0)	0 – 3 (1.03)
SATB2-associated	10	7 (70)	1.50 (2.0)	0 – 4 (1.27)
Smith Magenis	23	17 (74)	1.52 (1.0)	0 – 5 (1.47)
Tuberous Sclerosis	20	12 (60)	1.45 (1.0)	0 – 4 (1.57)
Wiedemann-Steiner	9	8 (89)	1.67 (1.0)	0 – 3 (1.12)
Wolf-Hirschhorn	6	3 (50)	0.67 (0.5)	0 – 2 (0.82)

Note. Self-restraint topography score calculated using Self-Restraint Checklist totals, maximum possible score = 8.

3.4.1.2 Demographic and behavioural correlates of self-restraint

Correlations between SRC topography scores and demographic and behavioural correlates are show in Table 3.5. Age moderately negatively correlated with SRC topography scores, suggesting younger age was related to showing a greater number of self-restraint topographies. SCQ scores positively correlated with SRC topography scores with a moderate-to-large effect, suggesting greater autism characteristics were related to showing a greater number of self-restraint topographies. There were no significant correlations between SRC topography scores and verbal ability, ability to walk unaided, or sex. The relation between

SRC topography scores and Wessex self-help scores was non-significant, however the small effect was notable, suggesting lower ability related to self-restraint. Self-help scores negatively correlated with SCQ scores with a moderate-to-large effect, suggesting that higher ability was related to lower autism traits. SRC topography scores positively correlated with SIB, aggression, and property destruction severity scores, with moderate to large effects. This suggested that higher BoC severity was related to showing a greater number of self-restraint topographies. Additionally, all BoC inter-correlated with large effects.

SRC topography scores strongly positively correlated with impulsivity, overactivity, impulsive speech, and overall activity scores. This suggested that higher impulsivity was related to showing a greater number of self-restraint topographies. SRC topography scores positively correlated with stereotyped behaviour[20], compulsive behaviour, insistence on sameness, restricted preferences, repetitive speech, and overall repetitive behaviour scores, with moderate to large effects. This suggested that higher repetitive and compulsive behaviour was related to showing a greater number of self-restraint topographies. Finally, SRC topography scores strongly positively correlated with anxiety and pain, and moderately correlated with low mood scores. This suggested that higher behavioural anxiety was related to showing a greater number of self-restraint topographies.

[20] Stereotypy scores measured by the CBQ and RBQ strong correlated, r_s=.56, suggesting measurement of a similar construct. Given that stereotypy may be better conceptualised as a repetitive behaviour rather than BoC, the RBQ subscale was used to measure stereotypy in this Chapter.

Table 3. 5 Spearman's rho correlations between self-restraint topography scores and demographic and behavioural variables

		1	2	3	4	5	6	7	8	9
1	SRC topography score	—								
Demographic variables										
2	Age years	**-.22***	—							
3	Verbal	-.01	.11	—						
4	Mobile	.04	.09	**.46***	—					
5	Sex	.02	-.03	-.10	**-.14***	—				
6	SCQ total	**.26***	.10	**-.22***	-.10	-.07	—			
7	Wessex self-help	-.10	**.21***	**.67***	**.46***	-.11	**-.27***	—		
CBQ severity scores										
8	SIB severity	**.22***	-.09	-.03	-.00	-.01	**.24***	**-.14***	—	
9	Aggression severity	**.31***	**-.20***	.06	.10	.01	.03	-.10	**.36***	—
10	Property destruction severity	**.20***	**-.16***	.13	**.17***	-.03	.08	.02	**.37***	**.57***

Note. SRC = Self-Restraint Checklist; SCQ = Social Communication Questionnaire; CBQ = Challenging Behaviour Questionnaire; SIB = self-injurious behaviour.

*Significant p-values after controlling for multiple comparisons in bold.

Table 3.5 cont.

		1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19
TAQ subscales and total score																				
11	Impulsivity	**.35***	**-.18***	.03	**.18***	-.13	**.23***	**-.14***	**.39***	**.50***	**.51***	—								
12	Overactivity	**.32***	**-.35***	-.10	.01	**-.22***	**.25***	**-.18***	**.24***	**.33***	**.32***	**.65***	—							
13	Impulsive speech	**.30***	.03	-	-.02	**-.16***	**.17***	.12	.10	.09	.10	**.44***	**.49***	—						
14	Total activity	**.37***	**-.26***	.09	**.16***	**-.20***	**.23***	-.07	**.30***	**.42***	**.43***	**.86***	**.91***	**.64***	—					
RBQ subscales and total score																				
15	Stereotypy	**.33***	-.10	-.09	-.00	**-.18***	**.43***	**-.19***	**.22***	.12	**.18***	**.38***	**.48***	**.27***	**.47***	—				
16	Compulsivity	**.24***	.01	**.30***	**.24***	-.07	**.31***	**.20***	.11	**.17***	**.14***	**.31***	**.25***	**.22***	**.34***	**.35***	—			
17	Insistence on sameness	**.22***	**.20***	**.29***	**.23***	-.09	**.29***	**.24***	.12	**.16***	**.14***	**.29***	**.15***	.13	**.26***	**.25***	**.62***	—		
18	Restricted preferences	**.40***	-.08	-	-.01	-.07	**.40***	**-.20***	**.36***	**.27***	**.31***	**.47***	**.40***	**.35***	**.48***	**.44***	**.44***	**.46***	—	
19	Repetitive speech	**.40***	-.13	-	.03	-.08	**.36***	**-.17***	**.27***	**.21***	**.27***	**.44***	**.50***	**.42***	**.54***	**.44***	**.33***	**.33***	**.64***	—
20	Total repetitive	**.38***	-.01	**.20***	**.20***	**-.13***	**.47***	.07	**.22***	**.20***	**.23***	**.47***	**.42***	**.30***	**.51***	**.71***	**.83***	**.71***	**.66***	**.56***

Note. TAQ = The Activity Questionnaire; RBQ = Repetitive Behaviour Questionnaire. Dash (-) indicates verbal ability was not correlated with variables that only included verbal participants.
*Significant p-values after controlling for multiple comparisons in bold.

Table 3.5 cont.

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19	20	21	22	23
ClASP-ID subscales																							
21 Behavioural anxiety	**.37***	-.10	**.18***	**.22***	-.12	**.29***	.06	**.41***	**.47***	**.39***	**.52***	**.38***	**.22***	**.49***	**.32***	**.40***	**.43***	**.53***	**.47***	**.52***	—		
22 Pain	**.40***	**-.24***	**-.17***	**-.16***	-.03	**.25***	**-.23***	**.38***	**.29***	**.17***	**.27***	**.39***	**.19***	**.33***	**.37***	**.22***	.12	**.37***	**.32***	**.35***	**.44***	—	
23 Low mood	**.22***	.11	.05	.11	-.02	**.29***	.01	**.19***	**.23***	**.15***	.13	-.00	.16	.07	.11	**.23***	**.23***	**.30***	.14	**.26***	**.47***	**.30***	—
24 Consolability	-.01	**.18***	.10	.06	-.10	.01	.10	-.02	**-.14***	-.08	-.02	-.02	-.12	-.03	.03	**.14***	**.20***	.03	.06	.12	-.01	-.06	-.11

Note. ClASP-ID = Clinical Anxiety Screen for People with Intellectual Disability.
*Significant p-values after controlling for multiple comparisons in bold.

3.4.2 Predictors of self-restraint

A five-step hierarchical multiple linear regression was conducted to predict self-restraint based on SIB, impulsivity, compulsivity and behavioural anxiety, controlling for age and ability. Results from the final model are presented in Table 3.6 (Appendix 3.12 displays the full five-step model). There were three significant predictors of self-restraint in the full hierarchical model: lower age, higher compulsivity and higher behavioural anxiety. The full hierarchical model explained 28.3% (R^2) of the variance in the presence of self-restraint, $F(1,242)=12.622$, $p<.001$, suggesting good model fit.

Table 3. 6 Hierarchical multiple regression of self-restraint predictors

Model and predictor variables	**β**	**SE**	**t**	***p***	**R^2**	**R^2 adj**	**Collinearity statistics**	
							Tol.	**VIF**
					.283	.265		
Age	-.226	.006	-3.911	**<.001***			.884	1.131
Self-help score	-.026	.046	-0.457	.648			.885	1.130
SIB severity	.019	.026	0.312	.755			.770	1.298
Impulsivity score	.122	.014	1.810	.072			.653	1.530
Compulsivity score	.168	.015	2.731	**.007***			.787	1.271
Anxiety score	.252	.007	3.553	**<.001***			.588	1.700
Constant		.381	1.730	.085				

Note. β = standardised beta coefficient; SE = standard error; R^2_{adj} = adjusted R^2, Tol. = tolerance, VIF = Variance Inflation Factor; SIB = self-injurious behaviour.
*Significant predictors ($p<.05$) in bold.

In summary, younger participants and those with lower ability showed a greater number of self-restraint topographies. Over-and-above these factors, compulsivity and anxiety were uniquely associated with self-restraint. In contrast, SIB severity and impulsivity did not uniquely predict self-restraint.

3.5 Discussion

This cross-syndrome study involving 272 participants with 23 neurogenetic syndromes revealed three key findings. First, the overall prevalence of self-restraint was 68%, and over one third of participants had shown both self-restraint and SIB in the past month. Second, higher levels of self-restraint were associated with younger age, autism characteristics, SIB severity, impulsivity, compulsivity and behavioural anxiety. Finally, compulsivity and behavioural anxiety made unique contributions to self-restraint when age and ability were taken into account. The theoretical and practical implications of these findings are now discussed.

This was the first study to describe self-restraint prevalence and topographies using a large sample of participants with neurogenetic conditions. Over two thirds of participants had shown self-restraint in the previous month (68%), and at least one topography was displayed in 19 of the 23 genetic syndromes examined. In total, 36% of participants had displayed both self-restraint and SIB in the previous month, with behaviours significantly associated. This was comparable with the meta-analytic prevalence estimate of showing both SIB and self-restraint (34%; section 2.4.5), however it is important to acknowledge the elevated prevalence of self-restraint in the current study in comparison to the meta-analytic estimate.

The prevalence of common self-restraint topographies across all participants with neurogenetic syndromes replicated estimates from the existing literature in autism and ID (e.g., Hyman et al., 2002; Powell et al., 1996; Richards et al., 2017). The most common behaviours were ‘holds or squeezes objects’ (43%) and ‘holds onto others or holds onto others clothing’ (36%). Pooled prevalence estimates of topographies identified by meta-analysis

revealed these two 'holding' behaviours were the most common forms of self-restraint (both 32%; section 2.4.4). Similarly, the least common behaviour in this study, 'chooses mechanical restraint' (3%), was the least prevalent pooled estimate (1%) in the meta-analysis. Thus, despite differences in sample composition and overall prevalence of self-restraint, within the current sample, 'holding' behaviours were relatively common, while seeking mechanical restraint was rare.

Individuals displayed between zero to eight self-restraint topographies, with a mean overall self-restraint topography score of 1.45. This replicated previous results using the SRC. For example, in a sample of individuals with ID who showed self-injury, Powell et al. (1996) reported a mean topography score of 1.98, while the mean score for individuals with CdLS was 1.74 (Hyman et al., 2002; section 1.4.5.1). Individuals with KBG syndrome displayed the maximum number of self-restraint topographies (eight), while individuals with Fragile X and Prader-Willi syndrome displayed up to seven forms. Displaying a greater number of self-restraint topographies was related to negative outcomes, including more severe SIB, impulsivity and compulsivity (discussed below). Therefore, the SRC is a short and efficient tool for examining the prevalence of multiple self-restraint topographies, which may reveal important correlates.

Self-restraint was more common among younger participants and among those with more autism characteristics. While replicating findings from individual studies (e.g., Fovel et al., 1989; section 2.5), this finding contradicts the meta-analytic result associating self-restraint with older age (section 2.4.3.1). Further research is required to assess the relation between age and self-restraint. Greater SIB severity was moderately associated with self-restraint

topography scores, replicating the pooled meta-analytic effect size (section 2.4.5). Interestingly, greater severity of aggressive and destructive behaviours also significantly correlated with self-restraint scores. This was the first cross-sectional study to test associations between self-restraint and other BoC, and findings contradict suggestion that self-restraint functions only to inhibit SIB (e.g., Fisher and Iwata, 1996). The relation between self-restraint and BoC is perhaps unsurprising, given the high co-occurrence of SIB and other BoC both within this study and in previous literature (e.g., Matson et al., 2008; section 5.2). A larger effect was identified between self-restraint and aggression than self-restraint and SIB. Current understanding of self-restraint is that behaviours emerge to compensate for painful, undesirable self-injury, however it is plausible that restraining behaviours also develop in response to other BoC. Future research is warranted to explore this association using increasingly stringent definitions of self-restraint.

Impulsivity was moderately associated with a greater number of self-restraint topographies. The latter finding directly replicated Laverty et al. (2020) who examined group differences between those with and without self-restraint in autistic children and adults. These findings support the impaired inhibitory control account, whereby deficits in inhibition lead to SIB that is unconstrained by environmental contingencies and is out of the individual's control, resulting in compensatory self-restraint (Oliver & Richards, 2015). However, once confounding variables and indicators of compulsivity were taken into account, impulsivity (alongside self-help scores and SIB severity) was not uniquely associated with self-restraint. This contrasts with a previous study showing that impulsivity and SIB predicted self-restraint two-fold in autistic children: Richards et al. (2017) recruited individuals with clinical autism diagnoses, while the present study found 64% of participants scored above the autism

threshold on an autism screening tool (SCQ). Therefore, differences may be explained by sample characteristics. Further, statistical approaches may account for distinct study findings, as confounding variables were accounted for in the present study, unlike previous research. To consider a theoretical implication, it may be that in the current heterogeneous sample of neurodevelopmental conditions, impulsivity was not a unique transdiagnostic risk marker for self-restraint. Importantly, given the correlation between impulsivity and self-restraint topographies, and the importance of impulse control for mental, physical and emotional health (Diamond, 2013), the impaired inhibitory control hypothesis is not refuted by these data. Instead, the link between impulsivity and self-restraint may not occur across the neurodevelopmental continuum. It is important to note that direct, performance-based measures of impulsivity may better tap into the EF construct more directly that informant-report measures (section 6.5.3). Thus, further research examining cognitive control using direct measures of EF in less heterogeneous samples is therefore needed.

Supporting the compulsivity account, compulsive behaviour and anxiety were associated with higher levels of self-restraint topographies. This replicated previous findings in individuals with ID (Hyman et al., 2002; Powell et al., 1996). Furthermore, this study examined anxiety-related behaviours using an informant-report tool designed for individuals with ID. This overcomes the challenge of assessing anxiety in populations who cannot verbalise internal states (35% of the current sample was minimally verbal). The present study found greater anxiety, pain and low mood were associated with self-restraint topography scores, replicating similar findings by Laverty et al. (2020) whereby autistic individuals who self-restrained were found to have lower mood, interest and pleasure than those who did not self-restrain. Altogether, these cross-sectional data are the first to link self-restraint with anxiety.

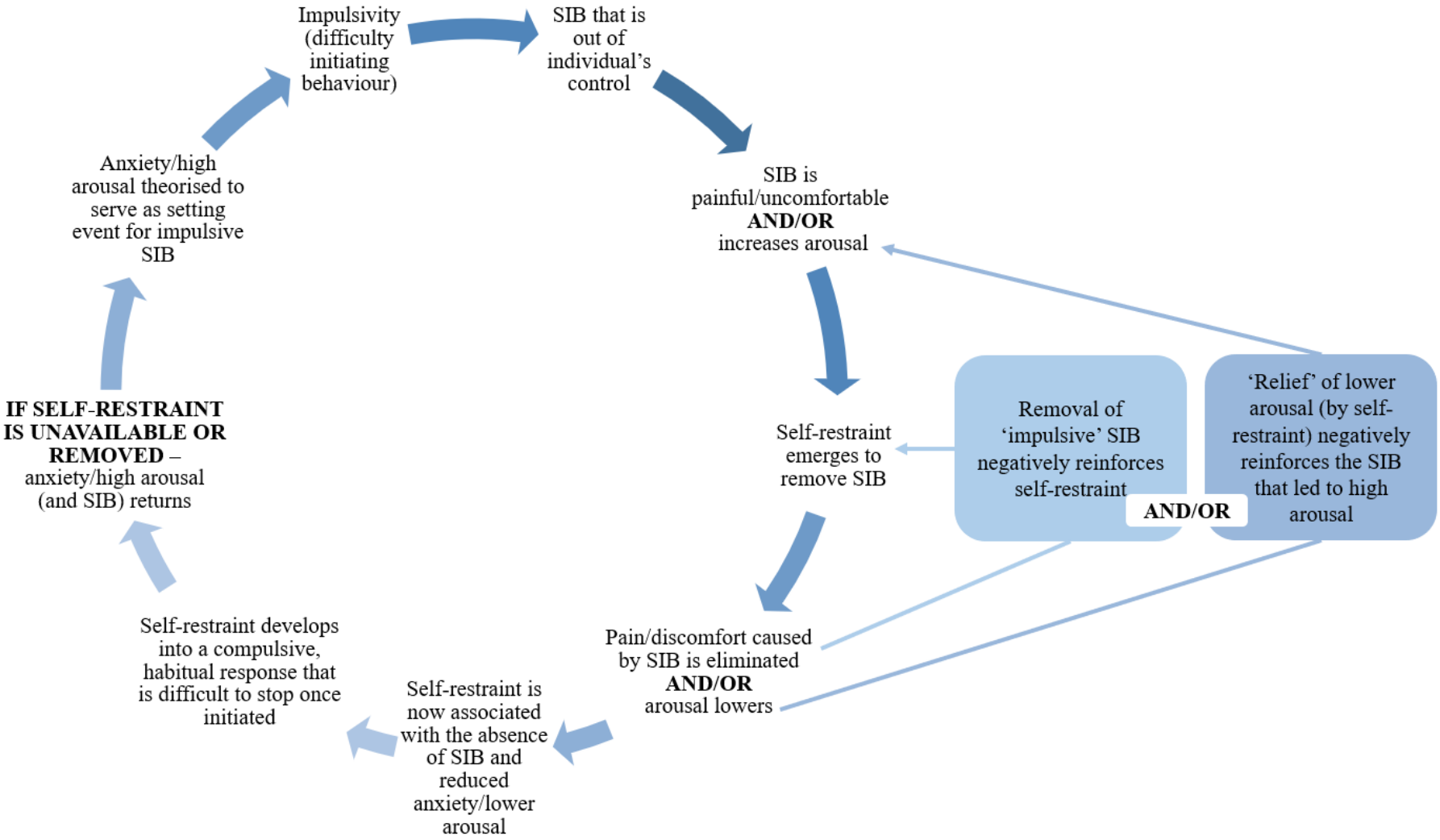
Lower age, higher compulsivity and higher anxiety scores were uniquely associated with self-restraint, once potential confounds were accounted for. This suggests that younger age, compulsivity and anxiety serve as important risk markers for self-restraint behaviour within neurogenetic conditions associated with autism and/or ID. The relation between younger age and self-restraint is perhaps unsurprising given the association between younger age and BoC (e.g., aggression r_s=-.20), and SIB severity with self-restraint. The unique contribution of compulsivity *and* anxiety, and the large effect correlation between compulsivity and anxiety, r_s=.40, supports the hypothesis that self-restraint is driven by a lack of behavioural control *and* high anxiety/arousal (King, 1993; Romanczyk et al., 1992; section 3.2). Compulsivity is considered the inability to stop repeating a behaviour (section 1.4.7.3), and anxiety/arousal refers to the apparent desperation to access the 'soothing' properties of self-restraint in those who cannot prevent the initiation of SIB. Thus, this study implicates compulsive behaviour and behavioural anxiety as important transdiagnostic factors of self-restraint across neurodevelopmental conditions, with implications for theoretical understanding.

The finding of compulsivity and behavioural anxiety predicting self-restraint serves as evidence in favour of a compulsive hypothesis of behaviour. Crucially, rather than competing with a purely operant model of self-restraint, whereby self-restraint is hypothesised to function to escape the aversive properties of painful SIB, the compulsive hypothesis *compliments* the operant account. As represented in Figure 3.1, the removal of SIB negatively reinforces self-restraint, and this may be in the context where SIB is perceived as painful, as suggested by the negative reinforcement account, *and/or* uncontrollable, as indicated by the compulsivity account. Whether self-restraint terminates painful SIB (negative reinforcement account), removes compulsive SIB (compulsivity account), and/or lowers arousal caused by

SIB (Romanczyk et al., 1992), it is perceived as soothing, relaxing and rewarding, leading to elevated anxiety/arousal upon self-restraint termination. It may therefore be implied that self-restraint emerges as an adaptive behaviour, however becomes concerning once established as a preferred behaviour (section 1.4.5.3). Future research directions include longitudinal examination of behavioural anxiety and/or psychobiological arousal in individuals who self-restrain and display environmentally- or automatically-reinforced SIB.

From these data, it is possible to propose (but not confirm) an adapted compulsivity account (original model hypothesised by King, 1993, is presented in Figure 3.1). An adapted model suggests that an individual may struggle to inhibit the initiation of self-injury (i.e., impulsive, not compulsive, SIB), causing anxiety. This may lead to the emergence of habitual (i.e., compulsive) self-restraint to reduce anxiety. This is akin to the Obsessive-Compulsive Disorder literature, which suggests that compulsions (e.g., repetitive hand washing; Gautam & Bhatia, 2015) occur to manage anxiety (which may be caused by obsessions; APA, 2013). This behavioural cycle is presented in Figure 3.3. Interestingly, Robbins et al. (2012) proposed that compulsions may occur in the absence of obsessions, with obsessions emerging as a "post hoc" attempt to rationalise unexplainable compulsions (p.85). This lends support for the adapted compulsivity account for individuals with severe/profound ID, where obsessions may not appear to drive compulsions, or may be challenging to assess. It is important to acknowledge that while compulsivity and anxiety were associated with self-restraint in the present study, the adapted compulsivity account was not fully tested and thus, cannot be confirmed. Therefore, future research should endeavour to test this potential pathway to self-injury and self-restraint, examining the perseverative components of compulsivity through direct assessments.

Figure 3. 3 An adapted compulsive account of self-injury and self-restraint



3.5.1 Limitations

Limitations of this study must be considered. First, many participants recruited into this study had participated in previous research (i.e., were identified through a database of participants known to the Cerebra Network), and/or were actively involved in syndrome-specific charities or support groups (which predominately advertised the study using social media). Therefore, results may be over-representative of individuals who actively seek out research and caregivers who care for individuals with greater support needs. Additionally, the study was more likely to reach individuals with access to the internet and familiarity with social media, potentially excluding a proportion of families who may not have such access. Thus, results of this study should not be interpreted as representative of all individuals with neurodevelopmental conditions. Despite this potential drawback, it is important to note that this is the first and largest cross-syndrome description of self-restraint, with findings broadly replicating those found by meta-analysis.

Second, while replicating previous studies and providing novel findings of the role of impulsivity and compulsivity in self-restraint in neurogenetic conditions, the indirect measures of behaviour may not capture EF directly (Snyder et al., 2015; section 6.2.3). Further research using direct measures of impulsivity and perseveration would inform explanatory theories of self-restraint and SIB.

Finally, while this study had sufficient statistical power to detect even modest effects (section 3.3.6), the size of each syndrome group was relatively small, precluding extensive cross-syndrome comparisons. For example, nine of the 23 syndromes recruited contained only one participant. While the focus of this study was not necessarily to classify self-restraint

phenomenology within genetic syndromes, rather to examine correlates and predictors of self-restraint along the neurodevelopmental continuum, findings and implications relating to syndrome-specific profiles of self-restraint are limited and should not be considered representative of all individuals with neurogenetic syndromes (which themselves are highly heterogeneous).

3.5.2 Conclusions

In conclusion, this cross-syndrome survey indicated that self-restraint occurred across a variety of neurodevelopmental conditions. A greater number of self-restraint topographies was linked with negative outcomes, including SIB severity, impulsivity and compulsivity, and anxiety and low mood. It is therefore suggested that the number of self-restraint topographies displayed by an individual (particularly if they have a genetic syndrome associated with multiple topographies) should be considered and monitored at the emergence of behaviour. The eight-item SRC is considered a quick and effective tool for gathering self-restraint topography data. Importantly, there were unique associations between compulsivity, anxiety and self-restraint within this heterogeneous sample of individuals with neurodevelopmental conditions, even when differences in age and ability were considered. This suggests that compulsivity and anxiety might be transdiagnostic risk markers for self-restraint within autistic individuals and/or those with ID. An adapted compulsivity account was proposed to account for these data. Future research directions include longitudinal examination of behavioural anxiety and/or psychobiological arousal in at-risk groups, and employing direct measures of impulsivity and compulsivity.

CHAPTER FOUR

METHODS AND PSYCHOMETRICS

4.1 Preface

Chapters 2 and 3 revealed a relatively high prevalence of self-restraint in populations at-risk of self-injury, including autistic individuals, individuals with intellectual disability and individuals with associated neurogenetic conditions. Self-injurious behaviour was a reliable correlate of self-restraint across Chapters, while the well-powered study in Chapter 3 found markers of impaired behavioural control, such as compulsivity, closely correlated with topographical self-restraint. Younger age, compulsivity and anxiety were suggested as potential transdiagnostic risk markers for self-restraint across the neurodevelopmental continuum. Importantly, hypotheses of behavioural control have only been examined using indirect informant-report measures, and thus far self-restraint has been measured only according to topography (i.e., form of behaviour). Therefore, this Chapter describes a variety of methods and assessment tools that will be used to measure self-restraint intensity (i.e., pervasiveness of behaviour). A novel battery of play-based assessments used to directly assess executive function is described. The psychometric suitability of self-restraint 'intensity' scores, and the developmental appropriateness, sensitivity and feasibility of the direct executive function measures is assessed, informing analyses of novel empirical data collected using these methods in Chapters 5 and 6.

4.2 Introduction

There are several approaches to measuring self-restraint, with different measures assessing different parameters (section 1.4.6). Self-restraint has been associated with self-injurious behaviour (SIB), with deficits in compulsivity and/or impulsivity purported to underpin behaviour (sections 1.4.7.3 and 3.5). Theoretical understanding, and by consequence implications for clinical practice, is limited by a lack of research measuring multiple parameters of self-restraint. Exclusion of autistic children with intellectual disability (ID) from research, a population at highest risk of SIB and maladaptive self-restraint, further limits understanding, especially as autism acts as a transdiagnostic correlate for self-restraint (section 3.4.2). Therefore, this Chapter describes the parameter-based approach to measure self-restraint, where behaviour is measured according to topography, intensity and/or preference for self-restraint. A protocol for administering direct (i.e., performance-based) and indirect measures (i.e., informant-report questionnaires) of executive function (EF) in this under-researched population is also described. The psychometric properties and suitability of measures within the population of interest were examined.

The systematic review and meta-analysis presented in Chapter 2 established that 39% of individuals studied in the existing self-restraint literature showed self-restraint, including autistic individuals and/or those with ID (section 2.4.3). However, examination of the participant characteristics in the included studies revealed a wide range of diagnostic labels used to describe samples (section 2.4.2), including “possible autism” (Forman, 2003), “autism-schizophrenia” (Bruhl et al., 1982) and “intellectual disability” (e.g., Rooker et al., 2020). Changes in terminology and diagnostic criteria over-time highlight the importance of defining the characteristics of the study sample. A recent meta-analysis revealed 38% of

autism studies fail to report information about ID severity (Russell et al., 2019). Poor characterisation of the study sample limits ability to generalise results to the full continuum of ID severity (i.e., mild to profound). Furthermore, a review of the quality of previous research found that standardised tests of autism and ID were rarely employed (section 2.4.2). Therefore, there is a need for researchers investigating self-restraint to document the diagnostic characteristics of participants, to improve the external validity of studies. This Chapter describes the assessments employed to measure autism and ID in the remaining empirical work.

A taxonomy of self-restraint measurement outlined in Chapter 1 (section 1.4.6) highlights the different parameters assessed by self-restraint measures. The most common tool, the Self-Restraint Checklist (SRC; Powell et al., 1996), captures one essential parameter of behaviour: topography. This informant-report[21] questionnaire efficiently describes phenomena within large samples, and described self-restraint in the large cross-syndrome study presented in Chapter 3. SRC-defined self-restraint has been associated with SIB within autistic (Laverty et al., 2020; Richards et al., 2017) and neurogenetic samples (section 3.4.1.2) and is predicted by elevated compulsivity and anxiety (section 3.4.2). Building upon the SRC, the Self-Restraint Questionnaire (SRQ; Oliver et al., 2003) measures *intensity*, the amount of time an individual spends engaging in self-restraint (section 1.4.5.2), which determines potentially maladaptive and pervasive self-restraint (section 3.4.1.2). No study has yet examined self-restraint intensity using the SRQ, thus associations with correlates such as SIB and compulsivity are unknown. Therefore, this Chapter outlines a novel scoring approach of the SRQ to identify intensity of self-restraint, allowing for examination of relations with putative correlates.

[21] For clarity, 'informant-report' refers to questionnaires completed by the primary caregiver.

The Self-Restraint Questionnaire 2 (SRQ2; Forman, 2003), an extension of the SRQ, adopts the most restrictive definition of self-restraint, capturing topographies that are intense, preferred and incompatible with SIB (section 1.4.6). This is arguably the most conclusive informant-report evidence of 'functional' self-restraint (Forman, 2003) over-and-above topography-only definitions (Oliver et al., 2003; section 1.4.4). Self-restraint that shares a maladaptive relation with SIB poses risks to physical health and adaptive functioning (section 1.4.2). This Chapter, therefore, describes the use of both the SRQ and SRQ2 to extend understanding of the properties of self-restraint. Additionally, the inter-rater reliability and internal consistency of the SRQ (Oliver et al., 2003) and SRQ2 (Forman, 2003) has thus far only been assessed in small samples known to self-injure. This Chapter examines the psychometric properties (i.e., internal consistency and factor structure) of the SRQ when used with autistic children with ID who may or may not show SIB.

Intensity of self-restraint can also be captured using observational methods. Meta-analytic investigations suggested informant-report and observational methods capture a similar construct (section 2.4.3.2), however approaches have not been directly compared within the same sample. It is therefore unclear whether questionnaire-based and observed self-restraint intensity are related, and correlate with putative risk markers. These data would reveal the potentially shared or distinct underlying construct between methods, with implications for assessing self-restraint in practice. Therefore, this Chapter describes a protocol for collecting video recordings of autistic children with ID to perform a naturalistic observational study of self-restraint, and compare informant-report and observed self-restraint intensity (Chapter 5). The recordings obtained through methods described in this Chapter served a second purpose: to test theoretical accounts of self-restraint (Chapter 6).

Self-restraint has been described as a compensatory behaviour that emerges in response to SIB (King, 1993; Oliver & Richards, 2015; sections 3.2 and 6.2.3) implicating compulsivity and impulsivity as important correlates of self-restraint. Empirical evidence has supported accounts: individuals who show both SIB and self-restraint display elevated compulsivity in comparison to those who show only one behaviour (e.g., Powell et al., 1996), while compulsivity and anxiety predicted self-restraint within a heterogeneous sample of individuals with neurogenetic conditions (section 3.4.2). Further, autistic individuals who self-restrain display poorer impulsivity than those who do not self-restrain (e.g., Laverty et al., 2020). However, findings have not been replicated within autistic individuals with ID, a high-risk population for SIB and self-restraint, and are reliant on indirect measures of EF (section 6.2.3). This Chapter therefore describes both indirect and direct measures of impulsivity to examine a hypothesis of impaired inhibitory control. Working memory and cognitive flexibility, two broader EF components, were also examined to provide an index of compulsivity, and examine the broader cognitive profiles of individuals who do and do not self-restrain.

Traditionally, EF tasks began as measures of adult frontal function. Over the past two decades, tasks have been adapted to study EF in children, infants and neurodiverse populations. Assessments of EF outlined in this Chapter build on this approach by adapting existing tasks to suit an under-studied population. In contrast to studies of EF in autism, there have been remarkably few investigations of EF among individuals with co-occurring autism and ID, despite recognition of unique impairment within this sample (McClain et al., 2022) and high co-occurrence of autism with ID (Tonnsen et al., 2016; section 1.4.7.3). This may be due to a lack of available tests suitable for heterogeneous ID populations. For example, many

EF tasks require non-EF skills for responding (Kristensen et al., 2022), such as verbal and/or motor skills (e.g., Khanna & Boland, 2010). Verbal and motor ability are highly variable within individuals with ID (Margolis et al., 2020; Vuijk et al., 2010), excluding many individuals from EF testing. This Chapter therefore measured EF in autistic individuals with ID using tasks designed to minimise verbal and motor demands, conferring a secondary advantage of generating data in which to examine the putative associations between EF, self-restraint and self-injury.

EF tasks designed for typically developing children and adults may be inappropriate for use with children with ID of the same chronological age, due to individual differences in instruction comprehension, motor development and language and communication skills (Fidler & Lanfranchi, 2022). A recent meta-analysis highlighted that individuals with ID (mean chronological age of 19 years) showed poorer EF than typically developing individuals matched on mental age (six years) across every EF component tested (Spaniol & Danielsson, 2022). This further highlights the need for unique assessments in this group. Assessments of EF described within this Chapter were therefore based on tasks developed and validated with typically developing infants, toddlers and pre-schoolers. A 'standardised' protocol of task adaptations was designed to suit a range of ID severities, including factoring in developmental age equivalents, and introducing reversal and stopping rules to adjust task difficulty. This Chapter sought to establish the developmental appropriateness (i.e., whether tasks suited children's developmental level, measured by observing the spread of scores around an expected value) and sensitivity (i.e., ability for measure to capture individual difference in the sample, measured by observing the range of scores around the mean; Petersen et al., 2016) of this novel battery of EF tasks in a large sample of autistic children with ID.

Perhaps reflecting a broader exclusion of children with ID from EF research (McClain et al., 2022), few questionnaire assessments have measured EF in autistic individuals with ID. Tsermentseli and colleagues (2018) administered The Behavior Rating Inventory of Executive Function–Teacher Report (BRIEF-TR; Gioia et al., 2000), while more recently McClain et al. (2022) utilised the pre-school version (BRIEF-P; Gioia et al., 2003). The BRIEF explores inhibition, shifting, working memory, planning and organisation, and emotional control. Indirect assessments of EF have revealed a profile of global executive dysfunction within autistic people with ID (Tsermentseli et al., 2018). McClain et al. (2022) extended this finding by comparing the EF performance of an autism and ID group to autism-only and ID-only groups. This study not only supported previous findings, revealing clinically significant EF difficulties across all core EF domains in the autism and ID group, but also revealed deficits unique to the cooccurring group. For example, the autism and ID group showed greater EF deficits than the autism-only group, and particular impairments in inhibitory control. Both Tsermentseli et al. (2018) and McClain et al. (2022) included 40 autistic individuals with ID, the largest sample sizes exploring EF in this population to date. However, whilst these studies provide valuable data revealing the unique EF profile of those with autism and ID, similar to the existing studies employing direct measures, both studies are limited by small samples. The present study therefore aims to replicate and extend unique findings of EF differences by recruiting the largest known sample of autistic children with ID. Additionally, there is suggestion that direct and indirect measures capture unique information about EF (Pérez-Pichardo et al., 2018; section 6.5.3), and therefore should be used in tandem to gain a comprehensive understanding of EF in populations with autism and ID. Therefore, this Chapter describes direct and indirect measures of EF.

In summary, a battery of tasks was used to assess both self-restraint and EF. Self-restraint was measured using a series of questionnaires and observational techniques. EF was measured using indirect and direct assessments. This Chapter describes the method for the studies reported in Chapters 5 and 6, and assesses two empirical aims: (1) examine the underlying factor structure of the SRQ and compare against existing subscales, and (2) conduct internal checks of the novel battery of play-based EF assessments to assess developmental appropriateness, sensitivity and feasibility.

4.3 Method

4.3.1 Design and development

This Chapter was nested within a larger MRC-funded study at the University of Birmingham called the 'Sleep-Impulsivity-Behaviour study', which aimed to examine mechanisms relating to SIB in autistic children with ID. The pre-registered protocol for the Sleep-Impulsivity-Behaviour study can be found on the Open Science Framework: https://osf.io/k5qpx, DOI:10.17605/OSF.IO/K5QPX.

Data collection (2021–2023) was undertaken remotely to eliminate risk of Covid-19 infection. Questionnaires were administered using an online survey platform (or on paper when requested). Existing, validated direct assessments of EF used with typically developing infants and pre-school children were adapted for administration via video conferencing. Tasks were presented to parents/caregivers[22] and children as games, and caregivers acted as 'co-researchers' by administering the tasks directly with their child at home under close supervision by the research team. This method was piloted with two typically developing 4-year-old males in April 2021. Piloting established the feasibility of methods and led to the refinement of written materials.

4.3.2 Recruitment and screening

Eligible participants for this study were identified through four routes: National Health Service (NHS) Trusts and services (*n*=111, 65%), social media or word of mouth (*n*=29, 17%), charities (*n*=22, 13%) and special educational needs schools (*n*=9, 5%). First, the study was advertised within special schools across England. For example, the study flyer was

[22] Henceforth 'caregivers', including biological, step, foster, adoptive and grandparents.

circulated on school websites and within newsletters. Second, healthcare professionals recruited families through 19 NHS Trusts across England. Third, charities such as Cerebra and local autism support groups aided advertisement. Finally, social media posts supported recruitment.

Upon expression of interest, caregivers confirmed eligibility[23] through an online screening questionnaire or through contact with a member of the research team. This screening stage confirmed that children were autistic, had an ID, did not have a known genetic syndrome and were aged between four and 15 years 9 months.

4.3.3 Participants

In total, 196 caregivers of autistic children with ID consented to participate. A total of 21 caregivers did not complete any study measures, and four participants were excluded due to ineligibility according autism scores below threshold on the Social Communication Questionnaire (SCQ; section 4.3.4.1.3). This left a total sample of 171 participants. Demographic information characterising the sample is reported in Table 4.1.

Covid-19 pandemic restrictions meant that direct assessments of autism using the Autism Diagnostic Observation Schedule, Second Edition (ADOS-2; Lord et al., 2012) were not completed as pre-registered. Caregivers instead provided a letter evidencing their child's autism diagnosis according to classification specified by the DSM-V (APA, 2013) or ICD-10 (World Health Organization, 1993). Alongside this, scores on the SCQ were compared against the clinical cut-off of 15, resulting in the exclusion of four participants. The mean

[23] Where possible, assent was sought directly from the child, with 'easy read information sheets' aiding study descriptions (Appendix 4.1).

SCQ score of the included sample, *M*=27.00, *SD*=5.17, was significantly higher than the clinical cut-off of 15, $t(170) = 30.345$, $p<.001$, 95%CI [11.22, 12.78]. ID was confirmed by informant-report both at the screening and questionnaire-collection stages. Vineland Adaptive Behavior Scale-Third Edition (VABS-III; section 4.3.4.1.4) Adaptive Behaviour Composite (ABC) scores also indicated the presence of ID (Table 4.1). Finally, during the EF session (section 4.3.5) it was requested that caregivers provide written evidence of autism and ID using a letter provided by healthcare professionals, such as results from diagnostic assessments[24] and/or diagnoses recognised within an Education, Health and Care Plan or similar. Out of the 126 children tested, 90 caregivers provided this additional confirmation of diagnosis (71%).

[24] Many children had not received formal IQ assessments clinically, and it was beyond the scope of this study to do this remotely.

Table 4. 1 Demographic characteristics of full study sample

Developmental variables	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Chronological age (years)	171		10.14 (10.19)	4.02 – 15.49 (2.86)
Sex (female/male/non-binary)	171	41[a]/129/1 (24/75/1)		
Mobility (walk unaided/ unable)	170	162/8 (95/5)		
SCQ total (all participants)	171		27.00 (28.00)	15 – 37 (5.17)
SCQ total (verbal [b])		105 (61)	27.24 (28.00)	15 – 37 (5.96)
SCQ total (non-verbal)		66 (39)	26.62 (27.00)	20 – 32 (3.58)
Autism description	169			
Autism		83 (49)		
Autism Spectrum Disorder		76 (45)		
High Functioning Autism		4 (2)		
Autistic Spectrum		3 (2)		
Asperger Syndrome		2 (1)		
Pervasive Dev. Disorder		1 (1)		
VABS-III ABC score	144		42.43 (40.00)	20 – 87 (14.59)
VABS-III receptive language age equivalent (months)	144		19.72 (19.00)	0 – 67 (10.16)
ID severity	169			
Mild		14 (8)		
Moderate		50 (30)		
Severe		71 (42)		
Profound		7 (4)		
Other [c]		5 (3)		
Unknown		22 (13)		
BPVS raw score [d]	70		70.67 (69.00)	2 – 146 (39.22)
BPVS standardised score [e]	70		74.14 (69.00)	69 – 118 (10.79)
BPVS age equivalent (months) [f]	41		84.51 (74.00)	45 – 162 (28.62)

Note. SCQ = Social Communication Questionnaire; VABS-III ABC = Vineland Adaptive Behavior Scales-Third Edition Adaptive Behaviour Composite scores; BPVS = British Picture Vocabulary Scale.

[a] *n*=1 self-described differently to gender assigned at birth. [b] 'Verbal' referred to ability to talk using short phrases or sentences. [c] Other = "estimated mild", "mild to moderate", "moderate-severe", "delay of around 2.5 years" and "not sure". [d] BPVS discontinued for *n*=56 and not attempted for *n*=10 due to child engagement or for safety. [e] Includes *n*=50 that scored lower than the average of a normed sample (70), indicated as '69'. [f] Age equivalents incalculable for *n*=29 as raw scores fell below normed sample scores.

4.3.4 Measures

4.3.4.1 Child characteristics and adaptive functioning

4.3.4.1.1 Background Questionnaire.

Demographic information about the child was collected, including sex, age, diagnoses and mobility (Appendix 4.2).

4.3.4.1.2 The British Picture Vocabulary Scales (BPVS-III; Dunn & Dunn, 2009).

The BPVS-III was administered with children via video conferencing to obtain an estimate of receptive language ability. The BPVS consists of 14 sets of 12 items and does not require expressive language. Norms identified from 3278 children aged 3-16 years with and without ID revealed excellent[25] internal reliability (α=.91, Dunn & Dunn, 2009). In the present study, BPVS items were displayed using a digital visualiser, allowing the child to see both the assessment items and the researcher. Children were oriented to the pictures displayed, labelled one to four. The researcher called out a target word (e.g., *"ball"*) and asked the child to select the picture that best represents the word's meaning. Responding to this task varied according to child preference and ability. Children were told they could point to the picture on their screen and their caregivers would state the number corresponding to their choice. Children could also verbally respond with the corresponding number or hold up the correct number of fingers for their chosen answer. According to the standardised instructions and scoring, a basal set was established when a child scored one or zero errors within a set. A ceiling was established when eight or more errors occurred within a set. A raw score was generated by

[25] According to general consensus within the literature, suggesting α values of .70 or greater are desirable (Taber, 2018).

subtracting the number of errors within each set from the highest item in the ceiling set. Standardised scores were calculated where possible.

4.3.4.1.3 The Social Communication Questionnaire (SCQ; Rutter et al., 2003).

The SCQ is an informant-report measure of autistic traits and was used to assess presence of autism according to an established clinical cut-off (15 or above; section 4.3.3). Participants who scored below this threshold were excluded. See section 3.3.4.1.1 for full SCQ description. Within the present sample, a reliability coefficient of ω=.80 suggested strong scale reliability[26].

4.3.4.1.4 The Vineland Adaptive Behavior Scales-Third Edition (VABS-III; Sparrow et al., 2016).

The VABS-III (Appendix 4.3) was used to assess adaptive functioning. This semi-structured interview was administered with the caregiver, and covered four domains: communication, daily living skills, socialisation, and motor skills. Interviewers determined a score of zero (*never* or *no*), one (*sometimes*) or two (*usually*, *often* or *yes*) across items in accordance with the manualised instructions. Subdomain items were administered until a basal (i.e., four consecutive twos) and ceiling (i.e., four consecutive zeros) were identified. Day-to-day ability is assessed, rather than abilities on 'good' or 'bad' days. Standardised scores across domains were generated, alongside an overall Adaptive Behaviour Composite (ABC) score. This assessment demonstrated excellent internal consistency across domains, ranging from α=.94-.99 when used to evaluate children with ID, autism, speech and language impairments and other disabilities (Pepperdine & McCrimmon, 2018). Within the present sample, domain

[26] See section 3.3.6 for interpretation of Omega.

reliability coefficients ranged from acceptable to strong: communication ω=.74, daily living skills ω=.78, socialisation ω=.83 and motor skills ω=.72.

4.3.4.2 Behaviour

4.3.4.2.1 Challenging Behaviour Questionnaire (CBQ; Hyman, Oliver & Hall, 2002).

The CBQ is an informant-report measure of the presence and severity of behaviours of concern (BoC) observed in the previous month. See section 3.3.4.2.1 for description and scoring. Within the present sample, there was acceptable internal consistency for SIB, aggression and property destruction subscales, ω=.77, .76, .78 respectively. Stereotypy internal consistency was below acceptable, ω=.53.

4.3.4.2.2 The Self-Restraint Questionnaire (SRQ; Oliver et al., 2003).

This 25-item questionnaire (Appendix 4.4) captures the prevalence and intensity of 23 named self-restraint topographies, such as sitting on hands and sitting/lying in a position that restricts arm movement. Pictorial cues illustrating behaviours aids informants, who indicated the frequency of behaviours on a scale from zero to four (zero = 'never', one = 'some', two = 'half', three = 'most' and four = 'all of the time'). Item 24 was a free-text box asking informants to further describe any behaviours the child was reported to show 'most of the time' or 'always'. The final item provided a free-text box for informants to list any 'other' possible self-restraint behaviours. Total 'SRQ intensity' scores were generated by summing items 1–23 (maximum score = 92). Higher scores indicate greater intensity. Internal consistency across all scores was strong, ω=.88.

The SRQ also generates four subscales: use of clothing/materials to restrict movement (sum items one to nine); use of own body (items 12, 13, 15 to 21); use of objects (items 10, 11 and 14), and preference for imposed restraint (items 22 and 23). Within a sample of individuals with developmental disabilities, Oliver et al. (2003) reported variable internal consistency across subscales, α=.87, .17, .72 and .61 respectively (Table 4.7 reports subscale internal consistency within the present sample), and high interobserver agreement (85%) between direct observations of self-restraint and definitions of self-restraint described by the SRQ (see Chapter 5 for comparisons within the present sample).

4.3.4.2.3 The Self-Restraint Questionnaire 2 (SRQ2; Forman, 2003).

The SRQ2 (Appendix 4.5) is a brief follow-up measure to the SRQ, completed by caregivers who indicated their child showed at least one topography of self-restraint at least 'some of the time' according to the SRQ. This questionnaire builds upon topography and intensity data collected by the SRQ to identify a third parameter; preference for behaviour (i.e., does the individual show distress or anxiety upon termination of self-restraint?). The SRQ2 also defines self-restraint as self-initiated (i.e., individual engages in behaviour of their own accord, without being told or asked to) and incompatible with self-injury/stimulus control of behaviour[27] (i.e., SIB is not possible when self-restraint occurs, and/or an object/item appear to exert behavioural control despite non-restrictive properties). Caregivers responded to a five-point Likert scale, with higher scores suggesting stronger evidence for meeting each parameter. Forman (2003) assessed SRQ2 inter-rater reliability as rated by two informants and reported intra-class correlations (ICC) ranging from .57 to .89 across the six items.

[27] The parameters of incompatibility with SIB and stimulus control were grouped together to account for instances where an individual uses a device to control behaviour, even if the device does not physically restrict movement (section 1.4.5.1).

Participants were categorised into 'self-restraint' or 'no self-restraint' groups. Children who scored 'never' across all SRQ items were automatically grouped as 'no self-restraint'. Forman (2003) generated a scoring procedure for the SRQ2 in children known to self-injure, based on scoring above a threshold on items relating to each parameter. Children who met criteria for all three parameters and definitions were grouped as 'self-restraint', replicating Forman (2003)'s scoring method for self-injurers. Inter-rater reliability for this scoring method from a sample of individuals with learning disabilities was reported by Forman (2003): Cohen's kappa (κ) for self-initiation κ=.78, incompatibility/stimulus control κ=.67, and preference κ=.93. In the present study, the SRQ2 was completed for children with *and without* self-injury (i.e., monthly SIB according to the CBQ), resulting in two scoring procedures. For children who did not self-injure, items one and two were not displayed (as they related to SIB). Thus, the 'incompatibility/stimulus control' parameter was not applicable. Children therefore were required to meet 'self-initiation' and 'preference for behaviour' parameters to be grouped as 'self-restraint'. Table 4.2 displays both scoring methods, alongside internal consistency statistics for each eligible subscale. Within the present sample, internal consistency of all SRQ2 items was excellent, ω=.98.

Table 4. 2 *SRQ2 scoring*

Item	Parameter/ descriptor of self-restraint	Scores required to meet parameter criteria		Internal consistency ω
		Child shows SIB	Child does not show SIB	
1 2	Incompatibility with SIB/ stimulus control	3 or above on item 1 *or* 2	*Items 1 and 2 not displayed*	.68
4	Self-initiation of behaviour	2 or above	2 or above	N/A
3 5	Preference for behaviour	3 or above on item 3 *or* 5	3 or above on item 3 *or* 5	.40
6	N/A[a]			

Note. [a] Item 6 of SRQ2 intended to measure whether the child could physically terminate self-restraint without another's help. This item was deemed 'invalid' by Forman (2003) due to variable interpretation by informants (e.g., caregivers considered item according to child motivation rather than physical incompatibility).

4.3.4.3 Executive function

4.3.4.3.1 The Activity Questionnaire (TAQ; Burbidge & Oliver, 2008).

The TAQ is an indirect measure of impulsivity. See section 3.3.4.2.4 for description and scoring. Within the present sample subscale reliability was strong, ω=.89.

4.3.4.3.2 The Childhood Executive Functioning Inventory (CHEXI; Thorell & Nyberg, 2008).

This 26-item questionnaire (Appendix 4.6) captured individual differences in EF. Subscales of working memory and planning are summed to generate a 'working memory factor score', and regulation and inhibition subscales combine to generate an 'inhibition factor score' (Thorell & Nyberg, 2008). Informants rated how 'true' statements were for their child using a Likert scale where one indicates 'definitely not true' and five 'definitely true'. This measure

has shown adequate internal consistency in a sample of 408 typically developing school children (α=.94; Trevisan et al., 2017), and good test-retest reliability, *r*=.89 when used with a sample of 28 neurotypical children (Thorell & Nyberg, 2008). Internal consistency across subscales was good (α=.85 and α=.89, respectively), and high test-retest reliability across subscales (*r*=.74 and *r*=.87, respectively) have been reported in neurotypical children aged 8 to 11 years (Catale et al., 2015). In the present sample subscale reliability was strong across the working memory factor, ω=.88, and inhibition factor, ω=.85.

4.3.4.3.3 Prohibition task

Inhibitory control was directly measured by two behavioural prohibition tasks (Devine et al., 2019; Friedman et al., 2011). Caregivers revealed an attractive magic wand toy hidden inside an envelope and engaged their child's attention by using verbal cues (*'Oh, look at this!'*) and physical prompts (e.g., twisting the wand upside down). Once engaged, the researcher asked the caregiver to put the wand down in front of the child, say *'Look, no don't touch'* and wag their index finger to emphasise 'no'. The caregiver was then asked to face away from their child and not engage with them for 30 seconds. For safety, caregivers were instructed to look away 'as much as they felt comfortable', such that children could remain within their vision while not engaging. Researchers ended the task either when the child touched the wand or after 30 seconds of waiting.

Dependent variables extracted from this trial were whether children resisted the urge to touch the toy for the full 30 seconds (pass/fail), and latency to touch the wand (seconds/milliseconds). Inter-rater reliability was established by two researchers

independently coding 37 recordings (29% of full sample). There was excellent[28] agreement on latency to touch the toys, ICC=.97, $p<.001$, and waiting outcome (0 = fail, 1 = pass), $\kappa=.95$, $p<.001$.

A second prohibition task (Devine et al., 2019; Friedman et al., 2011) was administered within the same session (Appendix 4.7, page 3 displays task order). Children were presented with an array of three toys revealed in a standardised order (cuddly toy > toy car > squeezy ball). Caregiver held all toys until the researcher was satisfied the child was engaged. This also prevented premature touching of stimuli. Caregivers then placed all three toys down and delivered the same 'don't touch' instruction as in the first prohibition task. After the caregiver had turned away, a waiting period of up to 60 seconds was initiated. This task was more challenging than the first prohibition task due to the greater number of attractive stimuli and longer 'waiting' period.

Dependent variables extracted from this task were the same as the wand task. An overall prohibition composite score was calculated by adding the number of passed trials from the wand and array prohibition tasks and dividing by two. For example, if a child passed the wand task then failed the array task (or vice versa), their prohibition composite score would be 0.5. Inter-rater reliability from 36 recordings (29%) revealed excellent agreement on waiting duration, ICC=.98, $p<.001$, and almost perfect agreement for pass/fail performance, $\kappa=.94$.

[28] See section 2.3.3 for κ interpretation.

4.3.4.3.4 Multilocation search

This task was used to assess working memory and provide an index of compulsivity. The protocol was developed by Devine et al. (2019) based on an earlier task by Miller and Marcovitch (2015). Children were required to find five stickers hidden under five cups following a five second delay. Children's attention was engaged, then caregivers hid coloured stickers under correspondingly coloured opaque cups, varying in size. A piece of white card was used to hide the cups from the child's view, as the caregiver counted down from five out loud. The card was then removed, and child asked '*Can you find a sticker?'* Children could point towards, pick up or verbalise which cup they would like to search under. The sticker found in the first-chosen cup was removed and hidden, and the child praised. The child was reminded that cup was now empty. The card was again used to obscure view of the cups, a five second delay initiated, and the card removed. This time, caregivers asked '*Can you find another sticker?'* If a child searched the empty cup (i.e., the sticker was retrieved in the previous search), caregivers said *'Oh no, this cup is empty. Let's try again',* showing the child the empty cup. The task was then re-set by covering the cups and beginning the countdown. Alternatively, children were congratulated for successful retrievals. Steps were repeated until either all five stickers had been retrieved, or until three consecutive errors were made (i.e., the child searched three empty cups in a row).

The order of cups searched, number of searches, and number of stickers retrieved were recorded. Two composite scores were calculated from this task. Working memory efficiency composite scores, calculated by dividing the total number of stickers retrieved by the total number of searches, ranged from zero to one, where one indicates 'perfect' efficiency (i.e., a sticker was retrieved upon every search). Perseveration composite scores, which provide an

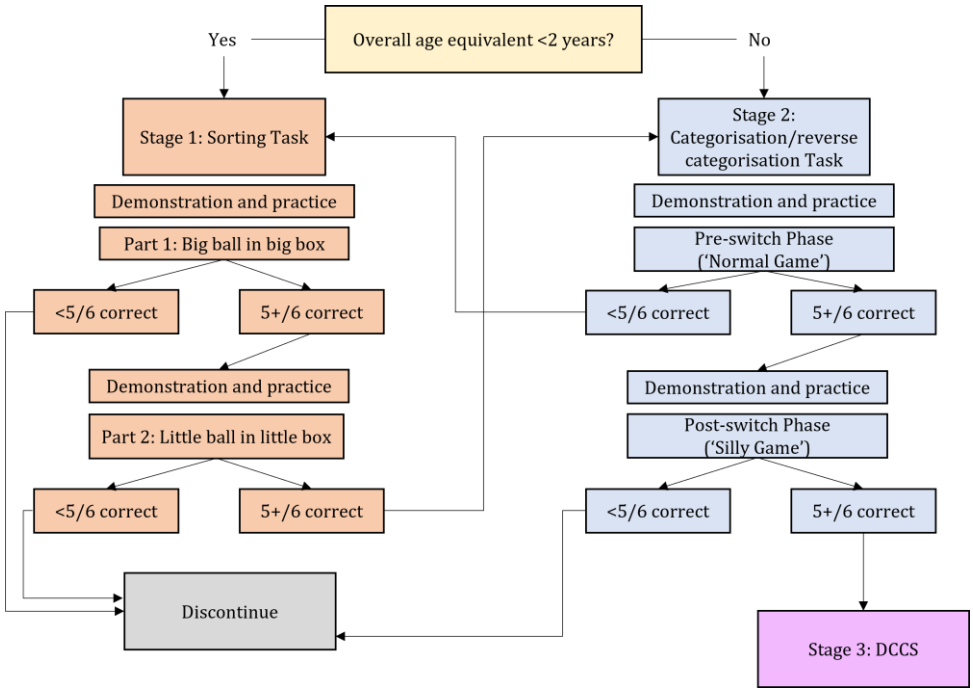
index of compulsivity, were calculated by summing the total number of perseverative errors (i.e., when child immediately repeats a search at an empty location) and dividing by the total number of searches. Perseveration scores ranged from zero to one, where zero indicated no perseveration (i.e., child did not repeatedly search an empty cup) and thus better performance (i.e., lower compulsivity). Double-coding of 35 recordings (28%) revealed excellent inter-rater agreement for number of stickers retrieved, ICC=.97, $p< .001$, and good agreement on number of searches, ICC=.81, $p<.001$.

4.3.4.3.5 Cognitive flexibility conflict scale

4.3.4.3.5.1 Protocol

Beck and colleagues' (2011) conflict scales were adapted for the current study to assess cognitive flexibility. Tasks increased in complexity and demand to capture variations in ability across the sample. Figure 4.1 demonstrates the cognitive flexibility task flow. Starting points were determined using the child's estimated overall adaptive age equivalent calculated by the VABS-III. Children who scored an estimated age equivalent of under two years started with the first simple sorting task (Devine et al., 2019). Children with an age equivalent of two or more years started at the more complex categorisation task (Carlson et al., 2004). If children started at and passed the simple sorting task, they moved onto the categorisation task. When children started at and failed the first stage of the categorisation task, the (less complex) simple sorting task was presented. Children who passed the categorisation task proceeded onto the Dimensional Change Card Sort (DCCS; Zelazo, 2006).

Figure 4. 1 *Cognitive flexibility protocol*



Each cognitive flexibility task had two phases: a pre-switch learning phase, and a post-switch demand phase. The pre-switch phase was considered less demanding on EF than the post-switch phase. A cognitive flexibility 'conflict' score was calculated for each participant by summing the number of *post*-switch phases the child passed. 'No-conflict' scores were generated by summing the number of passed *pre*-switch phases. Table 4.3 describes scoring procedure. Scores ranged from zero to three, with higher scores indicating better performance.

Table 4. 3 *Calculation of cognitive flexibility composite scores*

Score		**Description of scoring**
Conflict	0	Failed post-switch phase of sorting task
	1	Passed post-switch phase of sorting task
	2	Passed post-switch phase of categorisation task (even if sorting task not administered)
	3	Passed post-switch phase of DCCS
No-conflict	0	Failed pre-switch phase of sorting task
	1	Passed pre-switch phase of sorting task
	2	Passed pre-switch phase of categorisation task (even if sorting task not administered)
	3	Passed pre-switch phase of DCCS

4.3.4.3.5.2 Sorting task

Based on Devine et al.'s (2019) "Ball Run" task, this simple sorting task was shown to children with estimated age equivalents of less than two years. Children were presented with two target locations (one big box and one little box) with the lids off and placed shoulder-width apart (around 30cm). Caregivers were told the objective of this game was to see if children could sort an object (a ball) according to its size, with the task consisting of two phases. In the pre-switch phase, caregivers demonstrated placing a big ball in the big box, then said *'Now you try'*. The big ball was handed to the child centrally in-between the two target boxes, and the child's response was praised if correct or corrected if wrong. This task could be adapted such that children could point towards their chosen box or roll the ball towards it. The caregiver then handed the child the same big ball six further times without providing any feedback on performance, instead saying *'Let's try another'*. Thus, in this first phase, children had one stimulus object that remained constant across six trials (i.e., a large ball) and two possible target locations (i.e., a small box and a large box) for which to sort the

ball according to a simple learned rule. Children who correctly sorted the ball five or six times moved onto the second phase. In this post-switch phase, children needed to reverse the rule and place the little ball in the little box. This phase involved an executive demand because children had to override the simple rule learned in the first phase by ignoring the big box. Children who passed the second phase, by sorting the small ball correctly five or six times, moved onto the more complex categorisation task. The dependent variables were the total number of pre- and post-switch phase passed.

4.3.4.3.5.3 Categorisation/reverse categorisation (Carlson et al., 2004)

Children with an estimated age equivalent of two or more years, or who passed both phases of the sorting task started with this task. Two boxes were placed in front of the child around shoulder-width apart, with the lids on. Caregivers were told this was a 'sorting game' with two phases. In the pre-switch phase, caregivers taught a rule involving two stimuli (balls) varying on one dimension (size), and two targets (boxes) varying on one dimension (size). Caregivers demonstrated that the big ball goes in the big box and the little balls go in the little box. Before testing, children practiced sorting two big and two small balls with feedback (i.e., praise or correction). Balls were passed centrally to avoid cueing responses, and children could point to their chosen box if preferred. Lifting the lids required fine and gross motor skills, potentially disadvantaging children with poorer mobility, so a 'choice' was sufficiently made if the child clearly gestured towards a box. Six more balls were then presented to the child in a fixed, quasi-random order (little, big, little, big, big, little) and caregivers did not give any feedback. Rules were repeated as needed to reduce working memory load. Children who correctly sorted five or six balls proceeded to the categorisation phase, or the "Silly Game". In this post-switch phase, children's EF was challenged by caregivers teaching the

reversal rule (big balls > little box, small balls > big box). The practice and trial stages from the first phase were repeated with this new rule. Children who passed the categorisation phase by correctly sorting five or six balls later completed the most complex set-shifting task, the DCCS.

The dependent variables extracted from this scale were the score achieved in the pre- and post-switch phases. Double-coding of 27 recordings (21%) featuring the sorting and categorisation tasks revealed excellent inter-rater reliability across total number of correct trials, ICC=.99, p<.001.

4.3.4.3.5.4 Dimension Change Card Sort (DCCS; Zelazo, 2006)

The DCCS, the most complex set-shifting task, was administered to participants who passed the categorisation task. The DCCS was the most complex task because children were presented with two stimuli that varied on two dimensions (i.e., colour and shape), and two targets that varied on two dimensions (i.e., colour and shape). The DCCS was administered using PowerPoints slides and a script (Appendix 4.7, pages 21-22). Similar versions of this conflict task have been administered using tablets with typically developing children (Distefano et al., 2021). Based on the procedure outlined by Zelazo (2006) children attended to stimuli cards showing either a blue rabbit or a red boat, and categorised cards into one of the two target stimuli (a red rabbit or blue boat) according to a dimensional rule (e.g., colour). This pre-switch phases involved a demonstration and practice with feedback. Children were oriented to two target locations to 'sort' the cards, indicated by pointing or verbal choosing. Target cards differed from the sorting cards such that the rabbit was red and boat was blue, creating visual conflict in the learning phase. Caregivers or children called out location

number ‘one’ or ‘two’. Six target cards were then administered in a fixed order with no feedback. Children who passed this pre-switch phase (i.e., five or six correct trials) proceeded to the post-switch phase, where they sorted the same target cards according to a new dimensional rule (e.g., shape). Children did not receive a practice trial and feedback was withheld. Passing the post-switch phase required sorting five or six cards correctly. During both phases, children were reminded of the rule every time to reduce working memory load.

The total number of correct trials in each phase was recorded, and pass/fail was awarded for each pre- and post-switch phase. The dependent variable extracted was the number of phases passed. Inter-rater reliability of coding 21 recordings (32% of eligible participants) revealed near perfect agreement on total correct trials, ICC=.97, $p<.001$.

4.3.4.4 Valid administration

A coding scheme was developed to assess the validity EF task administration (Appendix 4.8). Following the session, video recordings were reviewed to extract the dependent variables and assess the validity of each task according to a checklist. Checks were developed to assess child engagement with tasks and interest in materials, standardisation of assessments, accuracy of task delivery by both the researcher and caregiver, and interference from primary or additional caregivers. Each task was assessed for its validity, and each validity check was marked as ‘pass’ (a score of one) or ‘fail’ (a score of zero). Scores were summed to provide an overall validity score, with each task receiving a final score between zero and eight. Higher scores reflected greater validity. Tasks with cumulative errors (i.e., validity scores of ≤ 3) were excluded, as over 50% of the validity checks for that task had ‘failed’, suggesting deviation from the standardised protocol designed to measure the EF component. Tasks with scores of four or above were removed from the data set where overarching issues were observed. For

example, if a child did not look at the prohibition task material(s) during the demonstration, and therefore the executive demand to inhibit touching the toy(s) was not initiated (i.e., failed validity check two), this trial would be excluded even if the overall validity score was above the cut-off of three. Two researchers independently assessed validity of task administration for 26 recordings (21%). Inter-rater reliability was observed using ICC. Total validity scores were compared, revealing good to excellent agreement across all direct assessments of EF, ranging from .73 to .99 (Table 4.4).

Table 4. 4 Inter-rater reliability of validity check procedure

EF component	**Task**	**Intraclass correlation**
Prohibition	Wand	.73
	Array	.95
Working memory	Multilocation search	.98
	Sorting (conflict 1)	.98
Cognitive flexibility	Categorisation (conflict 2)	.99
	DCCS (conflict 3)	.99

4.3.5 Procedure

Following consent (Appendix 4.9), caregivers were emailed a 'General Information' sheet (Appendix 4.10) providing a brief overview of the remote EF testing. The VABS-III interview was then completed over a video conference call. Researchers administered direct EF measures using standardised instructions. A bespoke pre-visit checklist and risk assessment (Appendix 4.11) was completed after the VABS-III, in advance of the EF testing (or "Games Session"). Caregivers were posted a laminated booklet with instructions to support administration (Appendix 4.12), and followed instructions spoken by researchers using wireless Bluetooth earphones. Caregivers were asked to not show their child the toys

until revealed during the Games Session.

At each session, two or more trained members of the research team, including at least one senior member of the team holding an undergraduate degree qualification or higher, administered and live-scored the EF tasks. Sessions were recorded. Participants were at home, in a quiet room with minimal disturbance, with at least one caregiver. All tasks except the BPVS and DCCS were administered by caregivers as researchers gave standardised instructions via the headphones that only the caregiver could hear. Researchers live-scored tasks to aid administration and informally assess reliability within the team (Appendix 4.13).

VABS-III and direct EF data were collected between 15th June 2021 and 26th January 2023. An average of 20 days elapsed between the VABS-III and EF session, ranging from four to 94 days. Best efforts were made to minimise time in-between sessions, with postponements mostly due to child illness. The VABS-III was completed across two sessions by 21 caregivers, while 11 families completed the EF assessments across two separate dates; all other VABS-III interviews and EF assessments were completed in a single session. EF visits lasted an average of 52 minutes, ranging from just under three minutes up to one hour 40 minutes (section 5.4.1).

Between tasks, children were rewarded with praise, playtime and an interactive 'junior scientist' certificate. Families kept all toys. Caregivers received a feedback report detailing descriptive and quantitative summaries of performance across all measures, informant-report and performance-based (Appendix 4.14). Health Research England and Bristol Research

Ethics Committee provided ethical approval for this study (IRAS Project ID 263048) (Appendix 4.15). Local R&D approval was obtained at all participating NHS sites.

4.3.6 Data analysis

To examine the factor structure of the SRQ, an Exploratory Factor Analysis (EFA) was conducted using Jamovi (v.2.2.5; jamovi, 2021; R Core Team, 2021). EFA seeks to identify underlying latent factors to describe the correlations between questionnaire items (Taherdoost et al., 2022). The sample size of 165 participants and at least 14 observations per item (i.e., above the recommendation of 10; Appendix 4.16 displays SRQ item distribution) indicated sufficient power for EFA (Costello & Osborne, 2005). A Shapiro-Wilk test revealed the data significantly deviated from a normal distribution, $p<.001$ across all items, indicating appropriateness of the principle axis factoring extraction method (Costello & Osborne, 2005). Bartlett's Test, the statistical probability that the correlation matrix has significant correlations among components, was significant, $\chi^2=1470$, $p<.001$, indicating the suitability of the data for EFA. The Kaiser-Meyer-Olkin (KMO) test was also performed, yielding an overall value of .813. KMO values above .80 are considered appropriate for EFA (Kaiser, 1974).

The number of factors was determined by parallel analysis (Horn, 1965), which uses both a random data simulation (i.e., artificial data generated by the Monte Carlo Simulation Technique) and the actual dataset. Eigenvalues are calculated for both datasets and compared. The number of eigenvalues generated by the artificial dataset that is significantly greater than that generated by the real dataset is determined to be the final number of factors (Çokluk & Kocak, 2016). Finally, a direct oblimin rotation method was used. This oblique rotation approach allows for inter-factor correlations (Zygmont & Smith, 2014) and is believed to produce more accurate solutions when used in social science (Costello & Osborne, 2005).

To establish the developmental appropriateness and sensitivity of the play-based EF assessments, task adherence and child performance was examined.

4.4 Results

4.4.1 Exploratory factor analysis of SRQ

Table 4.5 displays behavioural characteristics of the study sample, including self-restraint scores. To examine the underlying factor structure of the SRQ, an EFA was conducted and results compared against existing subscales reported by Oliver and colleagues (2003). Parallel analysis revealed a five-factor structure (Figure 4.2). Table 4.6 displays the results from the EFA. Factors explained 46.7% of the variance. All items loaded above the recommended 0.32 (Tabachnick & Fidell, 2001), suggesting suitability for the assigned factors. Items 8 and 18 cross-loaded across factors, and so were assigned to the factor they most strongly fit according to factor loadings. Factor 3 contained <3 items, resulting in a weak and unstable factor (Costello & Osborne, 2005). Items 4 and 5 were not assigned to a factor.

Table 4. 5 Behavioural characteristics of full study sample

Behavioural variables	**Possible score range**	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Self-restraint - SRQ		165			
Monthly self-restraint/no SR			158/7 (96/4)		
Total SRQ intensity score	0 – 92			16.20 (14)	0 – 84 (12.15)
Number of topographies	0 – 23			9.33 (9)	0 – 23 (5.49)
1 topography/2+ topographies			6/152 (4/96)		
Behaviours of concern – CBQ					
Monthly SIB/no current SIB		171	129/42 (75/25)		
Total SIB severity score	0 – 14			6.20 (7)	0 – 14 (4.38)
Monthly AGG/no current AGG		171	128/43 (75/25)		
Total AGG severity score	0 – 14			5.92 (6)	0 – 14 (4.21)
Monthly DoP/no current DoP		171	126/45 (74/26)		
Total DoP severity score	0 – 14			5.80 (7)	0 – 14 (4.24)
Monthly STPY/no current STPY		162	135/27 (83/17)		
Total STPY severity score	0 – 14			7.07 (8)	0 – 14 (3.97)

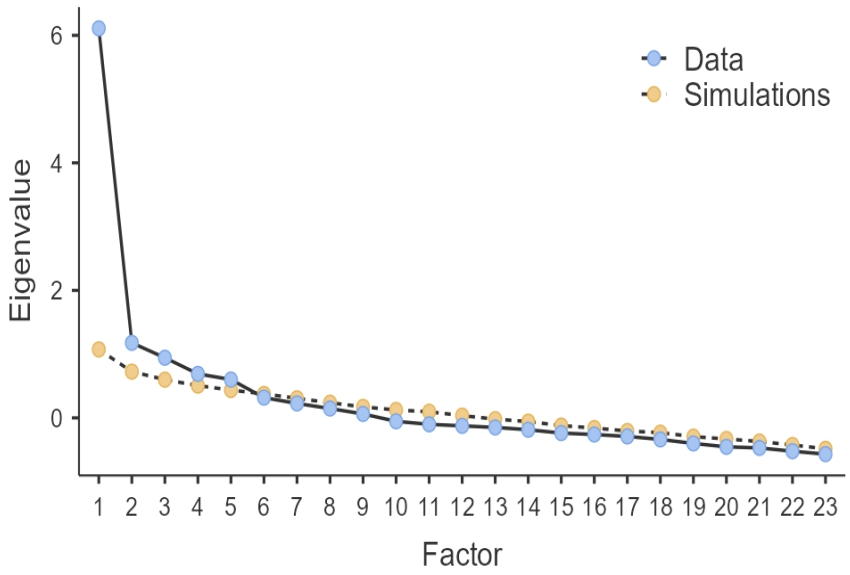
Note. SRQ = Self-Restraint Questionnaire; SR = self-restraint; CBQ = Challenging Behaviour Questionnaire; SIB = self-injurious behaviour; AGG = aggression; DoP = destruction of property; STPY = stereotypy.

Table 4.5 cont.

Behavioural variables	**Possible score range**	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Activity – TAQ					
Impulsivity subscale	0 – 24	168		20.45 (22)	0 – 24 (4.28)
Overactivity subscale	0 – 36	169		24.68 (27)	0 – 36 (8.61)
Impulsive speech subscale	0 – 12	101		6.61 (7)	0 – 12 (4.11)
Total score	0 – 72	168		48.74 (51)	9 – 72 (13.41)
Executive functions – CHEXI					
Inhibition factor	0 – 55	160		46.28 (55)	11 – 55 (7.51)
Working memory factor	0 – 65	159		53.99 (48)	13 – 65 (8.85)

Note. TAQ = The Activity Questionnaire; CHEXI = Childhood Executive Functioning Inventory.

Figure 4. 2 *Scree plot*



Note. Simulation line refers to artificial dataset generated by parallel analysis.

Table 4. 6 *Factor structure of SRQ items*

	Factor					
SRQ item	**1**	**2**	**3**	**4**	**5**	**Uniqueness**
23. Arms held	.718					.430
22. Hands held	.629					.600
12. Lying, hands behind back	.556					.467
13. Restrict arms	.427					.573
14. Carry object	.389					.787
4. Hands in trousers [a]						.779
2. Arms in clothing		.800				.386
1. Hands in clothing		.645				.394
3. Sleeves over hands		.552				.553
9. Wrap self in material		.383				.707
10. Arms around furniture		.355				.503
6. Wrap hands/arms together			.801			.359
7. Wrap material around arm(s)			.768			.363

Note. Principal axis factoring extraction method was used in combination with a direct oblimin rotation.

[a] Item was not assigned to a factor. [b] Item cross-loaded onto one other factor.

Table 4.6 cont.

	Factor					
SRQ item	**1**	**2**	**3**	**4**	**5**	**Uniqueness**
16. Hold hands behind back				.682		.318
15. Sit on hands				.509		.445
18. Hands between legs	.408[b]			.472		.459
19. Fold arms				.443		.669
17. Hold hands behind neck				.347		.645
5. Hands in pockets [a]						.806
20. Entwine arms other					.689	.418
11. Hands in tight spaces					.555	.416
8. Entwine hands other material			.364[b]		.371	.488
21. Interlock fingers					.364	.703

Note. Principal axis factoring extraction method was used in combination with a direct oblimin rotation.

[a] Item was not assigned to a factor. [b] Item cross-loaded onto one other factor.

Table 4.7 compares distribution of SRQ items across existing subscales and the new EFA factors. The EFA did not replicate Oliver et al. (2003)'s subscales, however similarities exist. Subscale 1 and Factor 1 share items involving 'using clothing or materials' to restrict movement, while Subscale 2 and Factor 2 involve items relating to 'use of own body'. Factor 3 contains two items from the original Subscale 1, grouping 'wrapping' behaviours. Factor 4 contains items grouped in Subscale 4, 'use of other people', however expands this into a broader category of using own or others' body to restrict hand/arm movement. Finally, Subscale 3 was not replicated in the EFA, and the EFA generated a novel fifth factor with no clear theme.

Table 4.7 displays the internal consistency of existing subscales and new factors. Given that subscales were not directly replicated by the EFA, it is challenging to compare coefficients across subscales and factors. Coefficients ranged from below acceptable (e.g., Subscale 3 ω=.57) to strong (e.g., Subscale 2 ω=.82) evidence of reliability.

Table 4. 7 Comparing items and internal consistency coefficients of existing SRQ subscales and new factors

Existing subscales		**EFA factors**	
Items	**Omega (ω)**	**Items**	**Omega (ω)**
Subscale 1. Use of clothing or materials	.77	*Factor 1. Use of clothing/materials*	.73
1. Entwine hands in front part of clothes		1. Entwine hands in front part of clothes	
2. Push arms into clothing		2. Push arms into clothing	
3. Pull sleeves over hands		3. Pull sleeves over hands	
4. Push hands inside trousers		9. Wrap blanket or other material around self	
5. Push hands inside pockets		10. Entwine arms around chair/furniture	
6. Wrap hands/arms together with material			
7. Wrap material around arm(s)			
8. Wrap or entwine hands in material (not clothes)			
9. Wrap blanket or other material around self			
Subscale 2. Use of own body	.82	*Factor 2. Use of own body*	.79
12. Lie on stomach with hands held behind back		15. Sit on hands	
13. Lie/sit in position restricting arm movement		16. Hold hands together behind back	
15. Sit on hands		17. Hold hands together behind neck	
16. Hold hands together behind back		18. Push hands between legs	
17. Hold hands together behind neck		19. Fold arms	
18. Push hands between legs			
19. Fold arms			
20. Entwine arms (other)			
21. Interlock fingers			

Note. EFA = exploratory factor analysis. Factors have been re-ordered to match the thematic subscales where possible.

Table 4.7 cont.

Existing subscales		EFA factors	
Items	**Omega (ω)**	**Items**	**Omega (ω)**
Subscale 3. Use of objects	.57	*Factor 3. Wrap material around arms*	.77
10. Entwine arms around chair/furniture 11. Push hands into tight spaces 14. Carry object without manipulating/using		6. Wrap hands/arms together with material 7. Wrap material around arm(s)	
Subscale 4. Preference for imposed restraint	.73	*Factor 4. Using own/others' body to restrict hand/arm movement*	.80
22. Indicate preference for hands to be held 23. Indicate preference for arms to be held		12. Lie on stomach with hands held behind back 13. Lie/sit in position restricting arm movement 14. Carry object without manipulating/using 22. Indicate preference for hands to be held 23. Indicate preference for arms to be held	
–		*Factor 5. No clear theme*	.71
		8. Wrap or entwine hands in material (not clothes) 11. Push hands into tight spaces 20. Entwine arms (other) 21. Interlock fingers	

Note. EFA = exploratory factor analysis. Factors have been re-ordered to match the thematic subscales where possible.

To assess the strength of the relationship between the five[29] factors generated by the EFA, correlations between factor scores were observed (Table 4.8). Results indicated small to medium inter-factor correlations[30], ranging from .175 (Factors 1 and 3) to .351 (Factors 1 and 2).

Table 4. 8 *Correlations between factor scores*

	1	**2**	**3**	**4**	**5**
1	—	.351	.175	.257	.301
2		—	.319	.223	.336
3			—	.202	.230
4				—	.257

In summary, the SRQ produced four stable factors, two of which were similar to the 'use of clothing/materials' and 'use of own body' subscales reported by Oliver et al. (2003). Factor internal consistency statistics were acceptable to strong, and factors positively intercorrelated with small to medium correlations.

[29] While five factors were identified by the EFA, Factor 3 was considered unstable due to containing only two items. Henceforth, Factors 1, 2, 4 and 5 are considered the four main EFA factors.

[30] See section 2.3.5 for correlation effect size interpretation.

4.4.2 Internal checks of direct EF tasks

Internal checks were conducted to test whether the direct EF tasks performed as expected. Developmental appropriateness and sensitivity were observed from child performance. Feasibility of tasks was assessed according to task adherence (i.e., data loss and reasons for task exclusion).

4.4.2.1 Child performance on EF tasks

Table 4.9 summarises task performance of the remote EF tasks, including the number of completed trials, number of children scoring at ceiling, pass/fail rates and mean composite scores of the original (i.e., non-imputed; section 6.3.3) dataset.

Table 4. 9 Descriptive statistics for direct executive function tasks

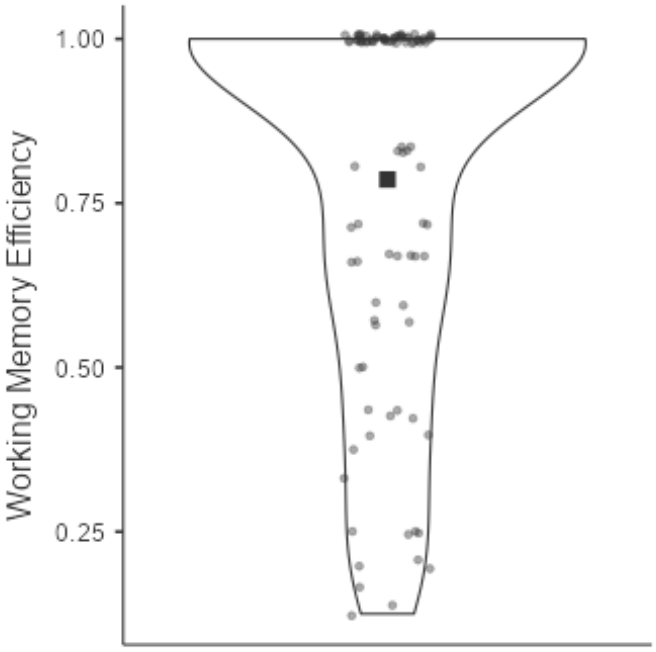
EF measure	***N* eligible**	***N* completed (%)**	***N* missing (%)**	**% ceiling**	***N* pass/fail**	**Mean (median)**	**Range (SD)**
Inhibition	126						
Prohibition wand		101 (80)	25 (20)	45.54	46/54		
Latency wand (ss.ms)		101 (80)	25 (20)	45.54		16.59 (15.30)	00.00 – 30.00 (13.02)
Prohibition array		111 (88)	15 (12)	28.82	32/79		
Latency array (ss.ms)		111 (88)	15 (12)	28.82		21.26 (4.85)	00.00 – 60.00 (25.83)
Prohibition composite [a]		96 (76)	30 (24)	29.17		0.40 (0.25)	0.00 – 1.00 (0.43)
Working memory	126						
Efficiency [a]		96 (76)	(30) 24	55.21		0.79 (1.00)	0.13 – 1.00 (0.28)
Perseveration [b]		96 (76)	(30) 24	70.83		0.09 (0.00)	0.00 – 0.75 (0.18)
Cognitive flexibility							
Sorting	40	11 (28)	29 (72)	18.18	2/9		
Categorisation	89	72 (81)	17 (19)	87.50	63/9		
DCCS	66	47 (71)	19 (29)	53.19	25/22		
Conflict score [c]	126	59 (47)	67 (53)	42.37		2.03 (2.00)	0 – 3 (1.07)
No-conflict score [c]	126	60 (48)	66 (52)	68.34		2.43 (3.00)	0 – 3 (0.98)

Note. Latency (ss.ms) = time taken to touch material(s) in seconds and milliseconds. DCCS = Dimension Change Card Sort.
[a] Min/max score = 0 – 1, higher scores indicate better performance. [b] Min/max score = 0 – 1, lower scores indicate better performance.
[c] Min/max score = 0 – 3, higher scores indicate better performance.

To assess whether the second prohibition 'array' task was more challenging than the first 'wand' prohibition task, task success was compared. The elevated fail rate of the array task compared to the fail rate of the wand task suggested it was more demanding (Table 4.9). An exact McNemar's test revealed that the difference in pass/fail rates from wand to array trials was statistically significant, $p<.001$. The mean prohibition composite score was over 20% and below 80% of the possible range of scores, suggesting developmental appropriateness (i.e., most scores clustered around a plausible range). Scores spanned the range of possible scores, with 50% participants passing one or both trials, while 50% participants failed both trials. These inter-individual differences suggested developmental sensitivity.

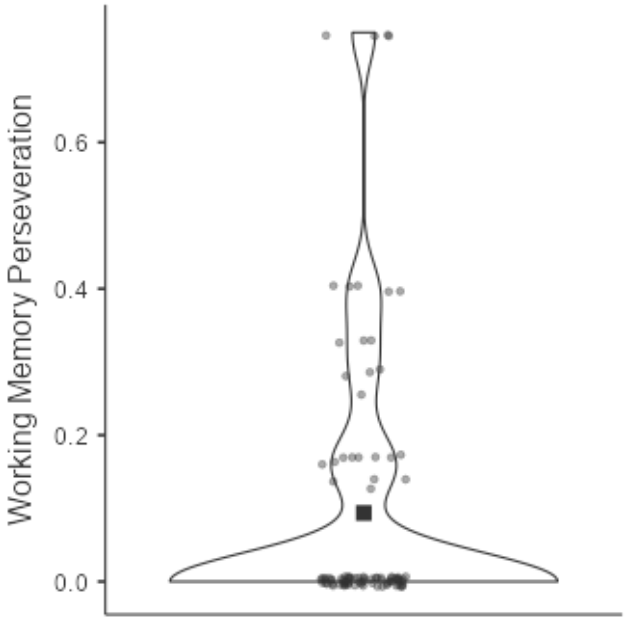
Second, the spread of working memory efficiency and perseveration scores was examined. Figures 4.3-4.4 display violin plots of the distribution of data. Developmental appropriateness was evidenced by the mean efficiency composite score sitting within 80% of the possible range of scores. Scores ranged from very low to high, with over 50% children reaching the ceiling. This suggested developmental sensitivity of this composite score. There was a wide range of perseveration composite scores, suggesting sensitivity. However, the mean perseveration score sat within the top 10% possible scores, and over 70% children reached the ceiling. This may suggest suitability of categorising perseveration scores into a dichotomous variable, with children split into 'perseveration' (n=28 with at least one perseveration) and 'no perseveration' (n=68 who reached the ceiling and achieved a perfect score of zero).

Figure 4. 3 Violin plot displaying spread of working memory efficiency scores



Note. Black square indicates mean score, grey dots represent individual data points and line shape represents density.

Figure 4. 4 Violin plot displaying spread of working memory perseveration scores



Note. Black square indicates mean score, grey dots represent individual data points and line shape represents density.

Finally, to test the validity of the conflict scale, pre-switch 'no-conflict' scores were compared against the post-switch 'conflict' scores. There was a statistically significant difference between pre-switch and post-switch scores, $t(58)=6.305$, $p<.001$. On average, pre-switch scores were .41 points higher than post-switch scores, 95%CI [.28, .54], indicating that children performed better during the pre-switch learning phase. This suggests that, as expected, the post-switch phase was more demanding. The mean conflict score sat above 20% and below 80% of the maximum possible score, suggesting developmental appropriateness, and the spread of scores suggested developmental sensitivity.

4.4.2.2 Task adherence

To assess feasibility of the EF tasks, task adherence was examined. In total, 573 EF tasks were attempted. Of these tasks, 438 (76%) were eligible for inclusion in analyses, and 135 tasks (24%) were excluded. Table 4.10 summarises reasons for data loss. The most common reason for task exclusion was discontinuation due to low child engagement or for safety (82 trials, 62%), while the least frequent reason was interference from additional caregivers (two trials, 1%).

In summary, the feasibility and validity analyses demonstrated that, as expected, tasks increased in difficulty such that children were more likely to fail task-phases that required more executive demand. Examination of mean composite scores and range of scores suggested tasks were developmentally-appropriate and sensitive. Finally, a low number of tasks were removed due to low validity scores, with the main reason for data loss due to task discontinuation.

Table 4. 10 Reasons for data loss on direct executive function tasks

Data loss reason		**Inhibition *n* (%)**		**Working memory *n* (%)**	**Cognitive flexibility *n* (%)**			**Total *N* (%)**
Broad reason	Specific reason	Wand	Array	Multilocation search	Sorting	Categorisation	DCCS	
Task/session discontinuation	Task discontinued	5 (4)	2 (1)	28 (21)	23 (17)	16 (12)	8 (6)	82 (61)
	Didn't attempt	-	1 (1)	1 (1)	2 (1)	-	4 (3)	8 (6)
Task delivery	Caregiver interference	2 (1)	1 (1)	-	2 (1)	1 (1)	4 (3)	10 (7)
	Caregiver administration error	4 (3)	3 (2)	-	1 (1)	-	-	8 (6)
	Child disinterested in materials	5 (4)	3 (2)	-	-	-	-	8 (6)
	Researcher administration error	2 (1)	1 (1)	-	1 (1)	-	2 (1)	6 (4)
	Interference from additional caregivers	2 (1)	-	-	-	-	-	2 (1)
Logistical challenges	Connectivity issues	4 (3)	2 (1)	-	-	-	1 (1)	7 (5)
Cumulative errors	Validity ≤ 3	1 (1)	2 (1)	1 (1)	-	-	-	4 (3)

Note. Dash (-) indicates no trials were excluded for listed reason. Percentages refer to the number of tasks excluded per reason divided by the total number of task exclusions.

4.5 Discussion

This Chapter describes the novel methods used to observe and assess self-restraint and EF in an under-represented population of autistic children with ID. The data collected using these methods formed the empirical basis of Chapters 5 and 6. The underlying factor structure of the SRQ was examined, and the developmental appropriateness, sensitivity and feasibility of a novel battery of direct EF tasks for autistic children with ID was assessed. There were three key findings. First, the SRQ produced four stable factors, with some replication of the original subscales reported by Oliver et al. (2003). The strong internal consistency of SRQ items and the variation in strength of factor correlations warrants discussion around the independence of constructs. Second, the mean composite scores generated from the play-based EF tasks fell within an expected range, suggesting that tasks were developmentally-appropriate, while the wide data spread suggested tasks were sufficiently sensitive to detect a range of abilities. Finally, the large proportion of EF data retained following a rigorous validity check procedure suggested tasks were feasible, and online administration suitable.

An EFA of the SRQ revealed four stable underlying factors. Factor internal consistency ranged from acceptable to strong. EFA factors did not replicate the original SRQ categories, however two similar factors involving ‘use of clothing or materials’ and ‘use of own body’ were produced. This lends psychometric support to these thematic subscales described by Oliver et al. (2003) in a well-characterised sample of autistic children with ID. Factors generated by the EFA significantly intercorrelated, suggesting that items measured a similar latent construct. This provides evidence in support of generating total scores from the SRQ to measure self-restraint. However, the strength of the inter-factor correlations ranged from small to medium, suggesting independence of constructs, lending support for generating

factor totals. However, this approach is complicated by the strong internal consistency of all 23 SRQ items. Overall, for simplicity a parsimonious approach of analysing according to the novel SRQ total scores, which incorporates topography and intensity, was employed in Chapters 5 and 6. Future research should endeavour to replicate the factor structure to establish reliability of factors, and further explore the clinical significance of the factors.

This Chapter described a novel, remote battery of play-based EF measures designed for autistic children with ID. Demographic characteristics revealed a high presence of autism and low adaptive functioning, suggesting the sample captured the population typically excluded from EF research. Internal checks indicated that average composite scores fell within 80% of the possible range suggesting developmental appropriateness, such that tasks were neither too easy nor hard, and the spread of scores suggested sensitivity of measures, such that individual differences were detected (Petersen et al., 2016). Balancing standardisation with adaptation to individual needs, an essential component of autism research (Giannadou et al., 2022), may have contributed to these results. Elevated fail-rates within more demanding trials suggested the tasks performed as theoretically expected and in accordance with typically developing infancy data (Devine et al., 2019). Therefore, tasks were considered suitable for autistic children with a range of ID severity.

A high level of data were retained across tasks ranging in executive demand. Task completion rates were comparable with those reported in the typically developing infancy literature. For example, Devine et al. (2019) retained 89% of prohibition trials when administered with 14-month-old typically developing children (80 and 88% in the present study). Devine et al. (2019)'s multilocation search task yielded 92% data retention within the infant group (76% in

the present study), and the scaled cognitive flexibility tasks were completed by 83% of the typically developing infants (81% in the present study). Finally, in the present study, the DCCS was completed by 71% eligible participants. This is comparable to findings by Jones et al. (2022) who reported a 61% DCCS completion rate in autistic children with 'below average IQ'. This demonstrates feasibility of administering play-based tasks remotely. The benefits of remote task administration are far reaching. Online testing eliminates the risk of transmitting infections such as Covid-19 to populations that are disproportionately impacted (i.e., autistic people/and or those with ID; Baweja et al., 2021; Turk et al., 2020). Participants remain in the familiarity of their home, minimising disruption. Elimination of researcher and participant travel increases time and cost-efficiency, providing opportunity to focus on recruiting a population that, despite significant clinical need, is systematically excluded from research. A recent meta-analysis estimated that 94% of autistic individuals included in the reviewed studies did not have ID, despite a tendency for research findings to be generalised to all autistic individuals (Russell et al., 2019). Around 50 to 55% autistic individuals have ID (Charman et al., 2011; Loomes et al., 2017), revealing a selection bias against the many autistic people with ID. This position may be reversed by using the remote methods presented in this Chapter. For example, flexibility afforded by online testing allows caregivers to schedule, and if necessary, reschedule, testing sessions to suitable times based on child and family need. In sum, remote testing offers a unique opportunity to collect rich EF data from an underrepresented population.

A validity checklist was designed to capture salient features of the tasks and exclude tasks with overarching issues. In total, 3% of all data exclusions resulted from cumulative errors captured by validity checklist scores. Logistical 'connectivity' issues, such as Wi-Fi or device

connectivity, accounted for 5% of data loss. Recognising challenges with sourcing expensive equipment, future research may endeavour to post video-capturing devices (e.g., tablets) and portable 'Wi-Fi USB adaptors' to reduce such data loss. Child disinterest in materials, errors in task administration, and interference from primary or additional caregivers resulted in 24% of data loss. More interesting stimuli, fewer or shorter trials, and caregiver training (e.g., video examples of correct and incorrect task administration) may reduce such data loss. Importantly, in the present study, task administration was shared across multiple researchers, indicating replicability of measures and satisfactory training. Finally, the largest data loss was due to task or session discontinuation (67% of lost data), perhaps due to low child engagement or emergence of distress or SIB. These exclusions highlight that despite the general feasibility and accessibility of the EF battery, future research is required to ensure methods suit the broad population. For example, despite caregivers choosing 'Games Session' times, visits were often conducted after school, when the child may be tired (a known correlate of self-injury; Khazaie et al., 2021). Future research capitalising on school-time data collection may increase data retention. Overall, minimal data were lost in the present study.

In summary, strong internal consistency of SRQ items and significant correlations between EFA factors suggested value in calculating total SRQ 'intensity' scores. On this basis, this approach will be adopted in Chapters 5 and 6, and implications discussed accordingly. Assessment of the feasibility, performance and validity of the battery of EF tasks indicated suitability with this under-represented population. Results and implications are discussed in Chapter 6.

CHAPTER FIVE

A NATURALISTIC OBSERVATIONAL STUDY OF SELF-RESTRAINT

5.1 Preface

The systematic review in Chapter 2 revealed that very few large-scale observational studies of self-restraint have been conducted. Observational studies provide rich data around intensity, an essential parameter of behavioural severity. Chapter 3 provided preliminary evidence for the relation between self-restraint and behaviours of concern, including self-injury and aggression, however this has not been studied beyond informant-report measures of self-restraint topography. The exploratory factor analysis in Chapter 4 suggested psychometric suitability of utilising total 'intensity' self-restraint scores. Therefore, to extend understanding of the potential pervasiveness of self-restraint behaviours and their relation with other clinical behaviours, Chapter 5 describes a naturalistic observational study of self-restraint intensity displayed by autistic children with intellectual disability. Using procedures described in Chapter 4, this Chapter examines the temporal relations between self-restraint and behaviours of concern, and compares informant-rated self-restraint intensity with observed behavioural intensity, with implications for the clinical application of measures.

5.2 Introduction

Self-restraint has been investigated using experimental functional analysis, manipulation of antecedents and consequences to identify behavioural function (Iwata et al., 1982; Neef & Iwata, 1994), alongside naturalistic observations, where contingencies are not experimentally manipulated. Observational and experimental studies have informed operant learning models of self-restraint and self-injurious behaviour (SIB; section 1.4.7). However, the existing literature is limited by small sample sizes, reducing ability to generalise findings to broader populations known to self-injure and self-restrain. Experimental functional analysis presents methodological and ethical challenges, overcome by adopting a naturalistic observation approach. Therefore, the overarching aim of this study was to observe the intensity of naturally-occurring self-restraint displayed by autistic children with co-occurring intellectual disability (ID).

Observations of self-restraint provide information about topography and intensity. Intensity, the amount of time an individual spends engaging in self-restraint (section 1.4.5.2), is an essential parameter of behaviour due to the adverse consequences of preferred and prolonged self-restraint (section 1.4.2; Forman, 2003). Existing literature describes individuals who display intense self-restraint, for example by trapping hands in clothing (e.g., Forman et al., 2002) and seeking restraint devices (e.g., a straightjacket; Kinnell, 1984, section 1.4.5.1). However, observations of intensity typically adopt a case study or series design, limiting generalisability of findings to broader neurodevelopmental conditions at high risk of self-injury and self-restraint (section 3.4.1.2). Participants included in these case descriptions typically have a severe to profound ID, display severe SIB and/or restraint-use that necessitates professional service support, and present with uniquely concerning behaviours

warranting research attention (e.g., Forman et al., 2002; Jennett et al., 2011; Pace et al., 1986). Furthermore, conducting research in laboratory settings, while advantageous for maintaining experimental control, may impact participant behaviour due to the unusual environment, interactions with unfamiliar researchers and disruption of routine (Giannadou et al., 2022). Such limitations reduce ability to generalise results to wider populations, such as autistic children and/or those with ID who do not access services for behaviours of concern (BoC), across real-life settings (Patino & Ferreira, 2018). Thus, to expand upon existing case descriptions, this study employed a large-scale observational approach to investigate individual differences in self-restraint, with methods permitting the possibility of examining correlates of observed self-restraint. Generalisability is also improved, for example autistic individuals display social behaviours that are more representative of real-world behaviours when observed at home and in the presence of a primary caregiver than in laboratory settings, due to lower stress (Grandgeorge et al., 2015). Thus, this study examined behaviour displayed in the natural home environment.

Descriptive observational studies of self-restraint shed light on the breadth of self-restraint topographies. This contrasts with experimental functional analysis, which focuses on the function of known self-injurious and self-restraint behaviours (Hanley et al., 2003). Nearly 100 different topographies of self-restraint have been reported in the literature (Forman, 2003; section 1.4.4), and neurodivergent individuals typically display more than one self-restraint topography. For example, 59% of participants with neurogenetic conditions who were reported to self-restrain had displayed two or more topographies in the previous month (section 3.4.1). Chapter 4 revealed 96% of autistic children with ID were reported to display two or more topographies, while 49% showed 10 or more topographies (section 4.4.1).

Restricted behavioural codes challenge ability to identify potential patterns of topography and intensity. Understanding intensity of common topographies and examining variation within topographies provides information around two essential parameters of self-restraint (i.e., topography and intensity). Therefore, this study observed over 23 topographies of self-restraint based on the Self-Restraint Questionnaire (SRQ; Oliver et al., 2003). Operationalised 'open' codes, topographies not included in the SRQ, were added once observed. Therefore, the first aim of this study was to describe the intensity of self-restraint topographies in a large community sample of autistic children with ID.

While functional analyses identify self-restraint function (e.g., Smith et al., 1992), the method presents practical and ethical challenges. Experimental manipulations rely on precise experimenter responding contingent upon behaviour. Intensive training and experience are required to ensure conditions are rigorous and safe (Poling et al., 2012). For example, a thorough medical exam is required to exclude participants at highest risk of severe behaviour, and consultation with caregivers is essential to obtain informed consent and to create a termination plan (Iwata et al., 1982; Neef & Iwata, 1994). Experimental functional analysis is further complicated by the presence of intense self-restraint. For example, removal of self-restraint during restraint-unavailable sub-conditions (e.g., Smith et al., 1992) may result in distress, anxiety and severe SIB, and lead to the premature termination of the experiment. Taken together, the resources required to train experimenters and obtain multidisciplinary assessments make experimental functional analysis a challenging and relatively inaccessible research technique. A less resource-intensive and ethically challenging alternative to describe observed self-restraint is naturalistic observation.

Naturalistic observation does not involve manipulation of the environment contingent upon behavioural presentation, and presents fewer ethical challenges than experimental functional analysis when studying self-restraint and SIB. Forman et al. (2002) conducted the first naturalistic observation of SIB and self-restraint with a 26-year-old man with profound ID and reported findings consistent with a hypothesis of negative reinforcement. Lag sequential analysis, examination of the temporal probability of behaviour, revealed significantly elevated rates of SIB before and after self-restraint (i.e., greater than chance). This study demonstrated how SIB and self-restraint interact when an individual is in their preferred environment and interacting with familiar caregivers. However, due to the lack of existing naturalistic observational data, understanding of the presence and intensity of self-restraint topographies displayed by those with varying severities of SIB is limited. Therefore, the second aim of this study was to examine the temporal relation between observed self-restraint and SIB using a naturalistic observation study design.

Meta-analysis of existing studies estimated a moderate correlation between self-restraint and SIB, r=.21 (section 2.4.5). Individuals with ID who show SIB are more likely to show other BoC, such as aggression and property destruction[31], than individuals who do not self-injure (Emerson et al., 2001; Matson et al., 2008). Autistic individuals are more likely to show self-injury, aggression and property destruction than non-autistic individuals (McClintock et al., 2003). Therefore, individuals with autism and/or ID who self-injure are likely to display other BoC. A cross-syndrome informant-report investigation into self-restraint revealed BoC were strongly intercorrelated and each correlated with self-restraint topography scores (section

[31] Aggression and property destruction are complex and potentially disabling behaviours. Emphasis is not placed on the presumption that the individual intends to display violence towards people or objects, rather, behaviours may reflect challenges with functional communication (Matson et al., 2005).

3.4.1.2). This novel finding implicated aggression and property destruction as key correlates of self-restraint, warranting discussion around the specificity of the relation between self-restraint and SIB (section 3.5). Furthermore, self-restraint is also associated with repetitive and compulsive behaviour (section 3.4.1.2). To date, the temporal relations between self-restraint and other BoC (and the temporal relations between different topographies of self-restraint) have yet to be examined using observational techniques. Therefore, this study examined the temporal relation between observed self-restraint and other BoC.

As described, observations provide data around self-restraint intensity, a clinically useful descriptor of behaviour. Intensity data can also be generated using informant-reported SRQ scores (section 1.4.6). Oliver et al. (2003) compared informant-report self-restraint with observed behaviour and found 100% of participants (n=14) displayed at least one form of self-restraint indicated by SRQ caregiver report. Beyond this small-scale investigation, no study has examined the potential association between intensity data drawn from observational techniques and informant report tools such as the SRQ. Behavioural (i.e., observational) measures record actual performance under specific circumstances, while informant-report questionnaires (i.e., SRQ) rely on caregiver interpretation, recall and overall assessment of how an individual 'usually' behaves (Dang et al., 2020). Behavioural and questionnaire measures typically share weak correlations (Dang et al., 2020; Toplak et al., 2013). A strong association between self-restraint measures would suggest observed and SRQ intensity measure a similar construct, indicating that both approaches are useful for identifying self-restraint intensity and may be used independently to capture the same phenomena. A weak association, however, may indicate that measures capture distinct constructs, with implications of measurement in clinical practice. Therefore, the final aim of this study was to

compare intensity of self-restraint observed in a naturalistic setting with informant-report self-restraint intensity. Further examination of relations between intensity scores and putative correlates, such as informant-reported BoC and executive function (EF) aim to further shed light on the theoretical and clinical utility of measures.

In summary, this study had three aims: (1) describe the intensity of different topographies of self-restraint observed in a sample of autistic children with ID, (2) describe temporal relations between BoC and observed self-restraint, and (3) examine links between observed self-restraint intensity and informant-report intensity, and compare relations with demographic, behavioural and cognitive variables.

5.3 Methods

5.3.1 Recruitment, participants and data collection

Participants were recruited using the procedure and methods outlined in Chapter 4 (section 4.3.2 describes recruitment, section 4.3.5 describes procedure). The behaviour of participants who completed the EF battery (*N*=126; section 4.3.4.3) was observed. Table 5.1 displays participant demographic information.

5.3.2 Measures

A range of questionnaires and direct assessments were used to describe demographic (e.g., age, gender, verbal ability), behavioural (e.g., self-restraint, SIB) and cognitive characteristics (i.e., EF). Section 4.3.4 describes measures in full.

5.3.3 Data coding

Videos were coded using Datavyu (v.1.5.3; Datavyu Team, 2014) in three stages, following criteria outlined in a bespoke Coding Manual (Appendix 5.1). Inter-rater reliability was calculated at each stage of coding, and behavioural definitions were refined following consensus meetings. Table 5.2 displays behavioural codes.

Table 5. 1 Demographic characteristics of experimental study sample

Developmental variables	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Chronological age (years)	126		9.95 (9.71)	4.02 – 15.49 (2.98)
Sex (female/male/non-binary)	126	28[a]/97/1 (22/77/1)		
Mobility (walk unaided/unable)	126	120/6 (95/5)		
SCQ total (all participants)	126		26.83 (27.00)	15 – 37 (5.08)
SCQ total verbal		74 (59)	27.12 (27.50)	15 – 37 (5.99)
SCQ total non-verbal		52 (41)	26.42 (27.00)	20 – 32 (3.42)
ASD/autism [b]		17/109 (13/87)		
Autism description	126			
Autism Spectrum Disorder		61 (48)		
Autism		58 (46)		
High Functioning Autism		3 (2)		
Autistic Spectrum		2 (2)		
Asperger Syndrome		2 (2)		
VABS-III ABC score	126		43.01 (40.50)	20 – 87 (14.89)
VABS-III RL AE (months)	126		19.57 (19.00)	0 – 67 (10.05)
Intellectual Disability	126			
Mild		9 (7)		
Moderate		40 (32)		
Severe		52 (41)		
Profound		5 (4)		
Unknown		18 (14)		
Other [c]		2 (2)		
BPVS raw score [d]	70		70.67 (69.00)	2 – 146 (39.22)
BPVS standardised score [e]	70		74.14 (69.00)	69 – 118 (10.79)
BPVS age equivalent (months) [f]	41		84.51 (74.00)	45 – 162 (28.62)

Note. SCQ = Social Communication Questionnaire; VABS-III ABC = Vineland Adaptive Behavior Scales, Third Edition Adaptive Behaviour Composite; VABS-III RL AE = receptive language age equivalents. BPVS = British Picture Vocabulary Scale, Third Edition.

[a] *n*=1 self-described gender different to that which was assigned at birth. [b] 'ASD' indicated by scoring 15 – 21 on SCQ, 'autism' indicated by scoring 22 or above. [c] Other = "estimated 'mild'", "delay of around 2.5 years". [d] BPVS discontinued for *n*=46 and not attempted for *n*=10 due to child engagement or for safety. [e] Includes *n*=50 who scored below average of a normed sample (70), indicated as '69'. [f] Age equivalents incalculable for *n*=29 as raw scores fell below normed sample scores.

Table 5. 2 *Description of behavioural codes*

Code number	Brief description and presence of behaviour in sample	
Self-restraint		
1	Entwine hand(s) in front part of clothes that are worn	+
2	Pull arm(s) out of sleeves and pushes them into main body of clothes that are worn	+
3	Pull sleeve(s) over hands	+
4	Push hand(s) inside trousers, skirt, etc	+
5	Push hand(s) inside pockets	+
6	Wrap hands or arms together using material	-
7	Wrap material around either or both arms (not tied together)	-
8	Wrap hand(s) in material other than clothes	+
9	Wrap blanket, sheet, etc, around self	+
10	Entwine arms around chair/other furniture	+
11	Push hands or body into tight space e.g., sofa cushion	+
12	Lie on stomach with hands held behind back	-
13	Lie/sit in position restricting movement of arm(s)	+
14	Carry object(s) around without manipulating, using or playing with it	+
15	Sit on hand(s)	+
16	Hold hands together behind back	+
17	Hold hands together behind neck	+
18	Push hands between legs	+
19	Fold arms	+
20	Entwine arms in any other way	+
21	Interlocks fingers or hands	+

Note. Plus (+) indicates behaviour was displayed by at least one participant, dash (-) indicates behaviour was not observed during respective coding stage.

Table 5.2 cont.

Code number	Brief description and presence of behaviour in sample	
22	Asks or indicates to others desire for hands to be held	+
23	Asks or indicates to others desire for arms to be held	+
24	Wears restraint device	+
25	Hold hands together above head	+
26	Entwine self in others' clothing	+
27	Grabs/holds another's body part	+
Self-injury		
1	Bite hard object using top and bottom teeth	-
2	Bite self by clenching jaw and teeth around body part	+
3	Forcefully push thumbs into cheek	-
4	Push fingers on eye, pulls or pokes eyelashes	+
5	Finger bite by clenching jaw and teeth on side of finger	+
6	Hand bite by clenching jaw and teeth on side of finger	-
7	Hand to hand by forcefully bring palms together	+
8	Hand to object hit by forcefully making contact with hard surface with palm	+
9	Head bang by forcefully making contact with head against hard surface	+
10	Head hit by forcefully making contact with fist and/or open palm on head	+
11	Hit self by forcefully making contact with fist and/or open palm on own body part	+
12	Leg slap by forcefully making contract on leg with palm	+
13	Mouth hit by forcefully makes contact on/around mouth with palm	+
14	Scratch self by dragging nails across or pinching own body	+
15	Scrape skin using forefinger at right angle to skin	+
16	Teeth hit by forcefully making contact with top teeth against object	-

Note. Plus (+) indicates behaviour was displayed by at least one participant, dash (-) indicates behaviour was not observed during respective coding stage.

Table 5.2 cont.

Code number	Brief description and presence of behaviour in sample	
17	Wrist hit by forcefully making wrist-to-wrist contact	+
18	Pull hair or skin	+
19	Choke self by wrapping hands around neck and squeezing	+
Aggression		
1	Forcefully hitting other using a body part	+
2	Strike foot out and kicking other, making contact	+
3	Clenching jaw and teeth around other's body part	-
5	Holding onto other's body or clothes and pulling	+
6	Pushing other	+
7	Rubbing hand against other's skin, pinching other's body part or dragging nails across other's body part	+
8	Spiting at or on other	-
9	Throwing object at other	+
10	Verbalising towards other using threatening language	+
Property destruction		
1	Biting or chewing small items (e.g., testing materials)	+
2	Tearing or ripping small items (e.g., books)	+
3	Throwing, kicking or smashing small items (e.g., testing materials)	+
4	Slamming, hitting or kicking doors, walls or windows	-
5	Tipping, smashing or throwing large items	+
6	Pulling items from walls or shelves	-

Note. Plus (+) indicates behaviour was displayed by at least one participant, dash (-) indicates behaviour was not observed during respective coding stage.

5.3.3.1 Stage one coding

Stage one focused on identifying clips where the child was visible using the 'onset' and 'offset' Datavyu functions. Due to the play-based nature of the session children were rarely in

full camera-view. Children were considered 'visible' when one or both of their hands were in view, or when the child was engaging in self-restraint (in this case hands may be hidden). This criterion was chosen after reviewing the most prevalent topographies of self-restraint identified by meta-analysis, 'holds or squeezes objects' and 'holds onto others or others clothing' (section 2.4.4), both of which involve the use of hand(s). Additionally, SRQ items involve use of one or both hands. Full criteria for whether a child was 'visible' or 'not visible' are reported in Appendix 5.1. In total, 108 hours of footage were reviewed at stage one. A total of 20 hours of footage (19% of footage reviewed at stage one) was coded by a second researcher. Table 5.3 reports inter-rater reliability of coding at all stages, with agreement ranging from moderate to excellent[32].

5.3.3.2 Stage two coding

At stage two, duration of self-restraint was coded using the 'onset' and 'offset' functions. Self-restraint codes represented topographies listed in the SRQ (Appendix 5.1). To generate an intensity score comparable with informant-reported SRQ intensity scores, wording was only altered to ensure codes were suitably operationalised. Four behaviours not included in the SRQ were observed during the early stage of stage two coding and added to the list of self-restraint codes (codes 24-27). In total 57 hours of footage was reviewed at stage two. A total of 12 hours of footage (21% of stage two footage) was double-coded.

[32] Intraclass correlation coefficients (ICC) were interpreted according to Koo & Li (2016)'s guidelines, see section 2.3.4. Cohen's kappa (κ) was interpreted according to McHugh (2012)'s guidelines, see section 2.3.3

5.3.3.3 Stage three coding

At stage three, to observe behaviours that occurred before self-restraint, the 30 seconds prior to the onset of self-restraint were observed and the frequency of self-injury, aggression, property destruction and other self-restraint[33] was recorded (Appendix 5.1). To observe behaviours that occurred after self-restraint, the 30 seconds immediately following the offset of self-restraint were observed and the frequency of behaviours recorded. Codes for BoC were developed from a previous study (Richards, 2012, p. 231) and items listed in the Challenging Behaviour Questionnaire (CBQ; section 4.3.4.2.1). 'Other self-restraint' codes were identical to those used at stage three. In total, 24 hours of footage was reviewed at stage three. In combination, 190 hours of footage was reviewed at stages one, two and three. The 30 seconds prior to and following 81 observations of self-restraint were double-coded.

Table 5. 3 *Inter-rater reliability of behavioural coding across all stages*

Coding stage	Comparisons	Coefficient (ICC/κ)
1	Total number of seconds child visible	.97
2	Total number of observations of self-restraint	.73
	Total duration of self-restraint	.90
3	Presence of self-injury before self-restraint	.93
	Presence of self-injury after self-restraint	.91
	Presence of aggression before self-restraint	.93
	Presence of aggression after self-restraint	.79
	Presence of property destruction before self-restraint	.90
	Presence of property destruction after self-restraint	.92

Note. 25% (*n*=32) of sessions were double coded for reliability across all three stages.

[33] 'Other self-restraint' refers to any self-restraint behaviour that occurred in the 30 seconds preceding or following self-restraint, and was not necessarily a different topography.

5.3.4 Data analysis

To address the first aim, to describe the intensity of self-restraint topographies, the proportion of time spent engaging in each topography was calculated for all participants. To account for variation in visible observation time between participants, percentages were calculated by dividing the total number of seconds a child spent engaging in a topography by the total number of seconds the child was visible and multiplying by 100.

The second aim was to examine associations between self-restraint and BoC and other instances of self-restraint. A total of eight 'conditions' were assessed: self-injury, aggression, property destruction and self-restraint occurring before and after self-restraint. Individual proportions were first calculated by summing the number of occasions BoC or self-restraint occurred before/after self-restraint per participant, dividing by the total number of self-restraint observations per participant and multiplying by 100. Mean proportions were generated by summing individual proportions per condition, dividing by the total number of participants reported to show the respective BoC in the previous month according to the CBQ (SIB n=82, aggression n=81 and property destruction n=81), and multiplying by 100. This was to reduce bias, as a child who was not reported to self-injure was unlikely to display SIB during natural observation. 'Other' self-restraint proportions involved all participants who displayed self-restraint (n=111; n=1 excluded due to missing data). Due to non-independence of data (i.e., more than one BoC could occur before/after the same occasion of self-restraint, and BoC could be counted multiple times due to the quick succession of self-restraint), it was not possible to conduct an inferential test of mean difference. Instead, standard error (SE) bars, an indication of variability (Cumming et al., 2007), were inspected to identify differences between means.

To examine intensity of observed self-restraint, 'total observation intensity scores' were calculated by converting intensity percentages to a score of zero (observation intensity of zero), one (0.01-49.99%), two (50-74.99%), three (75-94.99%) or four (95-100%). Thresholds were chosen to correspond to SRQ intensity descriptions (a score of zero = 'never', one = 'some', two = 'half', three = 'most' and four = 'all of the time'). Individual scores were summed to generate a total 'observation intensity score' for each participant. Observation intensity scores were correlated with informant-report SRQ intensity scores using SPSS (v.29). Shapiro-Wilk and Kolmogorov-Smirnov p-values of <.05 suggested non-normality of data, requiring the use of Spearman's rho (r_s), a non-parametric correlation. Relations between intensity scores and demographic, behavioural and cognitive variables putatively associated with self-restraint were examined. The Benjamini-Hochberg method was used to control for multiple comparisons (section 3.3.6).

5.4 Results

5.4.1 The intensity of observed self-restraint

Across the sample, 89% of participants displayed self-restraint. Table 5.4 displays behavioural characteristics.

In total, 6490 minutes and 39 seconds of video footage was recorded. An average of 51 minutes 51 seconds of footage was recorded for each child, *range* = 2 minutes 59 seconds – 99 minutes 53 seconds. An average of 77 'visible clips' were generated for each child, *range* = 1–225. Children were 'visible' for an average of 27 minutes and 13 seconds, *range* = 2 minutes 59 seconds – 70 minutes 14 seconds. Therefore, on average, children were visible for 53% of the recorded footage, *Mdn* (IQR [interquartile range])=54% (23.06), *range* = 17–100%.

Of the 126 participants, 112 children showed at least one self-restraint behaviour during the observable footage, while 14 children did not exhibit any self-restraint. A total of 1469 self-restraint behaviours were observed. On average children showed 11.66 self-restraint behaviours, *Mdn*=6, *range*=1–70, and 2.62 different topographies (Table 5.4). Therefore, children tended to show the same topography more than once during the session.

Table 5. 4 Behavioural characteristics of observation sample (N=126)

Behavioural variables	**Possible score range**	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Self-restraint – SRQ		125			
Monthly self-restraint/no SR			121/5 (96/4)		
Total SRQ intensity score	0 – 92			15.41 (13.00)	0 – 84 (12.06)
Number of topographies	0 – 23			8.89 (8.00)	0 – 23 (5.38)
1 topography/2+ topographies			6/114 (5/95)		
Self-restraint – observation		126			
Observed self-restraint/no SR			112/14 (89/11)		
Total observation intensity score	0 – 108			2.66 (2.00)	0 – 8 (1.91)
Number of topographies	0 – 27			2.62 (2.00)	0 – 8 (1.84)
1 topography/2+ topographies			26/86 (23/77)		
Behaviours of concern – CBQ					
Monthly SIB/no current SIB		126	93/33 (74/26)		
Total SIB severity score	0 – 14			5.94 (7.00)	0 – 14 (4.41)
Monthly AGG/no current AGG		126	93/33 (74/26)		
Total AGG severity score	0 – 14			5.72 (6.00)	0 – 14 (4.22)

Note. SRQ = Self-Restraint Questionnaire; CBQ = Challenging Behaviour Questionnaire; SIB = self-injurious behaviour; AGG = aggression.

Table 5.4 cont.

Behavioural variables	Possible score range	*N*	*n* (%)	Mean (median)	Range (SD)
Monthly DoP/no current DoP		126	93/33 (74/26)		
Total DoP severity score	0 – 14			5.85 (7.00)	0 – 14 (4.30)
Monthly STPY/no current STPY		118	96/22 (81/19)		
Total STPY severity score	0 – 14			6.91 (8.00)	0 – 14 (4.11)
Activity – TAQ					
Impulsivity subscale	0 – 24	126		20.23 (21.50)	0 – 24 (4.31)
Overactivity subscale	0 – 36	126		24.33 (25.50)	0 – 36 (8.73)
Impulsive speech subscale	0 – 12	73		6.12 (6.00)	0 – 12 (4.05)
Total score	0 – 72	126		48.12 (50.00)	9 – 72 (13.36)
Executive functions – CHEXI					
Inhibition factor	0 – 55	125		46.10 (47.00)	11 – 55 (7.38)
Working memory factor	0 – 65	124		54.26 (55.00)	13 – 65 (8.58)

Note. DoP = destruction of property; STPY = stereotypy; TAQ = The Activity Questionnaire; CHEXI = Childhood Executive Functioning Inventory.

Table 5.5 displays the number of participants who displayed each topography of self-restraint. Intensity proportion scores are reported alongside means and range of scores. Topography codes refer to the 27 self-restraint behaviours listed in Table 5.2. Proportion ranges are colour-coded to represent relative size of intensity: red represents very low intensity (participant engaged in topography <1% of time visible), orange represents low intensity (1–24.99%), yellow represents moderate intensity (25–49.99%), green represents high intensity (50–97.99%) and blue represents very high intensity (98–100%). Appendix 5.2 displays intensity proportions for each participant, and Appendix 5.3 represents the distribution of proportions using violin plots.

Table 5. 5 Descriptive statistics of intensity proportions

Topography	***n* (%)**	**Mean intensity % (median)**	**SD**	**Range**	
				Minimum	**Maximum**
1	17 (13)	1.13 (0.80)	1.05	0.10	3.70
2	1 (1)	3.50 (3.50)	-	3.50	3.50
3	5 (4)	1.12 (0.40)	1.31	0.10	3.30
4	5 (4)	1.44 (0.50)	1.42	0.30	3.40
5	5 (4)	0.39 (0.10)	0.62	0.04	1.50
6	0 (0)	-	-	-	-
7	0 (0)	-	-	-	-
8	11 (9)	8.52 (1.50)	14.63	0.20	47.60
9	9 (7)	33.87 (13.70)	39.68	1.50	97.60

Note. Dash (-) indicates no data. Red indicates very low intensity (<1%), orange = low intensity (1 – 24.99%), yellow = moderate intensity (25 – 49.99%), green = high intensity (50 – 97.99%) and blue = very high intensity (98 – 100%).

Table 5.5 cont.

Topography	*n* (%)	Mean intensity % (median)	SD	Range	
				Minimum	Maximum
10	1 (1)	3.10 (3.10)	-	3.10	3.10
11	6 (5)	2.30 (1.20)	2.87	0.30	7.90
12	0 (0)	-	-	-	-
13	40 (32)	4.78 (1.00)	15.87	0.10	100.00
14	5 (4)	11.20 (4.30)	18.05	0.50	43.10
15	7 (6)	1.28 (0.56)	1.22	0.20	2.80
16	6 (5)	0.76 (0.25)	1.12	0.15	3.00
17	13 (10)	0.75 (0.30)	1.19	0.02	4.20
18	16 (13)	2.04 (1.15)	3.05	0.08	11.30
19	37 (29)	1.96 (0.60)	4.16	0.05	23.76
20	1 (1)	4.14 (4.14)	-	4.14	4.14
21	61 (48)	1.68 (0.80)	2.45	0.02	12.80
22	29 (23)	2.15 (0.87)	3.45	0.10	14.40
23	6 (5)	3.07 (0.65)	5.82	0.12	14.89
24	2 (2)	99.84 (99.84)	0.23	99.68	100.00
25	5 (4)	0.38 (0.32)	0.31	0.08	0.84
26	9 (7)	1.29 (0.47)	1.98	0.20	6.41
27	24 (19)	2.26 (0.59)	4.03	0.06	15.76

Note. Dash (-) indicates no data. Red indicates very low intensity (<1%), orange = low intensity (1 – 24.99%), yellow = moderate intensity (25 – 49.99%), green = high intensity (50 – 97.99%) and blue = very high intensity (98 – 100%).

The most frequently observed behaviours were ‘interlocks fingers or hands’ (self-restraint code 21, 48% of participants), ‘lies or sits in a position restricting arm movement’ (code 13, 32%), and ‘folds arms’ (code 19, 29%). Regarding the intensity of common topographies, the mean intensity of ‘interlocks fingers or hands’ was low (2.45%) with proportions ranging from very low (0.02%) to low (12.08%). Mean intensity of ‘lies or sits in a position restricting arm movement’ was low (4.78%), ranging from very low (0.10%) to very high (100%). Finally, mean intensity of ‘folds arms’ was low (1.96%), ranging from very low (0.05%) to low (23.76%).

The following topographies were not observed: ‘wraps hands or arms together using material’ (code 6), ‘wraps a piece of material around either or both arms but not so that they are held together’ (code 7) and ‘lies on stomach with hands held behind back’ (code 12). Excluding these codes, the behaviours displayed by the fewest participants were ‘pulls arms out of sleeves and pushes into main body of clothing’ (code 2), ‘entwines arms around chair or other furniture’ (code 10) and ‘entwines arms in any other way’ (code 20; all n=1). The intensity of these less-common topographies was low (3.50, 3.10 and 4.14% respectively).

In addition to code 13 (described above), two topographies were observed at high or very high intensities: ‘wears restraint device’ (code 24) was displayed by two participants at intensities ranging from 99.68 – 100% (very high), with a mean intensity of 99.84%. ‘Wraps blanket or other large piece of material around self’ (code 9) was displayed by nine participants, three of whom displayed high (62.00%) to very high (97.60%) behavioural intensity. Mean intensity of this topography was moderate (33.87%).

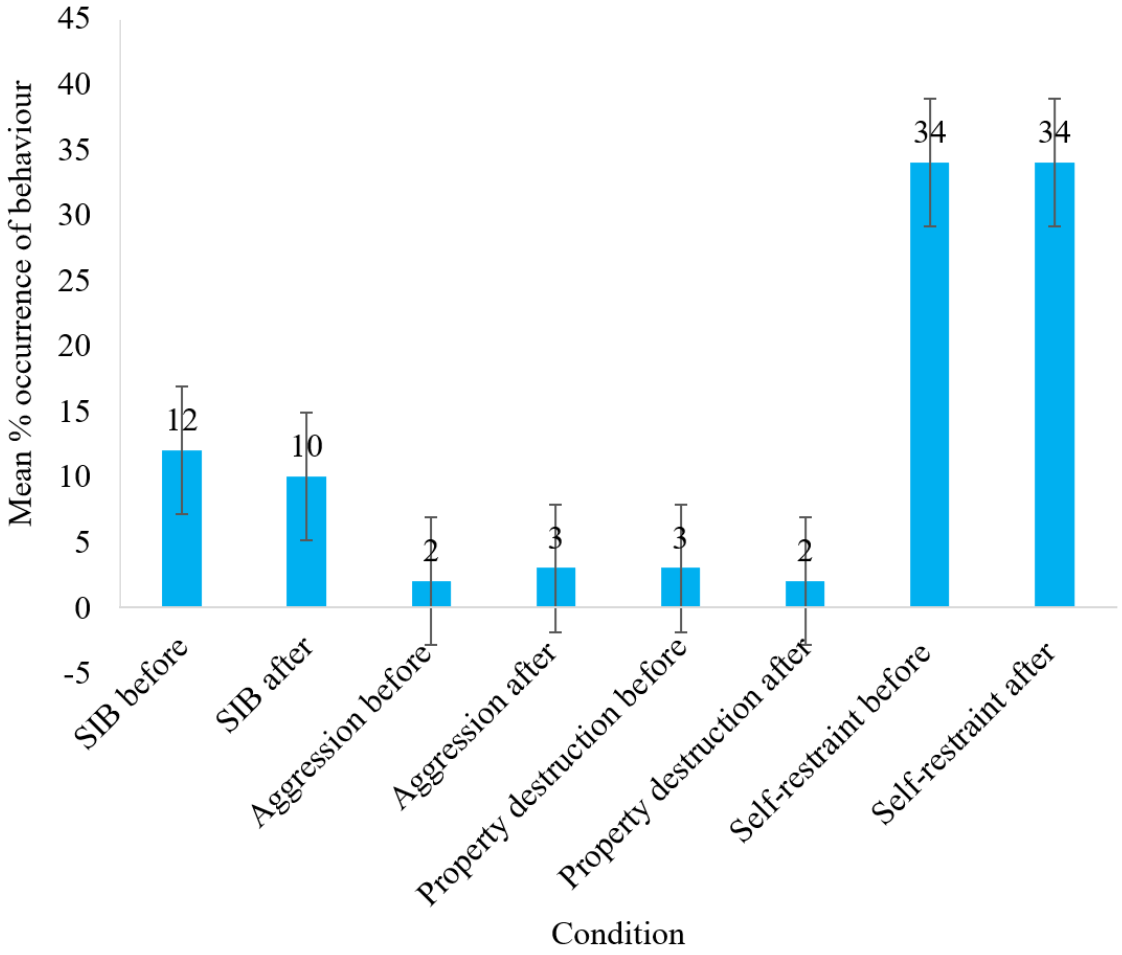
In summary, 24 of the 27 self-restraint behaviours of interest were displayed by participants, and the intensity of observed topographies of self-restraint ranged from 0.02 to 100%. Many topographies occurred at very low or low intensities, however the intensity of some topographies varied from very low/low to high/very high. Topographies observed at high or very high intensities were displayed by few participants (i.e., one to three individuals).

5.4.2 Temporal relations between self-restraint and behaviours of concern

A variety of BoC were observed before and after self-restraint (Table 5.2). Percentage of observations of self-restraint that were preceded and followed by SIB, aggression, destruction and other self-restraint is reported in Figure 5.1.

Other self-restraint means were higher than BoC means, suggesting other self-restraint occurred before and after self-restraint at a higher rate than SIB, aggression and property destruction. Aggression and destruction SE bars mostly overlapped, suggesting it was very unlikely that there was a significant difference in the presence of both behaviours before and after self-restraint. Aggression and property destruction SE bars crossed zero, suggesting means did not differ significantly from zero, while SIB SE bars did not cross zero. This, alongside the small overlap of SIB SE error bars with aggression and property destruction bars, suggested that SIB shared a different temporal relation with self-restraint than aggression and property destruction. Finally, within all conditions SE bars mostly overlapped, suggesting it was very unlikely that there was a significant difference in the presence of SIB, aggression, destruction or other self-restraint before *or* after self-restraint.

Figure 5. 1 Mean percentage of time behaviours occurred before and after self-restraint



Note. Lines indicate standard error. Groups include participants who showed behaviour in previous month according to CBQ and displayed self-restraint during observation time (section 5.3.4).

In summary, 34% of all observations of self-restraint were preceded and followed by other self-restraint, and evidence suggests this was greater than the presence of BoC. In total, 12% of self-restraint observations were preceded by SIB, and 10% followed by SIB. Comparisons with other BoC suggested SIB shared a different temporal relation with self-restraint than aggression and property destruction.

5.4.3 Observed self-restraint intensity and putative correlates

5.4.3.1 Description of observation intensity scores

Observation intensity scores were calculated based on intensity proportions. Table 5.4 displays descriptive statistics of observation intensity scores. The total number of topographies identified by observation did not correlate with the total number of topographies identified by the SRQ, $r_s(N=125)=-.005$, $p=.957$. Internal consistency of observation intensity scores was assessed using McDonald's Omega (section 3.3.6). A coefficient of .367 suggested below-acceptable internal consistency. Coefficients did not change following removal of outliers (e.g., 'wears restraint device', code 24; Appendix 5.4). Figures 5.2-5.3 display histograms for observation intensity and SRQ intensity scores respectively. Visual inspection revealed a similar pattern of distribution between scores, with a slight positive skew. This indicated that smaller values occurred at higher frequencies than larger values. Importantly, the scale of distribution was much smaller for the observation intensity scores (maximum score = 8) in comparison with SRQ intensity scores (maximum score = 84).

Figure 5. 2 Histogram of total observation intensity scores

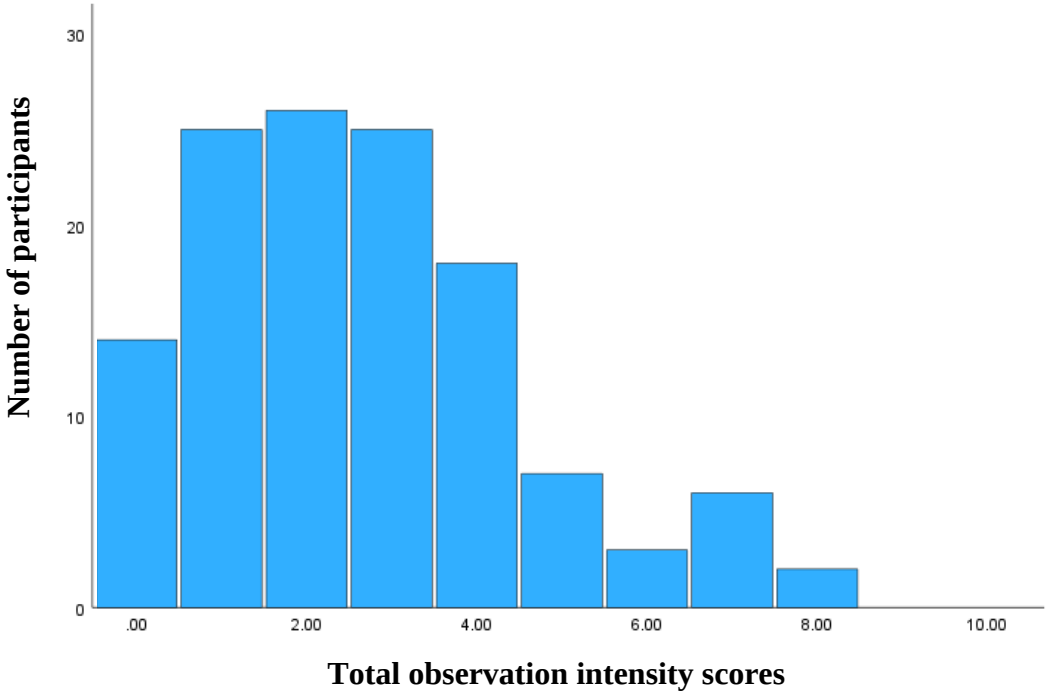
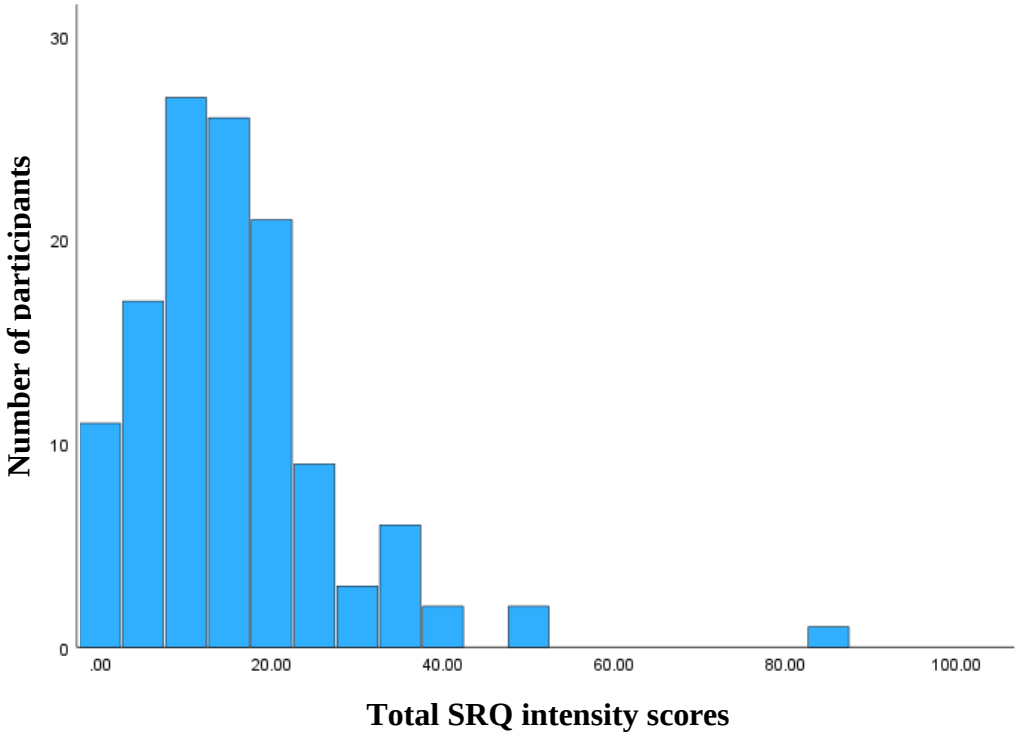


Figure 5. 3 Histogram of total SRQ intensity scores



5.4.3.2 Correlates of observed self-restraint intensity scores

Correlations between self-restraint scores and demographic, behavioural and cognitive variables are shown in Table 5.6. Results remain the same following exclusion of the outlier identified within the SRQ intensity distribution (*n*=1; Figure 5.3). Observation intensity and SRQ intensity scores did not significantly correlate, and a chi-square test revealed a non-significant association between scores, χ^2=237.720, *p*=.968.

Self-restraint observation intensity scores did not significantly correlate with demographic or behavioural variables. However, the medium effect between observation intensity scores and BPVS standardised scores was notable[34], suggesting greater self-restraint intensity (measured by observation) was linked to lower receptive language skills. Small-to-moderate positive correlations between observed self-restraint intensity and impulsivity and working memory indicated notable relations with EF.

SRQ intensity scores did not significantly correlate with demographic or behavioural variables, however small-to-moderate positive correlations were identified with SIB and property destruction severity and impulsivity.

In summary, self-restraint observation intensity scores were not significantly associated with SRQ intensity scores. SRQ intensity scores shared a closer relation with a range of EF scores than did observation intensity scores.

[34] See section 2.3.5 for effect size interpretation.

Table 5. 6 Correlations between self-restraint intensity scores and putative variables

		1	2	3	4	5	6	7	8	9
Self-restraint										
1	Observation intensity	—								
2	SRQ intensity	.054	—							
	N	*124*								
Demographics										
3	Age years	.117	-.109	—						
	N	*126*	*124*							
4	Gender	.001	-.005	.086	—					
	N	*125*	*124*	*125*						
5	Verbal ability	-.053	-.015	**.363***	.025	—				
	N	*125*	*124*	*125*	*125*					
6	Walk unaided	-.019	.062	.109	.031	**.266***	—			
	N	*125*	*124*	*125*	*125*	*125*				
7	SCQ total	.055	.050	.103	-.068	.053	-.009	—		
	N	*125*	*124*	*125*	*125*	*125*	*125*			
8	VABS-III ABC	-.166	.069	**-297***	.110	**.399***	.188	-.175	—	
	N	*125*	*124*	*125*	*125*	*125*	*125*	*125*		
9	VABS-III RL AE	-.105	-.076	**.418***	.104	**.661***	**.290***	-.128	**.505***	—
	N	*125*	*124*	*125*	*125*	*125*	*125*	*125*	*125*	
10	BPVS SS	-.236	.181	-.096	.021	.288		-.179	**.348***	.212
	N	*69*	*68*	*69*	*69*	*69*	-	*69*	*69*	*69*

Note. SRQ = Self-Restraint Questionnaire; SCQ = Social Communication Questionnaire; VABS-III ABC = Vineland Adaptive Behavior Scales, Third Edition Adaptive Behavior Composite; VABS-III RL AE = receptive language age equivalent; BPVS SS = British Picture Vocabulary Scale Standardised Scores; Dash (-) indicates variables that cannot correlate. *N*=1 outlier excluded from correlation matrix.
*Significant p-values after correcting for multiple tests in bold.

Table 5.6 cont.

		1	2	3	4	5	6	7	8	9	10	11	12	13	14	15
Behaviours of concern - CBQ																
11	SIB severity	.018	.165	.003	-.005	-.218	-.124	.175	**-.271***	**-.285***	-.056	—				
	N	*125*	*124*	*125*	*125*	*125*	*125*	*125*	*125*	*125*	*69*					
12	AGG severity	-.067	.145	.041	.030	-.020	-.052	.189	-.197	-.086	.041	**.598***	—			
	N	*125*	*124*	*125*	*125*	*125*	*125*	*125*	*125*	*125*	*69*	*125*				
13	DoP severity	.013	.216	.035	-.071	-.040	.026	.164	-.106	-.046	-.042	**.545***	**.622***	—		
	N	*125*	*124*	*125*	*125*	*125*	*125*	*125*	*125*	*125*	*69*	*125*	*125*			
14	STPY severity	.054	.099	-.103	-.127	**-.260***	-.086	.200	-.074	-.129	.040	**.266***	.163	**.267***	—	
	N	*117*	*116*	*117*	*117*	*117*	*117*	*117*	*117*	*117*	*66*	*117*	*117*	*117*		
Executive functions																
15	TAQ impulsivity	.169	.165	-.037	-.044	-.058	.051	**.285***	-.176	-.204	-.129	**.363***	**.414***	**.368***	.101	—
	N	*125*	*124*	*125*	*125*	*125*	*125*	*125*	*125*	*125*	*69*	*125*	*125*	*125*	*117*	
16	TAQ overactivity	.008	**.435***	-.204	-.076	-.174	.023	.105	-.012	-.170	-.076	**.298***	**.250***	**.366***	**.238***	**.545***
	N	*125*	*124*	*125*	*125*	*125*	*125*	*125*	*125*	*125*	*69*	*125*	*125*	*125*	*117*	*125*

Note. CBQ = Challenging Behaviour Questionnaire; SIB = self-injurious behaviour; AGG = aggression; DoP = destruction of property; STPY = stereotypy; TAQ = The Activity Questionnaire.
*Significant p-values after correcting for multiple tests in bold.

Table 5.6 cont.

		1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18	19
17	TAQ impulsive speech, *N*	.064 *73*	.265 *72*	-.047 *73*	.162 *73*	-	-	-.155 *73*	.157 *73*	.173 *73*	.192 *57*	**.036*** *73*	.176 *73*	.181 *73*	.069 *67*	**.347*** *73*	**.603*** *73*	—		
18	TAQ total scores, *N*	.037 *125*	**.373*** *124*	-.047 *125*	-.014 *125*	.113 *125*	.096 *125*	.122 *125*	.068 *125*	.032 *125*	.046 *69*	.253 *125*	**.301*** *125*	**.359*** *125*	.128 *117*	**.675*** *125*	**.897*** *125*	**.768*** *73*	—	
19	CHEXI inhibition, *N*	.077 *124*	.141 *123*	.148 *124*	.082 *124*	**.252*** *124*	.173 *124*	.077 *124*	.012 *124*	.159 *124*	.125 *69*	.017 *124*	.172 *124*	.199 *124*	-.011 *116*	**.487*** *124*	.372 *124*	**.417*** *73*	**.518*** *124*	—
20	CHEXI WM *N*	.117 *123*	.063 *122*	.222 *123*	.115 *123*	.047 *123*	.224 *123*	.108 *123*	-.169 *123*	-.043 *123*	.035 *68*	.107 *123*	.134 *123*	.181 *123*	.150 *115*	**.237*** *123*	**.211*** *123*	**.311*** *72*	**.111*** *123*	**.579*** *123*

Note. TAQ = The Activity Questionnaire; CHEXI = Childhood Executive Functioning Inventory. Dash (-) indicates variables that cannot correlate.
*Significant p-values after correcting for multiple tests in bold.

5.5 Discussion

This naturalistic observational study involving 126 autistic children with ID revealed three key findings. First, nearly 1500 observations of self-restraint were recorded across the visible footage, revealing a high prevalence and varying intensity of self-restraint both between and within the 24 topographies observed. Second, temporal investigations revealed that one third of all occurrences of self-restraint were preceded and followed by other self-restraint, while relatively few occurrences were preceded and followed by BoC. Finally, self-restraint observation intensity scores did not correlate with SRQ intensity scores, and the number of topographies observed and identified by informant-report did not correlate. No significant associations were identified between observation intensity scores and demographic, behavioural and cognitive variables. The theoretical and practical implications of these findings are now discussed.

Most of the sample (89%) displayed at least one form of self-restraint during the visible footage. On average, three different topographies were observed, while an average of nine topographies were identified by informant-report within the same sample. Alongside the lack of association between observed and informant-report intensity scores (discussed below), these findings suggest that the two approaches capture different phenomena. The SRQ examines a wide range of behaviours, indicated by the greater spread of scores, reflecting behaviours observed by the caregiver over-time. In contrast, observations record each instance of behaviour in fine-grained detail under specific conditions, accounting for the lower number of topographies observed. The most prevalent self-restraint topographies observed were ‘interlocks fingers/hands’ (48% of participants), ‘lies/sits in a position restricting arm movement’ (32%) and ‘folds arms’ (29%). Common topographies involved ‘using one’s own

body' to restrict movement and occurred at an elevated rate in comparison to existing literature (section 2.4.4). Differences in topographical prevalence may be explained by sample differences: the meta-analysis included a heterogeneous sample of individuals with autism and/or ID, where the overall prevalence estimate of self-restraint was 39% (section 2.4.3). The present study, however, observed self-restraint in 89% of a more homogenous sample of autistic children with ID. Thus, this finding may reflect an elevated prevalence of self-restraint and its topographies in autistic children with co-occurring ID, over-and-above individuals with autism *or* ID.

The intensity of observed self-restraint ranged from very low to very high between topographies. Some behaviours occurred at very low intensities, for example 'holds hands together above head' occurred for less than 1% of visible footage, with a mean intensity of 0.38%. Other topographies occurred at very high frequencies, for example the mean intensity of 'wears restraint device' was 99.84%. This was perhaps unsurprising, given the propensity for an individual to engage in mechanical restraint for prolonged periods of time (section 1.4.5.1). A crucial determinant of clinically concerning self-restraint is high or very high intensity of behaviour, as the risk of associated deleterious consequences, such as reduced physical and adaptive functioning, increases (section 1.4.2). Restraint fading, the systematic reduction of the restrictive properties of a restraint device, may be a suitable intervention to managing intense mechanical restraint.

Inspection of participant-level data revealed that some topographies occurred at varying intensities. For example, 'lies/sits in position restricting movement of arm(s)' ranged from very low (0.10%) to very high intensity (100%), while 'wraps blanket, sheet, etc, around self'

ranged from low (1.50%) to high intensity (97.60%). This finding reveals individual differences between participants. It is not possible to infer the function of behaviour based on these data alone, however within-topography intensity variation indicates that a person-centred approach should be adopted when assessing self-restraint in individuals who self-injure. This implication is supported by the experimental functional analysis literature, which identifies idiosyncratic functions of SIB and self-restraint between individuals (e.g., Kerth et al., 2009; Smith et al., 1992). It is important to note that very few children displayed high/very high intensity of self-restraint. Furthermore, many common topographies occurred at low intensities, for example although 'interlocks fingers/hands' was displayed by nearly half the sample, the mean intensity score was low (1.68%). This suggests that these behaviours, including prevalent topographies, are not behaviours of clinical concern. Currently, it is unclear whether a low intensity behaviour can develop into a high intensity behaviour over time, however operant learning models would support this possibility as experience and reinforcement shapes behaviour (section 1.4.7.2). Future research should therefore longitudinally assess the intensity of self-restraint to determine factors that contribute towards increasing intensity, and ultimately inform preventative strategies where these are indicated.

Tentative conclusions may be drawn regarding the temporal relations between BoC and self-restraint. Self-injury occurred at a higher frequency before (12%) and after (10%) self-restraint than aggression (2 and 3% respectively) and property destruction (3 and 2% respectively). Unlike SIB confidence intervals, aggression and property destruction confidence intervals crossed zero, suggesting SIB shared a relation with self-restraint that was different to other BoC. This preliminary finding counters previous results regarding BoC

(section 3.4.1.2) and may be explained by the unit of measurement. Observations of short time periods may capture different behaviours than broader informant ratings. Furthermore, the data in the present study suggest that it was very unlikely that self-injury (and other BoC) occurred at a different frequency before *or* after self-restraint, however conclusions are limited by the coding and analysis approach adopted (section 5.5.1). Negative reinforcement predicts that high levels of SIB will occur before self-restraint, but not necessarily after. Forman et al. (2002) observed significantly higher-than-chance SIB before *and* after self-restraint, referring to a hypothesis proposed by Romanczyk et al. (1992) to explain this finding. A 'vicious cycle' of behaviour is theorised to form, whereby self-restraint reduces arousal, and SIB functions to access the behaviour that leads to self-restraint (and subsequent reduced arousal). This behaviour is SIB itself (section 3.2). Further research using lag sequential analytic techniques is needed to investigate this theory.

Temporal investigations also revealed that one in three observations of self-restraint was preceded and followed by another observation of self-restraint. Evidence tentatively suggested that self-restraint occurred before and after other occurrences of self-restraint at a higher rate than BoC. This provides preliminary evidence that self-restraint is repetitive or compulsive in nature. Robbins et al. (2012) defined compulsive behaviours as repetitive and stereotypic. Previous informant-report literature has correlated self-restraint topography scores with repetitive and compulsive behaviours in individuals with neurodevelopmental conditions (section 3.4.1.2), and autistic individuals who self-restrain show significantly higher repetitive (but not compulsive) behaviour than those without self-restraint (Laverty et al., 2020). Therefore, this finding bolsters the adapted compulsive behaviour account proposed in section 3.5, implicating *self-restraint* itself as a compulsive behaviour over-and-above SIB. Further

research is required to examine the role of perseveration in autistic individuals with ID who do and do not self-restrain.

The prevalence of observed self-restraint (89%) was lower than informant-report self-restraint (96%). Observation intensity and informant-report intensity scores did not correlate with one another, warranting discussion. The meta-analysis in Chapter 2 indicated that differences in measurement technique (i.e., observational or questionnaire methods) did not significantly impact prevalence estimates, suggesting techniques capture a similar construct (i.e., topography; section 2.4.3.2). However, informant-report data may have been overrepresented in the meta-analysis. Dang et al. (2020) proposed that behavioural and self-report measures weakly correlate due to low reliability of behavioural methods. The present study demonstrated excellent inter-rater reliability at multiple stages to reduce error (e.g., under- or over-reporting behaviour). However, observation intensity scores yielded lower internal consistency than SRQ intensity scores (even after outliers were removed), and inspection of histograms revealed a lower presence of self-restraint identified by observation in comparison to informant-report. This may suggest that the observation period may not have been sufficient to detect a wide range and intensity of behaviours. Questionnaire measures cover a larger time frame, increasing likelihood of scoring across multiple items. Taken together, repeated observations at different times of day, or across multiple days is required to enhance reliability.

The lack of agreement between intensity scores may also be explained by scores capturing distinct constructs. Behavioural observations captured actual self-restraint intensity displayed on the day of observation, in structured conditions; playing games ranging in difficulty

(section 4.3.4.3) and unstructured free-play. The SRQ, however, captured caregivers' perception of children's behaviour displayed over the 'lifetime'. As such, SRQ intensity is better conceptualised as a caregivers' 'mental average' of self-restraint displayed across structured and unstructured settings (Dang et al., 2020). Informant-report measures rely on recall of past behaviour, and the risk of inaccurate recall (or 'recall bias') is exacerbated when implementing a long recall period (Althubaiti, 2016). On this basis, SRQ intensity may be more representative of typical self-restraint intensity over time and settings, while observation intensity reflects actual intensity under particular conditions. Furthermore, observation intensity scores did not correlate with any putative correlates, including age, SIB severity or impulsivity. However, while non-significant, SRQ intensity shared small-to-moderate effect sizes with SIB severity and impulsivity, and a significant relation with measures of activity. This replicated previous informant-report based findings (e.g., Richards et al., 2017) and suggests that SRQ intensity taps into the components of self-restraint studied within the wider literature.

There are clinical implications of these findings. Given its informant-report nature and links with self-injury and impulsivity, SRQ intensity scores may be best utilised in a preventative way at early emergence of SIB and self-restraint. SRQ total scores may be used as a quick and efficient technique to assess and continually monitor self-restraint intensity in conjunction with monitoring of self-injury. Elevated or increasing intensity over time may indicate a maladaptive relation between SIB and self-restraint has formed. Taken further, it is reasonable to suggest that the SRQ should be employed *before* self-injury emerges as a preventative strategy for high-risk populations for intense SIB and/or self-restraint (i.e., autism and ID). High levels of SRQ intensity in the absence of SIB may indicate potentially challenging

behaviour requiring investigation. Future research employing the SRQ in samples who do not self-injure, and assessing links with putative variables, is needed. Furthermore, to reduce recall error, it may be advisable to shorten the recall period of the SRQ, for example requesting that informants indicate behaviours shown 'in the previous month' or 'since self-injury emerged' (Althubaiti, 2016).

In contrast, observation intensity scores may be best utilised *after self-injury* has emerged. For example, in situations where SIB naturally occurs, a short observation of SIB *and* self-restraint (e.g., 10 minutes) may be suitable. Examination of the presence and intensity of self-restraint observed before or after self-injury would provide vital information around the role of behaviour. For example, high intensity of self-restraint following self-injury may suggest that self-restraint occurs as an attempt to escape from SIB, although this can only be confirmed with experimental functional analysis. To further understanding, future research directions include conducting a longitudinal intervention with an aim of reducing SIB (e.g., Functional Communication Training), while assessing the stability of self-restraint intensity.

5.5.1 Limitations

It is vital to acknowledge three main limitations of this study. First, the majority of participants were recruited through accessing National Health Services (65%), indicating possible sample bias towards individuals at higher risk of self-injury (section 4.3.2). However, to test theories and observe self-restraint and SIB, it was essential to recruit participants for whom such behaviours were present. Furthermore, varying intensity of self-restraint was observed, from very low to very high, indicating a more balanced sample than previous literature (e.g., when self-restraint occurred 96% time; Forman, 2003).

Second, to prioritise participant safety, safeguards were in place to prevent severe self-injury (and potentially by consequence, self-restraint). For example, pre-visit checks and risk assessments of BoC were conducted with caregiver(s) prior to testing, and sessions were terminated at the child, caregiver or researcher's discretion once any severe or unusual BoC began (section 4.3.5). Consequently, the prevalence of observed BoC may be under-representative of typical behaviour or may reflect contingencies present at lower levels of behaviour severity.

Finally, due to the coding approach, there was overlap in the number of BoC recorded, and not every occasion of SIB, aggression or property destruction was coded (only those observed 30 seconds prior to and following self-restraint). Therefore, lag sequential or contingency analysis, which would be able to provide the transitional probability of behaviours occurring before and after self-restraint, was not appropriate for these data (i.e., such analyses require frequency counts of BoC occurring the *absence* of self-restraint). Future research therefore should allow for such analyses by both accounting for 'over-coding' of behaviours, and coding for BoC throughout the entire video footage (i.e., not only in the 30 seconds prior to or following self-restraint).

5.5.2 Conclusion

In summary, the prevalence of self-restraint, both identified by informant-report and observation, was higher than previous studies, implicating autistic children with ID as a high-risk population for self-restraint. Varying intensities of self-restraint topographies indicated individual differences drive intensity of behaviour, favouring an idiosyncratic approach to understanding self-restraint and SIB. Many self-restraint behaviours were preceded and

followed by more self-restraint, indicating a compulsive and/or repetitive nature of behaviour. Further investigation is needed to unpack this novel finding. Finally, differences in informant-report and behavioural measures of intensity implicate unique clinical utility of each measure dependent on the emergence of SIB and/or self-restraint.

CHAPTER SIX

SELF-RESTRAINT, SELF-INJURIOUS BEHAVIOUR AND EXECUTIVE FUNCTIONS

6.1 Preface

Self-restraint is considered to emerge as a learned behaviour in response to self-injury. The observational data in Chapter 5 suggested that self-injury shares a different temporal relation with self-restraint than other behaviours of concern, however the cross-sectional data in Chapters 3 and 5 indicate aggression and property destruction are related to self-restraint. Hypotheses of impaired behavioural control suggest self-restraint emerges to compensate for 'uncontrollable' self-injury. Chapters 3 and 5 implicated compulsivity and impulsivity as important correlates of self-restraint, however findings are limited by the bounds of informant-rated methodology. Thus, using the developmentally-appropriate methods described in Chapter 4, this Chapter profiles self-restraint in autistic children with intellectual disability, and examines the relation between self-restraint and behaviours of concern. Chapter 6 aims to extend current understanding by employing novel direct, play-based measures of executive function. Finally, a stringent, multi-parameter definition of self-restraint was applied to capture intense, preferred and clinically concerning behaviours.

6.2 Introduction

Self-restraint is prevalent in conditions where self-injurious behaviour (SIB) is common, including within individuals with neurogenetic conditions (section 3.4.1), idiopathic autism (Laverty et al., 2020; Richards et al., 2017), intellectual disability (ID; Oliver et al., 2003) and co-occurring autism and ID (section 5.4.1). It is important to identify topographies of self-restraint that occur at high intensity and are associated with distress and/or self-injury due to physical health and adaptive functioning (section 1.4.2). Impaired behavioural control, evidenced by deficits in impulsivity (and broader executive functions [EF]) and compulsivity, is theorised to underpin self-restraint and SIB (King, 1993; Oliver & Richards, 2015). However understanding of self-initiated body restriction has been limited by inconsistent definitions of self-restraint and a reliance on indirect measures of EF. The overarching aim of this Chapter, therefore, was to examine associations between self-restraint defined using multiple parameters, SIB and direct (i.e., performance-based) and indirect (i.e., informant-report) measures of EF.

6.2.1 Continuous and categorical approaches to self-restraint measurement

Self-restraint topography (section 1.4.5.1) and intensity (section 1.4.5.2) are measured by the Self-Restraint Questionnaire (SRQ; Oliver et al., 2003). Previous research using the SRQ has adopted a grouping approach whereby individuals who score above zero are categorised as showing 'self-restraint'. However, Oliver et al. (2003) argued that the presence of self-restraining behaviours alone may not be concerning; rather the presence of additional parameters of behaviour, such as seeking self-restraint most or all the time (i.e., high intensity), indicates self-restraint of clinical concern. This is in line with transdiagnostic neurodevelopmental research that favours continuous, severity-based measures as opposed to

dichotomous categories (section 3.2; Astle et al., 2022). A factor analysis of SRQ items (section 4.4.1) suggested that continuous total scores reflecting 'SRQ intensity' can be measured reliably using total scores. This use of the SRQ has not yet been evaluated in relation to correlates of self-restraint, and so thus far cannot be used to inform models of mechanism that may underpin behaviour.

The Self-Restraint Questionnaire 2 (SRQ2; Forman, 2003) builds upon the SRQ to measure an additional parameter: preference for self-restraint (section 1.4.5.3). The SRQ2 further defines self-restraint as self-initiated and incompatible with self-injury (section 1.4.4). Individuals who fulfil restrictive SRQ2 criteria for 'multi-parameter self-restraint' may be at highest risk of developing 'vicious cycles' of behaviour with self-injury (section 1.4.7.2), as the presence of multiple parameters of self-restraint might indicate maladaptive behaviour that constrains adaptive functioning and interferes with activities of daily living (Oliver et al., 2003). The present study also used SRQ2 groupings to describe phenomena and test theoretical accounts.

There is a lack of consensus around the demographic characteristics associated with self-restraint. Identifying risk markers for severe self-restraint is vital for the prevention and treatment of the negative consequences of intense, preferred and persistent self-restraint (section 1.4.2). Studies defining self-restraint according to topography, for example using the Self-Restraint Checklist (SRC; Powell et al., 1996), suggest younger age (r_s=-.22; section 3.4.1.2), the presence of autism (r_s=.26; section 3.4.1.2), lower self-help skills (i.e., ability; relative risk statistic [RR]=1.54; Richards et al., 2017) and lower receptive language skills (r_s=-.24; section 5.4.3.2) are associated with self-restraint. Forman (2003) furthered

understanding by using SRQ2 groupings (i.e., accounting for topography, intensity, preference for behaviour, self-initiation of behaviour and incompatibility with SIB/stimulus control). Findings revealed individuals in the 'self-restraint' group were significantly younger and showed more severe SIB than those in the 'no self-restraint group' (Forman, 2003). No group differences were identified for gender or adaptive functioning. To date, the SRQ2 has been used with individuals with ID, of whom 30% had co-occurring autism (Forman, 2003). Therefore, the first aim of this study was to describe the prevalence, topographies and demographic correlates of self-restraint (measured using continuous severity scores and multi-parameter categorical groupings) in autistic children with ID.

6.2.2 Self-restraint and behaviours of concern

The relation between SIB and self-restraint is complex. Cross-sectional research suggests that the presence of self-injury increases the likelihood of self-restraint. For example, Richards et al. (2017) found the presence of severe SIB significantly increased risk of self-restraint in autistic children (RR=1.77) and adults (RR=2.17). The meta-analysis reported in Chapter 2 found a moderate positive correlation between SIB and self-restraint, r=.21 (section 2.4.5). A negative reinforcement account, however, predicts that individuals who self-restrain should show *lower* rates of self-injury, as the self-restraint functions to reduce (and is reinforced by a reduction in) SIB (section 1.4.7.2). Experimental functional analyses have supported this theory by reporting a decrease in self-injury when self-restraint is available (e.g., Forman et al., 2002). However, a large-scale naturalistic observational study of autistic children with ID reported no differences between the presence of SIB before *or* after self-restraint (section 5.4.2). Therefore, the relation between self-restraint and SIB varies according to study design and self-restraint measurement. Research is yet to examine the relation between self-restraint

and SIB using stringent, multi-parameter definitions of self-restraint (i.e., SRQ *and* SRQ2 within the same sample).

Previous literature suggests that self-restraint is unrelated to other behaviours of concern (BoC; Fisher & Iwata, 1996; section 3.2). Observations of autistic children with ID support this theory; Chapter 5 revealed aggression and property destruction were unlikely to occur before or after self-restraint (section 5.4.2). However, examination of BoC in a large sample of individuals with neurodevelopmental conditions revealed that self-restraint (measured by number of topographies) correlated with aggression, r_s=.31, property destruction, r_s=.20, and stereotypy[35], r_s=.33 (section 3.4.1.2). These contradictory findings may arise from differences in measurement (i.e., behavioural coding versus informant-report questionnaires), warranting further investigation using a multi-parameter definition of self-restraint. Therefore, the second aim of this study was to examine the relation between self-restraint (measured using continuous severity scores and multi-parameter categorical groupings), self-injury and other BoC in a large sample of autistic children with ID.

6.2.3 Self-restraint and executive functions

Not all individuals who self-injure also self-restrain (13%; section 2.4.5). Individual differences may be explained through differences in EF. The compulsivity (King, 1993) and inhibitory control accounts (Oliver & Richards, 2015) propose that compulsivity (i.e., tendencies to perform repetitive actions) and impulsivity (i.e., difficulties withholding from initiating an action; section 1.4.7.3), may indicate difficulties with behavioural control,

[35] For clarity, stereotypy is termed a 'BoC' throughout this Chapter, and is measured as such by the Challenging Behaviour Questionnaire. It is acknowledged that stereotypic behaviours such as 'rocking' may not be considered as concerning as self-injurious, aggressive or destructive behaviours.

leading to self-injury. Self-restraint is theorised to emerge as a compensatory tool to regain behavioural control. Chapter 3 implicated compulsivity as a transdiagnostic risk marker for self-restraint, and from these data an adapted compulsive account of behaviour was proposed, implicating self-restraint *itself* as a compulsive behaviour that emerges to manage anxiety that results from impulsive SIB (section 3.5). Within the heterogeneous sample of individuals with autism and/or ID, impulsivity and self-restraint correlated, however impulsivity was not uniquely associated with self-restraint after adjusting for potential confounds (section 3.4). Importantly, existing studies utilised a topography-based definition of self-restraint and a multi-parameter measure of self-restraint has not yet been adopted to test the compulsive and inhibitory control accounts. Furthermore, current understanding of the role of impulsivity and compulsivity in SIB and self-restraint is limited by an exclusive use of indirect (i.e., informant-report questionnaire-based) methods to measure EF.

Indirect measures of EF are thought to measure caregivers' perceptions of 'typical' performance, or *ability* to perform (Dang et al., 2020). Reflecting the wider developmental literature, links between indirect and direct measures of EF within autistic individuals and/or those with ID are unclear, further emphasising the need for developmentally-appropriate, direct methods. Direct measures capture performance under controlled, standardised conditions and are driven by the individual's motivation, effort and willingness to perform within the situation (Dang et al., 2020; Toplak et al., 2013). In a review drawing on the attention-deficit/hyperactivity disorder (ADHD) literature, Toplak et al. (2013) found that only 24% of the 286 correlations between direct and indirect measures of EF were statistically significant, ranging from very small to moderate effects (median r=.19). Low agreement suggests direct and indirect measures capture different aspects of EF, however it is unclear

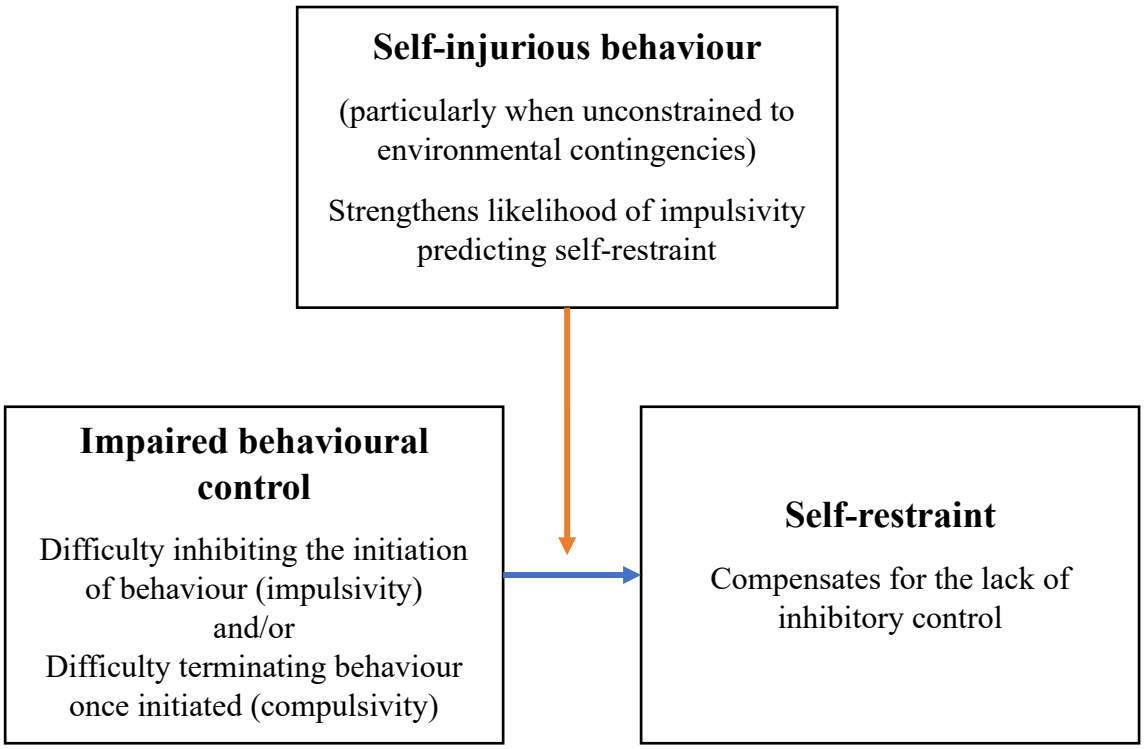
whether findings generalise to populations where SIB and self-restraint are present. Therefore, this study examined the correlations between direct and indirect measures of EF within a sample of autistic children with ID.

Altogether, to understand the role of executive control in self-injury and self-restraint, there is a need to go beyond indirect measures and examine EF directly using suitable tasks. Therefore, the third aim of this study was to examine the relation between self-restraint and direct and indirect measures of EF, including impulsivity, working memory (including a measure of perseveration, conceptualised to represent compulsivity) and cognitive flexibility.

Collectively, existing findings suggest deficits in impulse control may drive BoC, however no research has yet attempted to unpick the complex relation between self-injury, self-restraint intensity and broader components of EF. Richards et al. (2017) reported impulsivity/overactivity predicted the presence of self-restraint in autistic children (RR=2.35), while a cross-syndrome study found compulsivity predicted self-restraint in a large sample of individuals with neurogenetic conditions (section 3.4.2). While these data bolster theoretical accounts, it is unclear whether impulsivity and compulsivity operate in isolation in the presentation of SIB and self-restraint, or whether accounts may be complementary. Hypotheses of impaired inhibitory control and compulsivity propose that some individuals have less control over behaviour, and thus self-restraint emerges to compensate for the presence of 'undesirable' (and uncontrollable) behaviour. It is therefore expected that the presence of SIB moderates the relation between impulsivity and/or compulsivity (i.e., perseveration) and self-restraint, such that impulsivity/compulsivity will be more strongly associated with self-restraint in the context of SIB (Figure 6.1). No study has yet attempted to

examine this relation using direct and indirect measures of EF, despite the potential for implications to be far-reaching (e.g., beneficial use of EF training for BoC). It is important to include individuals at high risk of SIB and self-restraint to better understand potential drivers of behaviour. Therefore, the final aim of this study was to identify whether SIB severity moderated the relation between direct and indirect measures of EF (including impulsivity and compulsivity) and self-restraint intensity.

Figure 6. 1 Hypothesised relation between inhibitory control, self-restraint and self-injury



Note. Blue arrow represents hypothesised predictive relation, orange arrow represents hypothesised moderation.

6.2.4 Summary and aims

In summary, this study had four aims: (1) describe the prevalence, topographies and demographic correlates of self-restraint in autistic children with ID, (2) examine the relation between self-restraint and BoC, (3) examine the relation between self-restraint and direct and indirect measures of impulsivity, compulsivity and broader EFs, and examine the relations between direct and indirect measures of EF, and (4) identify whether SIB severity moderated the relation between self-restraint and impulsivity and/or compulsivity. Comparisons between SRQ intensity scores and SRQ2 multi-parameter groupings are examined within aims one, two and three.

6.3 Methods

6.3.1 Recruitment, participants and data collection

Participants included in this study were recruited using the procedure outlined in Chapter 4 (see section 4.3.2 for recruitment, section 4.3.5 for data collection). The full survey sample (*N*=171) is described in Chapter 4 (section 4.3.3 displays demographic characteristics and section 4.4.1 displays behavioural outcomes). Similarly, the final sample for the direct EF tasks (*N*=126) is described in Chapter 5 (section 5.3.1 displays demographic characteristics, section 5.4.1 displays behavioural characteristics).

Table 6.1 displays demographic characteristics of the SRQ subsample (*N*=165). Demographic characteristics of the SRQ2 subsample are reported in Appendix 6.1.

6.3.2 Measures and coding

Measured used to assess self-restraint, SIB and EF are described in full in section 4.3.4, alongside dependent variables extracted and calculation of EF composite scores.

Table 6. 1 Demographic characteristics of SRQ sample

Developmental variables	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Chronological age (years)	165		10.08 (10)	4.02 – 15.49 (2.86)
Sex (female/male/non-binary)	165	38[a]/126/1 (23/76/1)		
Mobility (walk unaided/unable)	164	157/7 (96/4)		
SCQ total (all participants)	165		27.18 (28)	15 – 37 (5.05)
SCQ total verbal		100 (61)	27.49 (28)	15 – 37 (5.82)
SCQ total non-verbal		65 (39)	26.69 (27)	20 – 32 (3.56)
Autism description	163			
Autism		80 (49)		
Autism spectrum disorder		73 (45)		
High functioning autism		4 (2)		
Autistic spectrum		3 (2)		
Asperger syndrome		2 (1)		
Pervasive dev. disorder		1 (1)		
VABS-III ABC score	142		42.24 (40)	20 – 87 (14.41)
VABS-III receptive language age equivalent (months)	142		19.53 (19)	0 – 67 (9.99)
ID severity	163			
Mild		11 (7)		
Moderate		50 (31)		
Severe		69 (42)		
Profound		7 (4)		
Unknown		25 (15)		
Other		1 (1)		
BPVS raw score [b]	69		70.51 (69)	2 – 146 (39.48)
BPVS standardised score [c]	69		74.22 (69)	69 – 118 (10.85)
BPVS age equivalent (months) [d]	40		84.98 (75)	45 – 162 (28.83)

Note. SCQ = Social Communication Questionnaire; VABS-III ABC score = Vineland Adaptive Behavior Scale, Third Edition Adaptive Behaviour Composite; BPVS = British Picture Vocabulary Scale.

[a] *n*=1 self-described differently to gender assigned at birth. [b] BPVS discontinued for *n*=46 and not attempted for *n*=10 due to child engagement or for safety. [c] Includes *n*=49 who scored below average of a normed sample (70), indicated as '69'. [d] Age equivalents incalculable for *n*=29 as raw scores fell below normed sample scores.

6.3.3 Data analysis

Analyses were conducted using SPSS (v.29). EF task missingness ranged from 12 to 72% (section 4.4.2.1) and missingness was related to age, verbal ability, mobility, VABS-III ABC scores and SIB severity scores. To maximise the sample size and obtain better parameter estimates, multiple imputation was performed (Woods et al., 2021). Data were considered missing not at random. Predictor variables included demographics related to missingness, and additional predictors theorised to be related to self-injury for which there were no missing data (Gastric Distress Questionnaire[36] total score, Social Communication Questionnaire [SCQ] score, The Activity Questionnaire [TAQ] impulsivity and overactivity scores). A total of 20 imputed datasets were created (Appendix 6.2 displays SPSS syntax). Imputed variables were direct composite scores (i.e., prohibition, working memory efficiency and perseveration, conflict, and no-conflict scores) and BPVS standardised scores. Thus, analyses including imputed variables represent the pooled (i.e., average) result from the 20 imputed datasets. Henceforth, unless stated otherwise, all analyses involving the direct EF composite variables used the imputed dataset.

Data normality of non-imputed variables (i.e., questionnaire and VABS-III data) was inspected using Shapiro-Wilk and Kolmogorov-Smirnov statistics. Non-parametric tests were employed where data were non-normal ($p<.05$). Where data were non-normal but the sample size was relatively large (i.e., >100) parametric t-test and linear regression analyses were considered appropriate (Lumley et al., 2002). Owing to SPSS capabilities, data normality of imputed variables was inspected through skew and kurtosis. Z values, calculated by dividing

[36] The Gastric Distress Questionnaire (GDQ; Oliver & Wilkie, 2005) examines behavioural markers of gastric distress displayed by individuals with ID, and was administered as part of the larger battery of Sleep-Impulsivity-Behaviour study questionnaires (section 4.3).

pooled values by the standard error, all lay between +/-3.29, suggesting normality (Kim, 2013).

The first aim was to describe the prevalence, topographies and demographic correlates of self-restraint. SRQ and SRQ2 descriptive statistics were reported, and differences in SRQ intensity scores between children who did and did not show current SIB were compared using t-tests. The proportion of individuals reported to show each self-restraint topography was compared across the SIB groups, and statistical associations examined using chi-square tests (χ^2).

The prevalence of each self-restraint topography was compared across the four SRQ2 groups ('no self-restraint/no SIB', 'no self-restraint/yes SIB', 'yes self-restraint/no SIB', and 'yes self-restraint/yes SIB'), and group differences examined using Fisher-Freeman-Halton exact tests, an extension of Fisher's exact test (Fisher, 1922) that allows for comparison across three or more groups (i.e., contingency table larger than 2x2).

Associations between SRQ intensity scores and continuous demographic variables were examined using Spearman's rho (r_s) and relations with dichotomous variables were examined using point-biserial correlations. Differences between the self-restraint and no self-restraint SRQ2 groups were examined using Mann-Whitney U and chi-square tests. Mann-Whitney U effect sizes were obtained by dividing the square root of N by the Z score. Across correlations and tests of group differences, the Benjamini-Hochberg method was used to control for multiple comparisons (section 3.3.6).

The second aim was to examine the relation between self-restraint and self-injury, and other

BoC. Relations between SRQ intensity scores and CBQ severity scores were examined using Spearman's rho. Differences in SIB severity scores between the 'self-restraint' and 'no self-restraint' SRQ2 groups were examined using Mann-Whitney U tests.

The third aim was to examine the relation between self-restraint and direct/indirect measures of EF. Relations between SRQ intensity scores and EF outcomes, intercorrelations between direct and indirect measures of EF, and relations between EF outcomes and demographic variables were examined used Spearman's rho. Differences in EF outcomes between the two SRQ2 groups ('self-restraint' and 'no self-restraint') were examined using Mann-Whitney U and t-tests. Differences in EF outcomes between the four SRQ2 groups were examined using Kruskal-Wallis analysis of variance (ANOVA) and one-way ANOVAs.

Finally, to examine the moderating role of SIB severity in the relation between self-restraint and EF, moderation analyses were performed using Hayes (2017) PROCESS Macro. Model 1 was computed using 5000 bootstrap samples to increase reliability and validity of results (Fritz & MacKinnon, 2008). The independent and moderating variables were mean-centered, to reduce multicollinearity. The level at which the moderating variable was significant (low, average or high) was identified by slope analysis (Aiken et al., 1991).

6.4 Results

6.4.1 Prevalence and profile of self-restraint

6.4.1.1 SRQ intensity and self-injury

Table 6.2 displays the behaviour characteristics of the study sample (Appendix 6.3 displays information for the SRQ2 subsample). Table 6.3 reports descriptive statistics for the imputed direct EF composite scores.

Table 6. 2 Behavioural characteristics of SRQ sample

Behavioural variables	**Possible score range**	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Self-restraint - SRQ		165			
Monthly self-restraint/no SR			158/7 (96/4)		
Total SRQ intensity score	0 – 92			16.20 (14)	0 – 84 (12.15)
Number of topographies	0 – 23			9.33 (9)	0 – 23 (5.49)
1 topography/2+ topographies			6/152 (4/96)		
Behaviours of concern – CBQ					
Monthly SIB/no current SIB		165	126/39 (76/24)		
Total SIB severity score	0 – 14			6.28 (7)	0 – 14 (4.36)
Monthly AGG/no current AGG		165	124/41 (75/25)		
Total AGG severity score	0 – 14			5.90 (6)	0 – 14 (4.17)
Monthly DoP/no current DoP		165	122/43 (74/26)		
Total DoP severity score	0 – 14			5.80 (7)	0 – 14 (4.23)
Monthly STPY/no current STPY		156	131/25 (84/16)		
Total STPY severity score	0 – 14			7.17 (8)	0 – 14 (3.93)

Note. SRQ = Self-Restraint Questionnaire; SR = self-restraint; CBQ = Challenging Behaviour Questionnaire; SIB = self-injurious behaviour; AGG = aggression; DoP = destruction of property; STPY = stereotypy.

Table 6.2 cont.

Behavioural variables	**Possible score range**	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Activity – TAQ					
Impulsivity subscale	0 – 24	164		20.44 (22)	0 – 24 (4.30)
Overactivity subscale	0 – 36	165		24.60 (27)	0 – 36 (8.68)
Impulsive speech subscale	0 – 12	98		6.12 (7)	0 – 12 (4.13)
Total score	0 – 72	164		48.60 (51)	9 – 72 (13.46)
Executive functions – CHEXI					
Inhibition factor	0 – 55	158		46.48 (48)	11 – 55 (7.13)
Working memory factor	0 – 65	157		54.22 (55)	13 – 65 (8.30)

Note. TAQ = The Activity Questionnaire; CHEXI = Childhood Executive Functioning Inventory.

Table 6. 3 Descriptive statistics for direct imputed composite scores (N=126)

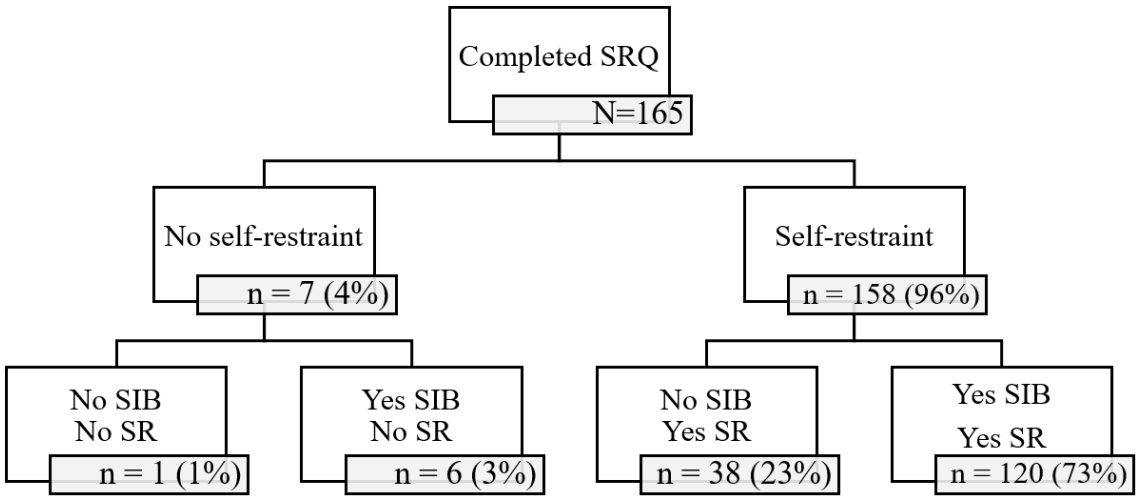
Composite score	***N* (%)**	**Pooled mean (SE)**	**Range**
Inhibition			
Prohibition score		0.75 (0.08)	0.00 – 2.00 [a]
Failed both trials	66 (52)		
Passed one trial	26 (21)		
Passed both trials	34 (27)		
Working memory			
Efficiency score		0.73 (0.03)	0.01 – 1.00
Perseveration score		0.13 (0.02)	0.00 – 0.75
Cognitive flexibility			
Conflict score		1.83 (0.40)	0.00 – 3.00
No-conflict score		1.73 (0.19)	0.00 – 3.00
BPVS standardised scores		75.68 (1.06)	69 – 118

Note. SE = Standard error; BPVS = British Picture Vocabulary Scale. Pooled means generated from the average of 20 imputed datasets.

[a] Imputed prohibition composite scores ranged from zero to two to allow for analysis: zero = failed both prohibition tasks, one = failed either wand *or* array task, two = passed both tasks.

A total of 165 caregivers completed the SRQ. Most participants (96%) were reported to self-restrain. Of this subsample, 73% showed current SIB (Figure 6.2). Of the 165 survey respondents, 126 children (76%) were reported to self-injure in the past month ('SIB group') and 39 (24%) did not ('no SIB group'). Children who showed monthly SIB had significantly higher SRQ intensity scores, M=17.33, SD=12.37, than the no SIB group, M=12.56, SD=10.77, $t(163)$=-2.163, p=.032. The effect size, Cohen's d=.411, was small-to-medium (Cohen, 1988). Therefore, children who self-injured were reported to show greater self-restraint intensity in comparison to those who did not self-injure.

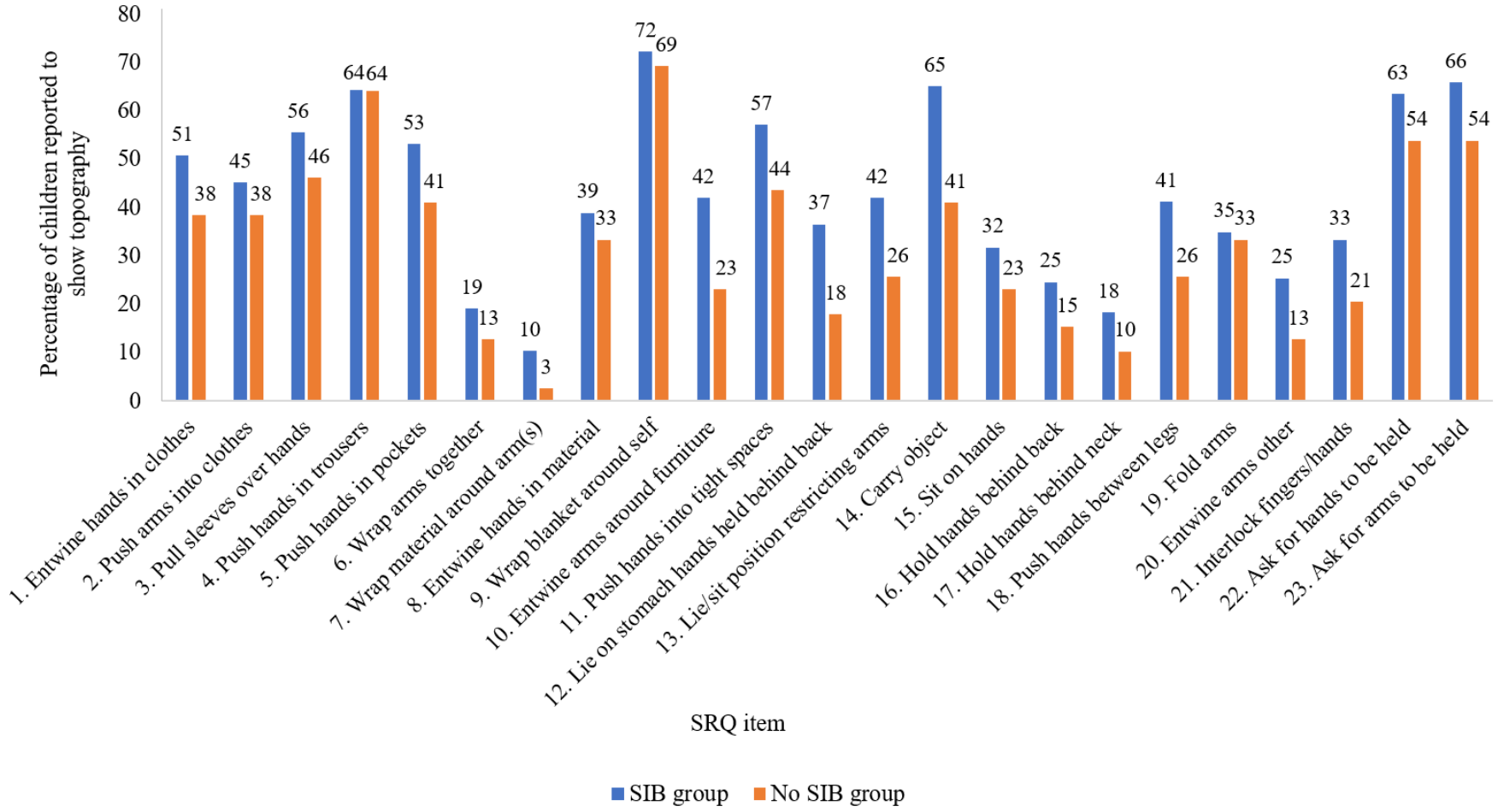
Figure 6. 2 Self-restraint and self-injury prevalence according to SRQ



Note. SRQ = Self-Restraint Questionnaire; SR = self-restraint; SIB = self-injurious behaviour.

Figure 6.3 depicts the proportion of children showing each self-restraint topography within the SIB (*N*=126) and no SIB (*N*=39) groups. All 23 topographies were observed in both groups. Children who showed SIB displayed a higher prevalence of most self-restraint topographies compared to the no SIB group, except for 'pushes hands in trousers', which was displayed by 64% of both groups. The most prevalent behaviour across both groups was 'wraps blanket around self' (72% SIB group, 69% no SIB group). The least prevalent behaviour was 'wraps material around arm(s)' (10% and 3% respectively). Amongst children without SIB, the prevalence of some self-restraint topographies was relatively high (e.g., wrapping material around self, 69%, pushing hands in trousers, 64%, and asking for arms or hands to be held, both 54%). A series of chi-square tests of independence revealed no significant associations between the SIB and no SIB groups on the presence of self-restraint topographies after correcting for multiple comparisons (Appendix 6.4).

Figure 6. 3 Prevalence of self-restraint topographies in children who did and did not show self-injury



6.4.1.2 SRQ2 groups and self-injury

A subsample of participants completed the SRQ2 (*n*=69, 42% of SRQ sample). Figure 6.4 displays the number of children reported by the SRQ2 to self-restrain and/or self-injure. Of this subsample, the prevalence of self-restraint when measured by the SRQ was 90%. When measured by the more stringent SRQ2 criteria, self-restraint prevalence lowered to 57%.

Of the children in the 'self-restraint' SRQ2 group, 56% (*n*=22) were reported to show monthly SIB while 44% (*n*=17) did not self-injure. Of this latter group, six children had reportedly showed SIB within the lifetime, while 11 had 'never' shown any SIB. Therefore, the SRQ2 identified a small sub-sample of participants who self-restrained according to multiple parameters in the absence of self-injury.

The prevalence of each self-restraint topography according to the four SRQ2 groups is displayed in Figure 6.5. According to this more stringent, multi-parameter definition of self-restraint, children who showed both self-restraint and SIB displayed the highest prevalence of 18 self-restraint topographies. Children who did not self-restrain nor self-injure demonstrated the highest prevalence of the remaining four behaviours: 'pushes hands in trousers', 'asks for arms to be held' (both 80%), 'pushes hands in pockets' and 'asks for hands to be held' (both 60%). A series of Fisher-Freeman-Halton exact tests revealed no statistically significant group associations on the presence of self-restraint topographies after correcting for multiple comparisons (Appendix 6.5).

Figure 6. 4 Prevalence of self-restraint according to the SRQ2

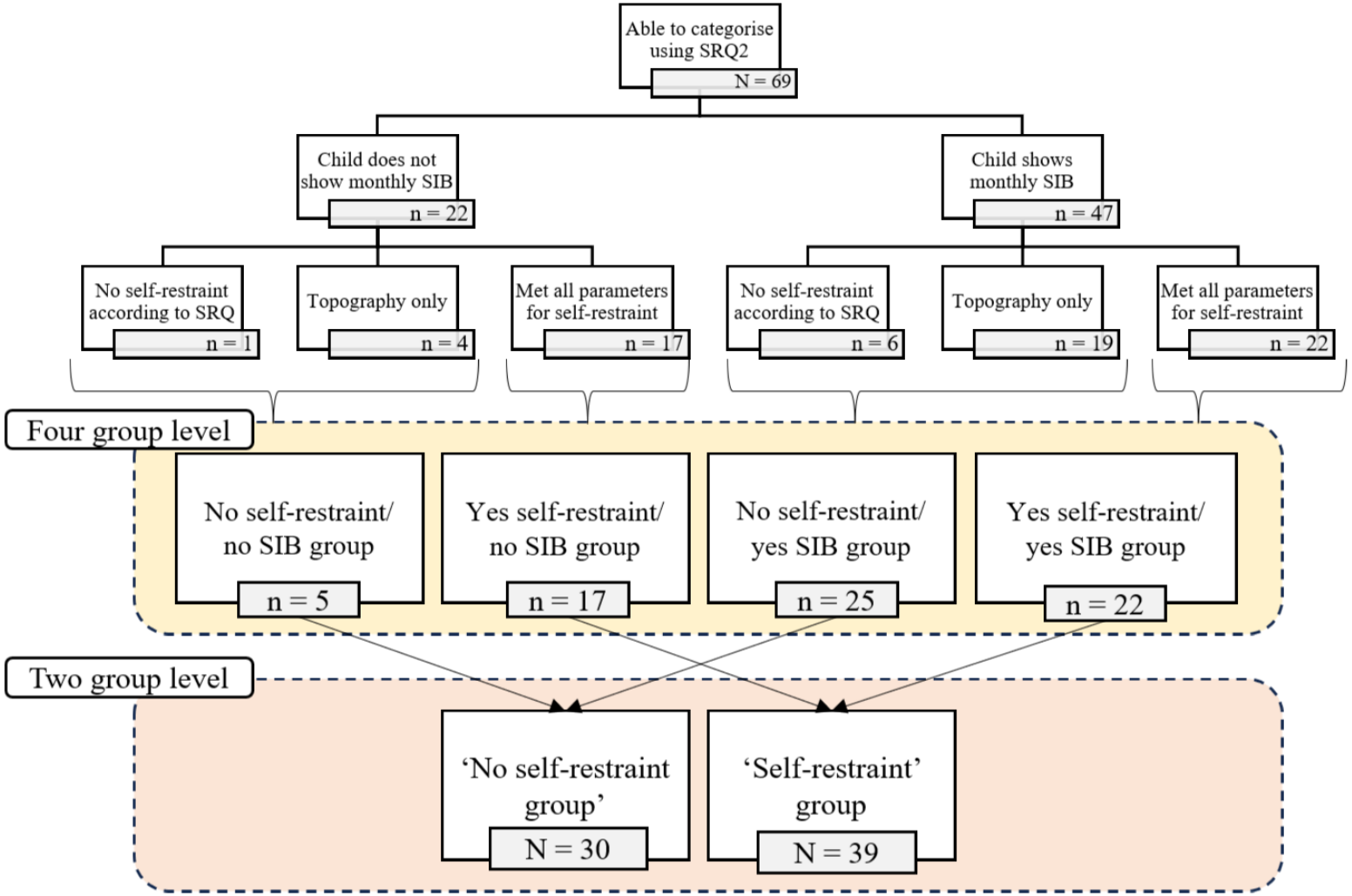
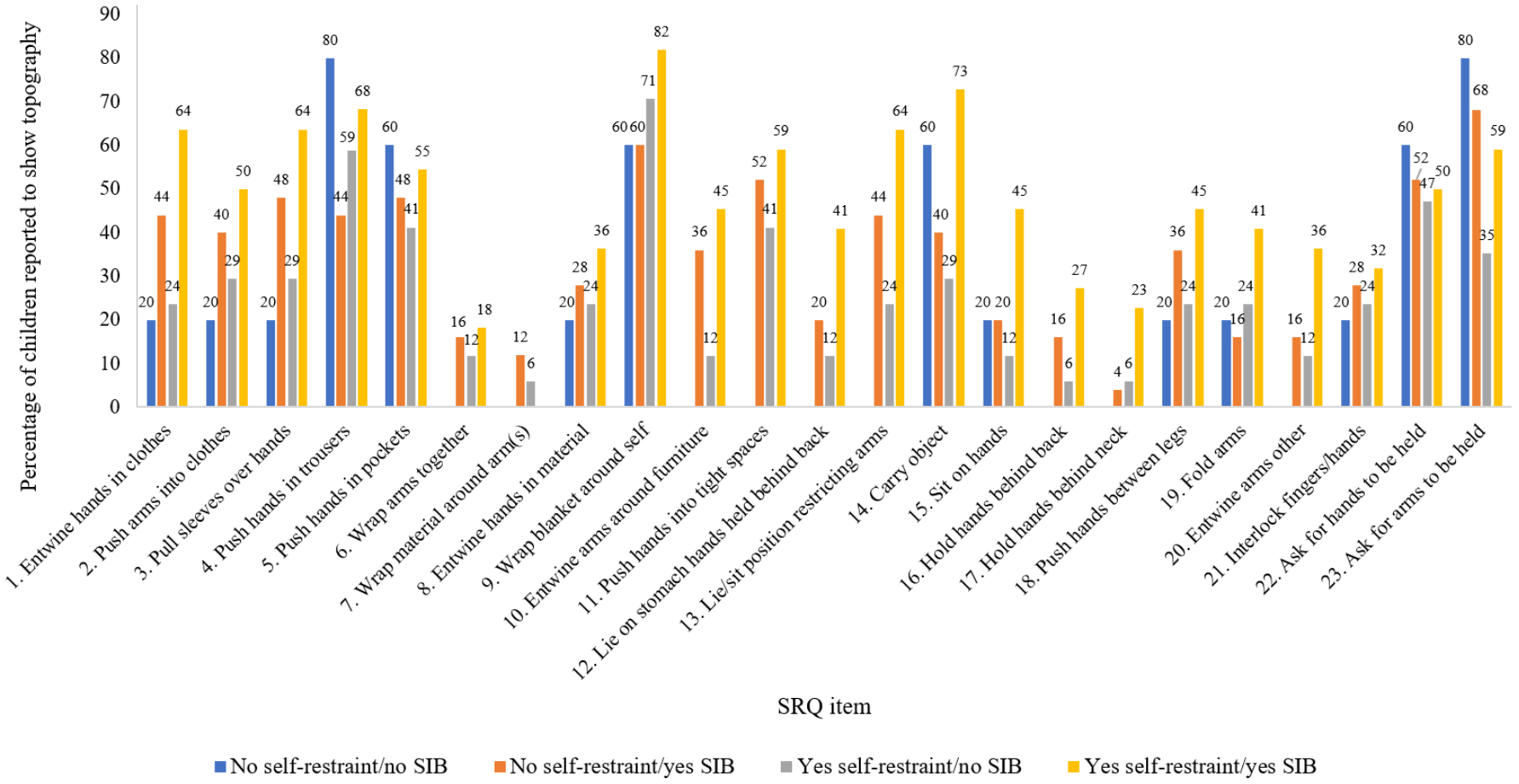


Figure 6. 5 Percentage of children reported to show each self-restraint topography when grouped by SRQ2



6.4.1.3 Demographic correlates of self-restraint

Correlations between SRQ intensity scores and demographic characteristics are shown in Table 6.4. There were no significant correlations after controlling for multiple comparisons, however the small effects between self-restraint intensity and younger age, lower verbal ability, lower receptive language and greater autism characteristics are notable.

Table 6. 4 Spearman's rho correlations between SRQ intensity scores and demographic characteristics

		1 SRQ intensity	*N*	*p*
1	SRQ intensity	—		
2	Exact age	-.122	165	.118
3	Verbal ability	-.111	165	.157
4	Gender	-.029	165	.709
5	VABS-III ABC	.043	142	.612
6	VABS-III RL AE	-.128	142	.128
7	BPVS SS [a]	.099	125	.296
8	SCQ total	.113	165	.148

Note. SRQ = Self-Restraint Questionnaire; VABS-III ABC = Vineland Adaptive Behavior Scales, Third Edition Adaptive Behaviour Composite scores; VABS-III RL AE = receptive language age equivalent; BPVS SS = British Picture Vocabulary Scale, standardised score; SCQ = Social Communication Questionnaire (Autism Traits).
[a] Pooled imputed BPVS scores.

'Self-restraint' and 'no self-restraint' SRQ2 groups were compared using Mann-Whitney U and chi-square tests (Table 6.5). Results revealed no statistically significant group differences across all demographic variables tested after controlling for multiple comparisons.

Table 6. 5 SRQ2 group comparisons of demographic characteristics

Independent samples t-tests							
Variable	**Group**	***N***	**Median**	***U***	***z***	***p***	***r***
Exact age	Self-restraint	39	9.70	570.0	-.182	.856	-0.022
	No self-restraint	30	9.76				
VABS-III ABC	Self-restraint	39	41.0	478.5	-.859	.359	-0.105
	No self-restraint	28	39.0				
VABS-III RL AE	Self-restraint	39	19.0	544.5	-.019	.985	-0.321
	No self-restraint	28	19.5				
BPVS SS	Self-restraint	18	69.0	133.5	-.782	.434	-0.132
	No self-restraint	17	69.0				
SCQ total	Self-restraint	39	27.0	556.0	-.352	.725	-0.042
	No self-restraint	30	25.5				

Chi-square associations					
		***N* non-verbal**	***N* verbal**	**χ^2 statistic**	***p***
Verbal ability	Self-restraint	14	25	2.098	.148
	No self-restraint	16	14		
		***N* male**	***N* female**	**χ^2 statistic**	***p***
Gender	Self-restraint	32	7	1.383	.240
	No self-restraint	21	9		

Note. VABS-III ABC = Vineland Adaptive Behavior Scale, Third Edition Adaptive Behaviour Composite scores; VABS-III RL AE = receptive language age equivalent; BPVS SS = British Picture Vocabulary Scale standardised scores; SCQ = Social Communication Questionnaire.

6.4.2 Behavioural correlates of self-restraint

Correlations between self-restraint intensity scores and BoC are shown in Table 6. SRQ intensity scores significantly correlated with SIB, aggression, property destruction and stereotypy severity scores, with small-to-medium effects[37].

Table 6. 6 Spearman's rho correlations between SRQ intensity scores and CBQ severity scores

		1	2	3	4
1	SRQ intensity scores	—			
	N				
2	SIB severity	**.201***	—		
	N	*165*			
3	Aggression severity	**.186***	**.582***	—	
	N	*165*	*171*		
4	Property destruction severity	**.246***	**.461***	**.584***	—
	N	*165*	*171*	*171*	
5	Stereotypy severity	**.179***	**.298***	**.233***	**.305***
	N	*156*	*162*	*162*	*162*

Note. SRQ = Self-Restraint Questionnaire. SIB = self-injurious behaviour.
*Significant p-values after correcting for multiple comparisons in bold.

Mann-Whitney U tests revealed children in the 'self-restraint' SRQ2 group displayed significantly lower SIB severity and stereotypy severity scores than the 'no self-restraint' group (Table 6.7). Aggression and property destruction severity scores did not significantly differ between groups.

[37] Section 2.3.5 for correlation effect size interpretation.

Table 6. 7 Mann-Whitney U results comparing SRQ2 groups across behaviours of concern

Behaviour	**Group**	***N***	**Median**	***U***	***z***	***p***	***r***
SIB	Self-restraint	39	5.0	397.5	-2.318	**.020***	-.279
	No self-restraint	30	8.0				
Aggression	Self-restraint	39	5.0	516.0	-0.852	.394	-.103
	No self-restraint	30	6.0				
Property destruction	Self-restraint	39	6.0	567.0	-0.221	.825	-.027
	No self-restraint	30	6.0				
Stereotypy	Self-restraint	36	7.5	346.0	-2.342	**.019***	-.290
	No self-restraint	29	9.0				

Note. *Significant p-values after correcting for multiple comparisons in bold.

6.4.3 Testing hypotheses of impaired behavioural control

6.4.3.1 Correlations between self-restraint and executive functions

Correlations between SRQ intensity scores and EF are shown in Table 6.8. Indirect measures of inhibitory control significantly correlated with each other and with indirect working memory scores. Regarding the relations between direct measures, both working memory measures negatively correlated (i.e., higher efficiency correlated with lower perseveration, both indicative of good performance). The direct inhibitory control task positively correlated with working memory efficiency, and negatively correlated with working memory perseveration (i.e., higher inhibitory control related to better working memory performance on the direct task). Working memory tasks significantly correlated with no-conflict scores ('pre-switch' conditions). Conflict scores did not significantly correlate with other direct measures of EF, however the moderate to large effect are notable.

Regarding the relations between indirect and direct measures of EF, neither indirect measure of inhibitory control correlated with the direct measure, and the indirect measure of working memory did not correlate with either direct measure. Therefore, indirect (i.e., questionnaire) and direct (i.e., performance-based) measures of EF appeared to be separable, arguably capturing distinct constructs.

Finally, SRQ intensity scores did not significantly correlate with any indirect nor direct measures of EF. However, the small-to-moderate correlation between self-restraint and TAQ impulsivity was notable. SRQ intensity scores also correlated with TAQ overactivity, impulsive speech and total activity scores, and TAQ subscales strongly inter-correlated (e.g., TAQ overactivity and total scores approached singularity), suggesting measurement of a similar construct. Therefore, greater self-restraint intensity was related to indirect measures of impulsivity.

Table 6. 8 Spearman's rho correlations between SRQ intensity scores and executive function outcomes

Executive function		**1**	**2**	**3**	**4**	**5**	**6**	**7**
	1. SRQ intensity	—						
Inhibitory control								
Indirect	**2.** TAQ impulsivity [a]	.171	—					
	N	*125*						
	3. TAQ overactivity [a]	**.447***	**.546***	—				
	N	*125*	*126*					
	4. TAQ impulsive speech [a]	**.265***	**.347***	**.603***	—			
	N	*72*	*73*	*73*				
	5. TAQ overall [a]	**.381***	**.675***	**.897***	**.768***	—		
	N	*125*	*126*	*126*	*73*			
Indirect	**6.** CHEXI inhibition [a]	.151	**.488***	**.376***	**.417***	**.521***	—	
	N	*124*	*125*	*125*	*73*	*125*		
Direct	**7.** Prohibition score [b]	.014	-.136	-.069	.268	.069	.147	—
	N	*125*	*126*	*126*	*73*	*126*	*125*	
Working memory								
Indirect	**8.** CHEXI WM factor [a]	.083	**.331***	**.227***	**.311***	**.298***	**.582***	.135
	N	*123*	*124*	*124*	*72*	*124*	*124*	*124*

Note. SRQ = Self-Restraint Questionnaire; TAQ = The Activity Questionnaire; CHEXI = Childhood Executive Functioning Inventory; WM = working memory.
[a] Higher scores indicate poorer EF performance. [b] Higher scores indicate better EF performance.
*Significant p-values after correcting for multiple comparisons in bold.

Table 6.8 cont.

Executive function			1	2	3	4	5	6	7	8	9	10	11
Direct	**9.**	WM efficiency [b]	.023	**-.225***	-.174	.066	-.027	.070	**.410***	-.035	—		
		N	*125*	*126*	*126*	*73*	*126*	*125*	*126*	*124*			
Direct	**10.**	WM perseveration [a]	-.067	.123	.062	-.084	-.069	-.165	**-.408***	-.046	**-.763***	—	
		N	*125*	*126*	*126*	*73*	*126*	*125*	*126*	*124*	*126*		
Cognitive flexibility													
Direct	**11.**	Conflict score [b]	.077	-.085	-.047	.231	.056	.031	.203	-.009	.299	-.307	—
		N	*125*	*126*	*126*	*73*	*126*	*125*	*126*	*124*	*126*	*126*	
Direct	**12.**	No-conflict score [b]	.030	-.125	-.098	.312	.051	.084	**.339***	-.019	**.396***	**-.341***	.389
		N	*125*	*126*	*126*	*73*	*126*	*125*	*126*	*124*	*126*	*126*	*126*

Note. SRQ = Self-Restraint Questionnaire; TAQ = The Activity Questionnaire; CHEXI = Childhood Executive Functioning Inventory; WM = working memory.
[a] Higher scores indicate poorer EF performance. [b] Higher scores indicate better EF performance.
*Significant p-values after correcting for multiple comparisons in bold.

6.4.3.2 Correlations between demographics and executive functions

Correlations between demographic characteristics and EF outcomes are displayed in Table 6.9. Age, verbal ability, adaptive functioning, autism characteristics and SIB severity correlated with EF. Receptive language, measured by imputed BPVS scores, did not significantly correlate with EF outcomes.

Higher age significantly correlated with higher prohibition composite scores, higher working memory efficiency and lower working memory perseveration composite scores. This suggested older age was related to better EF outcomes across direct measures of inhibition and working memory. However, higher age significantly correlated with higher CHEXI working memory scores, suggesting older age was related to poorer working memory as measured by this questionnaire.

Verbal ability was related to higher prohibition scores and working memory efficiency scores, as well as lower perseveration scores. This suggested children with greater verbal ability performed better across direct measures of EF. However, verbal ability was correlated with higher CHEXI working memory scores, suggesting children who were verbal showed poorer EF across this indirect measure.

Higher VABS-III ABC scores, indicating higher adaptive functioning, significantly correlated with higher prohibition scores, higher working memory efficiency and lower perseveration scores. This suggested better adaptive functioning was related to better EF performance across direct and indirect measures.

Higher SCQ scores were significantly related to higher TAQ impulsivity scores. This suggested that greater autism characteristics were related to poorer inhibition scores as measured by a questionnaire.

Higher SIB severity scores were significantly related to higher TAQ impulsivity scores, lower working memory efficiency and higher perseveration scores. This suggested that greater SIB severity was related to poorer performance across indirect measures of inhibition and direct measures of working memory.

Table 6. 9 Spearman's rho correlations between demographic variables and EF outcomes

	N	**1**	**2**	**3**	**4**	**5**	**6**
Demographic variables							
1. Age		—					
2. Verbal	*126*	**.358***	—				
3. VABS-III ABC [a]	*126*	**.412***	**.666***	—			
4. BPVS SS [a]	*126*	-.206	-.063	-.102	—		
5. SCQ [b]	*126*	.107	.044	-.136	-.163	—	
6. SIB severity [b]	*126*	.008	**-.230***	**-.296***	.019	.186	—
EF outcomes							
7. TAQ impulsivity [c]	*126*	-.036	-.064	-.207	-.016	**.289***	**.368***
8. CHEXI inhibition [c]	*125*	.150	**.244***	.150	-.054	.084	.027
9. Prohibition composite [d]	*126*	**.487***	**.396***	**.474***	-.016	.063	-.086
10. CHEXI WM [c]	*124*	**.223***	.031	-.059	.025	.123	.126
11. WM efficiency composite [d]	*126*	**.282***	**.554***	**.553***	-.071	.094	**-.282***
12. WM perseveration composite [c]	*126*	**-.230***	**-.567***	**-.544***	.107	-.109	**.262***
13. Conflict score [d]	*126*	.073	.277	.265	.134	-.003	-.142

Note. VABS-III ABC = Vineland Adaptive Behavior Scales, Third Edition Adaptive Behaviour Composite scores; BPVS SS = British Picture Vocabulary Scales Standardised Scores; WM = working memory; correlations between EF outcomes are displayed in Table 6.8.

[a] Higher scores indicate lower severity. [b] Higher scores indicate higher severity or characteristics. [c] Higher scores indicate poorer EF performance. [d] Higher scores indicate better EF performance.

*Significant p-values after correcting for multiple comparisons in bold.

6.4.3.3 Multi-parameter self-restraint and executive functions

6.4.3.3.1 Two groups: Self-restraint and no self-restraint

Mann-Whitney U tests compared 'self-restraint' and 'no self-restraint' SRQ2 groups on indirect measures of EF (Table 6.10). No significant differences between groups were identified after controlling for multiple comparisons, and effect sizes were small.

Table 6. 10 Mann-Whitney U results comparing indirect EF performance across two SRQ2 groups

EF	**SRQ2 group**	***N***	**Median**	***U***	***z***	***p***	***r***
TAQ impulsivity	Self-restraint	39	22.00	570.0	-.184	.854	-.022
	No self-restraint	30	21.50				
CHEXI inhibition	Self-restraint	38	48.00	546.5	-.291	.771	-.035
	No self-restraint	30	46.00				
CHEXI working memory	Self-restraint	38	56.00	534.5	-.439	.660	-.053
	No self-restraint	30	53.00				

Note. SRQ2 = Self-Restraint Questionnaire 2; TAQ = The Activity Questionnaire; CHEXI = Childhood Executive Functioning Inventory.

Independent samples t-tests revealed no significant differences between groups on any direct measure of EF after controlling for multiple comparisons (Table 6.11).

Table 6. 11 Independent samples t-tests comparing SRQ2 two-groups on mean executive function composite scores

Executive function	**SRQ2 group**	***N***	**Mean**	**SE mean**	***df***	***t***	***p***	***Hedges' g***
Prohibition score	Self-restraint	37	0.87	.16	62	-1.170	.241	-.31
	No self-restraint	27	0.60	.16				
WM efficiency	Self-restraint	37	0.72	.05	62	-0.235	.814	-.06
	No self-restraint	27	0.70	.06				
WM perseveration	Self-restraint	37	0.13	.04	62	0.313	.755	.09
	No self-restraint	27	0.15	.04				
Conflict score	Self-restraint	37	1.84	.44	62	0.224	.823	.07
	No self-restraint	27	1.92	.50				

Note. SRQ2 = Self-Restraint Questionnaire 2; WM = working memory.

6.4.3.3.2 Four groups: Self-restraint with and without self-injury

The SRQ2 produced four groups (see Figure 6.4): self-restraint and self-injury ('yes self-restraint/yes SIB'), self-restraint without self-injury ('yes self-restraint/no SIB'), self-injury without self-restraint ('no self-restraint/yes SIB'), and no self-restraint nor self-injury ('no self-restraint/no SIB'). The 'no self-restraint/no SIB' group was excluded from analyses due the small group size (n=5).

Kruskal-Wallis one-way ANOVAs revealed no significant group differences on indirect (i.e., non-normally distributed) EF variables (Table 6.12). One-way between subjects ANOVAs revealed no significant group differences on direct (i.e., imputed and normally-distributed) EF outcomes.

Table 6. 12 Kruskal-Wallis and one-way ANOVAs comparing EF outcomes across four SRQ2 groups

EF outcome	SRQ2 group	*N*	Mean rank	Test statistic Kruskal-Wallis H/ F(d)	*p*
Indirect measures					
TAQ impulsivity	-SR/+SIB	25	32.50	1.205	.547
	+SR/-SIB	17	28.82		
	+SR/+SIB	22	35.34		
CHEXI inhibition	-SR/+SIB	25	33.70	0.947	.623
	+SR/-SIB	17	33.41		
	+SR/+SIB	22	28.83		
CHEXI WM	-SR/+SIB	25	33.52	0.822	.663
	+SR/-SIB	17	33.41		
	+SR/+SIB	22	29.05		
Direct measures					
Prohibition composite	-SR/+SIB	22	0.63	0.835 (2)	.467
	+SR/-SIB	17	0.99		
	+SR/+SIB	20	0.77		
WM efficiency composite	-SR/+SIB	22	0.67	0.831 (2)	.468
	+SR/-SIB	17	0.78		
	+SR/+SIB	20	0.67		
WM perseveration composite	-SR/+SIB	22	0.17	1.959 (2)	.178
	+SR/-SIB	17	0.07		
	+SR/+SIB	20	0.18		
Conflict composite	-SR/+SIB	22	1.87	1.570 (2)	.397
	+SR/-SIB	17	2.01		
	+SR/+SIB	20	1.69		

Note. SRQ2 = Self-Restraint Questionnaire 2; TAQ = The Activity Questionnaire; CHEXI = Childhood Executive Functioning Inventory; SR = self-restraint, SIB = self-injurious behaviour; WM = working memory.

6.4.3.4 Does SIB severity moderate the relation between EF and self-restraint?

The potentially moderating role of SIB severity on the relation between EF and self-restraint was examined. Model 1 examined whether individual differences in self-injury moderated the relation between indirect measures of EF and self-restraint intensity scores. Model 2 examined whether individual differences in self-injury moderated the association between direct measures of EF and self-restraint intensity scores. Exact age, verbal ability and VABS-III ABC scores were entered into analyses as covariates as higher age, verbal ability, and greater ID severity correlated with poorer EF outcomes (section 6.4.3.2).

6.4.3.4.1 Model 1: Indirect measures of EF

Three moderator analyses were performed using PROCESS. The predictor variables in turn were TAQ impulsivity (Model 1a, *N*=142), CHEXI inhibition (Model 1b, *N*=135) and CHEXI working memory (Model 1c, *N*=134). The outcome variable was SRQ intensity scores. The moderator variable was SIB severity score.

Results revealed non-significant interactions for TAQ impulsivity (Table 6.13). The interaction between CHEXI inhibition and SIB severity was significant, indicating that the relation between indirect inhibition scores and self-restraint intensity was moderated by SIB severity. Figure 6.6 displays the simple slopes of low SIB severity (i.e., 16th percentile), b=.050, SE=.185, t=.268, *p*=.789, average SIB severity (50th percentile), b=.305, SE=.148, t=2.066, *p*=.041, and high SIB severity (84th percentile), b=.560, SE=.211, t=2.653, *p*=.009. Therefore, poor inhibition questionnaire scores predicted higher self-restraint intensity at both average and high levels of SIB severity.

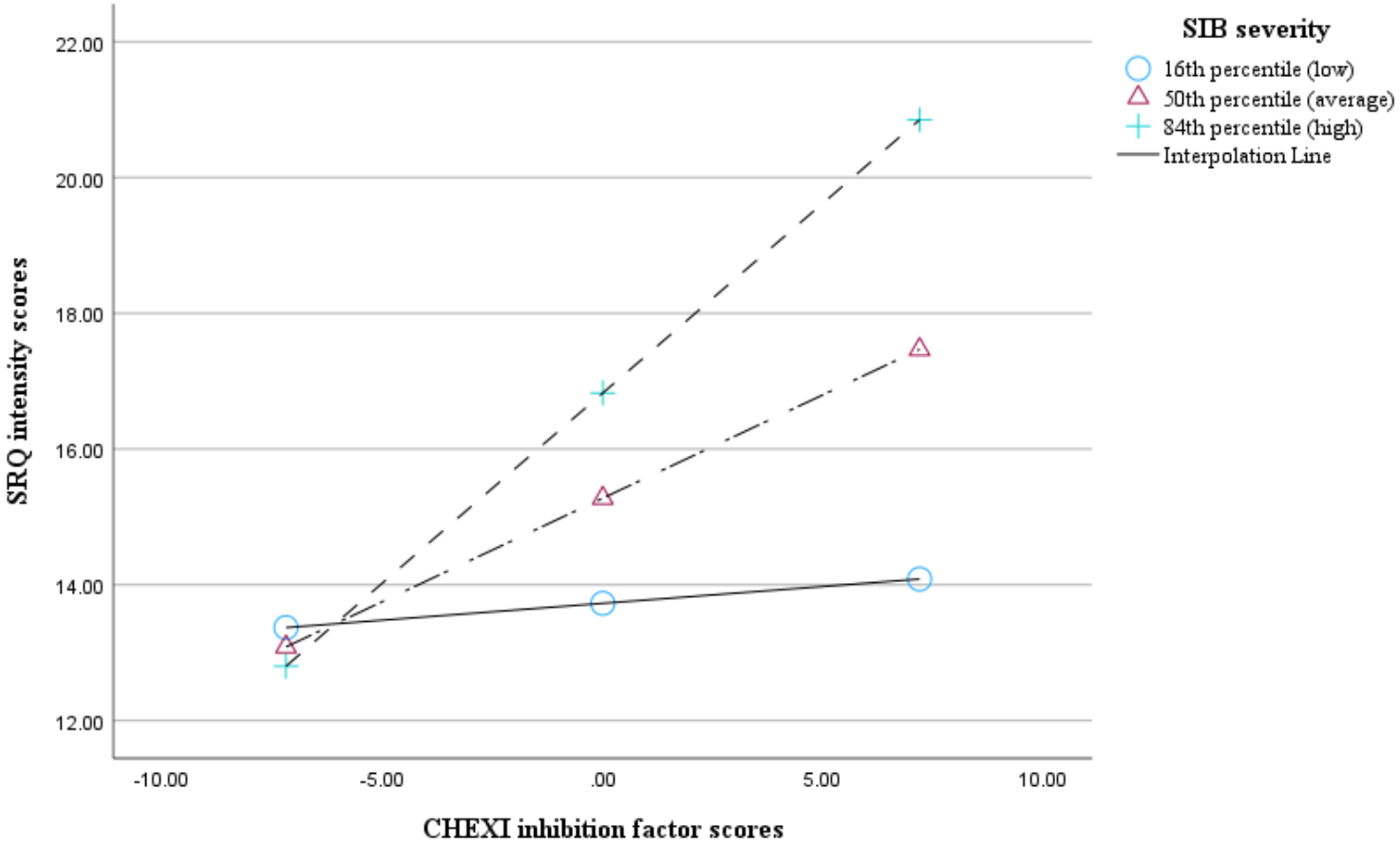
The interaction between CHEXI working memory and SIB severity scores was also significant, indicating that the relation between indirect working memory scores and self-restraint intensity was moderated by SIB severity. Figure 6.7 displays the simple slopes of low SIB severity, b=-.109, SE=.172, t=-0.633, *p*=.528, average SIB severity, b=.196, SE=.125, t=1.570, *p*=.119, and high SIB severity, b=.500, SE=.174, t=2.879, *p*=.005. Therefore, poor working memory questionnaire scores predicted self-restraint intensity at high SIB severity.

Table 6. 13 Moderating role of SIB severity on indirect measures of EF predicting self-restraint

	Model 1a: TAQ impulsivity			**Model 1b: CHEXI inhibition factor**			**Model 1c: CHEXI working memory factor**		
	B	(SE)	*p*	B	(SE)	*p*	B	(SE)	*p*
Covariates									
Exact age	-0.270	(0.422)	*.524*	-0.353	(0.415)	*.397*	-0.363	(0.420)	*.389*
Verbal ability	-2.370	(2.619)	*.367*	-2.381	(2.635)	*.368*	-1.067	(2.582)	*.681*
VABS-III ABC	0.014	(0.090)	*.877*	-0.010	(0.089)	*.909*	0.001	(0.089)	*.987*
Variables									
EF variable	0.383	(0.257)	*.138*	0.305	(0.148)	*.041*	0.196	(0.125)	*.119*
SIB severity	0.348	(0.248)	*.163*	0.356	(0.248)	*.154*	0.334	(0.248)	*.180*
EF x SIB	0.022	(0.059)	*.716*	0.059	(0.031)	***.057****	0.070	(0.027)	***.012****

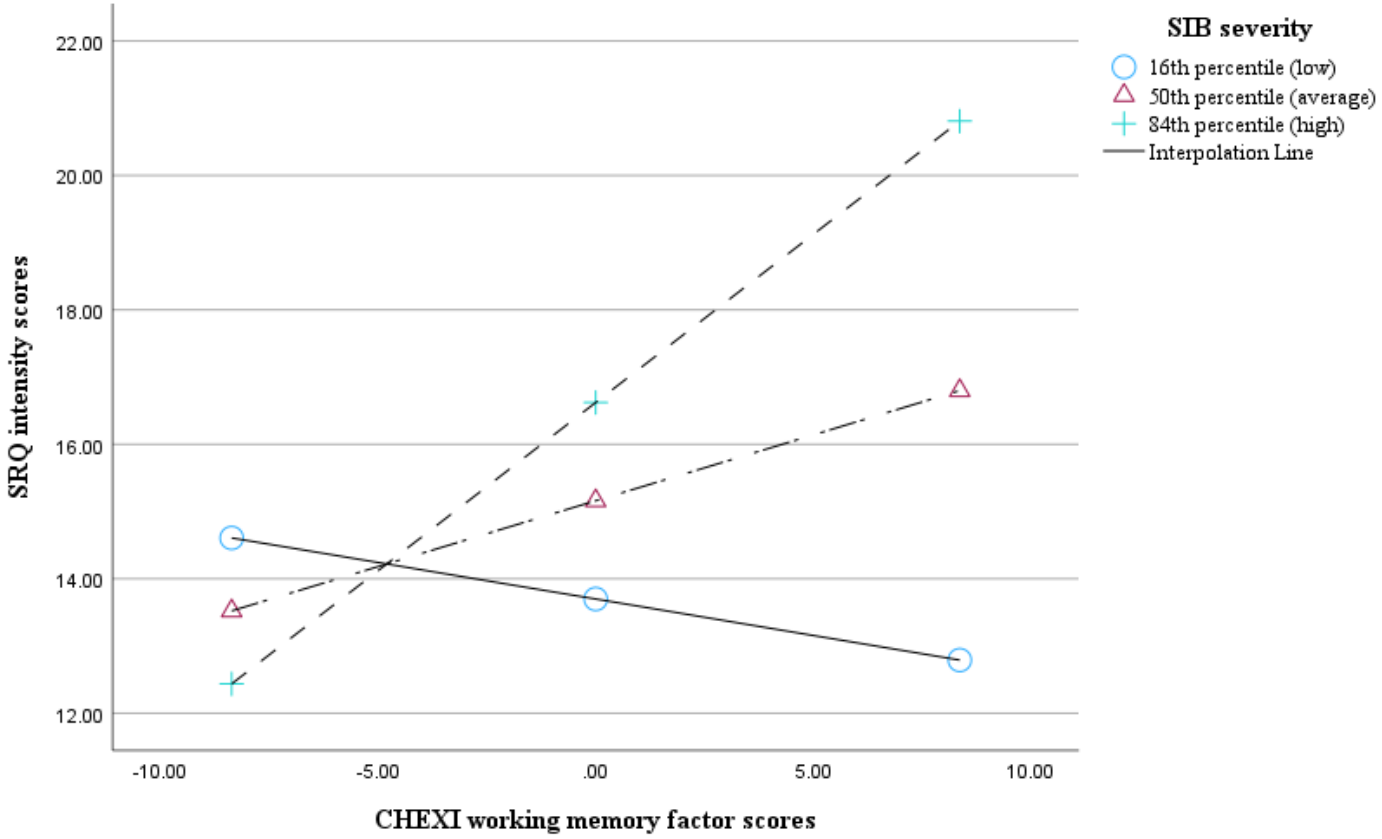
Note. B = unstandardised beta; SE = standard error; VABS-III ABC = Vineland Adaptive Behavior Scales, Third Edition Adaptive Behaviour Composite; EF = executive function; SIB = self-injurious behaviour.
*Interactions $p<.10$ probed by the model in bold.

Figure 6. 6 Interaction of SIB severity on CHEXI inhibition scores and self-restraint intensity



Note. 50th and 84th percentile interpolation lines statistically significant. Higher CHEXI scores indicate poorer inhibitory control.

Figure 6. 7 Interaction of SIB severity on CHEXI working memory scores and self-restraint intensity



Note. 84th percentile interpolation line statistically significant. Higher CHEXI scores indicate poorer working memory.

6.4.3.4.2 Model 2: Direct measures of EF

A further four moderator analyses were performed using PROCESS. The predictor variables in turn were prohibition composite scores (Model 2a, *N*=95), working memory efficiency composite scores (Model 2b, *N*=95), working memory perseveration composite scores (dichotomous scores, section 4.4.2.1; Model 2c, *N*=95) and conflict composite scores (Model 2d, *N*=58). The original (i.e., non-imputed) scores[38] were entered into the model (section 4.4.2.1). The predictor, moderator and control variables were identical to those entered into Model 1 (section 6.4.3.4.1). Results revealed non-significant interactions between direct measures of EF and SIB scores when controlling for covariates across all models (Table 6.14). Therefore, SIB severity did not moderate the relation between direct EF outcomes and self-restraint intensity scores.

[38] Owing to SPSS (version 29) capabilities, it was not possible to run the PROCESS macro on the imputed dataset.

Table 6. 14 Moderating role of SIB severity on direct measures of EF predicting self-restraint

	Model 2a: Prohibition scores			**Model 2b: WM efficiency scores**			**Model 2c: WM perseveration scores**			**Model 2d: Conflict scores**		
	B	(SE)	*p*	B	(SE)	*p*	B	(SE)	*p*	B	(SE)	*p*
Covariates												
Exact age	-0.139	(0.599)	*.817*	-0.204	(0.502)	*.685*	-0.341	(0.491)	*.489*	-0.635	(0.536)	*.242*
Verbal ability	-0.200	(3.320)	*.952*	0.098	(3.308)	*.977*	-0.819	(3.254)	*.802*	4.390	(3.967)	*.274*
VABS-III ABC	-0.043	(0.115)	*.708*	-0.084	(0.104)	*.419*	-0.096	(0.103)	*.354*	-0.185	(0.109)	*.094*
Variables												
EF variable	0.197	(3.922)	*.960*	2.636	(5.449)	*.630*	-2.870	(3.105)	*.358*	2.089	(1.660)	*.214*
SIB severity	0.594	(0.313)	*.061*	0.421	(0.290)	*.151*	0.480	(0.284)	*.094*	0.142	(0.310)	*.650*
EF x SIB	-1.037	(0.699)	*.142*	-0.960	(0.996)	*.338*	-0.550	(0.615)	*.374*	-0.248	(0.301)	*.414*

Note. B = unstandardised beta; SE = standard error; VABS-III ABC = Vineland Adaptive Behavior Scales, Third Edition Adaptive Behaviour Composite scores; SIB = self-injurious behaviour.

6.5 Discussion

This study involving 126 autistic children with ID revealed several key findings. First, individuals who self-injured displayed greater self-restraint intensity compared to those who did not self-injure, and over half of participants examined (57%) met criteria for multi-parameter self-restraint (i.e., topography, intensity and preference for behaviour). Second, self-restraint and SIB (and other BoC) were significantly associated, however the direction of association varied depending on whether continuous intensity scores or categorical multi-parameter assessments of self-restraint were used. Third, although indirect measures of EF converged with each other and direct performance-based measures of EF showed moderate correlations with each other, direct and indirect measures of EF did not correlate. Self-restraint correlated more closely with indirect EF than direct measures of EF. Finally, SIB severity moderated the relation between indirect measures of EF (inhibition and working memory) and self-restraint intensity. The theoretical and practical implications of these findings are discussed.

6.5.1 Prevalence, topographies and demographic correlates of self-restraint

The prevalence of self-restraint according to the SRQ was 96%, considerably higher than existing estimates using the SRC (i.e., scoring above zero), including estimates from individuals with neurodevelopmental conditions (68%; section 3.4.1.1), individuals with ID (65%; Forman, 2003), autistic children (41%; Richards et al., 2017) and autistic children and adults (29%; Laverty et al., 2020). This elevated prevalence may be attributed to the greater number of items on the SRQ (23) in comparison to the SRC (eight), making it more likely for an individual to be categorised as showing 'self-restraint'. In the present study, according to SRQ most children who showed self-restraint had displayed self-injury in the previous month

(73%), replicating previous findings involving autistic children (64%; Richards et al., 2017) and individuals with ID (77%; Hyman et al., 2002). Children who self-injured were reported to show significantly higher SRQ intensity scores than those who did not self-injure. The high self-restraint prevalence within this sample suggests that individuals with autism *and* ID are uniquely likely to display self-restraint *and* SIB in comparison to individuals with idiopathic autism or ID.

Within a subsample of participants, the prevalence of self-restraint as measured by the SRQ2 ('multi-parameter self-restraint') was lower, 57%, than SRQ-rated prevalence, 90%. This replicated Forman (2003)'s findings, who reported a lower prevalence of multi-parameter self-restraint (35%) in comparison to SRQ prevalence (65%) in individuals with ID. This finding was expected, as participants were required to meet a more restrictive definition of behaviour. Therefore, not all individuals who display self-restraint behaviours meet threshold for potentially 'concerning' behaviour.

Individuals classified as showing multi-parameter self-restraint and SIB according the SRQ2 (32% of the SRQ2 sample) displayed a greater prevalence of 18 self-restraint topographies than children who only showed self-restraint, SIB or neither behaviour. Prevalent topographies included 'wraps blanket or other material around self' (displayed by 82% of individuals who self-injured and self-restrained) and 'carries object without using, playing or manipulating it' (73%). The high prevalence of a variety of behaviours further supports previous suggestions that there is not one definite 'profile' of self-restraint (e.g., section 5.5). Nearly 100 topographies have been reported in the literature (Forman, 2003), and a naturalistic observation of self-restraint revealed variance in behavioural intensity both

between and within topographies (section 5.4.1). Thus, while identification of prevalent topographies is vital for improving recognition of self-restraint, a person-centred approach to understanding and managing behaviour is essential.

Individuals who did not self-restrain nor self-injure according to the SRQ2 displayed the highest prevalence of the remaining four topographies examined: 'pushes hands in trousers', 'asks for arms to be held' (both displayed by 80% of individuals in this group), 'pushes hands in pockets' and 'asks for hands to be held' (both 60%). This may suggest that autistic children with ID naturally display some self-restraining behaviours that appear unrelated to functional contingencies with SIB. For example, gesturing for imposed restraint may be reinforced by contingent caregiver attention, and pushing hands into clothing may be reinforced by sensory stimulation (Peterson & Peterson, 1968). An exploratory factor analysis of the SRQ further supports this suggestion, as items 'pushes hands into pockets/trousers' were not assigned to a factor, suggesting these behaviours did not strongly contribute towards measuring the underlying construct of 'self-restraint' (section 4.4.1). Therefore, while the function of behaviour cannot be confirmed using the cross-sectional methods, it is recognised that not all forms of self-restraint occur to manage SIB.

This was the first study to examine multi-parameter self-restraint in individuals who do and *do not* self-injure. Within the multi-parameter self-restraint subgroup, 44% of children (n=17) did not show 'current' SIB (i.e., self-injury in the previous month). This finding replicates existing findings of self-restraint occurring in the absence of SIB in autistic (35%; Richards et al., 2017) and ID populations (18%; Fovel et al., 1989) using a more fine-grained measurement approach and a more stringent definition of self-restraint than previous

literature. It is important to acknowledge that the multi-parameter assessment of self-restraint was only possible for a subsample of participants, meaning not all analyses can be replicated using the stringent SRQ2 criteria. Of the children grouped as showing multi-parameter self-restraint in the absence of SIB, six had 'previously' self-injured in the lifetime. Identification of this subgroup of individuals may lend support to an operant model of avoidance-motivated behaviour, whereby self-restraint functions so effectively that self-injury does not occur (section 1.4.7.2). However, this suggestion can only be confirmed using a longitudinal study design examining the stability of self-restraint and SIB over-time. Additionally, this theory cannot account for the 11 participants who showed multi-parameter self-restraint but had 'never' shown any form of self-injury in the lifetime. This suggests that for some autistic children with ID, self-restraint behaviours may become self-initiated, intense and preferred for reasons unrelated to SIB. Inspection of prevalent self-restraint topographies provides insight into the potential functions of such behaviours: children who showed self-restraint in the absence of SIB displayed a high prevalence of behaviours including 'wraps blanket or other material around body' (71%) and 'pushes hands in trousers' (68%). These behaviours may be sensory seeking, such as accessing warmth and comfort (Peterson & Peterson, 1968), supporting a theory of functional independence of SIB and self-restraint (section 1.4.7.2).

When measured according to intensity, self-restraint did not significantly correlate with demographic characteristics, including age, sex, autism characteristics and adaptive functioning. However, the small effects between self-restraint intensity and younger age, lower verbal ability, lower receptive language and greater autism characteristics are notable, replicating findings from individuals with neurogenetic syndromes (section 3.4.1.2). However, the relation between self-restraint and younger age contradicts results from a meta-

analysis (section 2.4.3.1), requiring further inspection (see section 7.2.3). Interestingly, there were no significant differences in demographic characteristics between the 'multi-parameter self-restraint' and 'no self-restraint' groups identified by the SRQ2, replicating previous findings (Forman, 2003). A lack of SRQ2 group differences in the present study may further indicate the variable profile of individuals who self-restrain. Findings may also be explained by a lack of statistical power, with the self-restraint and no self-restraint groups involving 39 and 30 participants respectively.

6.5.2 The relation between self-restraint and behaviours of concern

Higher self-restraint intensity was associated with higher SIB severity. This replicated previous findings using a topographical definition of self-restraint (section 3.4.1.2). However, when self-restraint was defined as intense *and* preferred, children who self-restrained displayed *lower* SIB severity. This finding mirrors research supporting a hypothesis of negative reinforcement, whereby individuals who self-restrain show less SIB, as self-restraint effectively avoids, delays or terminates self-injury (section 1.4.7.2). This provides tentative evidence that some autistic children with ID have developed self-restraint that 'successfully' controls SIB. This may suggest apparent benefits for teaching or modelling self-restraint as an intervention for pervasive SIB. However, children with lower SIB severity met threshold for intense and preferred self-restraint, which is associated with negative physical and emotional consequences (section 1.4.2). Therefore, this evidence suggests that while self-restraint may initially emerge as protective, through reinforcement and/or mechanisms of anxiety/arousal (Chapter 3) it may become maladaptive, at which stage restraint fading may be a suitable intervention.

Greater self-restraint intensity was related to greater aggression, property destruction and stereotypy severity. However, individuals who met criteria for multi-parameter self-restraint displayed significantly *lower* stereotypy severity than those who did not meet SRQ2 criteria. This novel finding may be explained by existing operant models of SIB: stereotypy is considered a risk marker and developmental precursor for SIB (e.g., Richman et al., 2013) and for some individuals, rhythmic repetitive or stereotypic behaviours may emerge as 'proto-SIB' (Oliver & Richards, 2015). Stereotypy is believed to evolve into self-injury through social reinforcement (Guess & Carr, 1991). Given this association between behaviours, the low presence of stereotypy *and* SIB in the multi-parameter self-restraint group is therefore perhaps unsurprising. Longitudinal research is required to assess the evolution of stereotypy, SIB and self-restraint.

6.5.3 Self-restraint and executive functions: direct and indirect measures

This study tested inhibitory control and compulsivity accounts of self-restraint using both direct and indirect measures of EF. Older age and verbal ability were related to better performance on direct inhibition and working memory tasks, while greater adaptive functioning skills correlated with better performance in direct and indirect measures of EF. Greater autism characteristics related to poorer informant-rated impulsivity, and higher SIB severity correlated with poorer informant-rated impulsivity and working memory. Therefore, greater ability was related to better EF performance, and greater autism traits was related to poorer EF performance, replicating previous findings (e.g. Clark et al., 2002; Demetriou et al., 2018; Fay-Stammbach & Hawes, 2019). Thus, alongside rigorous feasibility and validity checks (section 4.4.2), it is concluded that the novel battery of EF tasks functioned as expected.

Questionnaire measures of EF intercorrelated, and performance-based measures intercorrelated. Indirect and direct measures of EF, however, did not correlate in this sample of autistic children with ID. This replicates previous findings (e.g., Toplak et al., 2013), suggesting indirect and direct EF tasks capture different constructs. Indirect measures of EF are conceptualised to capture the 'reflective mind', with goals and beliefs driving behaviour (Toplak et al., 2013). Raters may interpret questionnaire items based on previous experiences within unstructured (Toplak et al., 2013) and complex day-to-day situations (Snyder et al., 2015). Such measures are advantageous in understanding 'real-world' behaviours (Barkley & Fischer, 2011), however informant-report also relies on contextual factors, such as recalling aspects of the environment and situation, that are unrelated to EF. Therefore, while indirect questionnaires provide vital insight into real-world behaviour and challenges, direct measures may better capture specific substrates of EF (Snyder et al., 2015). Direct measures of EF represent the 'algorithmic mind', the ability to process instructions under controlled circumstances (Toplak et al., 2013), where the individual is encouraged to perform to their 'best' ability (Dang et al., 2020). Thus, direct tasks better tap into the EF substrate, while indirect tasks reflect both EF and the broader context of the situation.

Self-restraint intensity scores did not significantly correlate with questionnaire measures of EF, however the small-to-medium effects between self-restraint and impulsivity are notable. Additionally, self-restraint correlated with overactivity, which in turn strongly correlated with impulsivity scores suggesting measurement of a similar construct. Altogether, results indicated an association between greater self-restraint intensity and poorer informant-rated impulsivity. This replicated previous findings within autistic samples (e.g., Richards et al., 2017) and provides evidence in favour of a hypothesis of impaired inhibition. Self-restraint

intensity scores, however, did not correlate with any direct measures of EF, and very small effect sizes (i.e., <0.1) were identified. This novel finding suggests that self-restraint was more closely related to markers of impaired inhibition displayed in the real-world environment, influenced by broader contextual factors, than the specific EF component. To further examine this finding, it is important to consider self-restraint according to multiple parameters.

There were no significant differences in EF outcomes between the multi-parameter self-restraint groups (at both the two- and four-group levels), including the direct measure of working memory perseveration, an index of compulsivity (i.e., the inability to override a prepotent response once initiated). This contradicts King (1993)'s theory of compulsive SIB and self-restraint, which has thus far solely been supported using indirect measures (e.g., Hyman et al., 2002; Laverty et al., 2020; section 3.4.2). Furthermore, regarding the inhibitory control hypothesis, no significant differences were found between groups on any measures of inhibition. Altogether, indirect measures of EF and compulsivity consistently correlate with self-restraint in individuals with neurodevelopmental conditions (e.g., section 3.4.1.2), while direct measures do not. This suggests that the caregiver's perception of EF *and* real-world circumstance is important when examining mechanisms of self-restraint, rather than the pure EF substrate. Thus, a holistic approach should be adopted when considering SIB and self-restraint, accounting both for the markers of impulsivity/compulsivity *and* supporting the caregiver to handle behaviours in real-world settings (e.g., psychoeducation). It is important to acknowledge that adopting a four-group SRQ2 approach considerably lowered group sizes (resulting in exclusion of the 'no self-restraint/no SIB' group, n=5), reducing likelihood of

yielding statistically significant differences (section 6.5.5). To examine the role of EF in self-restraint, it is important to account for potential confounding variables.

6.5.4 Self-injury moderates the relation between self-restraint and executive functions

A moderation analysis revealed that poorer informant-rated inhibition predicted greater self-restraint intensity scores at average and high levels of SIB severity, after controlling for confounding variables (age, verbal ability and adaptive functioning). Similarly, poorer informant-rated working memory predicted greater self-restraint intensity at high levels of SIB severity. Importantly, 'average' SIB severity within this sample was a CBQ score of six (where maximum severity was 14), with 76% children meeting criteria for SIB. This indicates a naturally high prevalence of SIB in the present sample (e.g., higher than estimates from idiopathic autism, 46%, Richards et al., 2017, and genetic syndromes, 45%, Wilde et al., 2017). Direct measures of inhibition or working memory did not predict self-restraint, further highlighting that direct and indirect measures capture different constructs (see above). This finding provides support for a model of impaired inhibitory control driving SIB and self-restraint.

Oliver and Richards (2015) proposed that impaired inhibition predisposes some individuals to developing repetitive 'proto-SIB' that evolves into SIB. Such individuals are less able to inhibit SIB in the presence of antecedents, leading to a gradual loss of behavioural control. At this stage, self-injury becomes reinforced by automatic contingencies (i.e., reinforced by non-environmental consequences, such as sensory stimulation), and self-restraint is theorised to emerge in attempt to regain control over 'impulsive' SIB. The current findings, whereby poor impulsivity predicted self-restraint in the presence of average and high levels of SIB severity, lend support to this hypothesis. Previous research has shown that self-restraint emerges in the

presence of automatically-reinforced SIB: Hagopian et al. (2015) discuss a unique SIB 'subtype' where self-injury is positively reinforced (e.g., by sensory stimulation) and self-restraint is negatively reinforced (e.g., by the removal of pain caused by SIB). Future research is needed to establish the functional role of SIB in those who show multiparameter self-restraint and poor informant-rated impulsivity and working memory.

6.5.5 Limitations

It is important to acknowledge limitations of this study. First, many participants were recruited though National Health Service trusts, therefore biasing the study sample towards those who met threshold for a health, emotional, sleep, behavioural or other need. It is therefore not appropriate to generalise this study's findings to all autistic children with ID.

Second, a subsample of participants were categorised by the SRQ2 (n=69), resulting in relatively small group sizes. For example, at the 'four group' level, only five children met criteria for 'no self-restraint nor SIB'. Future research employing larger sample sizes and well-powered statistical analyses would shed further light on theories tested.

Finally, the six items of the SRQ2 were not administered for every SRQ item indicated as 'yes' (as proposed by Forman, 2003), rather SRQ2 items represented caregivers consideration of 'all' self-restraint behaviours. While a level of nuance was lost using this approach (i.e., it is not possible to comment on the specific topographies that met criteria for multiple parameters), this approach was adopted to reduce risk of respondent fatigue. Participants displayed an average of nine self-restraint topographies, indicating an additional 54 questionnaire items would need displaying (i.e., completing the SRQ2 for all nine

behaviours). Given that the questionnaires were embedded in a larger study involving a caregiver interview and EF testing session (section 4.3.5), efforts were made to reduce 'paperwork' where possible. Reducing caregiver burden reduces attrition and risk of poor data quality.

6.5.6 Conclusions

In conclusion, self-restraint was extremely prevalent in this sample of autistic children with ID. Greater self-restraint intensity was associated with greater SIB severity, replicating previous findings. Stereotypy was revealed as a novel correlate of self-restraint, warranting future research attention. Individuals who showed multi-parameter self-restraint displayed lower SIB severity than non-self-restrainers, suggesting a potentially protective role of self-restraint. A novel developmentally-appropriate and sensitive battery of EF tasks indicated direct and indirect EF measures do not correlate, indicating measurement of disparate constructs. Self-restraint was related to indirect measures of EF, including impulsivity and working memory, and was not related to direct measures. Finally, poorer informant-rated impulsivity and working memory predicted greater self-restraint intensity only at average and high SIB severity, with implications for theory and intervention.

CHAPTER SEVEN

GENERAL DISCUSSION

7.1 Introduction

Despite the many reports of self-initiated restriction of own body movements in the literature, there are very few large-scale investigations testing theoretical accounts of self-restraint. Consequently, behaviours are not well understood. Self-restraint has been observed alongside self-injurious behaviour (SIB) in autistic individuals and/or those with intellectual disability (ID), and both SIB and self-restraint can evolve into persistent and clinically concerning behaviours. Self-restraint poses practical and theoretical challenges: intense self-restraint impacts physical health, adaptive functioning and quality of relationships with others, and the presence of self-initiated restraint contradicts a purely operant account of SIB, suggesting behaviours many wholly be under the individual's control.

The overarching aim of this thesis was to examine mechanisms driving self-restraint, and reconsider traditional accounts of operant learning to delineate associations with compulsivity, impulsivity and broader components of executive functioning (EF). To establish current understanding of self-restraint and generate pooled prevalence estimates of behaviour, a systematic review and meta-analysis was undertaken in Chapter 2. To describe self-restraint phenomenology and examine behaviour across the neurodevelopmental continuum, a large-scale cross-syndrome study was presented in Chapter 3. The data analysed in Chapters 5 and 6 were collected using the methods outlined in Chapter 4. This Chapter described the direct (performance/play-based) and indirect (informant-rated) measures that

assessed EF, examined the psychometric properties of the direct tasks, and assessed performance of the indirect EF tasks in an under-represented sample of autistic children with ID. Chapter 5 described a naturalistic observational study of self-restraint. The observed intensity of topographies, temporal relations with behaviours of concern (BoC) and associations with informant-rated self-restraint intensity were examined. Finally, Chapter 6 tested hypotheses of impaired inhibitory control and compulsivity under different parameter-based measures of self-restraint. Key findings are now summarised.

7.2 Main findings

7.2.1 Aim 1: Delineate the prevalence of self-restraint in populations known to self-injure

The prevalence of self-restraint varied according to population sampled and measurement approach. The systematic review and meta-analysis of existing data in Chapter 2 revealed 39% of individuals in the literature, including individuals with autism and/or ID, self-restrained. Further analysis revealed 34% of individuals who self-injured also showed self-restraint. Definitions (e.g., according to one parameter, Fovel et al., 1989, or several parameters, Forman, 2003) and measurement of self-restraint (i.e., questionnaire versus observation) varied across the 13 included studies. A subgroup analysis, however, revealed measurement approach did not influence the overall prevalence estimate. Chapter 3 revealed the prevalence of self-restraint across individuals with rare neurogenetic conditions was 68% (*n*=185).

Chapter 5 compared the prevalence of observed self-restraint and caregiver-reported self-restraint. Within a sample of 126 autistic children with ID, 89% of individuals (*n*=112)

displayed at least one topography during the observable footage. A larger proportion of individuals, 96% (*n*=121), were indicated by caregivers to show self-restraint on the Self-Restraint Questionnaire (SRQ; Oliver et al., 2003). The prevalence of self-restraint further varied in Chapter 6 when accounting for multi-parameter definitions of behaviour. A subsample of participants (*n*=69) were classified by the Self-Restraint Questionnaire 2 (SRQ2; Forman, 2003) to meet criteria for preference for behaviour (i.e., enjoyment of self-restraint, and anxiety/SIB upon self-restraint termination), self-initiation of self-restraint, and incompatibility of behaviour with SIB and/or symbolic restraint (alongside the prerequisite criteria identified by the SRQ). The prevalence of self-restraint according to this stringent, function-based definition of behaviour was 57% (*n*=39), in comparison to the SRQ prevalence of 90% (*n*=62).

Altogether, autistic children with ID had a higher prevalence of self-restraint in comparison to more heterogeneous samples, implicating this population as particularly at-risk of self-restraint. Autism was a correlate of self-restraint across neurogenetic conditions, suggesting that both autism diagnosis *and* autism traits may be important correlates of behaviour. The prevalence of multi-parameter self-restraint was lower than other estimates. Thus, the first key finding was that not all individuals who self-restrain may be doing so to attenuate SIB, and not all forms of self-restraint are considered concerning. Supporting ideas by Oliver et al. (2003) and Forman (2003), the presence of self-restraint *and* additional parameters, including high intensity and preference for behaviour, indicate ‘multi-parameter’ self-restraint.

7.2.1.1 Self-restraint in the absence of self-injury

Self-restraint consistently occurred in the *absence* of self-injury. The meta-analysis in Chapter 2 estimated 13% of individuals reported to self-restrain in the literature did not show co-occurring self-injury. Chapter 3 found 33% (*n*=83) of individuals with neurogenetic conditions had self-restrained but not self-injured in the previous month, and 23% (*n*=38) of the 165 autistic children with ID included in Chapter 6 self-restrained in the absence of SIB. Findings support previous literature indicating a small but substantial proportion of individuals who self-restrain in the absence of SIB (e.g., 13%, Hyman et al., 2002). The SRQ2 produced a multi-parameter 'self-restraint' group (*n*=39) in Chapter 6, of whom 44% (*n*=17) did not self-injure in the previous month. Of these 17 children, six had self-injured in the lifetime, while 11 had 'never' self-injured. Therefore, a second key finding was that for some individuals, self-restraint may emerge into the behavioural repertoire for reasons unrelated to self-injury.

7.2.2 Aim 2: Identify prevalent topographies of self-restraint

Of the 31 topographies meta-analysed in Chapter 2, the most common forms of self-restraint were 'holds or squeezes objects' and 'holds onto others/others' clothing', estimated to occur in 32% of individuals known to self-restrain. The cross-syndrome study in Chapter 3 replicated this finding within individuals with neurogenetic conditions: 43% of participants 'held or squeezed objects' (*n*=117) and 36% 'held onto others/others' clothing' (*n*=97). The observational study in Chapter 5 revealed 'interlocks fingers of hands' (48%, *n*=61), 'lies or sits in a position restricting arm movement' (32%, *n*=40) and 'folds arms' (29%, *n*=37) were frequently displayed by autistic children with ID. The least common topographies identified by meta-analysis (Chapter 2) *and* cross-syndrome comparison (Chapter 3) were 'chooses

mechanical restraint' (meta-analysis estimate 1%, cross-syndrome prevalence 3%, *n*=7) and 'other' behaviours (meta-analysis estimate 2%, cross-syndrome prevalence 4%, *n*=10). Therefore, within samples of individuals at-risk of SIB, 'holding' behaviours were prevalent and mechanical restraint was rare.

According to the multi-parameter definition of self-restraint measured by the SRQ2, autistic children with ID children who showed both self-restraint *and* SIB displayed the highest prevalence of 18 self-restraint topographies (Chapter 6). Common behaviours included 'wraps blanket around self' (82%) and 'carries object without using it' (71%). There were no significant differences in topography prevalence between the four groups classified by the SRQ2. Interestingly, children who did not self-restrain nor self-injure demonstrated the highest prevalence of the remaining four behaviours: 'pushes hands in trousers/pockets' and 'asks for arms /hands to be held'. Therefore, a third key finding was that some autistic children with ID may naturally display some self-restraint behaviours that are unrelated to functional contingencies with SIB. Altogether, common self-restraint topographies included holding onto people, clothing and objects, and restricting movement of fingers, hands or arms. Choosing mechanical restraint was rare.

7.2.2.1 Number of topographies

Chapter 3 revealed showing multiple self-restraint topographies associated with negative outcomes, including greater severity of BoC, compulsive and impulsive behaviour, and anxiety. Of the individuals with neurogenetic conditions reported to self-restrain in Chapter 3, 59% (*n*=109) had displayed two or more topographies in the previous month, with individuals showing up to eight topographies. In Chapter 5, 77% (*n*=86) of children who had displayed

self-restraint showed two or more topographies, *range*=0-8. Finally, of the autistic children with ID who self-restrained according to caregiver report in Chapter 6, 96% (*n*=152) displayed two or more topographies, with 49% (*n*=81) of the subsample showing 10 or more topographies. The mean topography score was 9.33, *range*=0-23. These are the largest figures in the known literature, surpassing Oliver et al. (2003)'s findings of individuals with ID displaying an average of 5.55 topographies. Thus, in accordance with the existing literature (section 1.4.5.1), individuals tended to exhibit more than one form of self-restraint.

7.2.2.2 Intensity of topographies

Chapter 5 measured intensity, the amount of time spent engaging in self-restraint. Of the common topographies observed (section 7.2.2), the mean intensity varied. 'Lies or sits in a position restricting arm movement' occurred, on average, at a low intensity (4.78% of the time visible), however participant-level data revealed intensity ranged from very low (0.10%) to very high (100%). 'Wears restraint device' occurred at high intensity a 99.84%. Finally, 'wraps blanket or other large piece of material around self' occurred at a moderate intensity (33.87%). Altogether, these data reveal a fourth key finding that there is not one topographical 'profile' of self-restraint. The relation between showing multiple topographies and negative outcomes, as well as the variance in behavioural intensity both between and within topographies, support this finding. Identification of common topographies is vital for improving recognition of self-restraint, however the number of behaviours displayed and the intensity of those behaviours should be considered. Altogether, a person-centred approach to understanding and managing behaviour is essential.

7.2.3 Aim 3: Describe demographic characteristics of participant groups that are associated with self-restraint

Age, autism and ability were important correlates of self-restraint. Meta-regressions undertaken in Chapter 2 implicated the presence of autism and older age as important demographic factors for self-restraint, while gender did not influence prevalence estimates. However, due to a lack of existing data analyses are interpreted with caution. Interestingly, within the heterogeneous neurogenetic sample in Chapter 3 and sample of autistic children with ID in Chapters 5 and 6, self-restraint was associated with *younger* age. These contradictory findings warrant discussion. A broader range of ages was pooled in Chapter 2, *range*=1-72 years, than Chapters 3 and 6, *range*=1-58 years and 4-15 years, respectively. The average age was also higher in the meta-analysis, *M*=21.78, than in Chapters 3 and 6, *M*=19.53 and *M*=10.08, respectively. Given that age shares a curve linear relation with SIB, such that SIB peaks in late childhood/early adolescence and reduces in older age (Oliver et al., 1987), it may be inferred that self-restraint peaks in childhood/adolescence, and reduces thereafter. Future empirical study using a large sample with full age representation is required to further examine this suggestion.

Chapter 3 reported greater autism characteristics and lower self-help skills were related to showing a greater number of self-restraint topographies across neurogenetic syndromes. The correlation between self-restraint and self-help skills was non-significant, however the small effect was notable. Within the autism and ID sample in Chapter 5, lower receptive language skills were associated with greater observed self-restraint intensity. The correlation was non-significant, however the small-to-moderate effect size were notable. Within the full sample of autistic children with ID described in Chapter 6, low verbal ability, lower adaptive

functioning skills and greater autism characteristics was related to greater self-restraint intensity. Despite non-significance of correlations, the small effects were notable and followed the pattern of existing data. Finally, group comparisons of the multi-parameter 'self-restraint' and 'no self-restraint' SRQ2 groups in Chapter 6 revealed no significant group differences across demographic variables.

Altogether, a key finding was that across most measurements of self-restraint, individuals who self-restrained tended to be younger, have a greater presence of autistic characteristics, had low verbal communication, and/or displayed lower adaptive functioning skills. Sex was not related to self-restraint. Correlates are broadly similar to those for SIB (Oliver et al., 1987), and future examination of the potential curve linear relation between self-restraint and age is required.

7.2.4 Aim 4: Examine the relation between self-restraint and self-injury

Synthesis of existing coefficients associating self-restraint with self-injury in Chapter 2 revealed a significant positive moderate pooled effect, r=.21. This effect was replicated across the neurodevelopmental continuum, with a moderate correlation coefficient reported in Chapter 3, r_s=.22. Interestingly, SIB severity did not predict self-restraint after controlling for demographic covariates. This may be explained by the compulsivity account (section 7.2.5), whereby self-restraint serves to reduce SIB.

Examination of the temporal relations between self-restraint and SIB (and other BoC) in Chapter 5 revealed 12% of the 1469 observations of self-restraint were preceded by at least one of the 19 topographies of SIB assessed, and 10% of observations were followed by SIB.

This was greater than other BoC (section 7.2.4.1), implicating a unique relation between self-restraint and SIB. Within the subsample of autistic children with ID in Chapter 5 (n=124), self-restraint intensity shared a small-to-moderate positive correlation with SIB severity. This relation was non-significant, however, in the full sample assessed in Chapter 6 (N=165), self-restraint intensity significantly correlated with SIB severity, r_s=.20. This replicated findings from Chapter 2 and 3. Thus, another key finding was that self-restraint and SIB reliably moderately correlate. While function of behaviour was not directly tested within the thesis (e.g., using experimental functional analysis) due to ethical and theoretical reasons, and so a negative reinforcement account, whereby self-restraint is theorised to function to escape painful SIB, was not directly assessed. However, the reliable positive relation between self-restraint and SIB lends support to the operant model.

Interestingly, Chapter 6 revealed children in the multi-parameter 'self-restraint' SRQ2 group displayed significantly *lower* SIB severity than the 'no self-restraint' group. This finding suggests that self-restraint measurement is important when assessing relations with putative variables, and reveals that individuals with intense, preferred self-restraint show *lower* levels of SIB. This tentatively suggests that some autistic children with ID have developed self-restraint as a protective behaviour in response to SIB, which may have evolved into a maladaptive (perhaps compulsive; section 7.2.5) behaviour. Further research assessing behavioural presence, severity and function longitudinally is required to confirm this hypothesis.

7.2.4.1 The relation between self-restraint and other behaviours of concern

Examination of this aim conferred a novel, secondary outcome, whereby relations between self-restraint and other BoC were established. Chapter 3 revealed a greater number of self-restraint topographies significantly correlated with aggression and property destruction, with moderate to large effects. Chapter 5 identified a moderate (non-significant) relation between self-restraint (SRQ) intensity and property destruction, while SRQ intensity scores significantly correlated with aggression and property destruction within the autistic children with ID in Chapter 6 with moderate to large effects. These are the first data associating self-restraint with clinical behaviours other than SIB. However, not all data point to an association.

Examination of temporal relations in Chapter 5 suggested SIB shared a different relation with self-restraint than other BoC, with aggression and property destruction preceding and following self-restraint at very low rates (2-3% of all occasions of behaviour). Chapter 6 reported no significant differences in aggression and destruction severity between the multi-parameter SRQ2 groups. Thus, while the correlational data suggests other BoC are potentially implicated in the development and maintenance of self-restraint, it is likely that SIB is the predominant BoC underpinning self-restraint. The presence of self-restraint specifically functioning to attenuate other BoC is likely rarer, and further research is required to unpick this relation.

7.2.5 Aim 5: Test the compulsivity hypothesis of self-restraint and self-injury

Meta-analysis in Chapter 2 revealed a very small, non-significant pooled effect between self-restraint and compulsivity, r=.003. This analysis is considered with caution as it included only

two samples, with an insufficient sample size (n=187) to detect small effects (N=614). Thus, the compulsivity hypothesis was best tested using the methods employed in Chapters 3, 5 and 6.

Within the sample of autistic children with ID (Chapter 6), self-restraint intensity significantly correlated with stereotypy, r_s=.18, and 'severity' of stereotypy was significantly lower in the multi-parameter 'self-restraint' group in comparison to the 'no self-restraint' group. This may be interpreted as a manifestation of repetitive/compulsive behaviour, and/or in regard to the 'proto-SIB' nature of stereotyped behaviour for some individuals (Oliver & Richards, 2015). Additionally, temporal relations in Chapter 5 suggested that self-restraint may occur as a repetitive/compulsive behaviour, with 34% of all observations of behaviour preceded and followed by more self-restraint. Thus, a key finding of this research is that across samples at-risk of pervasive SIB, self-restraint consistently associated with compulsive, repetitive and stereotyped behaviour.

The compulsivity account posits that high anxiety/arousal, either resulting directly from the 'uncontrollable' compulsive SIB, or as a predisposed mechanism, drives self-restraint (King, 1993; Romanczyk et al., 1992). Over-and-above confounding variables (section 7.2.3), compulsivity and behavioural anxiety uniquely predicted self-restraint. This serves as supportive evidence for the compulsivity account. Crucially, it was concluded that the compulsive hypothesis *compliments* rather than competes with the operant account. Whether self-restraint terminates painful SIB (negative reinforcement account), removes compulsive SIB (compulsivity account), and/or lowers arousal caused by SIB (Romanczyk et al., 1992), it is perceived by the individual as soothing, relaxing and rewarding, leading to elevated

anxiety/arousal upon self-restraint termination. This may imply that self-restraint initially emerges as an adaptive behaviour, however becomes concerning once established as intense and preferred.

The finding of compulsivity *and* anxiety uniquely predicting self-restraint allowed for the proposal of an adapted compulsive account. Chapter 6 revealed impulsivity and working memory predicted self-restraint at high levels of SIB severity. Taken together, these data give rise to the proposal of an integrated inhibitory control/compulsivity account (section 7.2.6.1).

7.2.6 Aim 6: Test the impaired inhibitory control hypothesis of self-injury and self-restraint

Meta-analysis in Chapter 2 revealed a very small, non-significant pooled effect between self-restraint and impulsivity/overactivity, r=.001. This analysis is considered with caution as it included only three samples, with an insufficient sample size (n=491) to detect small effects (N=614). Thus, a hypothesis of impaired inhibitory control was best tested using the methods employed in Chapters 3, 5 and 6.

Chapter 3 revealed self-restraint topography scores strongly positively correlated with informant-rated impulsivity within the heterogeneous sample. Interestingly, impulsivity did *not* uniquely predict self-restraint after controlling for confounds. It may be concluded that across individuals with heterogeneous causes of autism and/or ID, impulsivity is not a unique transdiagnostic risk marker for self-restraint. Within a less heterogeneous sample of autistic children with ID (Chapter 5), observed self-restraint intensity did not significant correlate with impulsivity, however the small-to-moderate effect was notable. Within the same sample,

informant-report self-restraint intensity shared a small-to-moderate non-significant correlation with impulsivity, while overactivity strongly correlated with self-restraint. Impulsivity and overactivity strongly correlated, suggesting measurement of a similar construct.

Chapter 6 employed both direct and indirect EF measures. Analyses in Chapter 4 revealed the novel direct tasks were feasible for remote administration, and were suitable for autistic children with ID. Direct and indirect measures of EF did not correlate, replicating previous findings (e.g., Toplak et al., 2013) and suggesting approaches captured distinct constructs. Controlled direct measures were conceptualised to capture the EF component more closely, while indirect measures captured broader contextual factors and real-world behaviours alongside EF.

Indirect measures of EF more closely correlated with self-restraint than direct measures. SRQ intensity scores shared a non-significant small-to-moderate association with impulsivity, and strongly correlated with poorer overactivity, impulsive speech and total activity. Impulsivity, overactivity and total activity scales approached singularity, suggesting measurement of a similar construct. The relations between self-restraint and direct measures of EF were very small (<.10). Thus, self-restraint more closely related to markers of impaired inhibition displayed in real-world environments as interpreted by caregivers, reflecting broader contextual factors alongside the EF component. Thus, a holistic approach should be adopted when considering SIB and self-restraint, accounting both for the markers of impulsivity/compulsivity (section 7.2.5) *and* supporting the caregiver to handle behaviours in real-world settings (e.g., psychoeducation).

When defining self-restraint according to multiple parameters by the SRQ2, no group differences we identified on any direct or indirect measures of EF (both at the two- and four-group levels). At first, this may suggest that impaired behavioural control (impulsivity and perseveration, conceptualised to represent habitual compulsivity) is not implicated in self-restraint. However, it is important to acknowledge that the SRQ2 was completed by a subsample of participants (42% of full sample), yielding relatively small groups and low statistical power (particularly at the four-group level, e.g., n=5). Further research replicating findings with larger samples is needed to further understanding.

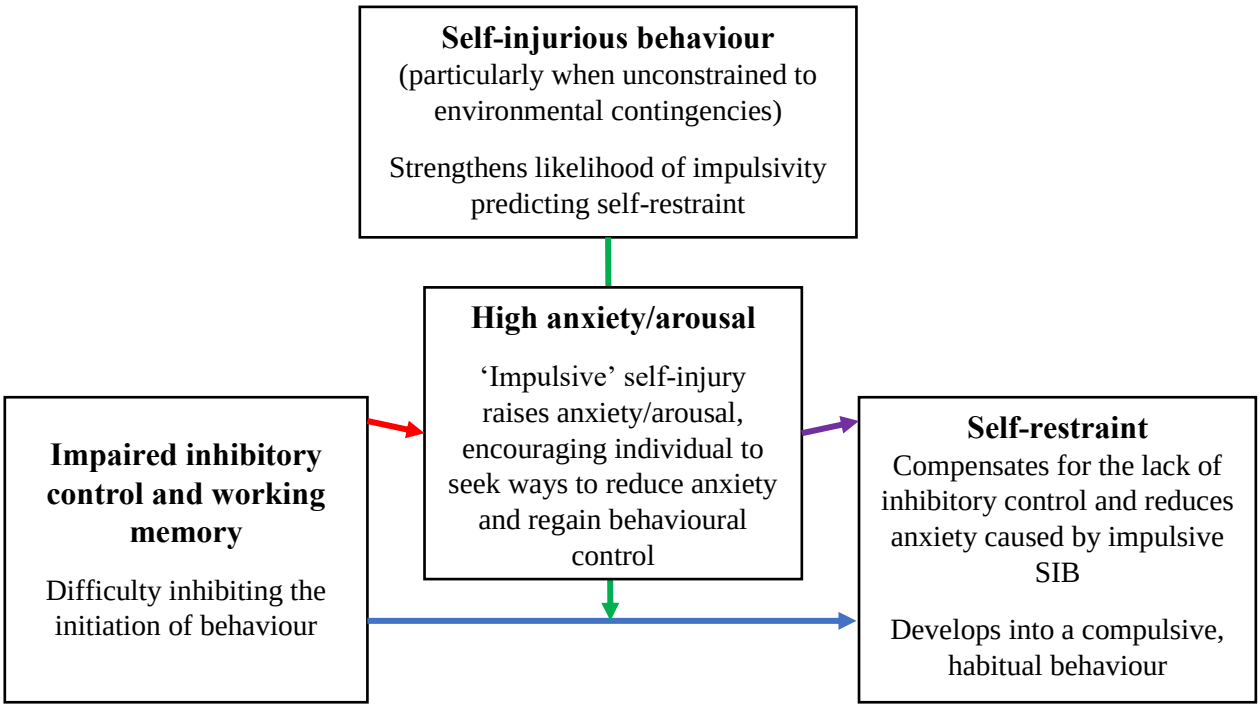
A moderation analysis controlling for age, verbal ability and adaptive functioning revealed poorer informant-rated impulsivity predicted higher self-restraint intensity at both average and high levels of SIB severity. Poorer informant-rated working memory predicted self-restraint intensity at high SIB severity. SIB severity did not moderate the relation between direct EF outcomes and self-restraint intensity scores. Importantly, the presence of SIB was disproportionately high in the present sample (e.g., in comparison to previous literature, Richards et al., 2017; Wilde et al., 2017). Altogether, these data support a hypothesis of impaired inhibitory control proposed by Oliver and Richards (2015), whereby for some individuals, self-restraint emerges in attempt to regain control over 'impulsive', automatically-reinforced SIB.

7.2.6.1 An integrated inhibitory control/compulsivity account

The adapted compulsivity account proposed in Chapter 3 suggested that self-restraint may develop into a habitual, compulsive behaviour that functions to reduce anxiety. The moderation analysis in Chapter 6, whereby impulsivity and working memory predicted self-

restraint only at high levels of SIB, suggest anxiety may be driven by self-injury that is out of the individual's control (i.e., impulsive). By integrating these two findings, a novel integrated inhibitory control/compulsivity account is proposed (represented in Figure 7.1). Further exploration of this hypothesis is warranted, including bringing together function-based assessments of self-restraint with the parameter-based approach to self-restraint measurement, and novel direct and indirect measures of EF.

Figure 7. 1 A novel integrated inhibitory control/compulsivity account



Note. Blue arrow represents predictive relation identified in Chapter 6, green arrow represents moderation identified in Chapter 6, purple arrow represents regression result in Chapter 3, and red arrow represents directions for future research.

7.2.7 Aim 7: Compare measurements of self-restraint according to different parameters

The review of key findings presented in sections 7.2.1 to 7.2.6 revealed the importance of self-restraint definition in outcomes. The final aim of this thesis was to examine the relations, or differences, between parameter-based definitions of self-restraint.

The eight-item SRC effectively gathers self-restraint topography data, while the 25-item SRQ assesses topography *and* intensity. An exploratory factor analysis (EFA) in Chapter 4 justified the psychometric suitability of 'total SRQ intensity scores'. Chapter 5 revealed a lack of association between informant-rated intensity and observed self-restraint intensity, indicating measures captured distinct constructs. SRQ intensity was considered to represent 'typical' behaviour over settings, while observation intensity reflected actual behaviour under controlled conditions. Questionnaires may be suitable for monitoring self-restraint intensity at the emergence of self-injury. Observational techniques may suitably examine functions of behaviour (e.g., the presence of high self-restraint intensity directly after SIB may suggest self-restraint functions to attenuate SIB). The SRQ and SRQ2 provide a quick measure of multi-parameter self-restraint. The SRQ2 may identify self-restraint that functions to attenuate SIB, as there was lower evidence of SIB severity as measured by the SRQ2 in comparison to the SRQ (section 7.2.4). This suggests that the additional parameter measured by the SRQ2, alongside confirmation of self-initiation of behaviour and incompatibility with SIB/symbolic restraint, is important for identifying 'functional' behaviour.

7.3 Methodological, theoretical and clinical contributions

The findings presented in this thesis provide methodological, theoretical and clinical contributions to the field of self-restraint. Regarding the methodological contributions, self-

restraint was measured according to five different approaches, with definitions increasing in restrictiveness. The clinical utility of measures is outlined in section 7.2.7. A novel, play-based battery measuring EF was developed and administered to an under-represented sample of autistic children with ID, and direct EF outcomes were compared with indirect methods. This addressed a significant gap in the autism literature, where the presence of ID typically results in study exclusion. The meticulous focus on methods and developmental appropriateness of the direct EF tasks in Chapter 4 strengthens confidence in findings, and allows for replication of methods in future populations.

Regarding the theoretical contributions, this thesis tested two accounts of self-restraint: the inhibitory control and compulsivity accounts. Chapter 6 revealed impulsivity predicted self-restraint only in the presence of average of high SIB, while Chapter 3 indicated that compulsivity and behavioural anxiety predicted self-restraint over-and-above covariates. These data provide evidence towards an integrated, adapted theory of self-restraint and self-injury, whereby 'impulsive' SIB leads to 'compulsive' self-restraint. The presence of intense and preferred self-restraint may indicate a lack of behavioural control. This may account for the emergence of a self-initiated regulatory behaviour in the presence of 'uncontrollable' (self-injurious) behaviour. Future longitudinal research integrating all model variables is required, however it is theorised that self-restraint is initially protective and goal driven (i.e., terminates SIB), however evolves into a habitual compulsive response over time. A second theoretical contribution is borne out of adopting a transdiagnostic and dimensional approach to understanding self-restraint. Continuous, severity-based variables were used to assess self-restraint, building upon the existing literature examining differences between arbitrarily-defined groups. The cross-syndrome study in Chapter 3 recognised the value of contemporary

transdiagnostic approaches to understanding behaviour, favouring trait severity over the genetic cause of autism and/or ID.

Finally, regarding clinical contributions, self-restraint was a prevalent behaviour in samples at-risk for SIB, including individuals with autism, ID and/or an associated neurogenetic syndrome. Identification of prevalent topographies, including clinging to people, people's clothing and objects, and restricting finger, hand and arm movement, aids recognition of potentially concerning behaviours. A proposed intervention for self-restraint topographies that involve use of objects, devices and/or symbolic restraints is restraint fading (e.g., Oliver et al., 1998). Future research is required to develop suitable interventions for more managing other forms of self-restraint (e.g., use of clothing/own body).

The intensity of topographies varied both across and within behaviours, indicating the importance of adopting a person-centred approach to intervention. Not all forms of self-restraint should be considered concerning, rather the presence of multiple topographies, high intensity of behaviour and anxiety or distress upon self-restraint termination indicate 'clinical' behaviours which may be occurring to attenuate SIB. Preliminary data suggested self-restraint may *also* function to manage other BoC, however the mechanism(s) underpinning the relation between self-restraint aggression and property destruction are limited to speculation. Future research should seek to examine the shared or separable relation between self-restraint and other BoC. Finally, potential risk markers for self-restraint included younger age, the presence of autism, lower adaptive functioning, low verbal ability and the presence of SIB. This implicates individuals with similar characteristics as at-risk for potentially clinical behaviours.

7.4 Overall conclusion

This thesis reconsidered the prevailing explanatory theory of self-restraint, such that behaviours function to escape painful self-injury, and implicated impaired behavioural control, evidenced by compulsivity and impulsivity, as important mechanisms underpinning behaviour. Crucially, not all self-restraining behaviours were considered 'functional', and the nuanced approach to self-restraint measurement indicated that the parameters chosen to define behaviour were important in understanding outcomes. It is concluded that while self-restraint may initially emerge as a protective, compensatory response to SIB, the emergence of multiple topographies, high behavioural intensity and a preference for behaviour may lead to clinically concerning self-restraint. Holistic, person-centred approaches to behavioural management are required in such instances. Regarding of the theoretical implications, results from this thesis suggest that there is not one 'pathway' to self-restraint, rather the interaction between operant learning paradigms and deficits in behavioural control may contribute towards the clinically concerning multi-parameter self-restraint.

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APPENDICES

Appendix 2.1: *Full database search strategy*

Name of database	Dates encompassed in search	Search strategy
Ovid Medline Ovid Medline In-Process & Other Non-Indexed Citations Ovid Embase APA PsycINFO APA PsycINFO Dissertations Abstracts International	1946 to February 05, 2021 1974 to February 05, 2021 1806 to February 05, 2021	(autism spectrum disorder* OR autism* OR autistic* OR autis* spectrum disorder* OR autis* OR ASD OR PDD-NOS OR PDDNOS OR unspecified PDD OR pervasive developmental disorder* OR (pervasive developmental disorder not otherwise specified) OR Asperger* OR Asperger* syndrome OR Kanner* OR Kanner* syndrome OR childhood autism OR classic* autism OR infantile autism) **OR** (intellectual disabilit* OR ID OR learning disabilit* OR intellectual impairment* OR mental retardation OR mental handicap* OR developmental disabilit* OR mental deficienc* OR intellectual developmental disorder* OR mental subnormalit* OR mental insufficienc* OR (intellectual and developmental disabilit*) OR IDD) **AND** (restrain* OR self restrain* OR self-restrain* OR restriction* OR impose* restrain* OR preference for imposed restrain* OR mechanical restrain* OR protective device* OR protective environment* OR self-protective device* OR SPD* OR protective appliance* OR protective equipment* OR protective helmet* OR human restrain* OR physical restrain* OR symbolic restrain* OR restrain* apparatus* OR environment* restriction* OR over restriction* OR physical prevention OR restrictive intervention OR restrict*)"

Appendix 2.1 cont.

Name of database	Dates encompassed in search	Search strategy
World Cat (restricted to thesis and dissertation content only)	Earliest available date to February 08, 2021	"(autism spectrum disorder# OR autism OR autistic OR autis* spectrum disorder* OR autis* OR ASD OR PDD-NOS OR PDDNOS OR unspecified PDD OR pervasive developmental disorder# OR (pervasive developmental disorder not otherwise specified) OR Asperger# OR Asperger# syndrome OR Kanner# OR Kanner# syndrome OR childhood autism OR classic? autism OR infantile autism) **OR** (intellectual disabilit* OR ID OR learning disabilit* OR intellectual impairment* OR mental retardation OR mental handicap# OR developmental disabilit* OR mental deficienc* OR intellectual developmental disorder* OR mental subnormalit* OR mental insufficienc* OR (intellectual and developmental disabilit*) OR IDD) **AND** (restrain* OR self restrain* OR self-restrain* OR restriction# OR impose# restrain* OR preference for imposed restrain* OR mechanical restrain* OR protective device# OR protective environment# OR self-protective device* OR SPD* OR protective appliance# OR protective equipment# OR protective helmet# OR human restrain* OR physical restrain* OR symbolic restrain* OR restrain* apparatus* OR environment* restriction* OR over restriction* OR physical prevention OR restrictive intervention OR restrict*)"

Appendix 2.2: *Additional data obtained for meta-analysis*

Sample	Description of additional data
Hagopian et al, 2017	Topography data
Laverty et al, 2020	Raw questionnaire data
Oliver et al, 2003 Devices sample	Main author's unpublished PhD thesis
Richards et al, 2017 (both samples)	Main author's unpublished PhD thesis + raw questionnaire data
Rojahn 1986	Unpublished manuscript

Appendix 2.3: *Amendments to quality assessment framework following reliability consensus meetings*

Domain	Change to criteria following consensus meetings
A	• Specified use of two samples
B	• Added "Diagnosis of autism obtained from official records"
C	• Added "Diagnosis of ID included in the study eligibility criteria" • Removed "Self/parent/teacher report using a well-validated measure of intellectual functioning" • Specified formal IQ and adaptive functioning measure can be administered by researcher

Appendix 2.4: *Justification for quality assessment framework ratings*

Sample	Domain	Score	Justification	Overall mean score
Bruhl et al., 1982	A	2	• Three non-random samples (Faribault State Hospital, Cambridge State Hospital and Brainerd State Hospital) (p. 221)	0.42
	B	1	• Obtained from records – "the etiological diagnosis of childhood autism and childhood schizophrenia had been made in four project participants" (p. 221)	
	C	1	• Recruited from specialist service	
	D	1	• Observation, inter-rater reliability not stated	
Favell et al., 1981	A	2	• "Two residential Units serving… Western Carolina Centre… [and] a unit at another mental retardation facility" (p. 426)	0.44
	B	N/A	-	
	C	1	• Recruited from service (Vineland age equivalents are provided but not clear if conducted within present study)	
	D	1	• Unvalidated survey – Friedin (1977) Rating Scale	
Forman, 2003	A	2	• Multiple non-random samples "Day services, council run, private and charity residential homes, clinical psychologists in the West Midlands and social services in three districts (Birmingham, Sandwell & Solihull)…schools for individuals with severe learning disabilities" (p. 126)	0.67
	B	1	• Screening instrument – DASH-II (Diagnostic Assessment of Severely Handicapped-II)	
	C	3	• Parent report of adaptive functioning – VABS (Vineland Adaptive Behavior Scale)	
	D	2	• Informant report (Self-Restraint Questionnaire + Self-Restraint Questionnaire 2)	

Appendix 2.4 cont.

Sample	Domain	Score	Justification	Overall mean score
Fovel et al., 1989	A	1	• Single restricted sample "state-supported school for retarded persons in Massachusetts" (p. 378) & recruited from previous study	0.22
	B	N/A	-	
	C	1	• Participants were residents of state-supported school for "retarded persons" for 3+ years (ID not confirmed in this study, no mention of previous IQ test)	
	D	0	• Measure not specified	
Hagopian et al., 2015	A	2	• Multi-region specialist clinic (Kennedy Krieger Institute)	0.50
	B	1	• "We obtained diagnoses of autism spectrum disorder… from review of official records" (p.526)	
	C	1	• "We obtained diagnoses of… level of intellectual disability… from review of official records" (p.526)	
	D	2	• Two trained observers, overall inter-rater reliability = 91.7%	
Hyman et al., 2002	A	2	• National non-random sampling (CdLS Foundation UK, national parent support group)	0.67
	B	N/A	-	
	C	2	• Parent report – Wessex Scale	
	D	2	• Informant report (Self-Restraint Checklist)	

Appendix 2.4 cont.

Sample	Domain	Score	Justification	Overall mean score
Iwata et al., 1994	A	2	• Multiple non-random samples – "Inpatients in a paediatric hospital or living in a state residential facility"	0.56
	B	N/A	-	
	C	1	• Recruitment from specialist services (not confirmed in this study)	
	D	2	• Two observers "accuracy percentages for experimenter behavior ranged from 72 to 100%" (p. 219	
Laverty et al., 2020	A	2	• National non-random sample – National Autistic Society	0.58
	B	1	• All participants had a clinical diagnosis of autism & were excluded if scored below cut off at two time points on SCQ	
	C	2	• The Wessex was used	
	D	2	• Informant report (Self-Restraint Checklist)	
Oliver et al., 2003 Devices	A	1	• Recruited from previous study (Oliver et al., 1987) which states participants were recruited from one health region	0.42
	B	1	• Judgement against "ICD 10 diagnostic criteria for atypical autism (F85.1) and childhood disintegrative disorder (F85.2) (World Health Organisation, 1986)" (p.546 thesis)	
	C	1	• Recruited from previous study (Oliver et al., 1987) which states participants were "people known to mental handicap services (IQ<70)" (p. 148)	
	D	2	• Informant report (Self-Restraint Questionnaire)	

Appendix 2.4 cont.

Sample	Domain	Score	Justification	Overall mean score
Oliver et al., 2003 No Devices	A	1	• Recruited from previous study (Oliver et al., 1987) which states participants were recruited from one health region	0.44
	B	N/A	-	
	C	1	• Recruited from previous study (Oliver et al., 1987) which states participants were "people known to mental handicap services (IQ<70)" (p. 148)	
	D	2	• Informant report (Self-Restraint Questionnaire)	
Powell et al., 1996	A	1	• Single regional sample "state developmental centre" (p. 42)	0.44
	B	N/A	-	
	C	1	• Recruited from a specialist ID service, inclusion criteria "had severe or profound mental retardation as determined by standardised intellectual and adaptive behaviour assessments" (p. 42) however measures were not named nor discussed any further	
	D	2	• Informant report (Self-Restraint Checklist)	
Richards et al., 2017 Adults	A	2	• National non-random sample – National Autistic Society	0.67
	B	2	• Previous diagnosis unconfirmed in present study "Service provision from NAS necessitates an ASD diagnosis from a qualified professional" (p. 703)	
	C	2	• The Self-injury, Aggression and Destruction Screening Questionnaire (SAD-SQ), containing items from the Wessex Scales	
	D	2	• Informant report (Self-Restraint Checklist)	

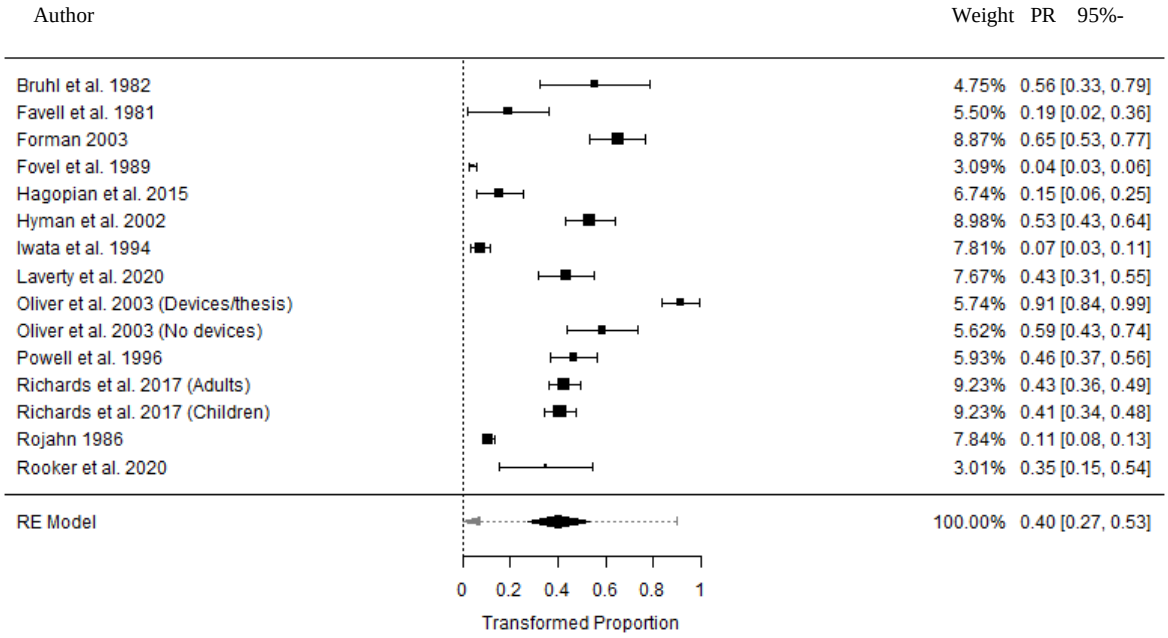
Appendix 2.4 cont.

Sample	Domain	Score	Justification	Overall mean score
Richards et al., 2017 Children	A	2	• National non-random sample – National Autistic Society	0.67
	B	2	• Previous diagnosis unconfirmed in present study "Service provision from NAS necessitates an ASD diagnosis from a qualified professional" (p. 703)	
	C	2	• The Self-injury, Aggression and Destruction Screening Questionnaire (SAD-SQ; Davies & Oliver, 2016), containing items from the Wessex Scales	
	D	2	• Informant report (Self-Restraint Checklist)	
Rojahn, 1986	A	2	• Multiple non-random samples "individuals registered in schools, training centres, workshops or group homes of the Life Support for the Mentally Retarded…estimated [to serve] 300,000 retarded persons across the Federal Republic of Germany" (p. 269)	0.56
	B	N/A	-	
	C	2	• Informant report of adaptive functioning – Vineland Social Maturity Scale (Doll, 1935)	
	D	1	• Unvalidated survey "four forms of stereotypy were to be scored present or absent…. self-restraint of the arms in own clothing" (p. 269)	
Rooker et al., 2020	A	1	• Single regional sample – inpatient unit	0.25
	B	0	• Unspecified	
	C	1	• Recruited from specialist service – "Inclusion criteria were… a diagnosis of IDD [intellectual and developmental disabilities]" (p. 1044)	
	D	1	• Observation, inter-rater reliability not stated	

Appendix 2.5: *Descriptions of genetic syndromes reported across samples*

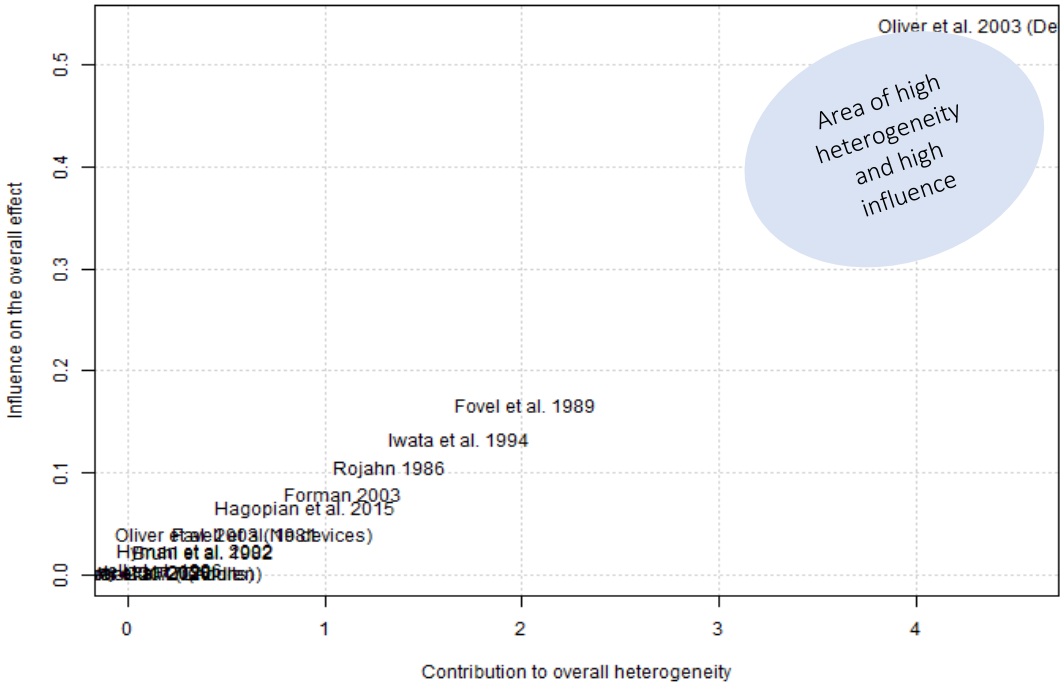
Genetic syndrome name	**Participants with diagnosis (*n*)**	**Sample(s)**
Congenital Rubella	1	Iwata et al.
Cornelia de Lange	91	Hyman et al. Iwata et al. Rooker et al.
Down syndrome	14	Bruhl et al. Iwata et al. Oliver et al. Devices
Fragile X	1	Forman
Genetic cause of ID	4	Oliver et al. Devices
Lesch-Nyhan	1	Iwata et al.
Lowe	1	Forman
Phenylketonuria (PKU)	4	Bruhl et al. Iwata et al.
Rett	3	Iwata et al.
Smith-Magenis	1	Forman
Tuberous Sclerosis Complex	3	Forman
Total	**124**	**6**

Appendix 2.6: *Forest plot for quality effects model for the overall prevalence of self-restraint*

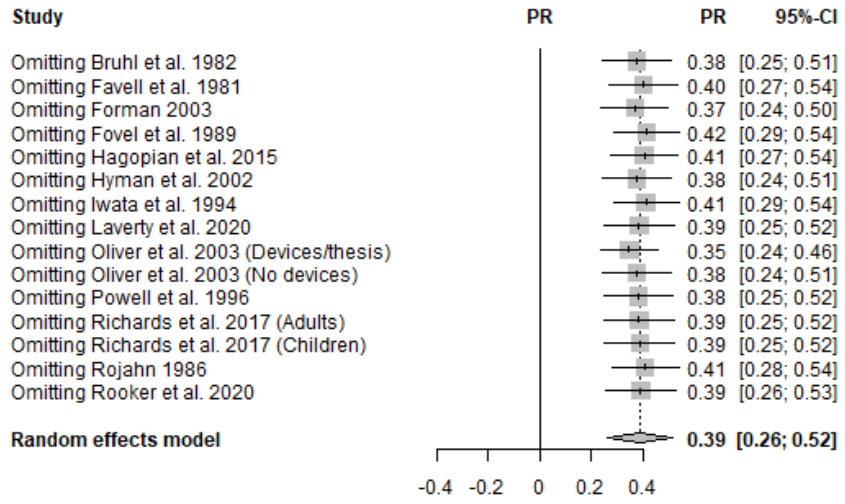


Appendix 2.7: *Overall prevalence of self-restraint heterogeneity checks*

a. Baujat chart of sources of heterogeneity for the overall prevalence of self-restraint



b. Overall pooled prevalence estimate for self-restraint "leave-one-out" analysis



Appendix 2.8: *Categorisation of observed self-restraint topographies across questionnaire items*

Questionnaire measure		**Observational measure**		
Measure and item	**Description of item**	**Study**	**Verbatim description of observed topography**	***n* engaged in topography**
SRC item 1	Wraps self in own clothing	Bruhl et al., 1982	Wraps himself up into shirt; Crawls into his T-shirt; Wraps herself up into her pajamas at night	4
		Favell et al., 1981	Wrapping his arms in whatever cloth was available (e.g., bed linen or his shirt)	
SRC item 2	Holds onto others, holds onto others' clothing	Bruhl et al., 1982	Hooks arms under staff's arms while near staff	2
		Hagopian et al., 2015	Holding onto others	
SRC item 3	Positions self to restrain	Hagopian et al., 2015	Placing BOTH arms under his body (includes under his leg)	1

Note. SRC = Self-Restraint Checklist.

Appendix 2.8 cont.

Questionnaire measure		**Observational measure**		
Measure and item	**Description of item**	**Study**	**Verbatim description of observed topography**	***n* engaged in topography**
SRC item 7	Chooses mechanical restraint	Bruhl et al., 1982	Constantly looked for his posey-blet and put himself into it or wrapped it around his neck, though not with any evident suicidal intent	4
		Favell et al., 1981	Jane enjoyed being restrained and consistently attempted to sit in the restraint chair; He had a 9-year history of being continuously restrained in a variety of devices, including a camisole, arm splints and a wrist-to-waist belt	
		Hagopian et al., 2015	Wearing the weighted blanket over her body	
SRQ item 1	Entwine his or her hands in the front of the clothes that are worn e.g. t-shirt, jumper?	Hagopian et al., 2015	Wrapping hand(s) in shirt	1
SRQ item 2	Pull his or her arms out of the sleeves and push them part or all of the way into the main body of the clothes that are worn	Bruhl et al., 1982	Pushes hands into his shirts; Wraps arms into his T-shirt; Hides arms high up under blouse; Restrains his right arm in his shirt at night	5
		Hagopian et al., 2015	Placing BOTH arms inside shirt	

Note. SRC = Self-Restraint Checklist; SRQ = Self-Restraint Questionnaire.

Appendix 2.8 cont.

Questionnaire measure		Observational measure		
Measure and item	**Description of item**	**Study**	**Verbatim description of observed topography**	***n* engaged in topography**
SRQ item 3	Pull the sleeves of his or her clothes over hands if clothes have long sleeves?	Hagopian et al., 2015	Covering her hands with her stockinet e.g., pulling her stockinet over her hand	1
SRQ item 4	If permitted, does he or she push hands inside trousers, if he or she is wearing them?	Bruhl et al., 1982	Puts hands in pants	1
SRQ item 6	Wrap his hands/arms together with any piece of long material e.g. sock, scarf when there is access to the material	Bruhl et al., 1982	Wraps towels around forearms	1
SRQ item 7	Wrap a long piece of material e.g. sock, scarf, around either or both arms but not so that they are held together, when there is access to the material	Hagopian et al., 2015	Wrapping arms in clothing	1

Note. SRQ = Self-Restraint Questionnaire.

Appendix 2.8 cont.

Questionnaire measure		Observational measure		
Measure and item	**Description of item**	**Study**	**Verbatim description of observed topography**	***n* engaged in topography**
SRQ item 8	Wrap or entwine hands in material other own clothes e.g. curtains, bed clothes, other clothing, if it is available	Hagopian et al., 2015	Wrapping hands in a towel, sheet or clothing; wrapping arms/hands in shoe laces	2
SRQ item 9	Wrap a blanket, sheet, towel or other large piece of material around himself or herself if it is available	Bruhl et al., 1982	Wraps himself up into blankets	1
SRQ item 10	Entwine arms around a chair or other piece of furniture	Bruhl et al., 1982	Hooks arms under arms of chair	2
		Hagopian et al., 2015	Putting arms through mat handles (will not include restraint on therapist attempts)	
SRQ item 13	Lie/sit in a position which restricts the movement of arms e.g. with them under the body, behind the back or pressed against a wall	Hagopian et al., 2015	Lying on hands/arms; Laying on arms/hands; Laying on top of one or both arms or hands	3
SRQ item 14	Carry an object or objects around but not manipulate, use or play with them	Bruhl et al., 1982	Carries pillow or blanket over his head	1

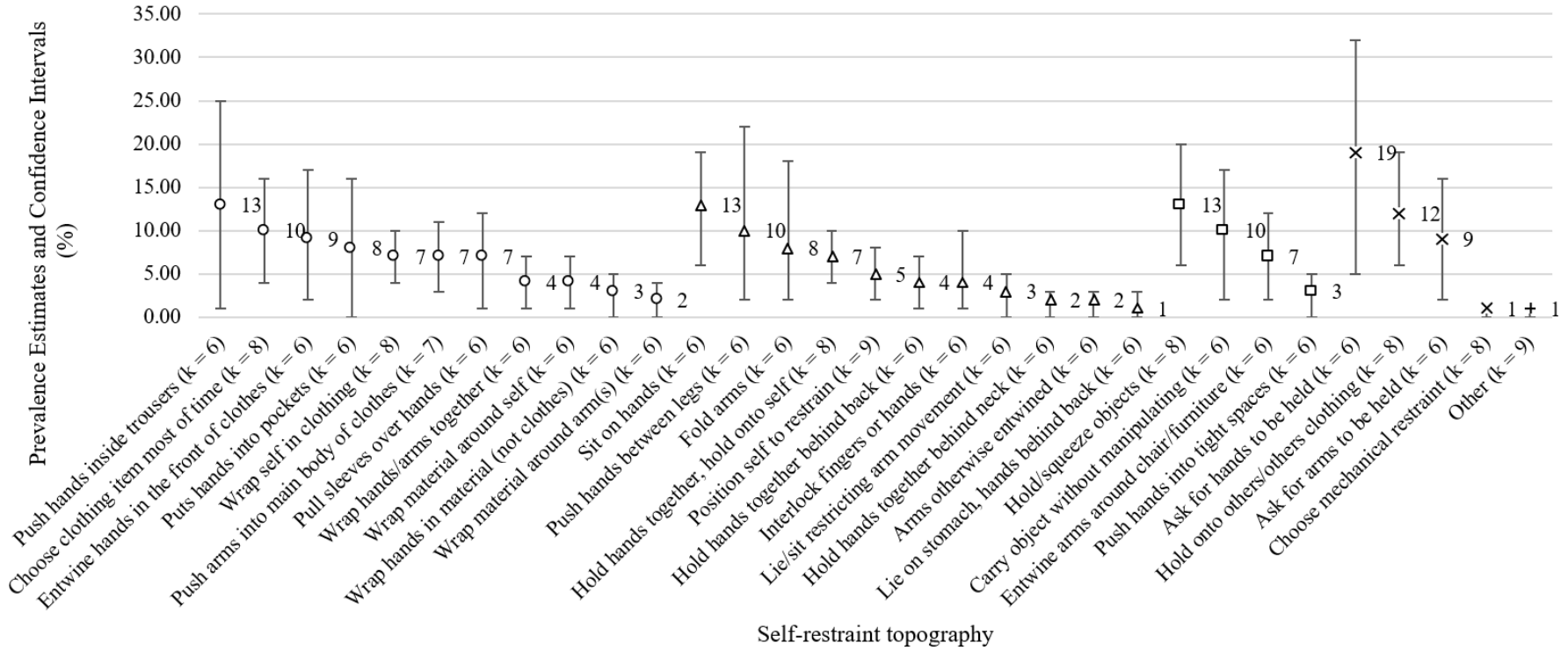
Note. SRQ = Self-Restraint Questionnaire.

Appendix 2.8 cont.

Questionnaire measure		**Observational measure**		
Measure and item	**Description of item**	**Study**	**Verbatim description of observed topography**	***n* engaged in topography**
SRQ item 15	Sits on hands	Bruhl et al., 1982	Sits on hands (at times); Sits on hands	7
		Hagopian et al., 2015	Sitting on her hands; Sitting on hands/arms; Sitting on arms/hands; Sitting on hands; Sitting on one or both hands	
SRQ item 18	Push hands between the legs	Hagopian et al., 2015	Placing one or both hands in between her legs	1
SRQ item 19	Folds arms	Hagopian et al., 2015	Folding arms	1
SRQ item 22	Does he or she ask or otherwise indicate that he or she wants hands held e.g. putting hands into another's or under another's arm?	Hagopian et al., 2015	Holding therapists hand	1
SRC item 8 / SRQ item 25	Other	Bruhl et al., 1982	Wraps fingers up into her necklace during day time; Hides hands behind apron	8
		Hagopian et al., 2015	Sitting or laying on others with her hands/arms between bodies; Placing a pillow over his body; Placing hands, arms or legs under pillow, Covering hands or head with items (i.e. mat, blanket), Crossing legs	

Note. SRC = Self-Restraint Checklist; SRQ = Self-Restraint Questionnaire.

Appendix 2.9: *Prevalence estimates and confidence intervals of each topography of self-restraint for all participants*



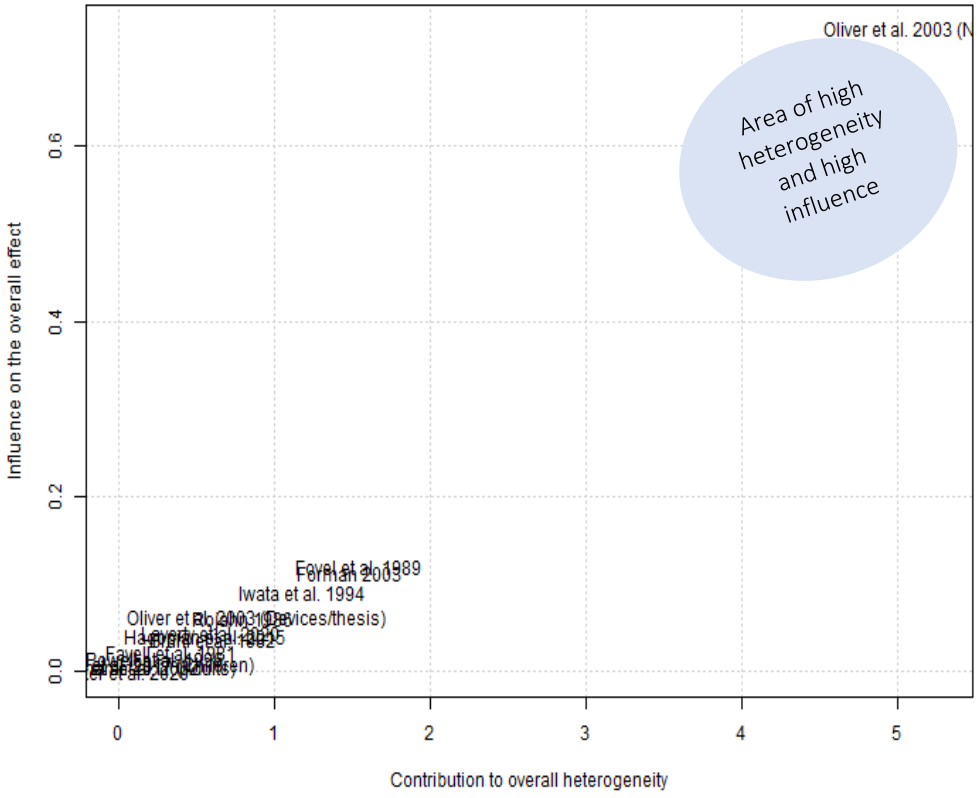
○ Estimates for topographies involving the use of clothing or materials
△ Estimates for topographies involving the use of body
□ Estimates for topographies involving the use of objects
✕ Estimates for topographies involving a preference for imposed restraint
+ Estimates representing other forms of self-restraint

Appendix 2.10: *Qualitative descriptions of "other" forms of self-restraint*

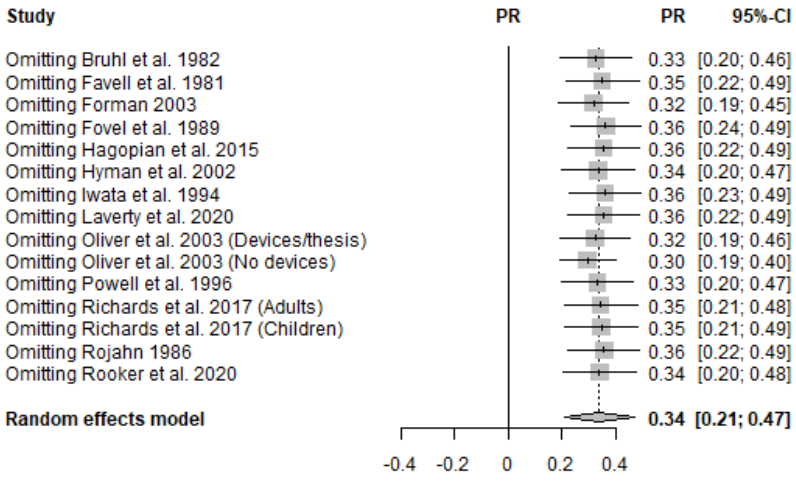
Author	***n* "other" topography**	**Description of self-restraint**
Bruhl et al.	2	• "hides hands behind apron" (p. 228) • "constantly looked for posey-belt and put himself into it or wrapped it around his neck, though not with any evident suicidal intent" (p. 228)
Hagopian et al.	3	• "placing a pillow over his body, or placing hands, arms or legs under pillow" • "crossing legs" • "covering hands or head with items (i.e. mat, blanket)" (author-sent observational data)
Laverty et al.	2	• "Sits under beanbag" • "Wraps self in sofa throw" (author-sent data, parent report)
Richards et al. Adults	1	• "Locks doors" (author-sent data, parent report)
Richards et al. Children	1	• Pushes body against walls (author-sent data, parent report)

Appendix 2.11: *Prevalence of self-restraint in the presence of self-injurious behaviour heterogeneity checks*

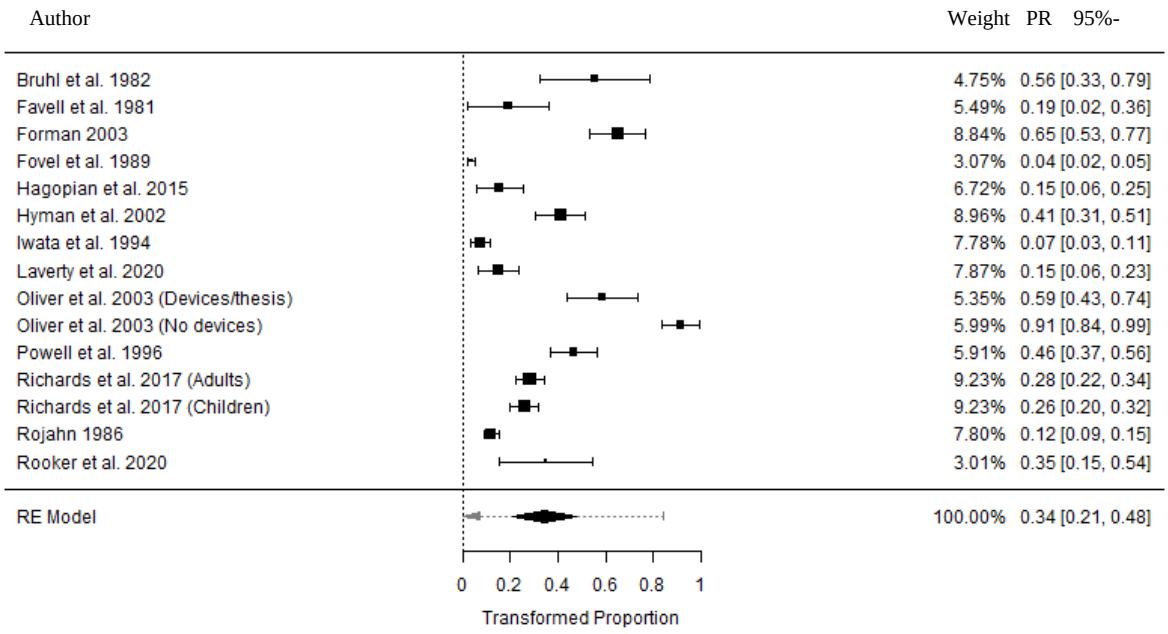
a. Baujat chart of sources of heterogeneity for the prevalence of self-restraint in individuals who self-injure



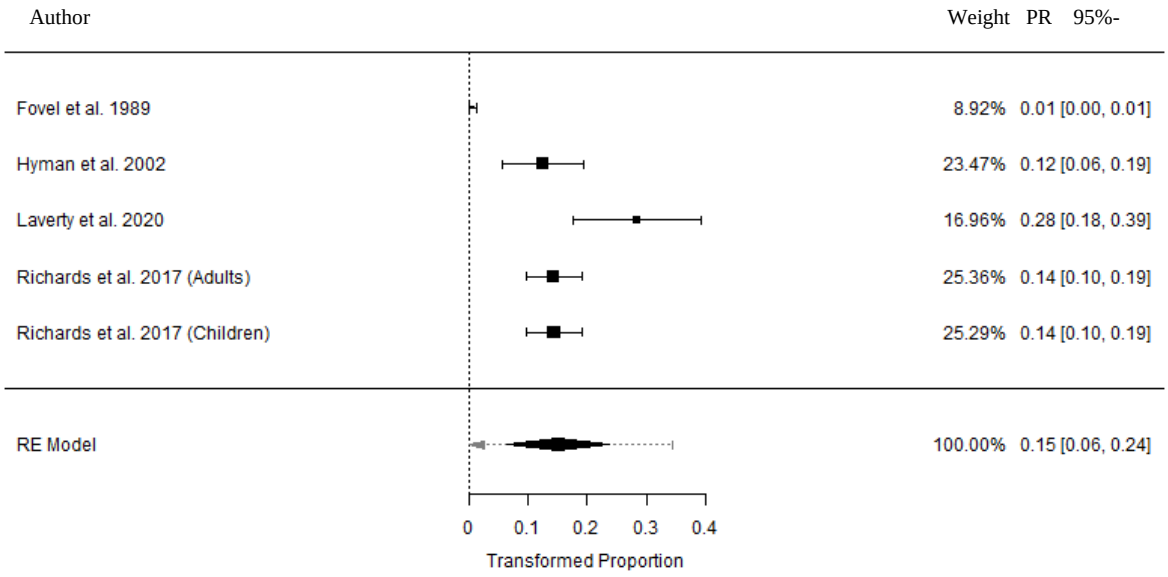
b. "Leave-one-out" analysis for the prevalence of self-restraint in the presence of self-injury



Appendix 2.12: *Quality-rated forest plot for the prevalence of self-restraint that co-occurs with self-injury*

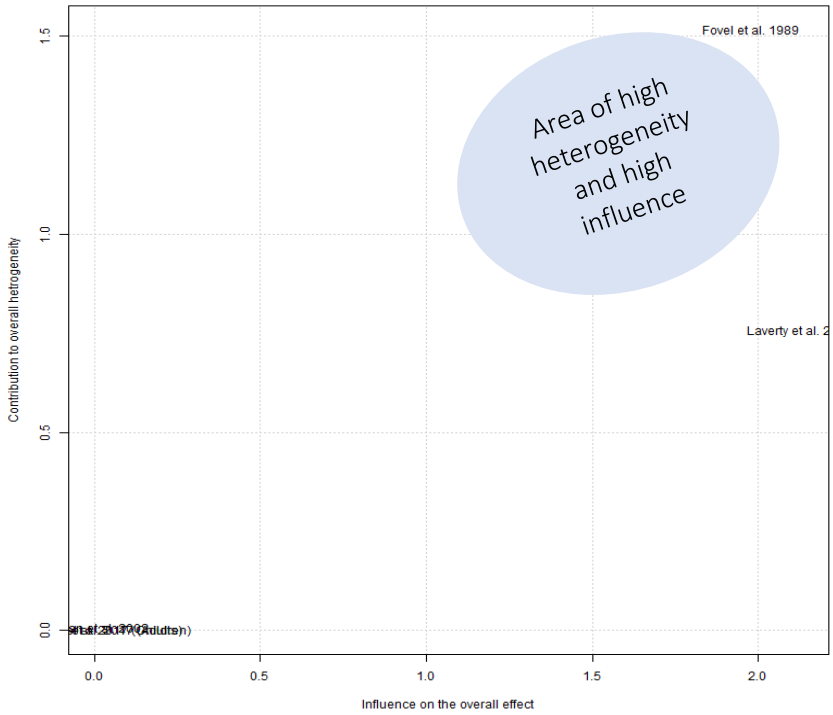


Appendix 2.13: *Quality-rated forest plot for the prevalence of self-restraint in the absence of self-injury*

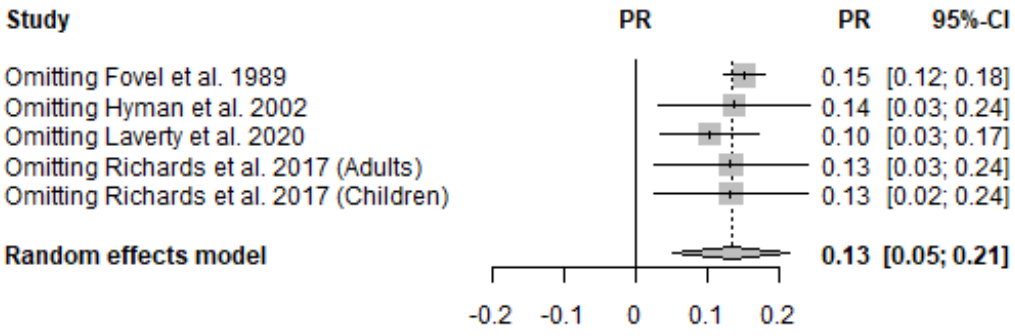


Appendix 2.14: *Prevalence of self-restraint in the absence of self-injurious behaviour heterogeneity checks*

a. Baujat chart of sources of heterogeneity for the prevalence of self-restraint in the absence of self-injury

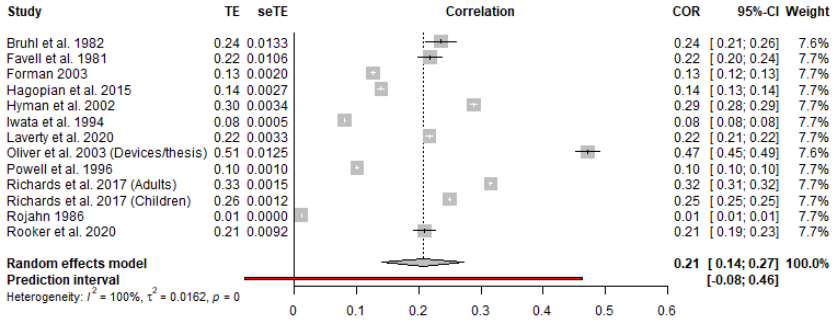


b. Leave-one-out" analysis for the prevalence of self-restraint in the absence of self-injury

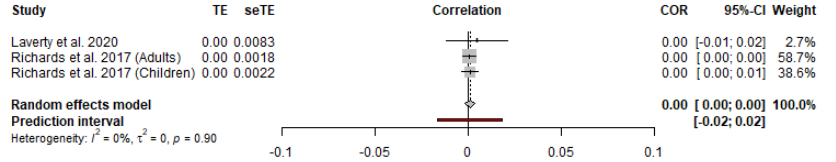


Appendix 2.15: *Random effects forest plots for each correlation analysis*

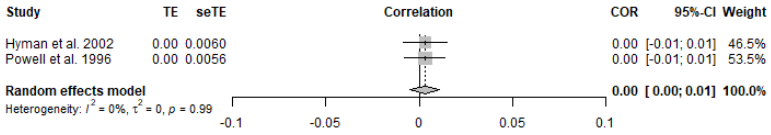
a. Self-injurious behaviour and self-restraint



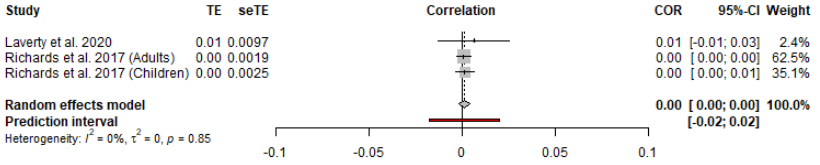
b. Restricted/repetitive behaviours and self-restraint



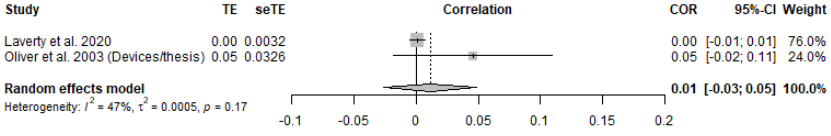
c. Compulsive behaviours and self-restraint



d. Impulsivity/overactivity and self-restraint

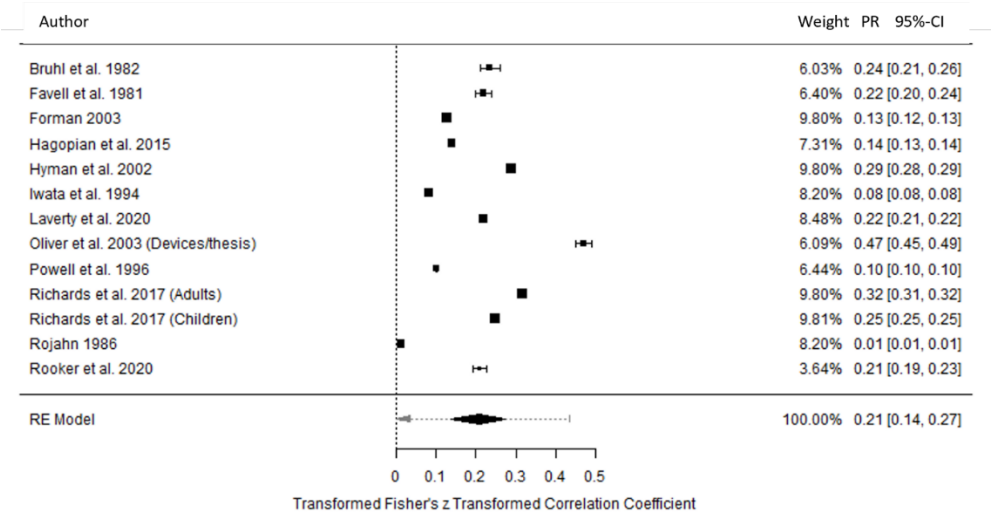


e. Stereotyped behaviours and self-restraint

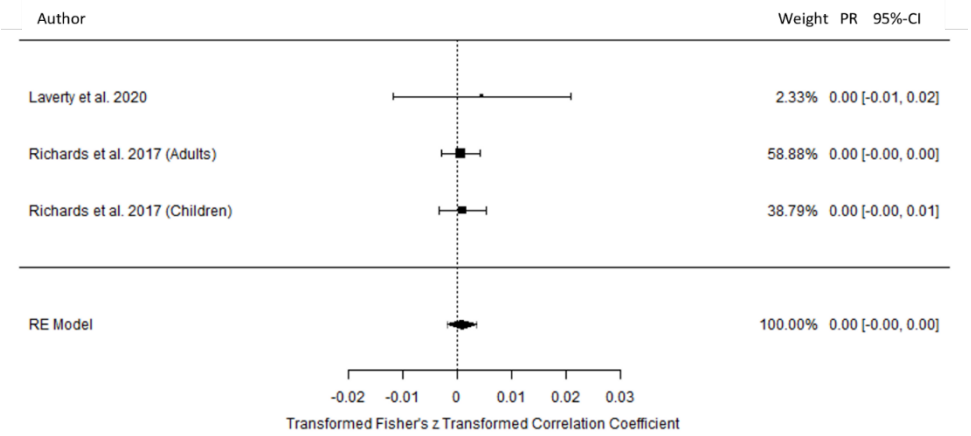


Appendix 2.16: *Forest plots for quality-rated random effects model for each correlation analysis*

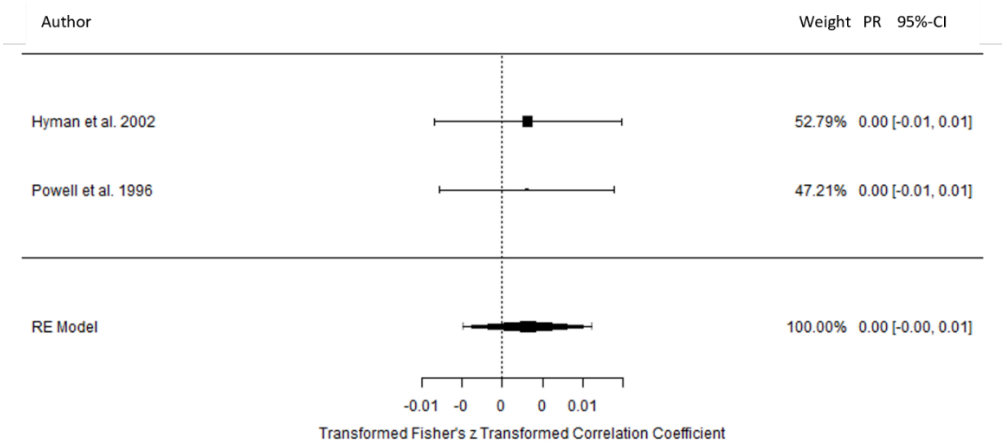
a. Self-injurious behaviour and self-restraint



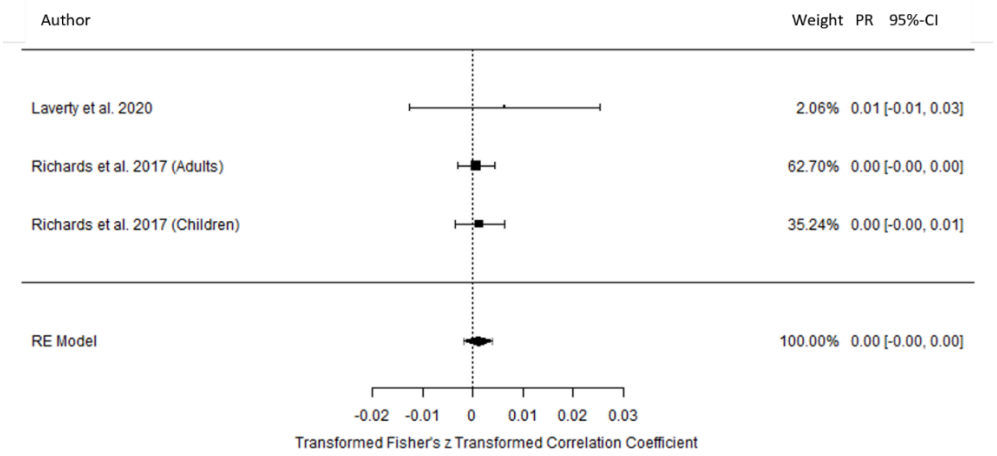
b. Restricted/repetitive behaviours and self-restraint



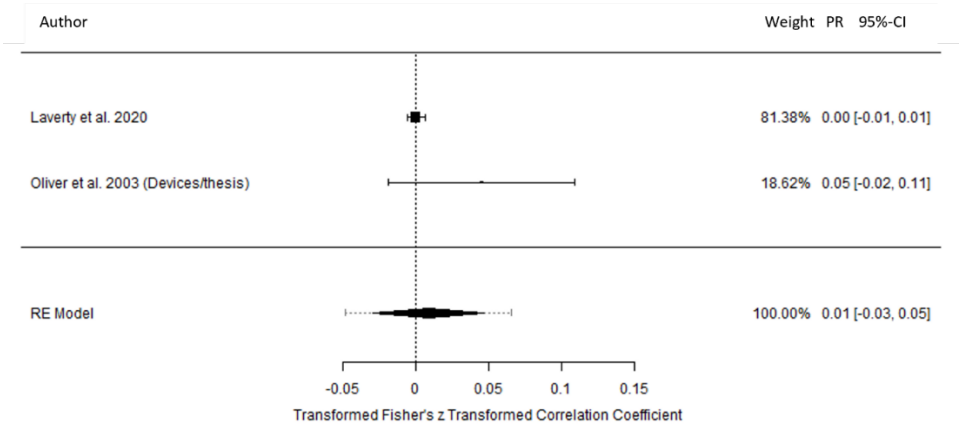
c. Compulsive behaviours and self-restraint



d. Impulsivity/overactivity and self-restraint



e. Stereotyped behaviours and self-restraint



Appendix 3.1: *BEOND study consent form*

Behavioural and Emotional Outcomes in individuals with Neurodevelopmental Disorders (BEOND)

Study Directors: Dr Caroline Richards, Dr Hayley Crawford, Dr Jane Waite & Dr Jo Moss

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Consent Form for Parents/Caregivers and Individuals Under 16 Years Old

Version 2, 08/09/2022

PLEASE READ THIS SECTION FIRST

Now it is up to you to decide whether or not you and your child/the person you care for would like to participate. The decision about whether or not to take part in the study must be 'informed'. This means that anyone making the decision must understand exactly what is involved in the study, what will be required from participants and why. They should have read and understood both the **BEOND Information Sheet** *(v2 07/09/2022)* and the **GDPR Privacy Statement** *(v2 05/09/2022)*.

As the person completing the survey we require consent from the parent/caregiver who intends to complete the survey. However, because the survey collects a lot of information about your child/the person you care for, it is also important to consider how they feel about participation.

A symbol information sheet is provided in this pack to help you explain the study to your child/the person you care for. If you are unsure whether or not your child/the person you care for is able to understand enough about the BEOND study to make a decision independently then the study team can provide you with support in how to assess this.

If your child/the person you care for is **unable** to understand the study information then as parent/caregiver you can provide consent for both of you. If your child/the person you care for is **able** to understand the study information then we would also seek assent from them that they are happy for you to share their information with us.

Please make sure you have read and understood the information sheet and thought about whether your child/the person you care for would be able to understand the BEOND study. Please select the appropriate option and complete the sections as instructed:

To the best of my knowledge I believe my child/the person I care for has understood the study information and is capable of making a decision about whether they would like to participate in the study. *Please complete **section 1** and then have your child/the person you care for complete **section 2***	☐
To the best of my knowledge I believe my child/the person I care for is unable to understood the BEOND study information and would not be capable of making a decision about whether they would like to participate in the study. *You as the parent/caregiver will need to provide consent for your child/the person you care for, please **complete section 1 only**.* **By ticking this box you acknowledge that in consenting yourself you are also providing consent on behalf of your child/the person you care for.**	☐

SECTION 1 For the person completing the survey

Please indicate your response to the following statements by circling 'yes' or 'no' as appropriate:

1.	I confirm that I have read and understood the participant information sheet for the BEOND study (*v2 07/09/2022*). I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	YES NO
2.	I agree to my personal data and that of my child/person I care for and data relating to me and that of my child/person I care for collected during the study being processed as described in the Participant Information Sheet.	YES NO
3.	I understand that if during the study I tell the research team something that causes them to have concerns in relation to my health and/or welfare or that of my child/person I care for they may need to breach my confidentiality.	YES NO
4.	I understand that my participation and that of my child/person I care for is voluntary and that I am free to withdraw at any time without giving any reason, without my or that of my child's/person I care for's medical care or legal rights being affected.	YES NO
5.	I agree to take part in the above study.	YES NO

*Optional clauses, the statements below are **optional**:*

6.	I agree to the Cerebra Network for Neurodevelopmental Disorders sharing my research data with any professionals or clinicians working with me and the person I care for should they request to see them.	YES NO
7.	I agree to a member of the research team contacting the person I care for's GP to confirm any current diagnosis. I understand that if such information is unavailable then a member of the research team will contact me to arrange genetic testing the in form of a saliva swab.	YES NO
8.	I agree to my anonymised data being used by research teams outside of the Cerebra Network for future research.	YES NO
9.	I would like to receive a personalised feedback report detailing the results from my child/the person I care for.	YES NO

Signature _____ Date _____

These are the details that we will use for all future contact relating to BEOND.
Please complete in block capitals and ensure that all details are accurate before continuing.

Your name:

_____ _____
First name(s) *Surname*

Name of your child/person you care for:

_____ _____
First name(s) *Surname*

Your relationship to your child/the person you care for:

_____

Contact details

Address: _____

_____

_____

Postcode: _____

Telephone number: _____

Email address: _____

Optional *if you have consented to point 7 above, please complete GP details below*

Name of your child/the person you care for's GP: _____

GP Address: _____

GP Telephone Number: _____

If you have medical letters or documents that confirm your child/the person you care for's diagnosis that you are happy to share with us, then you may include a photocopy when you return this pack. Otherwise, email k.a.wade@bham.ac.uk to discuss secure ways of sharing documentation with us digitally.
DO NOT EMAIL MEDICAL DOCUMENTATION AS AN ATTACHMENT.

SECTION 2 For the person being reported on

If your child/the person you care for is able to understand the information provided in the information sheet, either independently or with your support, then please get them to fill out this section.

Please let us know if you agree with each of the below statements by circling 'yes' if you agree, or 'no' if you disagree:

1. Has somebody else explained the project to you or have you read the information?	YES	NO
2. Do you understand what the project is about?	YES	NO
3. Have you asked all of the questions you want?	YES	NO
4. Have you had your questions answered in a way you understand?	YES	NO
5. Do you understand it is OK to stop taking part at any time?	YES	NO
6. Are you happy to take part?	YES	NO

If you circled '**no**' for **any** of the statements, or if you don't want to take part, then **stop here** and don't fill out any more of the form.

If you circled '**yes**' for **all** of the statements then please answer these extra statements and then write your name in the space below.
These statements are **optional**. You can choose if you want to say 'yes' to these. Even if you say 'no to these you can still take part and we will not do anything you say 'no' to:

7. If your doctor/other medical professional asks to see your results from this project, it is OK for us to share this with them?	YES	NO
8. Are you happy for us to look at your DNA test results, or to send you a swab so we can find out about your DNA?	YES	NO
9. Are you happy for us to contact you again in the future	YES	NO

Your name: _____ Date: _____

Please also tell us who explained the study to you:

Print name: _____ Date: _____

Please double check the details you have provided before moving on





Behavioural and Emotional Outcomes in individuals with Neurodevelopmental Disorders (BEOND)

Study Directors: Dr Caroline Richards, Dr Hayley Crawford, Dr Jane Waite & Dr Jo Moss

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Consent Form for Parents/Caregivers and Individuals Over 16 Years Old

PLEASE READ THIS SECTION FIRST

Now it is up to you to decide whether or not you and your child/the person you care for would like to participate in the BEOND study. The decision about whether or not to take part in the study must be 'informed'. This means that anyone making the decision must understand exactly what is involved in the study, what will be required from participants and why. They should have read and understood both the **BEOND Information Sheet** *(v2 07/09/2022)* and the **GDPR Privacy Statement** *(v2 05/09/2022)*.

As the person completing the survey we require consent from the parent/caregiver who intends to complete the survey. However, because the survey collects a lot of information about your child/the person you care for, it is also important to consider how they feel about participation.

A symbol information sheet is provided in this pack to help you explain the study to your child/the person you care for. If you are unsure whether or not your child/the person you care for is able to understand enough about the BEOND study to make a decision independently then the study team can provide you with support in how to assess this.

Please make sure you have read and understood the information sheet and thought about whether your child/the person you care for would be able to understand the BEOND study. Please select the appropriate option and complete the sections as instructed:

To the best of my knowledge I believe my child/the person I care for has understood the BEOND study information and is capable of making a decision about whether they would like to participate in the study. *Please complete **section 1** and then have your child/the person you care for complete **section 2***	☐
To the best of my knowledge I believe my child/the person I care for unable to understand the BEOND study information and would not be capable of making a decision about whether they would like to participate in the study. *You will need to identify an appropriate consultee – please refer to the consultee information sheet for guidance. The person completing the survey should complete **section 1.** If someone else is acting as consultee they should complete **section 3**.*	☐

SECTION 1 For the person completing the survey

Please indicate your response to the following statements by circling 'yes' or 'no' as appropriate:

1. I confirm that I have read and understood the participant information sheet for the BEOND study (*v2 07/09/2022*). I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. YES NO
2. I agree to my personal data and that of my child/person I care for and data relating to me and that of my child/person I care for collected during the study being processed as described in the Participant Information Sheet. YES NO
3. I understand that if during the study I tell the research team something that causes them to have concerns in relation to my health and/or welfare or that of my child/person I care for they may need to breach my confidentiality. YES NO
4. I understand that my participation and that of my child/person I care for is voluntary and that I am free to withdraw at any time without giving any reason, without my or that of my child's/person I care for's medical care or legal rights being affected. YES NO
5. I agree to take part in the above study. YES NO

*Optional clauses, the statements below are **optional**:*

6. I agree to the Cerebra Network for Neurodevelopmental Disorders sharing my research data with any professionals or clinicians working with me and the person I care for should they request to see them. YES NO
7. I agree to a member of the research team contacting the person I care for's GP to confirm any current diagnosis. I understand that if such information is unavailable then a member of the research team will contact me to arrange genetic testing the in form of a saliva swab. YES NO
8. I agree to my anonymised data being used by research teams outside of the Cerebra Network for future research. YES NO
9. I would like to receive a personalised feedback report detailing the results from my child/the person I care for. YES NO

If you are also acting as consultee on behalf of your child/the person you care for please read and respond to points 10 & 11 below.
If you are NOT in a position to act as consultee, please leave points 10 & 11 blank and have the person acting as consultee complete section 3.

10. I confirm that I have read and understood the consultee information sheet (*v2 07/09/2022*) and that I am an appropriate person to act as consultee to my child / the person I care for, for the purposes of the BEOND study. YES NO
11. It is my opinion as consultee that my child/the person I care for would have no objection to participating in the BEOND study YES NO

_____ _____

Signature Date

These are the details that we will use for all future contact relating to BEOND.
Please complete in block capitals and ensure that all details are accurate before continuing.

Your name:

_____ _____
First name(s) *Surname*

Name of your child/person you care for:

_____ _____
First name(s) *Surname*

Your relationship to your child/the person you care for:

_____

Contact details

Address: _____

_____

_____

Postcode: _____

Telephone number: _____

Email address: _____

Optional *if you have consented to point 7 above, please complete GP details below*

Name of your child/the person you care for's GP: _____

GP Address: _____

GP Telephone Number: _____

If you have medical letters or documents that confirm your child / the person you care for's diagnosis that you are happy to share with us, then you may include a photocopy when you return this pack. Otherwise, email k.a.wade@bham.ac.uk to discuss secure ways of sharing documentation with us digitally.
DO NOT EMAIL MEDICAL DOCUMENTATION AS AN ATTACHMENT.

SECTION 2 For the person being reported on

If your child/the person you care for is able to understand the information provided in the information sheet, either independently or with your support, then please get them to fill out this section.

Please let us know if you agree with each of the statements below by circling 'yes' if you agree, or 'no' if you disagree:

1. Has somebody else explained the project to you or have you read the information?	YES	NO
2. Do you understand what the project is about?	YES	NO
3. Have you asked all of the questions you want?	YES	NO
4. Have you had your questions answered in a way you understand?	YES	NO
5. Do you understand it is OK to stop taking part at any time?	YES	NO
6. Are you happy to take part?	YES	NO

If you circled '**no**' for **any** of the statements, or if you don't want to take part, then **stop here** and don't fill out any more of the form.

If you circled '**yes**' for **all** of the statements then please answer these extra statements and then write your name in the space below.
These statements are **optional**. You can choose if you want to say 'yes' to these. Even if you say 'no to these you can still take part and we will not do anything you say 'no' to:

7. If your doctor/other medical professional asks to see your results from this project, it is OK for us to share this with them?	YES	NO
8. Are you happy for us to look at your DNA test results, or to send you a swab so we can find out about your DNA?	YES	NO
9. Are you happy for us to contact you again in the future	YES	NO

Your name: _____ Date: _____

Please also tell us the name of the person who explained the study to you:

Print name: _____ Date: _____

Appendix 3.2: *BEOND study background questionnaire*



The Background Questionnaire

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SECTION 1

The following questions are about your child/the person you care for:

1. Which of the following best describes their gender?
   - ☐ Male
   - ☐ Female
   - ☐ Prefer to self-describe as: _____
     (e.g. non-binary, gender fluid, agender)
   - ☐ Prefer not to say

   Does their current gender identity match the gender they were assigned at birth?
   - ☐ Yes
   - ☐ No - assigned male at birth
   - ☐ No - assigned female at birth

2. Date of Birth: ____/____/_____ Age in years: _____
   Due date: ____/____/_____ ☐ *Tick if due date is not known*

3. Does your child/the person you care for use at least 30 different signs/words in their vocabulary?
   - ☐ Yes
   - ☐ No

4. Is your child/the person you care for able to walk unaided?
   - ☐ Yes
   - ☐ No

5. Which of the following best describes your child/the person you care for's ethnic group?
   - ☐ White
   - ☐ Mixed or multiple ethnic groups
   - ☐ Asian or Asian British
   - ☐ Black, African, Caribbean, or Black British
   - ☐ Not listed: _____

6. Has your child/the person you care for been diagnosed with a genetic syndrome?
   - ☐ Yes – **Please indicate which syndrome below and answer questions 7-9**
   - ☐ No – **Please move on to question 10**

☐ 19p13	☐ Pallister-Killian Syndrome
☐ 1p36	☐ Pitt-Hopkins Syndrome
☐ 8p23 deletion	☐ Phelan McDermid Syndrome
☐ Angelman Syndrome	☐ Potocki-Lupski Syndrome
☐ CHARGE Syndrome	☐ Prader Willi Syndrome
☐ Cri du Chat Syndrome	☐ Rubenstein Taybi Syndrome
☐ Cornelia de Lange Syndrome	☐ SATB2 Syndrome
☐ Down Syndrome	☐ Smith Magenis Syndrome
☐ Fragile X Syndrome	☐ Soto Syndrome
☐ Kleefstra Syndrome	☐ Tuberous Sclerosis Complex
☐ Lowe Syndrome	☐ Williams Syndrome

☐ Not listed: _____

7. What is the genetic mechanism causing your child/the person you care for's syndrome?

☐ Uni-Parental Disomy	☐ Translocation
☐ Deletion	☐ Unknown
☐ Sequence repetition	

☐ Not listed: _____

8. At what age was your child/the person you care for diagnosed?

_____

9. Who diagnosed your child/the person you care for?
   - ☐ Pediatrician
   - ☐ GP
   - ☐ Clinical Geneticist
   - ☐ Other: _____

10. Has your child/the person you care for been diagnosed with an intellectual disability, learning disability or global developmental delay?
    - ☐ Yes – **Please indicate the level of disability below**
    - ☐ No – **Please move on to question 11**

☐ Mild	☐ Profound
☐ Moderate	☐ Unknown
☐ Severe	☐ Other: _____

11. Has your child/the person you care for been diagnosed with autism?
    - ☐ Yes – **Please indicate their diagnosis below**
    - ☐ No – **Please move on to question 12**

☐ Autism	☐ Autism Spectrum Disorder
☐ Asperger Syndrome	☐ High Functioning Autism
☐ Autistic Features	☐ Autistic (like) Traits
☐ Autistic Continuum	☐ Pervasive Developmental Disorder
☐ Atypical Autism	☐ Autistic Spectrum

12. Has your child/the person you care for been diagnosed with ADHD?
    - ☐ Yes
    - ☐ No

13. Which of the following best describes your child/the person you care for's living arrangement?
    - ☐ Lives with caregivers at least 50% of the time
      Please complete section 2
    - ☐ Lives away from caregivers at least 50% of the time *(either independently or in a supported setting)*
      Please complete section 3

SECTION 2

The following questions are about you and your household:

1. How would you describe your gender?
   - ☐ Male
   - ☐ Female
   - ☐ Prefer to self-describe as: _____
     (e.g. non-binary, gender fluid, agender)
   - ☐ Prefer not to say

2. What was your age in years on your last birthday? _____ years

3. Which of the following best describes your ethnic group?
   - ☐ White
   - ☐ Mixed or multiple ethnic groups
   - ☐ Asian or Asian British
   - ☐ Black, African, Caribbean, or Black British
   - ☐ Not listed: _____

4. Please select the option which best describes your highest level of formal education.
   - ☐ No formal education
   - ☐ Secondary school, GCSEs or equivalent
   - ☐ College, sixth form, A levels or equivalent
   - ☐ University, undergraduate degree or equivalent
   - ☐ University, postgraduate degree or equivalent
   - ☐ Not listed: _____

5. Who else, aside from yourself and your child/the person you care for, lives with you?

Relationship to the person you care for	**Age**	**Gender**
_____	_____	_____
_____	_____	_____
_____	_____	_____
_____	_____	_____
_____	_____	_____

6. Does the person you care for stay overnight away from home? *(Tick all that apply)*

☐ No	
☐ Shared custody arrangement	How often? _____
☐ Overnight visits with another relative	How often? _____
☐ Respite Care	How often? _____
☐ Residential School	How often? _____
☐ Not listed:_____	How often? _____

Recent data from research with families of children with special needs has shown that a family's financial resources are important in understanding family member's views and experiences. With this in mind, we would be very grateful if you could answer the additional question below. We are not interested in exactly what your family income is, but we would like to be able to look at whether those with high versus lower levels of financial resources have different experiences.

7. How does your total household income compare to the national average? (£29,000 in the UK) Please include a rough estimate of total salaries and other income (including benefits) before tax and national insurance/pensions.
(If you are responding from outside the UK, please respond according to your national median income.)

☐ Below the national average
☐ Roughly the same as the national average
☐ Above the national average
☐ Would prefer not to answer

Please check your answers and move on to the next questionnaire

SECTION 3

The following questions are about the placement that your child/the person you care for resides in:

1. What kind of placement does your child/the person you care for reside in?

   _____

   (e.g. residential school, secure facility, supported living)

2. Which of the following best categorises the service providing the placement?
   - ☐ Learning disability service
   - ☐ Autism service
   - ☐ Mental health service
   - ☐ Unsure/don't know
   - ☐ N/A
   - ☐ Other:_____

3. Excluding staff members, approximately how many other people does your child/the person you care for share their lodgings/living space with?

   _____

4. On an average day shift, how many support staff are on shift?

   _____

5. Does your child/the person you care for have an allocated key worker?
   - ☐ Yes
   - ☐ No

6. How long has your child/the person you care for lived here?
   - ☐ Less than a year
   - ☐ 1-3 years
   - ☐ 3-5 years
   - ☐ More than 5 years

7. Does your child/the person you care for have regular visits with their family?
   - ☐ Overnight stays at family home
   - ☐ Day trips with family *(Either to family home or elsewhere)*
   - ☐ Family members visit at placement
   - ☐ No/limited contact with family

Please check your answers and move on to the next questionnaire

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The MASS-P

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1. In the past 2 years, has your child/the person you care for received support from services/professionals for any of their physical, emotional and/or behavioural needs?
   - ☐ Yes
   - ☐ No **(Please skip to question 7)**

2. Which of the following professionals did your child/the person you care for see in order to address their specific physical, emotional and/or behavioural needs?
   (*Please tick where applicable*)

Professional/Service	***Challenging behaviour***	***Sleep***	***Mental Health***	***Medical concerns****
General Practitioner (GP)				
Psychiatrist				
Clinical Psychologist				
Educational Psychologist				
Occupational Therapist				
Speech and Language Therapist				
Syndrome-specific Support Group				
Social Worker				
Nurse				
General Paediatrician				
Teacher /Teaching Assistant				
Dietician/ Nutritionist				
Specialist medical professional *e.g. neurologist, gastroenterologist (Please specify)* _____				
Other *(Please specify)*: _____				

*Please briefly outline your child / the person you care for's medical concerns:

3. Following access to the following professionals, have you noticed a change in your child/the person you care for's **challenging behaviour**?
Please tick where applicable.

CHALLENGING BEHAVIOUR ***Professional/Service***	***Major worsening***	***Slight worsening***	***No change***	***Slight improvement***	***Major improvement***
General Practitioner (GP)					
Psychiatrist					
Clinical Psychologist					
Educational Psychologist					
Occupational Therapist					
Speech and Language Therapist					
Syndrome-specific Support Group					
Social Worker					
Nurse					
General Paediatrician					
Teacher /Teaching Assistant					
Dietician/ Nutritionist					
Specialist medical professional *e.g. neurologist, gastroenterologist (Please specify)* _____					
Other *(Please specify)*: _____					

4. Following access to the following professionals, have you noticed a change in your child/the person you care for's **sleep**?
Please tick where applicable.

SLEEP ***Professional/Service***	***Major worsening***	***Slight worsening***	***No change***	***Slight improvement***	***Major improvement***
General Practitioner (GP)					
Psychiatrist					
Clinical Psychologist					
Educational Psychologist					
Occupational Therapist					
Speech and Language Therapist					
Syndrome-specific Support Group					
Social Worker					
Nurse					
General Paediatrician					
Teacher /Teaching Assistant					
Dietician/ Nutritionist					
Specialist medical professional *e.g. neurologist, gastroenterologist (Please specify)* _____					
Other *(Please specify)*: _____					

5. Following access to the following professionals, have you noticed a change in your child/the person you care for's **mental health?**
Please tick where applicable.

MENTAL HEALTH ***Professional/Service***	***Major worsening***	***Slight worsening***	***No change***	***Slight improvement***	***Major improvement***
General Practitioner (GP)					
Psychiatrist					
Clinical Psychologist					
Educational Psychologist					
Occupational Therapist					
Speech and Language Therapist					
Syndrome-specific Support Group					
Social Worker					
Nurse					
General Paediatrician					
Teacher /Teaching Assistant					
Dietician/ Nutritionist					
Specialist medical professional *e.g. neurologist, gastroenterologist (Please specify)* _____					
Other *(Please specify)*: _____					

6. Following access to the following professionals, have you noticed a change in your child/the person you care for's **medical needs**?
Please tick where applicable.

MEDICAL CONCERNS ***Professional/Service***	***Major worsening***	***Slight worsening***	***No change***	***Slight improvement***	***Major improvement***
General Practitioner (GP)					
Psychiatrist					
Clinical Psychologist					
Educational Psychologist					
Occupational Therapist					
Speech and Language Therapist					
Syndrome-specific Support Group					
Social Worker					
Nurse					
General Paediatrician					
Teacher /Teaching Assistant					
Dietician/ Nutritionist					
Specialist medical professional *e.g. neurologist, gastroenterologist (Please specify)* _____					
Other *(Please specify)*: _____					

7. Does your child/ the person you care for currently take prescribed medication for their specific physical/medical, emotional and/or behavioural needs?
   - ☐ Yes
   - ☐ No (**Please skip to question 9)**

8. Please list current medication, dose and primary reason for prescription.

Name of medication	***Dose***	***Primary reason for prescription***

9. Do you consider that your child/the person you care for's physical, emotional and behavioural needs have been met by your local services?
   - ☐ Yes (**Please skip to end of form)**
   - ☐ No
   - ☐ Partially met
   - ☐ Not sure
   - ☐ My child/the person I care for does not have any specific physical, emotional, or behavioural needs

10. What are the reasons for the unmet needs?
    (Tick all that apply)
    - ☐ A lack of service provision in your local area
    - ☐ A lack of appropriate services for your child/the person you care for's needs
    - ☐ Long waiting lists
    - ☐ Covid-19-related restrictions to service access
    - ☐ Financial/personal reasons
    - ☐ Accessibility reasons
    - ☐ Other
      (*please specify*): _____

Please check your answers and move on to the next questionnaire

Appendix 3.3: *Description of service access for challenging behaviour (N=260)*

Service	***n* yes / no**	**% yes**
Teacher/ teaching assistant	48 / 212	18
Speech and language therapist	48 / 212	18
Occupational therapist	45 / 217	17
Clinical psychologist	39 / 223	15
Psychiatrist	37 / 225	14
General paediatrician	31 / 231	12
Social worker	28 / 234	11
Specialist	24 / 238	9
Educational psychologist	23 / 239	9
GP	22 / 240	8
Nurse	16 / 246	6
Syndrome-specific support group	16 / 246	6
Other	10 / 252	4
Dietician/nutritionist	8 / 254	3

Appendix 3.4: *The Social Communication Questionnaire*



The SCQ-C

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Directions: Please answer each question by circling *yes* or *no*. A few questions ask about several related types of behavior; please circle yes if any of these behaviors were present during the **past 3 months**. Although you may be uncertain about whether some behaviors were present or not, please answer yes or no to every question on the basis of what you think.

1.	Is she/he now able to talk using short phrases or sentences? **If *no*, skip to question 8**	**Yes**	**No**
2.	Do you have a to and fro "conversation" with her/him that involves taking turns or building on what you have said?	**Yes**	**No**
3.	Does she/he ever use odd phrases or say the same thing over and over in almost exactly the same way (either phrases she/he has heard other people use or ones that she/he makes up)?	**Yes**	**No**
4.	Does she/he ever use socially inappropriate questions or statements? For example, does she/he ever regularly ask personal questions or make personal comments at awkward times?	**Yes**	**No**
5.	Does she/he ever get her/his pronouns mixed up (e.g. saying you or she/he instead of I)?	**Yes**	**No**
6.	Does she/he ever use words that she/he seems to have invented or made up her/himself; put things in odd, indirect ways; or use metaphorical ways of saying things (e.g. saying hot rain for steam)?	**Yes**	**No**
7.	Does she/he ever say the same thing over and over in exactly the same way, or insist that you say the same thing over and over again?	**Yes**	**No**
8.	Does she/he ever have things that she/he seems to have to do in a very particular way or order, or rituals that she/he insists that you go through?	**Yes**	**No**
9.	Does her/his facial expression usually seem appropriate to the particular situation, so far as you can tell?	**Yes**	**No**
10.	Does she/he ever use your hand like a tool, or as if it were part of her/his own body (e.g. pointing with your finger, putting your hand on a doorknob to get you to open the door)?	**Yes**	**No**
11.	Does she/he ever have any interests that preoccupy her/him and might seem odd to other people (e.g. traffic lights, drainpipes or timetables)?	**Yes**	**No**
12.	Does she/he ever seem to be more interested in parts of a toy or an object (e.g. spinning the wheels of a car), rather than using the object as it was intended?	**Yes**	**No**
13.	Does she/he ever have any special interests that are unusual in their intensity but otherwise appropriate for her/his age and peer group (e.g. trains, or dinosaurs)?	**Yes**	**No**
14.	Does she/he ever seem to be unusually interested in the sight, feel, sound, taste or smell of things or people?	**Yes**	**No**
15.	Does she/he ever have any mannerisms or odd ways of moving her/his hands or fingers, such as flapping or moving her/his fingers in front of her/his eyes?	**Yes**	**No**
16.	Does she/he ever have any complicated movements of her/his whole body, such as spinning or repeatedly bouncing up and down?	**Yes**	**No**
17.	Does she/he ever injure her/himself deliberately, such as by biting her/his arm or banging her/his head?	**Yes**	**No**

18.	Does she/he ever have any objects (other than a soft toy or comfort blanket) that she/he has to carry around?	**Yes**	**No**
19.	Does she/he have any particular friends or a best friend?	**Yes**	**No**
20.	Does she/he ever talk with you just to be friendly (rather than to get something)?	**Yes**	**No**
21.	Does she/he ever spontaneously copy you (or other people) or what you are doing (such as vacuuming, gardening or mending things)?	**Yes**	**No**
22.	Does she/he ever spontaneously point at things around her/him just to show you things (not because she/he wants them)?	**Yes**	**No**
23.	Does she/he ever use gestures, other than pointing or pulling your hand, to let you know what she/he wants?	**Yes**	**No**
24.	Does she/he nod her/his head to indicate yes?	**Yes**	**No**
25.	Does she/he shake her/his head to indicate no?	**Yes**	**No**
26.	Does she/he usually look at you directly in the face when doing things with you or talking with you?	**Yes**	**No**
27.	Does she/he smile back if someone smiles at her/him?	**Yes**	**No**
28.	Does she/he ever show you things that interest her/him to engage your attention?	**Yes**	**No**
29.	Does she/he ever offer to share things other than food with you?	**Yes**	**No**
30.	Does she/he ever seem to want you to join in her/his enjoyment of something?	**Yes**	**No**
31.	Does she/he ever try to comfort you if you are sad or hurt?	**Yes**	**No**
32.	If she/he wants something or wants help, does she/he look at you and use gestures with sounds or words to get your attention?	**Yes**	**No**
33.	Does she/he show a normal range of facial expressions?	**Yes**	**No**
34.	Does she/he ever spontaneously join in and try to copy the actions in social games, such as The Mulberry Bush or London Bridge is Falling Down?	**Yes**	**No**
35.	Does she/he play any pretend or make-believe games?	**Yes**	**No**
36.	Does she/he seem interested in other children of approximately the same age whom she/he does not know?	**Yes**	**No**
37.	Does she/he respond positively when another child approaches her/him?	**Yes**	**No**
38.	If you come into a room and start talking to her/him without calling her/his name, does she/he usually look up and pay attention to you?	**Yes**	**No**
39.	Does she/he ever play imaginative games with another child in such a way that you can tell that each child understands what the other is pretending?	**Yes**	**No**
40.	Does she/he play cooperatively in games that need some form of joining in with a group of other children, such as hide-and-seek or ball games?	**Yes**	**No**

Please check your answers and move on to the next questionnaire

Appendix 3.5: *The Wessex Questionnaire*

UNIVERSITY OF BIRMINGHAM · WARWICK · The Cerebra Network for Neurodevelopmental Disorders · Aston University BIRMINGHAM UK · UNIVERSITY OF SURREY

The Wessex Questionnaire

© Albert Kushlick, Psychological Medicine

Please provide the following information for your child/the person you care for. It is important that you respond to every item. Please tick the most appropriate response.

A) Wetting (nights)	☐ 1. Frequently	☐ 2. Occasionally	☐ 3. Never
B) Soiling (nights)	☐ 1. Frequently	☐ 2. Occasionally	☐ 3. Never
C) Wetting (days)	☐ 1. Frequently	☐ 2. Occasionally	☐ 3. Never
D) Soiling (days)	☐ 1. Frequently	☐ 2. Occasionally	☐ 3. Never
E) Walk with help*	☐ 1. Not at all	☐ 2. Not upstairs	☐ 3. Upstairs & elsewhere
	***note**: if this person walks by himself/herself upstairs and elsewhere, please also tick '3. Upstairs and elsewhere' for 'Walk with help'*		
F) Walk by himself	☐ 1. Not at all	☐ 2. Not upstairs	☐ 3. Upstairs & elsewhere
G) Feed himself	☐ 1. Not at all	☐ 2. With help	☐ 3. Without help
H) Wash himself	☐ 1. Not at all	☐ 2. With help	☐ 3. Without help
I) Dress himself	☐ 1. Not at all	☐ 2. With help	☐ 3. Without help
J) Vision	☐ 1. Blind or almost	☐ 2. Poor	☐ 3. Normal
K) Hearing	☐ 1. Deaf or almost	☐ 2. Poor	☐ 3. Normal

L) Speech	☐ 1. Never a word	☐ 2. Odd words only	☐ 3. Sentences & normal	☐ 4. Can talk but doesn't

If this person talks in sentences, is his/her speech:

☐ 1. Difficult to understand even by acquaintances, impossible for strangers?
☐ 2. Easily understood for acquaintances, difficult for strangers?
☐ 3. Clear enough to be understood by anyone?

M) Reads	☐ 1. Nothing	☐ 2. A Little	☐ 3. Newspapers and/or books
N) Writes	☐ 1. Nothing	☐ 2. A Little	☐ 3. Own correspondence
O) Counts	☐ 1. Nothing	☐ 2. A Little	☐ 3. Understands money values

Please check your answers and move on to the next questionnaire

Appendix 3.6: *The Challenging Behaviour Questionnaire*



The CBQ

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Section A – Self-injurious Behaviour

Has the person ever shown self-injurious behaviour (e.g. head banging, head-punching or slapping, removing hair, self-scratching, body hitting, eye poking or pressing)?

☐ Yes ☐ No

Has the person shown self-injurious behaviour in the last month (e.g. head banging, head-punching or slapping, removing hair, self-scratching, body hitting, eye poking or pressing)?

☐ Yes ☐ No

If the behaviour has not occurred, please go to section B.
If the behaviour occurred in the past month, please answer questions 1a to 1d:

1a) Place a tick next to the item for any of the following list of behaviours which the person displays in a repetitive manner (repeats the same movement/ behaviour twice or more in succession):

Hits self with body part (e.g. slaps head or face)	☐
Hits self against surface or object (e.g. bangs head on floor or table)	☐
Hits self with object	☐
Bites self (e.g. bites hand on wrist or arm)	☐
Pulls (e.g. pulls hair or skin)	☐
Rubs or scratches self (e.g. rub marks on arm or leg)	☐
Inserts finger or objects (e.g. eye poking)	☐
Other form of self-injury, (please specify): _____	☐

1b) In the last month, for how long did the **longest** episode or burst of this behaviour last? *(Please circle one number)*

1	**2**	**3**	**4**	**5**
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

1c) In the last month as a result of this behaviour, has physical contact or prevention or restraint by others been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm? *(Please circle one number)*

0	**1**	**2**	**3**	**4**
Never	At least once a month	At least once a week	At least once a day	At least once an hour

1d) Think about how often this behaviour occurred in the last month. If there was no change and you watched the person now, then would you definitely see the behaviour:

1	**2**	**3**	**4**	**5**
By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

Section B – Aggression

Has the person ever shown aggression (e.g. punching, pushing, kicking, pulling hair, grabbing other's clothing)?

☐ Yes ☐ No

Has the person shown aggression in the last month (e.g. punching, pushing, kicking, pulling hair, grabbing other's clothing)?

☐ Yes ☐ No

If the behaviour has not occurred, please go to section C.
If the behaviour occurred in the past month, please answer questions 2a to 2d.

2a) Place a tick next to the item for any of the following list of behaviours which the person displays:

Hits other with body part (e.g. slapping, punching, kicking, head-butting)	☐
Hits other with an object (e.g. throwing object or using a weapon)	☐
Bites other	☐
Pulls or grabs other (e.g. hair-pulling, grabbing clothing)	☐
Rubs, pinches or scratches other	☐
Spits at other	☐
Verbal aggression (e.g. threatening, swearing)	☐
Other form of aggression, (please specify): _____	☐

2b) In the last month, for how long did the **longest** episode or burst of this behaviour last? *(Please circle one number)*

1	2	3	4	5
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

2c) In the last month as a result of this behaviour, has physical contact or prevention or restraint by others been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm? *(Please circle one number)*

0	1	2	3	4
Never	At least once a month	At least once a week	At least once a day	At least once an hour

2d) Think about how often this behaviour occurred in the last month. If there was no change and you watched the person now, then would you definitely see the behaviour:

1	2	3	4	5
By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

Section C – Disruption and Destruction of Property

Has the person ever shown disruption and destruction of property or the environment (e.g. tearing or chewing own clothing, tearing newspapers, breaking windows or furniture, slamming doors, spoiling a meal)?

☐ Yes ☐ No

Has the person shown disruption and destruction of property or the environment in the last month? (e.g. tearing or chewing own clothing, tearing newspapers, breaking windows or furniture, slamming doors, spoiling a meal)?

☐ Yes ☐ No

If the behaviour has not occurred, please go to section D.
If the behaviour occurred in the past month, please answer questions 3a to 3d.

3a) Place a tick next to the item for any of the following list of behaviours which the person displays:

Biting or chewing small objects (e.g. books, clothing)	☐
Tearing or ripping small items (e.g. books, clothing)	☐
Throwing, kicking or smashing small items (e.g. cups, mobile phones)	☐
Slamming, hitting, or kicking doors, walls or windows	☐
Tipping, smashing or throwing large items (e.g. furniture, televisions)	☐
Pulling items from walls or shelves	☐
Verbal aggression (e.g. threatening, swearing)	☐
Other form of disruption (please specify): _____	☐

3b) In the last month, for how long did the **longest** episode or burst of his behaviour last? *(Please circle one number)*

1	**2**	**3**	**4**	**5**
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

3c) In the last month as a result of this behaviour, has physical contact or prevention or restraint by others been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm? *(Please circle one number)*

0	**1**	**2**	**3**	**4**
Never	At least once a month	At least once a week	At least once a day	At least once an hour

3d) Think about how often this behaviour occurred in the last month. If there was no change and you watched the person now, then would you definitely see the behaviour:

1	**2**	**3**	**4**	**5**
By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

Section D – Stereotyped Behaviours

Has the person ever shown stereotyped behaviours? (e.g. rocking, twiddling objects, patting or tapping part of the body, constant hand movements, eye pressing)?

☐ Yes ☐ No

Has the person shown stereotyped behaviours in the last month? (e.g. rocking, twiddling objects, patting or tapping part of the body, constant hand movements, eye pressing)?

☐ Yes ☐ No

If the behaviour has not occurred, please go to section E.
If the behaviour occurred in the past month, please answer questions 4a to 4d:

4a) Place a tick next to the item for any of the following list of behaviours which the person displays:

Full body movements (e.g. rocking, bouncing)	☐
Movement of an object (e.g. twiddling or spinning object)	☐
Movement of isolated body part (e.g. hand flapping, head shaking)	☐
Eye-pressing or visual regard of movement (e.g. strobing, spinning object)	☐
Mouthing or sucking on body part or object	☐
Other form of disruption, (please specify): _____	☐

4b) In the last month, for how long did the **longest** episode or burst of his behaviour last? *(Please circle one number)*

1	**2**	**3**	**4**	**5**
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

4c) In the last month as a result of this behaviour, has physical contact or prevention or restraint by others been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm? *(Please circle one number)*

0	**1**	**2**	**3**	**4**
Never	At least once a month	At least once a week	At least once a day	At least once an hour

4d) Think about how often this behaviour occurred in the last month. If there was no change and you watched the person now, then would you definitely see the behaviour:

1	**2**	**3**	**4**	**5**
By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

Section E – Other forms of challenging behaviour

Has the person ever shown any other form of challenging behaviour?

☐ Yes ☐ No

Has the person shown any other form of challenging behaviour in the last month?

☐ Yes ☐ No

If the behaviour has not occurred, please skip to the end of this form
If the behaviour occurred in the past month, please answer questions 5a to 5d:

5a) Place a tick next to the item for any of the following list of behaviours which the person displays:

Pica (e.g. eating paper, leaves, discarded food)	☐
Inappropriate vocalisations (e.g. screaming, shouting)	☐
Removal of clothing (not for purpose of washing changing or toileting)	☐
Sexual behaviour (e.g. public masturbation, inappropriate sexual contact)	☐
Anal poking	☐
Smearing of bodily substance (non-accidental)	☐
Stealing	☐
Self-induced vomiting	☐
Other form of challenging behaviour (please specify): _____	☐

5b) In the last month, for how long did the **longest** episode or burst of his behaviour last? *(Please circle one number)*

1	**2**	**3**	**4**	**5**
Less than a minute	Less than 5 minutes	Less than 15 minutes	Less than an hour	More than an hour

5c) In the last month as a result of this behaviour, has physical contact or prevention or restraint by others been necessary e.g. blocking, taking objects from an individual, temporary restraint of an arm? *(Please circle one number)*

0	**1**	**2**	**3**	**4**
Never	At least once a month	At least once a week	At least once a day	At least once an hour

5d) Think about how often this behaviour occurred in the last month. If there was no change and you watched the person now, then would you definitely see the behaviour:

1	**2**	**3**	**4**	**5**
By this time next month	By this time next week	By this time tomorrow	In the next hour	In the next 15 minutes

Please check your answers and move on to the next questionnaire

Appendix 3.7: *The Clinical Anxiety Screen for People with Intellectual Disability*



CIASP-ID

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This questionnaire is going to ask you about behaviours you may or may not have seen in the person you care for, over the last MONTH.

For each item, you will be asked to rate how frequently the behaviours have occurred over the last month. For some of the questions, we will also ask you to think about whether the behaviour has been occurring more or less than is usual for the person you care for, over the last month.

Please try to answer every question.
If you are unsure whether you have seen the behaviour, please select 'almost never' and move onto the next question

Has he/she...

1. Seemed withdrawn or 'vacant'?

☐	☐	☐	☐	☐	☐	☐
Almost never	Less than a quarter of the time	Less than half of the time	About half of the time	More than half of the time	About three quarters of the time	All of the time

Over the past one month, has the behaviour been…

- ☐ Occurring a lot more than is typical of him/her
- ☐ Occurring a bit more than is typical of him/her
- ☐ Occurring at the same rate that is typical of him/her
- ☐ Occurring a bit less than is typical of him/her
- ☐ Occurring a lot less than is typical of him/her

	Almost never	Once a month	Less than once a week	Once or twice a week	At least 3-4 times per week	Every day	More than once a day
Does he or she…							
2. Ever make negative or frustrated vocalisations? *(e.g. whining, grumbling growling, shouting, screaming)*	☐	☐	☐	☐	☐	☐	☐
3. Appear on edge OR on the lookout for danger?	☐	☐	☐	☐	☐	☐	☐
4. Ever seem protective of a particular part of his/her body? *(e.g. holding it, guarding it, flinching)*	☐	☐	☐	☐	☐	☐	☐
5. Pace around the room	☐	☐	☐	☐	☐	☐	☐
6. Ever appear restless or agitated	☐	☐	☐	☐	☐	☐	☐
7. Ever run away or hide from certain objects or situations	☐	☐	☐	☐	☐	☐	☐
8. Ever cover himself/herself with a blanket or try to place a barrier between himself/herself and others or a situation	☐	☐	☐	☐	☐	☐	☐
9. Ever have watery eyes that is different from crying	☐	☐	☐	☐	☐	☐	☐
10. Ever freeze suddenly (stick to the spot) in response to specific situations	☐	☐	☐	☐	☐	☐	☐
11. Ever grind his/her teeth	☐	☐	☐	☐	☐	☐	☐
12. Avoid (or try to avoid) certain objects or places	☐	☐	☐	☐	☐	☐	☐
13. Ever take sharp intakes of breath or gasp	☐	☐	☐	☐	☐	☐	☐
14. Ever look very worried or anxious	☐	☐	☐	☐	☐	☐	☐
15. Have an angry look on his/her face	☐	☐	☐	☐	☐	☐	☐
16. Startle easily, or is easily alarmed	☐	☐	☐	☐	☐	☐	☐
Over the past month, have you noticed…							
17. Increased or different leg movements *(e.g. restlessness, tense, tremors, kicking, drawing legs up, jerking)*	☐	☐	☐	☐	☐	☐	☐
18. That he/she shakes or trembles	☐	☐	☐	☐	☐	☐	☐
19. His/her face looks tense	☐	☐	☐	☐	☐	☐	☐
Over the past month, has he/she…							
20. Been hitting, holding or touching a part of their body	☐	☐	☐	☐	☐	☐	☐
Over the past month, have his/her…							
21. Movements ever become jerky?	☐	☐	☐	☐	☐	☐	☐
22. Lips ever become tight, pout or quiver?	☐	☐	☐	☐	☐	☐	☐

Has he/she…

23. Lost interest in activities that he/she used to enjoy?

Almost never	Less than a quarter of the time	Less than half of the time	About half of the time	More than half of the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

Over the past one month, has the behaviour been…

- ☐ Occurring a lot more than is typical of him/her
- ☐ Occurring a bit more than is typical of him/her
- ☐ Occurring at the same rate that is typical of him/her
- ☐ Occurring a bit less than is typical of him/her
- ☐ Occurring a lot less than is typical of him/her

Does he/she…

24. Lack energy?

Almost never	Less than a quarter of the time	Less than half of the time	About half of the time	More than half of the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

25. Get tired for no apparent reason?

Almost never	Less than a quarter of the time	Less than half of the time	About half of the time	More than half of the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

Is he/she…

26. Spending more time asleep than usual? *(e.g. not waking in the morning, sleep during the day)*

Almost never	Less than a quarter of the time	Less than half of the time	About half of the time	More than half of the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

Over the past one month, has the behaviour been…

- ☐ Occurring a lot more than is typical of him/her
- ☐ Occurring a bit more than is typical of him/her
- ☐ Occurring at the same rate that is typical of him/her
- ☐ Occurring a bit less than is typical of him/her
- ☐ Occurring a lot less than is typical of him/her

Is he/she…

27. Quiet and spending time alone?

Almost never	Less than a quarter of the time	Less than half of the time	About half of the time	More than half of the time	About three quarters of the time	All of the time
☐	☐	☐	☐	☐	☐	☐

Over the past one month, has the behaviour been…

- ☐ Occurring a lot more than is typical of him/her
- ☐ Occurring a bit more than is typical of him/her
- ☐ Occurring at the same rate that is typical of him/her
- ☐ Occurring a bit less than is typical of him/her
- ☐ Occurring a lot less than is typical of him/her

How often are these statements true for your child/the person you support?	Almost never	Less than a quarter of the time	Less than half of the time	About half of the time	More than half of the time	About three quarters of the time	All of the time
28. Preparing him/her before things happen helps to reduce his/her distress.	☐	☐	☐	☐	☐	☐	☐
29. Removing the person I care for from a situation or removing an item/object generally calms them.	☐	☐	☐	☐	☐	☐	☐
30. When the person I care for is distressed, I am able to calm or comfort him/her.	☐	☐	☐	☐	☐	☐	☐
31. When in certain preferred environments *(e.g. home, their bedroom)* the person I care for generally appears calm and relaxed.	☐	☐	☐	☐	☐	☐	☐
32. We are unable to do 'typical' day to day activities because of the emotional distress that would cause him/her *(e.g. holidays, visiting friends, going for meals, general days out.)*	☐	☐	☐	☐	☐	☐	☐
33. We are unable to do activities we used to do with the person I care for because of the emotional distress he/she would experience.	☐	☐	☐	☐	☐	☐	☐

34. Have you noticed any other changes in mood or behaviour not covered in this questionnaire? *(if yes, please give details…)*

Please check your answers and move on to the next questionnaire

Appendix 3.8: *The Repetitive Behaviour Questionnaire*



The RBQ

© Jo Moss & Chris Oliver, Journal of Autism and Developmental Disorders

Instructions:

1. The questionnaire asks about 19 different behaviours.
2. Each behaviour is accompanied by a brief definition and examples. The examples given for each behaviour are not necessarily a complete list but may help you to understand the definitions more fully.
3. Please read the definitions and examples carefully and circle the appropriate number on the scale to indicate how frequently the person you care for has engaged in each of the behaviours **within the last month**
4. If a particular behaviour does not apply to the person you care for because they are not mobile or verbal, please circle the number 0 on the scale.

		Never	Once a month	Once a Week	Once a day	More than once a day
1.	**Object stereotypy:** Repetitive, seemingly purposeless movement of objects in an unusual way *E.g., twirling or twiddling objects, twisting or shaking objects, banging or slapping objects.*	0	1	2	3	4
2.	**Body stereotypy:** Repetitive, seemingly purposeless movement of whole body or part of body (other than hands) in an unusual way. *E.g., body rocking, or swaying or spinning, bouncing, head shaking, body posturing.* Does not include self-injurious behaviour)	0	1	2	3	4
3.	**Hand stereotypy:** Repetitive, seemingly purposeless movement of hands in an unusual way. *E.g., finger twiddling, hand flapping, wiggling or flicking fingers, hand posturing.* Does not include self-injurious behaviour.	0	1	2	3	4
4.	**Cleaning:** Excessive cleaning, washing or polishing of objects or parts of the body *E.g., polishes windows and surfaces excessively, washes hands and face excessively.*	0	1	2	3	4
5.	**Tidying up:** Tidying away any objects that have been left out. This may occur in situations when it is inappropriate to put the objects away. Objects may be put away into inappropriate places. *E.g., putting cutlery left out for dinner in the bin, removes all objects from surfaces.*	0	1	2	3	4
6.	**Hoarding:** Collecting, storing or hiding objects to excess, including rubbish, bits of paper, and pieces of string or any other unusual items.	0	1	2	3	4
7.	**Organising objects:** Organising objects into categories according to various characteristics such as colour, size, or function. *E.g., ordering magazines according to size, ordering toy cars according to colour, ordering books according to topic.*	0	1	2	3	4
8.	**Attachment to particular people:** Continually asking to see, speak or contact a particular 'favourite' person. *E.g., continually asks to see or speak to particular friend, carer, babysitter or schoolteacher.*	0	1	2	3	4

		Never	Once a month	Once a Week	Once a day	More than once a day
9.	**Repetitive questions**: Asking specific questions over and over. *E.g., always asking people what their favourite colour is, asking who is taking them to school the next day over and over.*	0	1	2	3	4
10.	**Attachment to objects:** Strong preference for a particular object to be present at all times. *E.g., carrying a particular piece of string everywhere, taking a particular red toy car everywhere, attachment to soft toy or particular blanket.*	0	1	2	3	4
11.	**Repetitive phrases/signing:** Repeating particular sounds, phrases or signs that are unrelated to the situation over and over. *E.g., repeatedly signing the word 'telephone'.*	0	1	2	3	4
12.	**Rituals:** Carrying out a sequence of unusual or bizarre actions before, during or after a task. The sequence will always be carried out when performing this task and will always occur in the same way. *E.g., turning round three times before sitting down, turning lights on and off twice before leaving a room, tapping door frame twice when passing through it.*	0	1	2	3	4
13.	**Restricted conversation:** Repeatedly talks about specific, unusual topics in great detail. *E.g., conversation restricted to trains, buses, dinosaurs, particular film, country or sport.*	0	1	2	3	4
14.	**Echolalia:** Repetition of speech that has either just been heard or has been heard more than a minute earlier. *E.g., Mum: 'Jack don't do that' Jack: 'Jack don't do that'.*	0	1	2	3	4
15.	**Preference for routine:** Insists on having the same household, school or work schedule every day. *E.g., likes to have the same activities on the same day at the same time each week, prefers to eat lunch at exactly the same time every day, wearing the same jumper every day.*	0	1	2	3	4
16.	**Lining up or arranging objects***:* Arrangement of objects into lines or patterns. *E.g., placing toy cars in a symmetrical pattern, precisely lining up story books.*	0	1	2	3	4
17.	**Just right behaviour:** Strong insistence that objects, furniture and toys always remain in the same place. *E.g., all chairs, pictures and toys have a very specific place that cannot be changed.*	0	1	2	3	4
18.	**Completing behaviour:** Insists on having objects or activities 'complete' or 'whole'. *E.g., must have doors open or closed not in between, story must be read from beginning to end, not left halfway through.*	0	1	2	3	4
19.	**Spotless behaviour:** Removing small, almost unnoticeable pieces of lint, fluff, crumbs or dirt from surfaces, clothes and objects. *E.g., picking fluff off a jumper, removing crumbs from the kitchen table.*	0	1	2	3	4

Please check your answers and move on to the next questionnaire

Appendix 3.9: *The Activity Questionnaire*



The TAQ

© C Burbidge and C Oliver (2003)

- Please read each item carefully and circle the appropriate number on the scale, for the person you care for.
- Please ensure that you indicate a response for every item. If the particular behaviour does not apply, for example, if the person is not verbal or not mobile, please circle 0 on the scale.

		Never/ almost never	Some of the time	Half of the time	A lot of the time	Always/ almost all the time
1.	Does the person wriggle or squirm about when seated or lying down?	0	1	2	3	4
2.	Does the person fidget or play with their hands and/or feet when seated or lying down?	0	1	2	3	4
3.	Does the person find it difficult holding still?	0	1	2	3	4
4.	Does the person find it difficult to remain in their seat even when in situations where it would be expected?	0	1	2	3	4
5.	Does the person prefer to be moving around or become frustrated if left in one position for too long?	0	1	2	3	4
6.	When the person is involved in a leisure activity (e.g. watching TV, playing a game etc.) do they make a lot of noise?	0	1	2	3	4
7.	When the person is involved in an activity, are they boisterous and/or rough?	0	1	2	3	4
8.	Does the person act as if they are "driven by a motor" (i.e. often very active)?	0	1	2	3	4
9.	Does the person seem like they need very little rest to recharge their battery?	0	1	2	3	4
10.	Does the person often talk excessively?	0	1	2	3	4
11.	Does the person's behaviour seem difficult to manage/contain whilst out and about (e.g. in town, in supermarkets etc.)?	0	1	2	3	4
12.	Do you feel that you need to "keep an eye" on the person at all times?	0	1	2	3	4
13.	Does the person you care for seem to act/do things without stopping to think first?	0	1	2	3	4
14.	Does the person blurt out answers before questions have been completed?	0	1	2	3	4
15.	Does the person start to respond to instructions before they have been fully given or without seeming to understand them?	0	1	2	3	4
16.	Does the person want things immediately?	0	1	2	3	4
17.	Does the person find it difficult to wait?	0	1	2	3	4
18.	Does the person disturb others because they have difficulty waiting for things or waiting their turn?	0	1	2	3	4

Please check your answers and move on to the next questionnaire

Appendix 3.10: *The Self-Restraint Checklist*



The SRC

Taken from: (Powell et al., 1996)

The following items are about self-restraint. This refers to self-initiated movement/behaviour performed to maintain a set position which appears to stop or prevent movement of body parts/areas.

Please respond accordingly to the following list of behaviours which the individual has displayed **IN THE LAST MONTH:**

	Yes	No
1. Wraps self in own clothing	☐	☐
2. Holds onto others or holds onto others clothing	☐	☐
3. Positions self to restrain (e.g., sitting on hands, wraps arms in chair)	☐	☐
4. Holds hands together, holds onto self	☐	☐
5. Holds or squeezes objects	☐	☐
6. Chooses to wear a particular item of clothing most of the time	☐	☐
7. Chooses mechanical restraint	☐	☐
8. Other form of self-restraint (*please specify*):	☐	☐
_____		
_____		
_____		
_____		

Please check your answers and move on to the next questionnaire

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Dr Caroline Richards
University of Birmingham
Edgbaston
Birmingham
B15 2TT

Email: approvals@hra.nhs.uk
HCRW.approvals@wales.nhs.uk

09 May 2022

Dear Dr Richards

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**Behavioural and Emotional Outcomes in individuals with Neurodevelopmental Disorders**
IRAS project ID:	**299757**
Protocol number:	**RG_21-140**
REC reference:	**22/WA/0086**
Sponsor	**University of Birmingham**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **299757**. Please quote this on all correspondence.

Yours sincerely,
Penny Beresford

Approvals Specialist

Email: HCRW.approvals@wales.nhs.uk

Copy to: *Dr Birgit Whitman*

Appendix 3.12: *Full hierarchical multiple regression model*

Model and predictors	**β**	**SE**	**t**	***p***	**R^2**	**R^2_{adj}**	**Collinearity statistics**	
							Tol.	**VIF**
Step 1					.099	.092		
Age	-.319	.007	-5.161	**<.001***			.956	1.046
Self-help score	.024	.049	0.388	.698			.956	1.046
Constant		.320	6.490	**<.001***				
Step 2					.137	.127		
Age	-.301	.007	-4.942	**<.001***			.948	1.055
Self-help score	.048	.048	0.780	.436			.943	1.061
SIB severity	.198	.025	3.290	**.001***			.973	1.028
Constant		.336	5.008	**.041***				
Step 3					.196	.183		
Age	-.236	.007	-3.871	**<.001***			.887	1.128
Self-help score	.050	.047	0.839	.402			.942	1.061
SIB severity	.103	.026	1.648	.101			.845	1.183
Impulsivity score	.271	.014	4.207	**<.001***			.797	1.254
Constant		.387	2.057	**.041***				
Step 4					.245	.230		
Age	-.237	.006	-4.004	**<.001***			.887	1.128
Self-help score	-.001	.046	-0.012	.991			.899	1.112
SIB severity	.083	.025	1.358	.176			.840	1.191
Impulsivity score	.208	.014	3.225	**.001***			.750	1.334
Compulsivity score	.238	.014	4.000	**<.001***			.878	1.139
Constant		.378	2.599	.085				
Step 5					.283	.265		
Age	-.226	.006	-3.911	**<.001***			.884	1.131
Self-help score	-.026	.046	-0.457	.648			.885	1.130
SIB severity	.019	.026	0.312	.755			.770	1.298
Impulsivity score	.122	.014	1.810	.072			.653	1.530
Compulsivity score	.168	.015	2.731	**.007***			.787	1.271
Anxiety score	.252	.007	3.553	**<.001***			.588	1.700
Constant		.381	1.730	.085				

Note. β = standardised beta coefficient; SE = standard error; R^2_{adj} = adjusted R^2, Tol. = tolerance, VIF = Variance Inflation Factor; SIB = self-injurious behaviour.
*Significant predictors (p<.05) in bold.

Appendix 4.1: *SIB study easy read information sheet*

Easy Read Information Study 1 v2.0 04.03.21
SIB Study
IRAS ID: 263048

Richards Lab

Medical Research Foundation

UNIVERSITY OF BIRMINGHAM

MRC Medical Research Council

CEREBRA Working wonders for children with brain conditions

Sleep - Impulsivity - Behaviour (SIB) Study

Hello,

We are a team from the University of Birmingham.



Dr Caroline Richards



Dr Georgie Agar



Katherine Marlow



Daniel Sutherland



Magda Skubera



Page 1 of 3

We work with children with autism and learning disabilities.





We would like you to take part in a study to find out about behaviour.

If you take part then we will:



- Ask your parents/the person who looks after you about you to find out about the things that you do and play with you

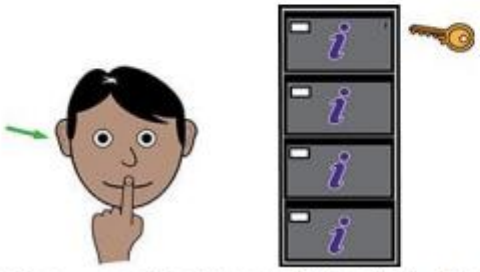




- Play some games with you and do some activities on the computer or tablet



- Take a short video of you



The video and information we collect is confidential – that means only we will look at it, we won't tell anyone else that you took part in the study.



Thank you.

Appendix 4.2: *SIB study background questionnaire*

ID number:
Date:

Background Questionnaire v1.0 05.04.19
SIB Study
IRAS ID: 263048

Sleep - Impulsivity - Behaviour (SIB) Study

Background Questionnaire

Please tick or write your response to these questions concerning background details of the person you care for:

1. **Today's date:** _____
2. **Gender:** Male ☐ Female ☐
3. **Date of Birth:** ___/___/____ **Age:**_____
4. **Is the person you care for verbal?** (i.e. more than 30 signs/words in their vocabulary)

   Yes/No (delete as appropriate)

5. **Is the person you care for able to walk unaided?**

   Yes/No (delete as appropriate)

6. **Has the person you care for been diagnosed with autism?** Yes/No (delete as appropriate)

   Please indicate which diagnosis the person you care for has been given:

Autism	☐	Autism Spectrum Disorder	☐
Asperger Syndrome	☐	High Functioning Autism	☐
Autistic Features	☐	Autistic (like) traits	☐
Autistic Continuum	☐	Pervasive Developmental Disorder	☐
Atypical Autism	☐	Autistic Spectrum	☐

7. **Has the person you care for been diagnosed with intellectual disability, learning disability or global developmental delay?** Yes/No (delete as appropriate)

   Please indicate the level of intellectual disability/learning disability of the person you care for:

Mild	☐	Moderate	☐
Severe	☐	Profound	☐
Unknown	☐		

   Other _____

8. **When was the person you care for diagnosed?** _____

Page 1 of 4

9. Who diagnosed the person you care for?

Paediatrician ☐ Clinical Geneticist ☐
GP ☐

Other _____

10. Has the person you care for ever been diagnosed with a biological sleep disorder? Yes/No (delete as appropriate)

If yes, please indicate below which diagnosis the person you care for has been given. If no, please move on to question 11.

Sleep-related breathing disorder (e.g. obstructive sleep apnea syndromes, central sleep apnea syndromes, sleep-related hypoventilation or hypoxemia disorders) ☐
Periodic limb movement disorder or restless leg syndrome ☐
Narcolepsy ☐

Other _____

11. Does the person you care for take any medication relating to sleep. If yes, please give details:

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

12. Has the person you care for ever been diagnosed with epilepsy? Yes/No (delete as appropriate)

If yes, please answer questions 13 to 16b. If no, please move on to question 17.

13. When was the person you care for diagnosed with epilepsy? _____

14. If known, please indicate the cause of the person you care for's epilepsy

_____

15. When did the person you care for last have a seizure? _____

_____

16. Is the person you care for currently taking medication to control their epilepsy?
Yes/No (delete as appropriate)

16a. If no, has the person you care for ever taken medication to control their epilepsy?
Yes/No (delete as appropriate)

16b. If yes to either of these, please give details below and indicate whether the person you care for is currently taking the medication or has stopped and for what reason.

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

17. Has the person you care for had any medical/health difficulties in the last six months? If yes, please give details:

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

18. Does the person you care for take any medication relating to behaviour? If yes, please give details:

_____

_____

_____

_____

_____

_____

_____

_____

_____

_____

In the information sheet and consent form we informed you that we may wish to contact your child's/person you care for's GP (or alternative health professional e.g. Paediatrician) in order to communicate to them information regarding your child's health and diagnostic status (see consent form and information sheet for more information). If you have already indicated on the consent form that you are happy for us to do this, please complete the relevant details below:

19. Name of the person you care for's GP_____

GP Address_____

_____

GP Telephone number_____

Appendix 4.3: *The Vineland Adaptive Behaviour Scale-Third Edition*



Vineland-3

Vineland Adaptive Behavior Scales™–Third Edition

Comprehensive Interview Form

Examinee's Name: _____ Today's Date: _____ Examinee's Birth Date: _____

Examinee's Age: _____ Examinee's Sex: ☐ Female ☐ Male Respondent's Name: _____

Respondent's Relationship to the Examinee: ☐ Mother ☐ Father ☐ Other _____ Interviewer's Name: _____

DIRECTIONS

Complete instructions for administering this form are provided in Chapter 2 of the Vineland-3 Manual. The Manual also includes a list of suggested interview questions and scoring criteria for the items in Appendix G, which you may wish to have available when conducting the interview.

Keep in mind that the Coping Skills subdomain does not begin until age 2; the Written, Domestic, and Community subdomains do not begin until age 3; and the optional Maladaptive Behavior domain does not begin until age 3.

SCORING THE ITEMS

Score each item 2, 1, or 0 as follows:

Circle 2 if, when the behavior is needed or appropriate, the individual *usually* performs it without help or prompting (or if he/she performed the behavior when younger, but has now outgrown it).

Circle 1 if, when the behavior is needed or appropriate, the individual *sometimes* performs it without help or prompting.

Circle 0 if the individual *never* performs the behavior, or never performs it without help or prompting. Whether he/she hasn't learned the behavior, is not physically able to perform it, is not expected or allowed to perform it, or chooses not to perform it, circle 0.

Some items use a different scoring system: 2 = Yes, 0 = No. This is indicated in the booklet.

If the respondent has not had the opportunity to observe a behavior, ask him/her to estimate a score for that item. In addition to marking the item score, also check the box to the right of the item score, in the column labeled **Check if Est.**

If the individual performed a behavior when he/she was younger, but has now outgrown it, circle 2. Do not check the Estimated box.

Some items include a Scoring Tip, indicated with the symbol ☑. Use the tips to help you score those items.

BASAL AND CEILING RULES

A basal is established when there are *four consecutive items* with scores of 2. A ceiling is established when there are *four consecutive items* with scores of 0. Continue administering Interview Topics until you have established a basal and a ceiling, and all items between the basal and ceiling have been scored. If there is no basal, you must score all items from the first item in the subdomain up to the ceiling. If there is no ceiling, you must score all items from the last item in the subdomain back to the basal.

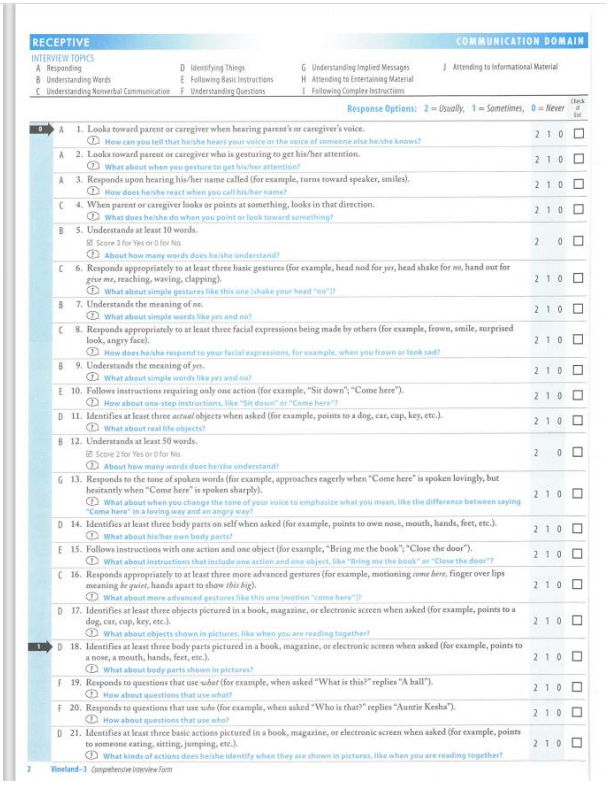
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PsychCorp

Product Number 31321



RECEPTIVE — COMMUNICATION DOMAIN

INTERVIEW TOPICS

A Responding; B Understanding Words; C Understanding Nonverbal Communication; D Identifying Things; E Following Basic Instructions; F Understanding Questions; G Understanding Implied Messages; H Attending to Entertaining Material; I Following Complex Instructions; J Attending to Informational Material

Response Options: 2 = *Usually*, 1 = *Sometimes*, 0 = *Never* — Check if Est

0 A 1. Looks toward parent or caregiver when hearing parent's or caregiver's voice. — 2 1 0
ⓘ How can you tell that he/she hears your voice or the voice of someone else he/she knows?

A 2. Looks toward parent or caregiver who is gesturing to get his/her attention. — 2 1 0
ⓘ What about when you gesture to get his/her attention?

A 3. Responds upon hearing his/her name called (for example, turns toward speaker, smiles). — 2 1 0
ⓘ How does he/she react when you call his/her name?

C 4. When parent or caregiver looks or points at something, looks in that direction. — 2 1 0
ⓘ What does he/she do when you point or look toward something?

B 5. Understands at least 10 words. — 2 0
☑ Score 2 for Yes or 0 for No.
ⓘ About how many words does he/she understand?

C 6. Responds appropriately to at least three basic gestures (for example, head nod for *yes*, head shake for *no*, hand out for *give me*, reaching, waving, clapping). — 2 1 0
ⓘ What about simple gestures like this one (shake your head "no")?

B 7. Understands the meaning of *no*. — 2 1 0
ⓘ What about simple words like yes and no?

C 8. Responds appropriately to at least three facial expressions being made by others (for example, frown, smile, surprised look, angry face). — 2 1 0
ⓘ How does he/she respond to your facial expressions, for example, when you frown or look sad?

B 9. Understands the meaning of *yes*. — 2 1 0
ⓘ What about simple words like yes and no?

E 10. Follows instructions requiring only one action (for example, "Sit down"; "Come here"). — 2 1 0
ⓘ How about one-step instructions, like "Sit down" or "Come here"?

D 11. Identifies at least three *actual* objects when asked (for example, points to a dog, car, cup, key, etc.). — 2 1 0
ⓘ What about real life objects?

B 12. Understands at least 50 words. — 2 0
☑ Score 2 for Yes or 0 for No.
ⓘ About how many words does he/she understand?

G 13. Responds to the tone of spoken words (for example, approaches eagerly when "Come here" is spoken lovingly, but hesitantly when "Come here" is spoken sharply). — 2 1 0
ⓘ What about when you change the tone of your voice to emphasize what you mean, like the difference between saying "Come here" in a loving way and an angry way?

D 14. Identifies at least three body parts on self when asked (for example, points to own nose, mouth, hands, feet, etc.). — 2 1 0
ⓘ What about his/her own body parts?

E 15. Follows instructions with one action and one object (for example, "Bring me the book"; "Close the door"). — 2 1 0
ⓘ What about instructions that include one action and one object, like "Bring me the book" or "Close the door"?

C 16. Responds appropriately to at least three more advanced gestures (for example, motioning *come here*, finger over lips meaning *be quiet*, hands apart to show *this big*). — 2 1 0
ⓘ What about more advanced gestures like this one (motion "come here")?

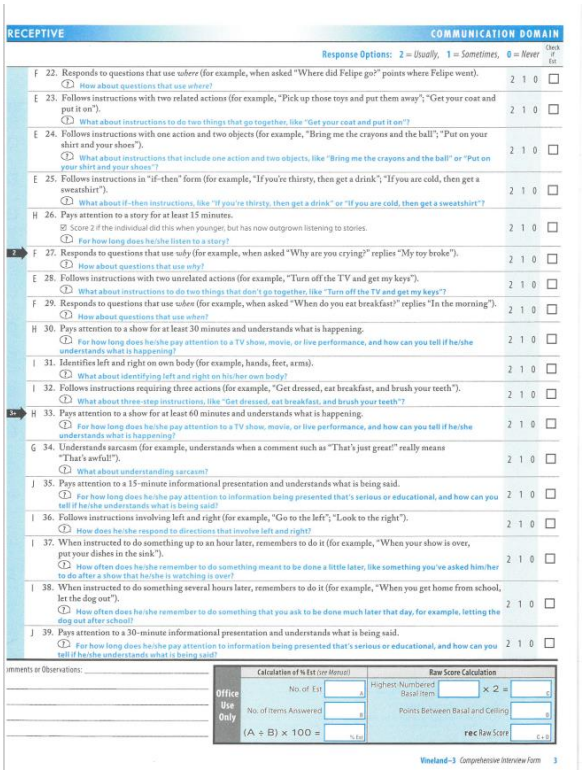
D 17. Identifies at least three objects pictured in a book, magazine, or electronic screen when asked (for example, points to a dog, car, cup, key, etc.). — 2 1 0
ⓘ What about objects shown in pictures, like when you are reading together?

1 D 18. Identifies at least three body parts pictured in a book, magazine, or electronic screen when asked (for example, points to a nose, a mouth, hands, feet, etc.). — 2 1 0
ⓘ What about body parts shown in pictures?

F 19. Responds to questions that use *what* (for example, when asked "What is this?" replies "A ball"). — 2 1 0
ⓘ How about questions that use what?

F 20. Responds to questions that use *who* (for example, when asked "Who is that?" replies "Auntie Kesha"). — 2 1 0
ⓘ How about questions that use who?

D 21. Identifies at least three basic actions pictured in a book, magazine, or electronic screen when asked (for example, points to someone eating, sitting, jumping, etc.). — 2 1 0
ⓘ What kinds of actions does he/she identify when they are shown in pictures, like when you are reading together?



RECEPTIVE — COMMUNICATION DOMAIN

Response Options: 2 = *Usually*, 1 = *Sometimes*, 0 = *Never* — Check if Est

F 22. Responds to questions that use *where* (for example, when asked "Where did Felipe go?" points where Felipe went). — 2 1 0
ⓘ How about questions that use where?

E 23. Follows instructions with two related actions (for example, "Pick up those toys and put them away"; "Get your coat and put it on"). — 2 1 0
ⓘ What about instructions to do two things that go together, like "Get your coat and put it on"?

E 24. Follows instructions with one action and two objects (for example, "Bring me the crayons and the ball"; "Put on your shirt and your shoes"). — 2 1 0
ⓘ What about instructions that include one action and two objects, like "Bring me the crayons and the ball" or "Put on your shirt and your shoes"?

E 25. Follows instructions in "if-then" form (for example, "If you're thirsty, then get a drink"; "If you are cold, then get a sweatshirt"). — 2 1 0
ⓘ What about if-then instructions, like "If you're thirsty, then get a drink" or "If you are cold, then get a sweatshirt"?

H 26. Pays attention to a story for at least 15 minutes. — 2 1 0
☑ Score 2 if the individual did this when younger, but has now outgrown listening to stories.
ⓘ For how long does he/she listen to a story?

2 F 27. Responds to questions that use *why* (for example, when asked "Why are you crying?" replies "My toy broke"). — 2 1 0
ⓘ How about questions that use why?

E 28. Follows instructions with two unrelated actions (for example, "Turn off the TV and get my keys"). — 2 1 0
ⓘ What about instructions to do two things that don't go together, like "Turn off the TV and get my keys"?

F 29. Responds to questions that use *when* (for example, when asked "When do you eat breakfast?" replies "In the morning"). — 2 1 0
ⓘ How about questions that use when?

H 30. Pays attention to a show for at least 30 minutes and understands what is happening. — 2 1 0
ⓘ For how long does he/she pay attention to a TV show, movie, or live performance, and how can you tell if he/she understands what is happening?

I 31. Identifies left and right on own body (for example, hands, feet, arms). — 2 1 0
ⓘ What about identifying left and right on his/her own body?

I 32. Follows instructions requiring three actions (for example, "Get dressed, eat breakfast, and brush your teeth"). — 2 1 0
ⓘ What about three-step instructions, like "Get dressed, eat breakfast, and brush your teeth"?

3+ H 33. Pays attention to a show for at least 60 minutes and understands what is happening. — 2 1 0
ⓘ For how long does he/she pay attention to a TV show, movie, or live performance, and how can you tell if he/she understands what is happening?

G 34. Understands sarcasm (for example, understands when a comment such as "That's just great!" really means "That's awful!"). — 2 1 0
ⓘ What about understanding sarcasm?

J 35. Pays attention to a 15-minute informational presentation and understands what is being said. — 2 1 0
ⓘ For how long does he/she pay attention to information being presented that's serious or educational, and how can you tell if he/she understands what is being said?

I 36. Follows instructions involving left and right (for example, "Go to the left"; "Look to the right"). — 2 1 0
ⓘ How does he/she respond to directions that involve left and right?

I 37. When instructed to do something up to an hour later, remembers to do it (for example, "When your show is over, put your dishes in the sink"). — 2 1 0
ⓘ How often does he/she remember to do something meant to be done a little later, like something you've asked him/her to do after a show that he/she is watching is over?

I 38. When instructed to do something several hours later, remembers to do it (for example, "When you get home from school, let the dog out"). — 2 1 0
ⓘ How often does he/she remember to do something that you ask to be done much later that day, for example, letting the dog out after school?

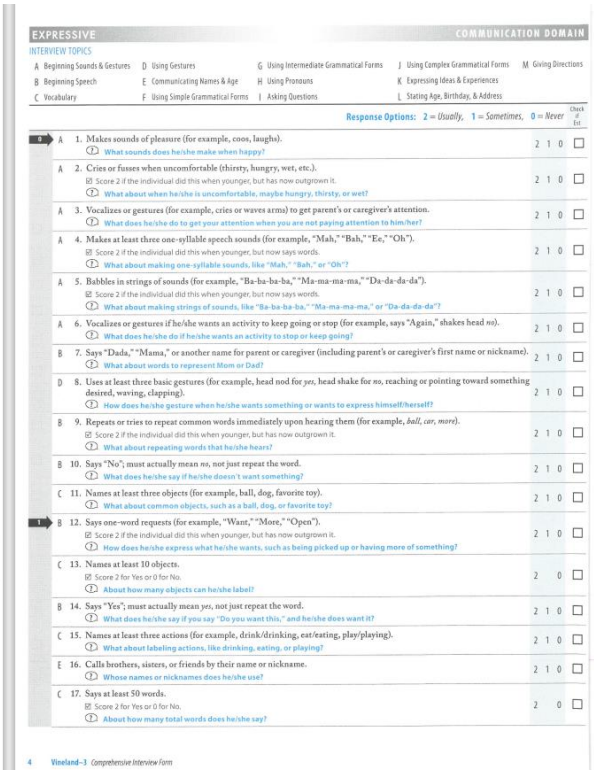
J 39. Pays attention to a 30-minute informational presentation and understands what is being said. — 2 1 0
ⓘ For how long does he/she pay attention to information being presented that's serious or educational, and how can you tell if he/she understands what is being said?

Comments or Observations: _____

Office Use Only

Calculation of % Est (*see Manual*): No. of Est [A] ÷ No. of Items Answered [B] × 100 = % Est

Raw Score Calculation: Highest Numbered Basal Item × 2 = [C]; Points Between Basal and Ceiling [D]; rec Raw Score [C + D]



EXPRESSIVE — COMMUNICATION DOMAIN

INTERVIEW TOPICS

A Beginning Sounds & Gestures; B Beginning Speech; C Vocabulary; D Using Gestures; E Communicating Names & Age; F Using Simple Grammatical Forms; G Using Intermediate Grammatical Forms; H Using Pronouns; I Asking Questions; J Using Complex Grammatical Forms; K Expressing Ideas & Experiences; L Stating Age, Birthday, & Address; M Giving Directions

Response Options: 2 = *Usually*, 1 = *Sometimes*, 0 = *Never* — Check if Est

0 A 1. Makes sounds of pleasure (for example, coos, laughs). — 2 1 0
ⓘ What sounds does he/she make when happy?

A 2. Cries or fusses when uncomfortable (thirsty, hungry, wet, etc.). — 2 1 0
☑ Score 2 if the individual did this when younger, but has now outgrown it.
ⓘ What about when he/she is uncomfortable, maybe hungry, thirsty, or wet?

A 3. Vocalizes or gestures (for example, cries or waves arms) to get parent's or caregiver's attention. — 2 1 0
ⓘ What does he/she do to get your attention when you are not paying attention to him/her?

A 4. Makes at least three one-syllable speech sounds (for example, "Mah," "Bah," "Ee," "Oh"). — 2 1 0
☑ Score 2 if the individual did this when younger, but now says words.
ⓘ What about making one-syllable sounds, like "Mah," "Bah," or "Oh"?

A 5. Babbles in strings of sounds (for example, "Ba-ba-ba-ba," "Ma-ma-ma-ma," "Da-da-da-da"). — 2 1 0
☑ Score 2 if the individual did this when younger, but now says words.
ⓘ What about making strings of sounds, like "Ba-ba-ba-ba," "Ma-ma-ma-ma," or "Da-da-da-da"?

A 6. Vocalizes or gestures if he/she wants an activity to keep going or stop (for example, says "Again," shakes head *no*). — 2 1 0
ⓘ What does he/she do if he/she wants an activity to stop or keep going?

B 7. Says "Dada," "Mama," or another name for parent or caregiver (including parent's or caregiver's first name or nickname). — 2 1 0
ⓘ What about words to represent Mom or Dad?

D 8. Uses at least three basic gestures (for example, head nod for *yes*, head shake for *no*, reaching or pointing toward something desired, waving, clapping). — 2 1 0
ⓘ How does he/she gesture when he/she wants something or wants to express himself/herself?

B 9. Repeats or tries to repeat common words immediately upon hearing them (for example, *ball*, *car*, *more*). — 2 1 0
☑ Score 2 if the individual did this when younger, but has now outgrown it.
ⓘ What about repeating words that he/she hears?

B 10. Says "No"; must actually mean *no*, not just repeat the word. — 2 1 0
ⓘ What does he/she say if he/she doesn't want something?

C 11. Names at least three objects (for example, ball, dog, favorite toy). — 2 1 0
ⓘ What about common objects, such as a ball, dog, or favorite toy?

1 B 12. Says one-word requests (for example, "Want," "More," "Open"). — 2 1 0
☑ Score 2 if the individual did this when younger, but has now outgrown it.
ⓘ How does he/she express what he/she wants, such as being picked up or having more of something?

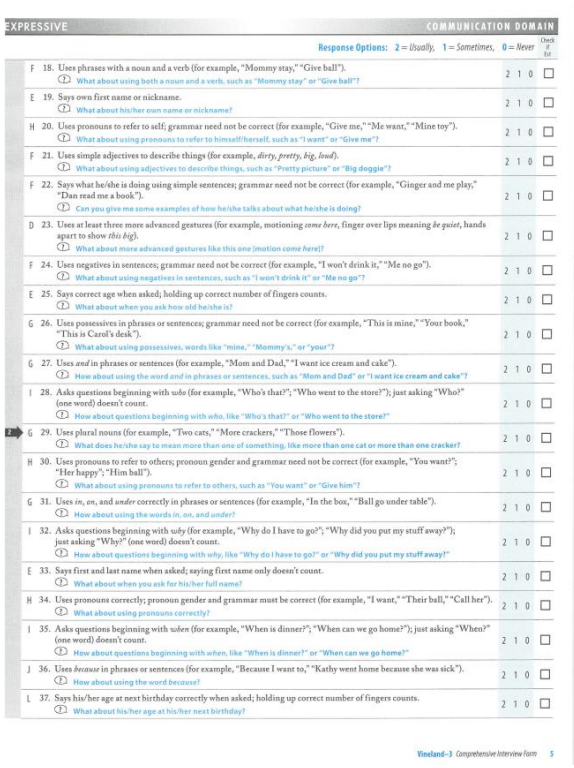
C 13. Names at least 10 objects. — 2 0
☑ Score 2 for Yes or 0 for No.
ⓘ About how many objects can he/she label?

B 14. Says "Yes"; must actually mean *yes*, not just repeat the word. — 2 1 0
ⓘ What does he/she say if you say "Do you want this," and he/she does want it?

C 15. Names at least three actions (for example, drink/drinking, eat/eating, play/playing). — 2 1 0
ⓘ What about labeling actions, like drinking, eating, or playing?

E 16. Calls brothers, sisters, or friends by their name or nickname. — 2 1 0
ⓘ Whose names or nicknames does he/she use?

C 17. Says at least 50 words. — 2 0
☑ Score 2 for Yes or 0 for No.
ⓘ About how many total words does he/she say?



EXPRESSIVE — COMMUNICATION DOMAIN

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

F 18. Uses phrases with a noun and a verb (for example, "Mommy stay," "Give ball").
What about using both a noun and a verb, such as "Mommy stay" or "Give ball"? 2 1 0

E 19. Says own first name or nickname.
What about his/her own name or nickname? 2 1 0

H 20. Uses pronouns to refer to self; grammar need not be correct (for example, "Give me," "Me want," "Mine toy").
What about using pronouns to refer to himself/herself, such as "I want" or "Give me"? 2 1 0

F 21. Uses simple adjectives to describe things (for example, *dirty, pretty, big, loud*).
What about using adjectives to describe things, such as "Pretty picture" or "Big doggie"? 2 1 0

F 22. Says what he/she is doing using simple sentences; grammar need not be correct (for example, "Ginger and me play," "Dan read me a book").
Can you give me some examples of how he/she talks about what he/she is doing? 2 1 0

D 23. Uses at least three more advanced gestures (for example, motioning *come here*, finger over lips meaning *be quiet*, hands apart to show *this big*).
What about more advanced gestures like this one [motion *come here*]? 2 1 0

F 24. Uses negatives in sentences; grammar need not be correct (for example, "I won't drink it," "Me no go").
What about using negatives in sentences, such as "I won't drink it" or "Me no go"? 2 1 0

E 25. Says correct age when asked; holding up correct number of fingers counts.
What about when you ask how old he/she is? 2 1 0

G 26. Uses possessives in phrases or sentences; grammar need not be correct (for example, "This is mine," "Your book," "This is Carol's desk").
What about using possessives, words like "mine," "Mommy's," or "your"? 2 1 0

G 27. Uses *and* in phrases or sentences (for example, "Mom and Dad," "I want ice cream and cake").
How about using the word *and* in phrases or sentences, such as "Mom and Dad" or "I want ice cream and cake"? 2 1 0

I 28. Asks questions beginning with *who* (for example, "Who's that?"; "Who went to the store?"); just asking "Who?" (one word) doesn't count.
How about questions beginning with *who*, like "Who's that?" or "Who went to the store?" 2 1 0

2 G 29. Uses plural nouns (for example, "Two cats," "More crackers," "Those flowers").
What does he/she say to mean more than one of something, like more than one cat or more than one cracker? 2 1 0

H 30. Uses pronouns to refer to others; pronoun gender and grammar need not be correct (for example, "You want?"; "Her happy"; "Him ball").
What about using pronouns to refer to others, such as "You want" or "Give him"? 2 1 0

G 31. Uses *in, on,* and *under* correctly in phrases or sentences (for example, "In the box," "Ball go under table").
How about using the words *in, on,* and *under*? 2 1 0

I 32. Asks questions beginning with *why* (for example, "Why do I have to go?"; "Why did you put my stuff away?"); just asking "Why?" (one word) doesn't count.
How about questions beginning with *why*, like "Why do I have to go?" or "Why did you put my stuff away?" 2 1 0

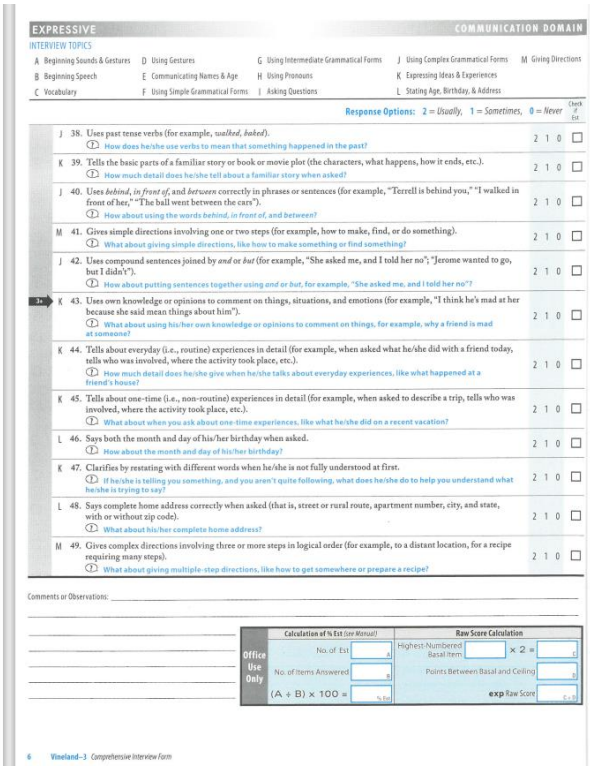
E 33. Says first and last name when asked; saying first name only doesn't count.
What about when you ask for his/her full name? 2 1 0

H 34. Uses pronouns correctly; pronoun gender and grammar must be correct (for example, "I want," "Their ball," "Call her").
What about using pronouns correctly? 2 1 0

I 35. Asks questions beginning with *when* (for example, "When is dinner?"; "When can we go home?"); just asking "When?" (one word) doesn't count.
How about questions beginning with *when*, like "When is dinner?" or "When can we go home?" 2 1 0

J 36. Uses *because* in phrases or sentences (for example, "Because I want to," "Kathy went home because she was sick").
How about using the word *because*? 2 1 0

L 37. Says his/her age at next birthday correctly when asked; holding up correct number of fingers counts.
What about his/her age at his/her next birthday? 2 1 0



EXPRESSIVE — COMMUNICATION DOMAIN

INTERVIEW TOPICS

A Beginning Sounds & Gestures · B Beginning Speech · C Vocabulary · D Using Gestures · E Communicating Names & Age · F Using Simple Grammatical Forms · G Using Intermediate Grammatical Forms · H Using Pronouns · I Asking Questions · J Using Complex Grammatical Forms · K Expressing Ideas & Experiences · L Stating Age, Birthday, & Address · M Giving Directions

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

J 38. Uses past tense verbs (for example, *walked, baked*).
How does he/she use verbs to mean that something happened in the past? 2 1 0

K 39. Tells the basic parts of a familiar story or book or movie plot (the characters, what happens, how it ends, etc.).
How much detail does he/she tell about a familiar story when asked? 2 1 0

J 40. Uses *behind, in front of,* and *between* correctly in phrases or sentences (for example, "Terrell is behind you," "I walked in front of her," "The ball went between the cars").
How about using the words *behind, in front of,* and *between*? 2 1 0

M 41. Gives simple directions involving one or two steps (for example, how to make, find, or do something).
What about giving simple directions, like how to make something or find something? 2 1 0

J 42. Uses compound sentences joined by *and* or *but* (for example, "She asked me, and I told her no"; "Jerome wanted to go, but I didn't").
How about putting sentences together using *and* or *but*, for example, "She asked me, and I told her no"? 2 1 0

3+ K 43. Uses own knowledge or opinions to comment on things, situations, and emotions (for example, "I think he's mad at her because she said mean things about him").
What about using his/her own knowledge or opinions to comment on things, for example, why a friend is mad at someone? 2 1 0

K 44. Tells about everyday (i.e., routine) experiences in detail (for example, when asked what he/she did with a friend today, tells who was involved, where the activity took place, etc.).
How much detail does he/she give when he/she talks about everyday experiences, like what happened at a friend's house? 2 1 0

K 45. Tells about one-time (i.e., non-routine) experiences in detail (for example, when asked to describe a trip, tells who was involved, where the activity took place, etc.).
What about when you ask about one-time experiences, like what he/she did on a recent vacation? 2 1 0

L 46. Says both the month and day of his/her birthday when asked.
How about the month and day of his/her birthday? 2 1 0

K 47. Clarifies by restating with different words when he/she is not fully understood at first.
If he/she is telling you something, and you aren't quite following, what does he/she do to help you understand what he/she is trying to say? 2 1 0

L 48. Says complete home address correctly when asked (that is, street or rural route, apartment number, city, and state, with or without zip code).
What about his/her complete home address? 2 1 0

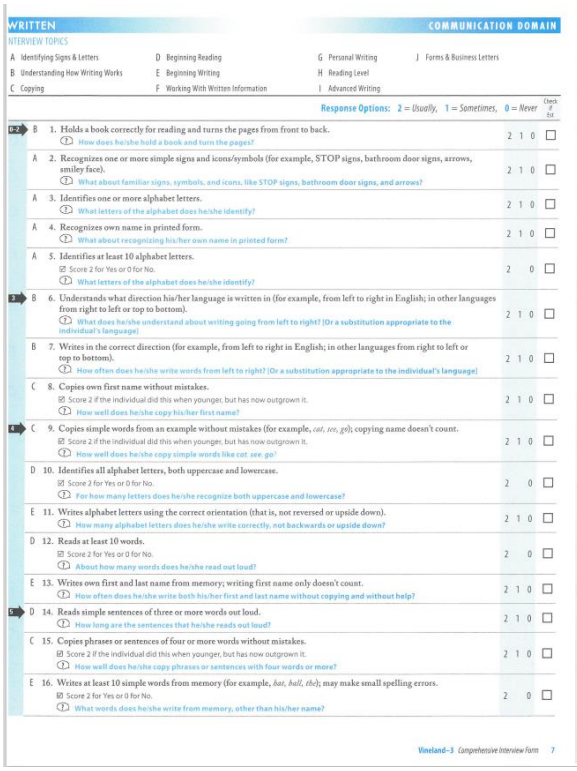
M 49. Gives complex directions involving three or more steps in logical order (for example, to a distant location, for a recipe requiring many steps).
What about giving multiple-step directions, like how to get somewhere or prepare a recipe? 2 1 0

Comments or Observations: _____

Office Use Only

Calculation of % Est (see Manual)
No. of Est [A]
No. of Items Answered [B]
(A ÷ B) × 100 = % Est

Raw Score Calculation
Highest Numbered Basal Item [ ] × 2 = [C]
Points Between Basal and Ceiling [D]
exp Raw Score C + D



WRITTEN — COMMUNICATION DOMAIN

INTERVIEW TOPICS

A Identifying Signs & Letters · B Understanding How Writing Works · C Copying · D Beginning Reading · E Beginning Writing · F Working With Written Information · G Personal Writing · H Reading Level · I Advanced Writing · J Forms & Business Letters

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

0-2 B 1. Holds a book correctly for reading and turns the pages from front to back.
How does he/she hold a book and turn the pages? 2 1 0

A 2. Recognizes one or more simple signs and icons/symbols (for example, STOP signs, bathroom door signs, arrows, smiley face).
What about familiar signs, symbols, and icons, like STOP signs, bathroom door signs, and arrows? 2 1 0

A 3. Identifies one or more alphabet letters.
What letters of the alphabet does he/she identify? 2 1 0

A 4. Recognizes own name in printed form.
What about recognizing his/her own name in printed form? 2 1 0

A 5. Identifies at least 10 alphabet letters.
Score 2 for Yes or 0 for No.
What letters of the alphabet does he/she identify? 2 0

3 B 6. Understands what direction his/her language is written in (for example, from left to right in English; in other languages from right to left or top to bottom).
What does he/she understand about writing going from left to right? [Or a substitution appropriate to the individual's language] 2 1 0

B 7. Writes in the correct direction (for example, from left to right in English; in other languages from right to left or top to bottom).
How often does he/she write words from left to right? [Or a substitution appropriate to the individual's language] 2 1 0

C 8. Copies own first name without mistakes.
Score 2 if the individual did this when younger, but has now outgrown it.
How well does he/she copy his/her first name? 2 1 0

4 C 9. Copies simple words from an example without mistakes (for example, *cat, see, go*); copying name doesn't count.
Score 2 if the individual did this when younger, but has now outgrown it.
How well does he/she copy simple words like *cat, see, go*? 2 1 0

D 10. Identifies all alphabet letters, both uppercase and lowercase.
Score 2 for Yes or 0 for No.
For how many letters does he/she recognize both uppercase and lowercase? 2 0

E 11. Writes alphabet letters using the correct orientation (that is, not reversed or upside down).
How many alphabet letters does he/she write correctly, not backwards or upside down? 2 1 0

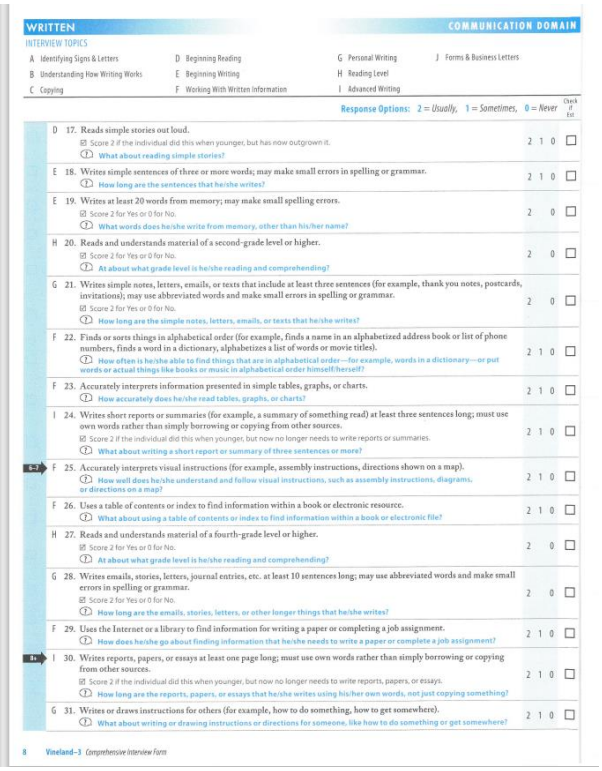
D 12. Reads at least 10 words.
Score 2 for Yes or 0 for No.
About how many words does he/she read out loud? 2 0

E 13. Writes own first and last name from memory; writing first name only doesn't count.
How often does he/she write both his/her first and last name without copying and without help? 2 1 0

5 D 14. Reads simple sentences of three or more words out loud.
How long are the sentences that he/she reads out loud? 2 1 0

C 15. Copies phrases or sentences of four or more words without mistakes.
Score 2 if the individual did this when younger, but has now outgrown it.
How well does he/she copy phrases or sentences with four words or more? 2 1 0

E 16. Writes at least 10 simple words from memory (for example, *bat, ball, the*); may make small spelling errors.
Score 2 for Yes or 0 for No.
What words does he/she write from memory, other than his/her name? 2 0



WRITTEN — COMMUNICATION DOMAIN

INTERVIEW TOPICS

A Identifying Signs & Letters · B Understanding How Writing Works · C Copying · D Beginning Reading · E Beginning Writing · F Working With Written Information · G Personal Writing · H Reading Level · I Advanced Writing · J Forms & Business Letters

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

D 17. Reads simple stories out loud.
Score 2 if the individual did this when younger, but has now outgrown it.
What about reading simple stories? 2 1 0

E 18. Writes simple sentences of three or more words; may make small errors in spelling or grammar.
How long are the sentences that he/she writes? 2 1 0

E 19. Writes at least 20 words from memory; may make small spelling errors.
Score 2 for Yes or 0 for No.
What words does he/she write from memory, other than his/her name? 2 0

H 20. Reads and understands material of a second-grade level or higher.
Score 2 for Yes or 0 for No.
At about what grade level is he/she reading and comprehending? 2 0

G 21. Writes simple notes, letters, emails, or texts that include at least three sentences (for example, thank you notes, postcards, invitations); may use abbreviated words and make small errors in spelling or grammar.
Score 2 for Yes or 0 for No.
How long are the simple notes, letters, emails, or texts that he/she writes? 2 0

F 22. Finds or sorts things in alphabetical order (for example, finds a name in an alphabetized address book or list of phone numbers, finds a word in a dictionary, alphabetizes a list of words or movie titles).
How often is he/she able to find things that are in alphabetical order—for example, words in a dictionary—or put words or actual things like books or music in alphabetical order himself/herself? 2 1 0

F 23. Accurately interprets information presented in simple tables, graphs, or charts.
How accurately does he/she read tables, graphs, or charts? 2 1 0

I 24. Writes short reports or summaries (for example, a summary of something read) at least three sentences long; must use own words rather than simply borrowing or copying from other sources.
Score 2 if the individual did this when younger, but now no longer needs to write reports or summaries.
What about writing a short report or summary of three sentences or more? 2 1 0

6+ F 25. Accurately interprets visual instructions (for example, assembly instructions, directions shown on a map).
How well does he/she understand and follow visual instructions, such as assembly instructions, diagrams, or directions on a map? 2 1 0

F 26. Uses a table of contents or index to find information within a book or electronic resource.
What about using a table of contents or index to find information within a book or electronic file? 2 1 0

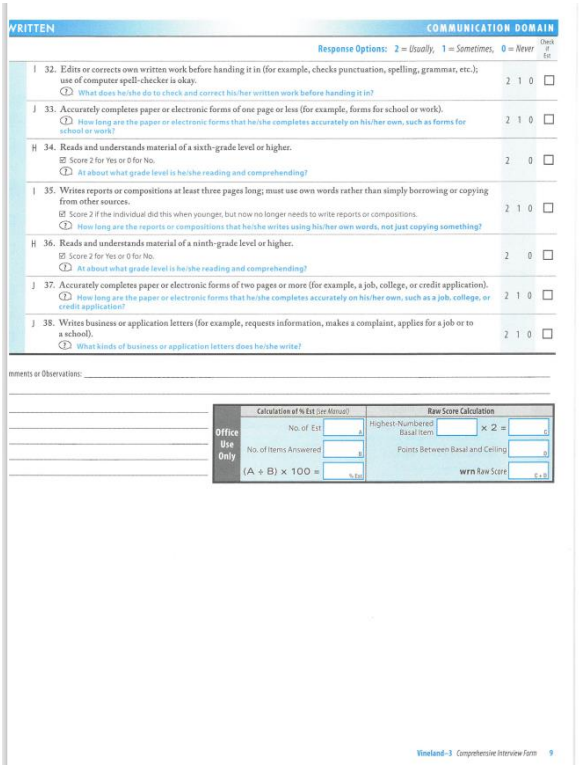
H 27. Reads and understands material of a fourth-grade level or higher.
Score 2 for Yes or 0 for No.
At about what grade level is he/she reading and comprehending? 2 0

G 28. Writes emails, stories, letters, journal entries, etc. at least 10 sentences long; may use abbreviated words and make small errors in spelling or grammar.
Score 2 for Yes or 0 for No.
How long are the emails, stories, letters, or other longer things that he/she writes? 2 0

F 29. Uses the Internet or a library to find information for writing a paper or completing a job assignment.
How does he/she go about finding information that he/she needs to write a paper or complete a job assignment? 2 1 0

8+ I 30. Writes reports, papers, or essays at least one page long; must use own words rather than simply borrowing or copying from other sources.
Score 2 if the individual did this when younger, but now no longer needs to write reports, papers, or essays.
How long are the reports, papers, or essays that he/she writes using his/her own words, not just copying something? 2 1 0

G 31. Writes or draws instructions for others (for example, how to do something, how to get somewhere).
What about writing or drawing instructions or directions for someone, like how to do something or get somewhere? 2 1 0



WRITTEN — COMMUNICATION DOMAIN

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

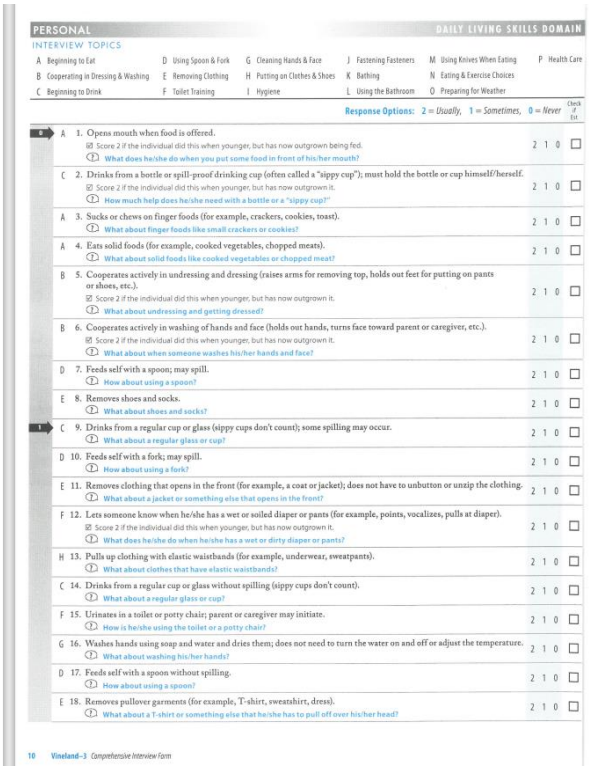
			Check if Est
I	32. Edits or corrects own written work before handing it in (for example, checks punctuation, spelling, grammar, etc.); use of computer spell-checker is okay. ⓘ What does he/she do to check and correct his/her written work before handing it in?	2 1 0	☐
J	33. Accurately completes paper or electronic forms of one page or less (for example, forms for school or work). ⓘ How long are the paper or electronic forms that he/she completes accurately on his/her own, such as forms for school or work?	2 1 0	☐
H	34. Reads and understands material of a sixth-grade level or higher. ☑ Score 2 for Yes or 0 for No. ⓘ At about what grade level is he/she reading and comprehending?	2 0	☐
I	35. Writes reports or compositions at least three pages long; must use own words rather than simply borrowing or copying from other sources. ☑ Score 2 if the individual did this when younger, but now no longer needs to write reports or compositions. ⓘ How long are the reports or compositions that he/she writes using his/her own words, not just copying something?	2 1 0	☐
H	36. Reads and understands material of a ninth-grade level or higher. ☑ Score 2 for Yes or 0 for No. ⓘ At about what grade level is he/she reading and comprehending?	2 0	☐
J	37. Accurately completes paper or electronic forms of two pages or more (for example, a job, college, or credit application). ⓘ How long are the paper or electronic forms that he/she completes accurately on his/her own, such as a job, college, or credit application?	2 1 0	☐
J	38. Writes business or application letters (for example, requests information, makes a complaint, applies for a job or to a school). ⓘ What kinds of business or application letters does he/she write?	2 1 0	☐

mments or Observations: _____

Office Use Only

Calculation of % Est (see Manual)		Raw Score Calculation	
No. of Est	A	Highest-Numbered Basal Item	× 2 = C
No. of Items Answered	B	Points Between Basal and Ceiling	D
(A ÷ B) × 100 =	% Est	wrn Raw Score	C + D

Vineland-3 Comprehensive Interview Form 9



PERSONAL — DAILY LIVING SKILLS DOMAIN

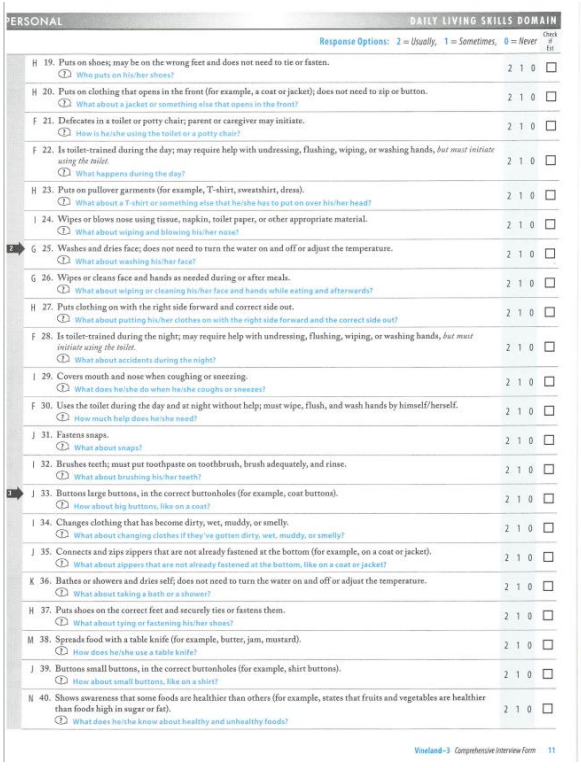
INTERVIEW TOPICS

A Beginning to Eat	D Using Spoon & Fork	G Cleaning Hands & Face	J Fastening Fasteners	M Using Knives When Eating	P Health Care
B Cooperating in Dressing & Washing	E Removing Clothing	H Putting on Clothes & Shoes	K Bathing	N Eating & Exercise Choices	
C Beginning to Drink	F Toilet Training	I Hygiene	L Using the Bathroom	O Preparing for Weather	

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

				Check if Est
0	A	1. Opens mouth when food is offered. ☑ Score 2 if the individual did this when younger, but has now outgrown being fed. ⓘ What does he/she do when you put some food in front of his/her mouth?	2 1 0	☐
	C	2. Drinks from a bottle or spill-proof drinking cup (often called a "sippy cup"); must hold the bottle or cup himself/herself. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ How much help does he/she need with a bottle or a "sippy cup?"	2 1 0	☐
	A	3. Sucks or chews on finger foods (for example, crackers, cookies, toast). ⓘ What about finger foods like small crackers or cookies?	2 1 0	☐
	A	4. Eats solid foods (for example, cooked vegetables, chopped meats). ⓘ What about solid foods like cooked vegetables or chopped meat?	2 1 0	☐
	B	5. Cooperates actively in undressing and dressing (raises arms for removing top, holds out feet for putting on pants or shoes, etc.). ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ What about undressing and getting dressed?	2 1 0	☐
	B	6. Cooperates actively in washing of hands and face (holds out hands, turns face toward parent or caregiver, etc.). ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ What about when someone washes his/her hands and face?	2 1 0	☐
	D	7. Feeds self with a spoon; may spill. ⓘ How about using a spoon?	2 1 0	☐
	E	8. Removes shoes and socks. ⓘ What about shoes and socks?	2 1 0	☐
1	C	9. Drinks from a regular cup or glass (sippy cups don't count); some spilling may occur. ⓘ What about a regular glass or cup?	2 1 0	☐
	D	10. Feeds self with a fork; may spill. ⓘ How about using a fork?	2 1 0	☐
	E	11. Removes clothing that opens in the front (for example, a coat or jacket); does not have to unbutton or unzip the clothing. ⓘ What about a jacket or something else that opens in the front?	2 1 0	☐
	F	12. Lets someone know when he/she has a wet or soiled diaper or pants (for example, points, vocalizes, pulls at diaper). ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ What does he/she do when he/she has a wet or dirty diaper or pants?	2 1 0	☐
	H	13. Pulls up clothing with elastic waistbands (for example, underwear, sweatpants). ⓘ What about clothes that have elastic waistbands?	2 1 0	☐
	C	14. Drinks from a regular cup or glass without spilling (sippy cups don't count). ⓘ What about a regular glass or cup?	2 1 0	☐
	F	15. Urinates in a toilet or potty chair; parent or caregiver may initiate. ⓘ How is he/she using the toilet or a potty chair?	2 1 0	☐
	G	16. Washes hands using soap and water and dries them; does not need to turn the water on and off or adjust the temperature. ⓘ What about washing his/her hands?	2 1 0	☐
	D	17. Feeds self with a spoon without spilling. ⓘ How about using a spoon?	2 1 0	☐
	E	18. Removes pullover garments (for example, T-shirt, sweatshirt, dress). ⓘ What about a T-shirt or something else that he/she has to pull off over his/her head?	2 1 0	☐

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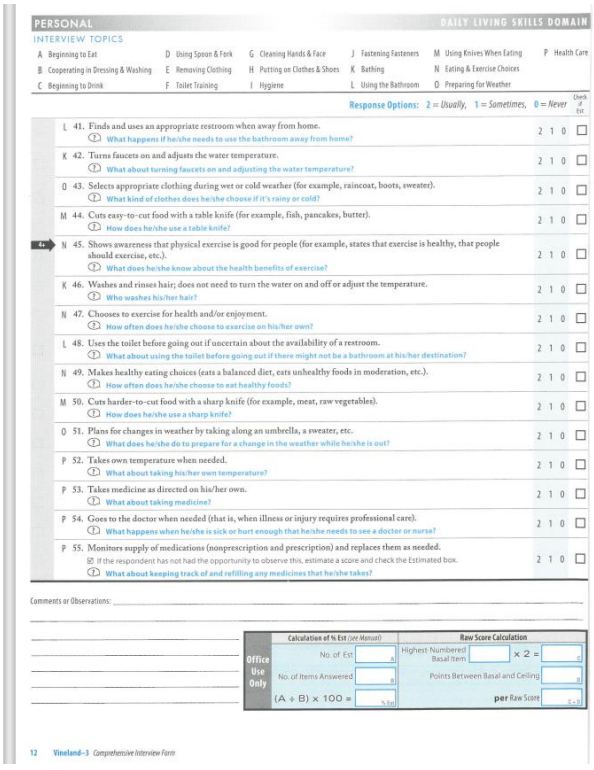


PERSONAL — DAILY LIVING SKILLS DOMAIN

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

				Check if Est
	H	19. Puts on shoes; may be on the wrong feet and does not need to tie or fasten. ⓘ Who puts on his/her shoes?	2 1 0	☐
	H	20. Puts on clothing that opens in the front (for example, a coat or jacket); does not need to zip or button. ⓘ What about a jacket or something else that opens in the front?	2 1 0	☐
	F	21. Defecates in a toilet or potty chair; parent or caregiver may initiate. ⓘ How is he/she using the toilet or a potty chair?	2 1 0	☐
	F	22. Is toilet-trained during the day; may require help with undressing, flushing, wiping, or washing hands, *but must initiate using the toilet.* ⓘ What happens during the day?	2 1 0	☐
	H	23. Puts on pullover garments (for example, T-shirt, sweatshirt, dress). ⓘ What about a T-shirt or something else that he/she has to put on over his/her head?	2 1 0	☐
	I	24. Wipes or blows nose using tissue, napkin, toilet paper, or other appropriate material. ⓘ What about wiping and blowing his/her nose?	2 1 0	☐
2	G	25. Washes and dries face; does not need to turn the water on and off or adjust the temperature. ⓘ What about washing his/her face?	2 1 0	☐
	G	26. Wipes or cleans face and hands as needed during or after meals. ⓘ What about wiping or cleaning his/her face and hands while eating and afterwards?	2 1 0	☐
	H	27. Puts clothing on with the right side forward and correct side out. ⓘ What about putting his/her clothes on with the right side forward and the correct side out?	2 1 0	☐
	F	28. Is toilet-trained during the night; may require help with undressing, flushing, wiping, or washing hands, *but must initiate using the toilet.* ⓘ What about accidents during the night?	2 1 0	☐
	I	29. Covers mouth and nose when coughing or sneezing. ⓘ What does he/she do when he/she coughs or sneezes?	2 1 0	☐
	F	30. Uses the toilet during the day and at night without help; must wipe, flush, and wash hands by himself/herself. ⓘ How much help does he/she need?	2 1 0	☐
	J	31. Fastens snaps. ⓘ What about snaps?	2 1 0	☐
	I	32. Brushes teeth; must put toothpaste on toothbrush, brush adequately, and rinse. ⓘ What about brushing his/her teeth?	2 1 0	☐
3	J	33. Buttons large buttons, in the correct buttonholes (for example, coat buttons). ⓘ How about big buttons, like on a coat?	2 1 0	☐
	I	34. Changes clothing that has become dirty, wet, muddy, or smelly. ⓘ What about changing clothes if they've gotten dirty, wet, muddy, or smelly?	2 1 0	☐
	J	35. Connects and zips zippers that are not already fastened at the bottom (for example, on a coat or jacket). ⓘ What about zippers that are not already fastened at the bottom, like on a coat or jacket?	2 1 0	☐
	K	36. Bathes or showers and dries self; does not need to turn the water on and off or adjust the temperature. ⓘ What about taking a bath or a shower?	2 1 0	☐
	H	37. Puts shoes on the correct feet and securely ties or fastens them. ⓘ What about tying or fastening his/her shoes?	2 1 0	☐
	M	38. Spreads food with a table knife (for example, butter, jam, mustard). ⓘ How does he/she use a table knife?	2 1 0	☐
	J	39. Buttons small buttons, in the correct buttonholes (for example, shirt buttons). ⓘ How about small buttons, like on a shirt?	2 1 0	☐
	N	40. Shows awareness that some foods are healthier than others (for example, states that fruits and vegetables are healthier than foods high in sugar or fat). ⓘ What does he/she know about healthy and unhealthy foods?	2 1 0	☐

Vineland-3 Comprehensive Interview Form 11



PERSONAL — DAILY LIVING SKILLS DOMAIN

INTERVIEW TOPICS

A Beginning to Eat	D Using Spoon & Fork	G Cleaning Hands & Face	J Fastening Fasteners	M Using Knives When Eating	P Health Care
B Cooperating in Dressing & Washing	E Removing Clothing	H Putting on Clothes & Shoes	K Bathing	N Eating & Exercise Choices	
C Beginning to Drink	F Toilet Training	I Hygiene	L Using the Bathroom	O Preparing for Weather	

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

				Check if Est
	L	41. Finds and uses an appropriate restroom when away from home. ⓘ What happens if he/she needs to use the bathroom away from home?	2 1 0	☐
	K	42. Turns faucets on and adjusts the water temperature. ⓘ What about turning faucets on and adjusting the water temperature?	2 1 0	☐
	O	43. Selects appropriate clothing during wet or cold weather (for example, raincoat, boots, sweater). ⓘ What kind of clothes does he/she choose if it's rainy or cold?	2 1 0	☐
	M	44. Cuts easy-to-cut food with a table knife (for example, fish, pancakes, butter). ⓘ How does he/she use a table knife?	2 1 0	☐
4a	N	45. Shows awareness that physical exercise is good for people (for example, states that exercise is healthy, that people should exercise, etc.). ⓘ What does he/she know about the health benefits of exercise?	2 1 0	☐
	K	46. Washes and rinses hair; does not need to turn the water on and off or adjust the temperature. ⓘ Who washes his/her hair?	2 1 0	☐
	N	47. Chooses to exercise for health and/or enjoyment. ⓘ How often does he/she choose to exercise on his/her own?	2 1 0	☐
	L	48. Uses the toilet before going out if uncertain about the availability of a restroom. ⓘ What about using the toilet before going out if there might not be a bathroom at his/her destination?	2 1 0	☐
	N	49. Makes healthy eating choices (eats a balanced diet, eats unhealthy foods in moderation, etc.). ⓘ How often does he/she choose to eat healthy foods?	2 1 0	☐
	M	50. Cuts harder-to-cut food with a sharp knife (for example, meat, raw vegetables). ⓘ How does he/she use a sharp knife?	2 1 0	☐
	O	51. Plans for changes in weather by taking along an umbrella, a sweater, etc. ⓘ What does he/she do to prepare for a change in the weather while he/she is out?	2 1 0	☐
	P	52. Takes own temperature when needed. ⓘ What about taking his/her own temperature?	2 1 0	☐
	P	53. Takes medicine as directed on his/her own. ⓘ What about taking medicine?	2 1 0	☐
	P	54. Goes to the doctor when needed (that is, when illness or injury requires professional care). ⓘ What happens when he/she is sick or hurt enough that he/she needs to see a doctor or nurse?	2 1 0	☐
	P	55. Monitors supply of medications (nonprescription and prescription) and replaces them as needed. ☑ If the respondent has not had the opportunity to observe this, estimate a score and check the Estimated box. ⓘ What about keeping track of and refilling any medicines that he/she takes?	2 1 0	☐

Comments or Observations: _____

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Calculation of % Est (see Manual)		Raw Score Calculation	
No. of Est	A	Highest-Numbered Basal Item	× 2 = C
No. of Items Answered	B	Points Between Basal and Ceiling	D
(A ÷ B) × 100 =	% Est	per Raw Score	C + D

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Vineland–3 Comprehensive Interview Form 13

DOMESTIC — DAILY LIVING SKILLS DOMAIN

INTERVIEW TOPICS

A Beginning Home Safety; B Cleaning Up After Self; C Putting Clothes, etc. Where They Belong; D Food Preparation Basics; E Advanced Home Safety; F Basic Household Chores; G Cleaning Up After Meals; H More Complex Food Preparation; I Advanced Household Chores

Response Options: 2 = *Usually*, 1 = *Sometimes*, 0 = *Never* — Check If Est

0-2	A	1. Is careful around hot objects (for example, the stove or oven, an open fire). What about safety concepts like being careful around hot objects?	2 1 0	☐
	A	2. Is careful when using sharp objects (for example, scissors, knives). What about sharp things like knives or scissors?	2 1 0	☐
	B	3. Wipes up own spills; must get wiping material and clean the spill adequately. What does he/she do if he/she spills something?	2 1 0	☐
	C	4. Puts dirty clothes in the proper place to be washed (for example, a laundry basket or chute). What about his/her dirty clothes?	2 1 0	☐
3-4	B	5. Removes dirty shoes or wipes them on a doormat before entering a residence. What does he/she do if his/her shoes are dirty when he/she is going into someone's house?	2 1 0	☐
	B	6. Puts away his/her books, toys, etc. when done using them. What about after he/she is done using things like toys, books, or magazines?	2 1 0	☐
	D	7. Washes hands before preparing food. What about washing his/her hands before preparing food?	2 1 0	☐
	B	8. Fully clears own dishes, utensils, napkins, cups, etc. after eating. What about clearing his/her own dishes and things after eating?	2 1 0	☐
	F	9. Does at least two simple household chores (for example, dusts, empties trash cans, feeds pet). What simple chores does he/she do?	2 1 0	☐
5-6	D	10. Prepares a simple snack or meal (for example, a sandwich, cheese and crackers, microwave foods). What about preparing a simple snack or meal like a sandwich, cheese and crackers, or microwave food?	2 1 0	☐
	E	11. Understands what to do in dangerous situations (for example, when to get help, when to call 911, how to distance self from danger). What does he/she understand about what to do in dangerous situations, for example, when to get help or call 911?	2 1 0	☐
	C	12. Hangs wet towel on a towel rack or hook, or puts in the proper place to be washed. What about his/her wet towels?	2 1 0	☐
	C	13. Puts clean clothes away where they belong (for example, in drawers or closet, on hooks). What about his/her clean clothes after they've been washed?	2 1 0	☐
	D	14. Uses at least two simple kitchen appliances (for example, toaster, microwave, electric can opener). What kitchen appliances does he/she use, things like a toaster, microwave, or electric can opener?	2 1 0	☐
	D	15. Washes fruits and vegetables before eating or cooking them. What about washing fruits and vegetables before eating or cooking them?	2 1 0	☐
	E	16. Secures home against intruders when home (keeps doors locked, finds out who is at the door before opening it, etc.). What does he/she do to keep intruders out when he/she is home, things like locking the doors and asking who's at the door before opening it?	2 1 0	☐
	D	17. Uses at least three kitchen utensils to prepare food (for example, knives, tongs, spatula, vegetable peeler). What kitchen utensils does he/she use, things like knives, spatulas, and vegetable peelers?	2 1 0	☐
7-8	E	18. Is careful when operating household appliances or equipment (for example, vacuum cleaner, lawnmower, iron, power tools). How careful is he/she when using household appliances or equipment like a vacuum cleaner, lawnmower, iron, or power tools?	2 1 0	☐
	D	19. Prepares and eats leftovers. What about preparing leftovers?	2 1 0	☐

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DOMESTIC — DAILY LIVING SKILLS DOMAIN

Response Options: 2 = *Usually*, 1 = *Sometimes*, 0 = *Never* — Check If Est

	F	20. Uses household products correctly (for example, laundry detergent, furniture polish, glass cleaner). What about using household products like cleaners correctly?	2 1 0	☐
	G	21. Puts dishes away when clean and dry. What about putting away clean dishes when he/she is supposed to?	2 1 0	☐
	E	22. Secures home against intruders when leaving home (locks doors, closes windows, turns on alarm, etc.). What does he/she do to keep intruders out when he/she leaves home?	2 1 0	☐
	G	23. Washes dishes (either by hand, or by loading and running the dishwasher when needed). What about washing dishes when he/she is supposed to?	2 1 0	☐
	F	24. Cleans floors thoroughly (sweeps, vacuums, mops, etc.). What about cleaning floors when he/she is supposed to?	2 1 0	☐
7-8	G	25. Puts leftover food away (for example, in plastic bags or wrap, in containers). What about putting away leftover food when he/she is supposed to?	2 1 0	☐
	H	26. Uses the stove or oven for cooking or baking (turns burners on and off, sets oven temperature, etc.). What about using the stove or oven?	2 1 0	☐
	I	27. Recognizes when simple maintenance tasks need to be done and does them (for example, replaces light bulbs, batteries, filters, or vacuum cleaner bag). What about noticing when things like light bulbs or batteries need changing and doing it on his/her own?	2 1 0	☐
	I	28. Cleans bathroom (toilet, sink, tub or shower, etc.). What about cleaning a bathroom when he/she is supposed to?	2 1 0	☐
	I	29. Does laundry; must wash, dry, and fold/hang. What about doing laundry when he/she is supposed to?	2 1 0	☐
	H	30. Prepares a full meal consisting of three or more food items. What's an example of a big meal that he/she prepares by himself/herself?	2 1 0	☐

mments or Observations: _____

Office Use Only

Calculation of % Est (see Manual)		Raw Score Calculation	
No. of Est	A	Highest-Numbered Basal Item × 2 =	C
No. of Items Answered	B	Points Between Basal and Ceiling	D
(A ÷ B) × 100 =	% Est	dom Raw Score	C + D

Vineland–3 Comprehensive Interview Form 15

COMMUNITY — DAILY LIVING SKILLS DOMAIN

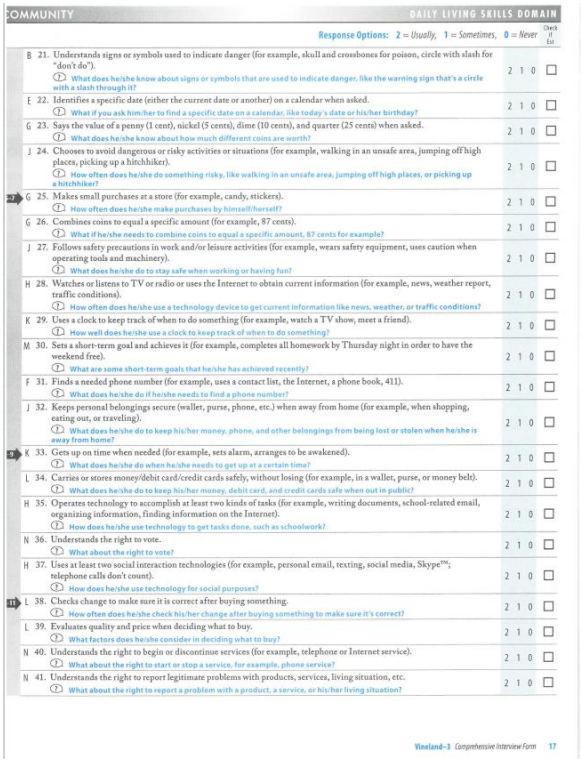
INTERVIEW TOPICS

A Basic Money Skills; B Basic Safety Outside the Home; C Telling Time; D Basic Rights & Responsibilities; E Calendar Basics; F Telephone Use; G Using Money; H Using Technology; I Eating Out; J Advanced Safety Outside the Home; K Being On Time; L Shopping Skills; M Achieving Goals; N Understanding Rights; O Traveling; P Earning Money; Q Managing Money

Response Options: 2 = *Usually*, 1 = *Sometimes*, 0 = *Never* — Check If Est

	F	1. Talks to a familiar person using a phone, computer, or other electronic device; does not need to place the call. How often does he/she talk with someone he/she knows?	2 1 0	☐
0-2	A	2. Understands that money is used to buy things. How can you tell that he/she understands what money is used for?	2 1 0	☐
	A	3. Counts at least 10 objects, one by one. To use money, you have to be able to count things. How many things like blocks or other objects does he/she count correctly?	2 1 0	☐
	B	4. Remains within safe distance of caregiver when in public places; being carried, pushed in a stroller, etc. doesn't count. ☑ Score 2 if the individual did this when younger, but has now outgrown it. What about staying within a safe distance from you when you are out in public?	2 1 0	☐
	B	5. Understands and follows safety precautions while riding in a car (for example, keeps seat belt on, refrains from unnecessarily distracting the driver). What about staying safe in the car, for example, keeping his/her seatbelt on and not distracting the driver?	2 1 0	☐
	C	6. Understands that a clock is used to tell time. How can you tell if he/she knows what a clock is used for?	2 1 0	☐
	H	7. Operates at least two technology devices for entertainment (for example, a television, DVD player, music player, handheld game, computer used for entertainment). What electronic devices does he/she use for entertainment?	2 1 0	☐
	I	8. Uses appropriate manners when eating in public (for example, uses utensils, sits properly, doesn't disrupt others). How are his/her manners when eating out?	2 1 0	☐
	E	9. Says all seven days of the week in order when asked. If you ask him/her to say the days of the week in order, how often does he/she get it right?	2 1 0	☐
3-4	B	10. Looks both ways when crossing streets or roads. What does he/she do in terms of safety when crossing streets or roads?	2 1 0	☐
	D	11. Respects the right to personal privacy for self and others (for example, while using the restroom or changing clothes; not opening others' mail). What about people's right to privacy?	2 1 0	☐
	A	12. Names a penny, nickel, dime, and quarter when asked; does not need to know the value of the coins. What coins does he/she identify?	2 1 0	☐
5	E	13. Says the current day of the week when asked. What if you ask him/her "What day of the week is it today?"	2 1 0	☐
	A	14. Understands that some items cost more than others (for example, might say "I have enough money to buy stickers but not a book" or "Which pens cost less?"). What does he/she understand about some things costing more than others?	2 1 0	☐
	F	15. Knows what phone number to call in an emergency and how to make the call (for example, knows how to call 911 or an emergency contact). What does he/she know about making a call in an emergency?	2 1 0	☐
	B	16. Obeys traffic lights and *Walk/Don't Walk* signs when crossing streets or roads. What about crossing when there's a light or a *Walk/Don't Walk* sign?	2 1 0	☐
	C	17. Tells time using a digital clock or watch. What kinds of clocks or watches can he/she tell time on?	2 1 0	☐
	D	18. Understands and follows community rules and laws (for example, rules and laws regarding littering, pet control, respecting others' property, etc.). What about community rules and laws, like those regarding littering, pet control, and respecting others' property?	2 1 0	☐
	F	19. Makes calls to others using a phone, computer, or other electronic device. How often does he/she call someone on his/her own?	2 1 0	☐
	G	20. Knows the difference between bills of different values ($1, $5, $10 bills, etc.). What does he/she know about the values of different bills, for example, the difference between a one-dollar bill and a ten-dollar bill?	2 1 0	☐

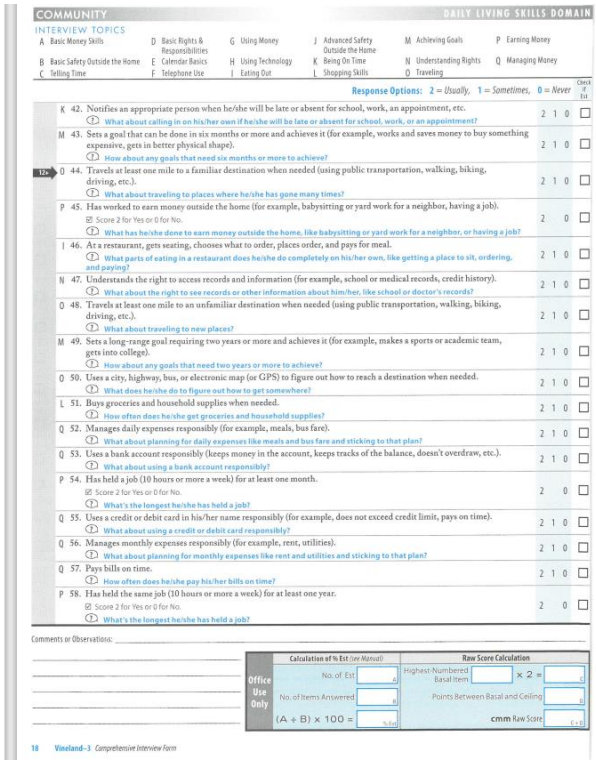
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COMMUNITY — DAILY LIVING SKILLS DOMAIN

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

					Check if Est
	B	21.	Understands signs or symbols used to indicate danger (for example, skull and crossbones for poison, circle with slash for "don't do"). ⓘ What does he/she know about signs or symbols that are used to indicate danger, like the warning sign that's a circle with a slash through it?	2 1 0	☐
	E	22.	Identifies a specific date (either the current date or another) on a calendar when asked. ⓘ What if you ask him/her to find a specific date on a calendar, like today's date or his/her birthday?	2 1 0	☐
	G	23.	Says the value of a penny (1 cent), nickel (5 cents), dime (10 cents), and quarter (25 cents) when asked. ⓘ What does he/she know about how much different coins are worth?	2 1 0	☐
	J	24.	Chooses to avoid dangerous or risky activities or situations (for example, walking in an unsafe area, jumping off high places, picking up a hitchhiker). ⓘ How often does he/she do something risky, like walking in an unsafe area, jumping off high places, or picking up a hitchhiker?	2 1 0	☐
27	G	25.	Makes small purchases at a store (for example, candy, stickers). ⓘ How often does he/she make purchases by himself/herself?	2 1 0	☐
	G	26.	Combines coins to equal a specific amount (for example, 87 cents). ⓘ What if he/she needs to combine coins to equal a specific amount, 87 cents for example?	2 1 0	☐
	J	27.	Follows safety precautions in work and/or leisure activities (for example, wears safety equipment, uses caution when operating tools and machinery). ⓘ What does he/she do to stay safe when working or having fun?	2 1 0	☐
	H	28.	Watches or listens to TV or radio or uses the Internet to obtain current information (for example, news, weather report, traffic conditions). ⓘ How often does he/she use a technology device to get current information like news, weather, or traffic conditions?	2 1 0	☐
	K	29.	Uses a clock to keep track of when to do something (for example, watch a TV show, meet a friend). ⓘ How well does he/she use a clock to keep track of when to do something?	2 1 0	☐
	M	30.	Sets a short-term goal and achieves it (for example, completes all homework by Thursday night in order to have the weekend free). ⓘ What are some short-term goals that he/she has achieved recently?	2 1 0	☐
	F	31.	Finds a needed phone number (for example, uses a contact list, the Internet, a phone book, 411). ⓘ What does he/she do if he/she needs to find a phone number?	2 1 0	☐
	J	32.	Keeps personal belongings secure (wallet, purse, phone, etc.) when away from home (for example, when shopping, eating out, or traveling). ⓘ What does he/she do to keep his/her money, phone, and other belongings from being lost or stolen when he/she is away from home?	2 1 0	☐
9	K	33.	Gets up on time when needed (for example, sets alarm, arranges to be awakened). ⓘ What does he/she do when he/she needs to get up at a certain time?	2 1 0	☐
	L	34.	Carries or stores money/debit card/credit cards safely, without losing (for example, in a wallet, purse, or money belt). ⓘ What does he/she do to keep his/her money, debit card, and credit cards safe when out in public?	2 1 0	☐
	H	35.	Operates technology to accomplish at least two kinds of tasks (for example, writing documents, school-related email, organizing information, finding information on the Internet). ⓘ How does he/she use technology to get tasks done, such as schoolwork?	2 1 0	☐
	N	36.	Understands the right to vote. ⓘ What about the right to vote?	2 1 0	☐
	H	37.	Uses at least two social interaction technologies (for example, personal email, texting, social media, Skype™; telephone calls don't count). ⓘ How does he/she use technology for social purposes?	2 1 0	☐
11	L	38.	Checks change to make sure it is correct after buying something. ⓘ How often does he/she check his/her change after buying something to make sure it's correct?	2 1 0	☐
	L	39.	Evaluates quality and price when deciding what to buy. ⓘ What factors does he/she consider in deciding what to buy?	2 1 0	☐
	N	40.	Understands the right to begin or discontinue services (for example, telephone or Internet service). ⓘ What about the right to start or stop a service, for example, phone service?	2 1 0	☐
	N	41.	Understands the right to report legitimate problems with products, services, living situation, etc. ⓘ What about the right to report a problem with a product, a service, or his/her living situation?	2 1 0	☐



COMMUNITY — DAILY LIVING SKILLS DOMAIN

INTERVIEW TOPICS

A Basic Money Skills	D Basic Rights & Responsibilities	G Using Money	J Advanced Safety Outside the Home	M Achieving Goals	P Earning Money
B Basic Safety Outside the Home	E Calendar Basics	H Using Technology	K Being On Time	N Understanding Rights	Q Managing Money
C Telling Time	F Telephone Use	I Eating Out	L Shopping Skills	O Traveling	

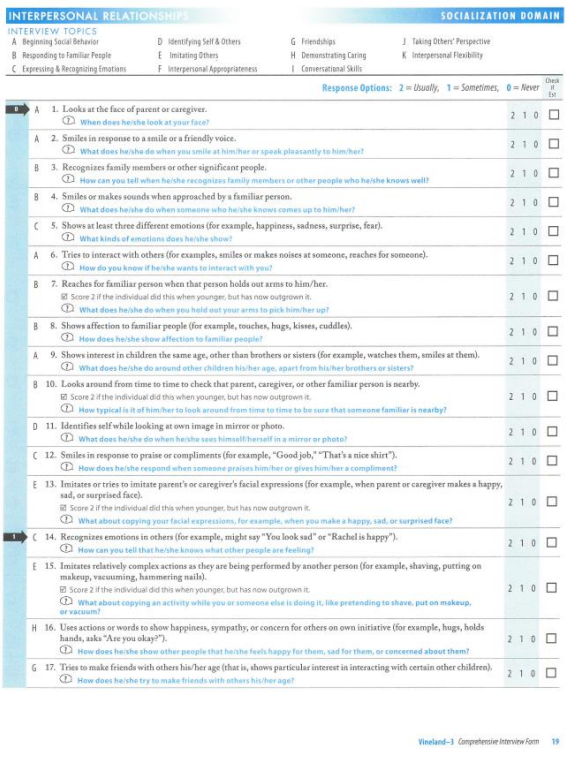
Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

					Check if Est
	K	42.	Notifies an appropriate person when he/she will be late or absent for school, work, or an appointment, etc. ⓘ What about calling in on his/her own if he/she will be late or absent for school, work, or an appointment?	2 1 0	☐
	M	43.	Sets a goal that can be done in six months or more and achieves it (for example, works and saves money to buy something expensive, gets in better physical shape). ⓘ How about any goals that need six months or more to achieve?	2 1 0	☐
12+	O	44.	Travels at least one mile to a familiar destination when needed (using public transportation, walking, biking, driving, etc.). ⓘ What about traveling to places where he/she has gone many times?	2 1 0	☐
	P	45.	Has worked to earn money outside the home (for example, babysitting or yard work for a neighbor, having a job). ☑ Score 2 for Yes or 0 for No. ⓘ What has he/she done to earn money outside the home, like babysitting or yard work for a neighbor, or having a job?	2 0	☐
	I	46.	At a restaurant, gets seating, chooses what to order, places order, and pays for meal. ⓘ What parts of eating in a restaurant does he/she do completely on his/her own, like getting a place to sit, ordering, and paying?	2 1 0	☐
	N	47.	Understands the right to access records and information (for example, school or medical records, credit history). ⓘ What about the right to see records or other information about him/her, like school or doctor's records?	2 1 0	☐
	O	48.	Travels at least one mile to an unfamiliar destination when needed (using public transportation, walking, biking, driving, etc.). ⓘ What about traveling to new places?	2 1 0	☐
	M	49.	Sets a long-range goal requiring two years or more and achieves it (for example, makes a sports or academic team, gets into college). ⓘ How about any goals that need two years or more to achieve?	2 1 0	☐
	Q	50.	Uses a city, highway, bus, or electronic map (or GPS) to figure out how to reach a destination when needed. ⓘ What does he/she do to figure out how to get somewhere?	2 1 0	☐
	L	51.	Buys groceries and household supplies when needed. ⓘ How often does he/she get groceries and household supplies?	2 1 0	☐
	Q	52.	Manages daily expenses responsibly (for example, meals, bus fare). ⓘ What about planning for daily expenses like meals and bus fare and sticking to that plan?	2 1 0	☐
	Q	53.	Uses a bank account responsibly (keeps money in the account, keeps tracks of the balance, doesn't overdraw, etc.). ⓘ What about using a bank account responsibly?	2 1 0	☐
	P	54.	Has held a job (10 hours or more a week) for at least one month. ☑ Score 2 for Yes or 0 for No. ⓘ What's the longest he/she has held a job?	2 0	☐
	Q	55.	Uses a credit or debit card in his/her name responsibly (for example, does not exceed credit limit, pays on time). ⓘ What about using a credit or debit card responsibly?	2 1 0	☐
	Q	56.	Manages monthly expenses responsibly (for example, rent, utilities). ⓘ What about planning for monthly expenses like rent and utilities and sticking to that plan?	2 1 0	☐
	Q	57.	Pays bills on time. ⓘ How often does he/she pay his/her bills on time?	2 1 0	☐
	P	58.	Has held the same job (10 hours or more a week) for at least one year. ☑ Score 2 for Yes or 0 for No. ⓘ What's the longest he/she has held a job?	2 0	☐

Comments or Observations: _____

Office Use Only

Calculation of % Est (see Manual)		Raw Score Calculation	
No. of Est	A	Highest-Numbered Basal Item	× 2 = C
No. of Items Answered	B	Points Between Basal and Ceiling	D
(A + B) × 100 =	%Est	cmm Raw Score	Est



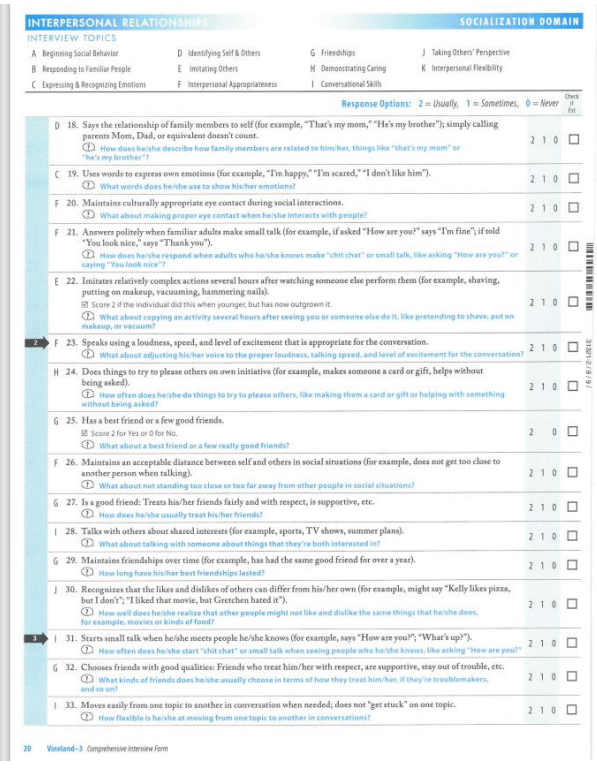
INTERPERSONAL RELATIONSHIPS — SOCIALIZATION DOMAIN

INTERVIEW TOPICS

A Beginning Social Behavior	D Identifying Self & Others	G Friendships	J Taking Others' Perspective		
B Responding to Familiar People	E Imitating Others	H Demonstrating Caring	K Interpersonal Flexibility		
C Expressing & Recognizing Emotions	F Interpersonal Appropriateness	I Conversational Skills			

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

					Check if Est
0	A	1.	Looks at the face of parent or caregiver. ⓘ When does he/she look at your face?	2 1 0	☐
	A	2.	Smiles in response to a smile or a friendly voice. ⓘ What does he/she do when you smile at him/her or speak pleasantly to him/her?	2 1 0	☐
	B	3.	Recognizes family members or other significant people. ⓘ How can you tell when he/she recognizes family members or other people who he/she knows well?	2 1 0	☐
	B	4.	Smiles or makes sounds when approached by a familiar person. ⓘ What does he/she do when someone who he/she knows comes up to him/her?	2 1 0	☐
	C	5.	Shows at least three different emotions (for example, happiness, sadness, surprise, fear). ⓘ What kinds of emotions does he/she show?	2 1 0	☐
	A	6.	Tries to interact with others (for examples, smiles or makes noises at someone, reaches for someone). ⓘ How do you know if he/she wants to interact with you?	2 1 0	☐
	B	7.	Reaches for familiar person when that person holds out arms to him/her. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ What does he/she do when you hold out your arms to pick him/her up?	2 1 0	☐
	B	8.	Shows affection to familiar people (for example, touches, hugs, kisses, cuddles). ⓘ How does he/she show affection to familiar people?	2 1 0	☐
	A	9.	Shows interest in children the same age, other than brothers or sisters (for example, watches them, smiles at them). ⓘ What does he/she do around other children his/her age, apart from his/her brothers or sisters?	2 1 0	☐
	B	10.	Looks around from time to time to check that parent, caregiver, or other familiar person is nearby. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ How typical is it of him/her to look around from time to time to be sure that someone familiar is nearby?	2 1 0	☐
	D	11.	Identifies self while looking at own image in mirror or photo. ⓘ What does he/she do when he/she sees himself/herself in a mirror or photo?	2 1 0	☐
	C	12.	Smiles in response to praise or compliments (for example, "Good job," "That's a nice shirt"). ⓘ How does he/she respond when someone praises him/her or gives him/her a compliment?	2 1 0	☐
	E	13.	Imitates or tries to imitate parent's or caregiver's facial expressions (for example, when parent or caregiver makes a happy, sad, or surprised face). ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ What about copying your facial expressions, for example, when you make a happy, sad, or surprised face?	2 1 0	☐
1	C	14.	Recognizes emotions in others (for example, might say "You look sad" or "Rachel is happy"). ⓘ How can you tell that he/she knows what other people are feeling?	2 1 0	☐
	E	15.	Imitates relatively complex actions as they are being performed by another person (for example, shaving, putting on makeup, vacuuming, hammering nails). ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ What about copying an activity while you or someone else is doing it, like pretending to shave, put on makeup, or vacuum?	2 1 0	☐
	H	16.	Uses actions or words to show happiness, sympathy, or concern for others on own initiative (for example, hugs, holds hands, asks "Are you okay?"). ⓘ How does he/she show other people that he/she feels happy for them, sad for them, or concerned about them?	2 1 0	☐
	G	17.	Tries to make friends with others his/her age (that is, shows particular interest in interacting with certain other children). ⓘ How does he/she try to make friends with others his/her age?	2 1 0	☐



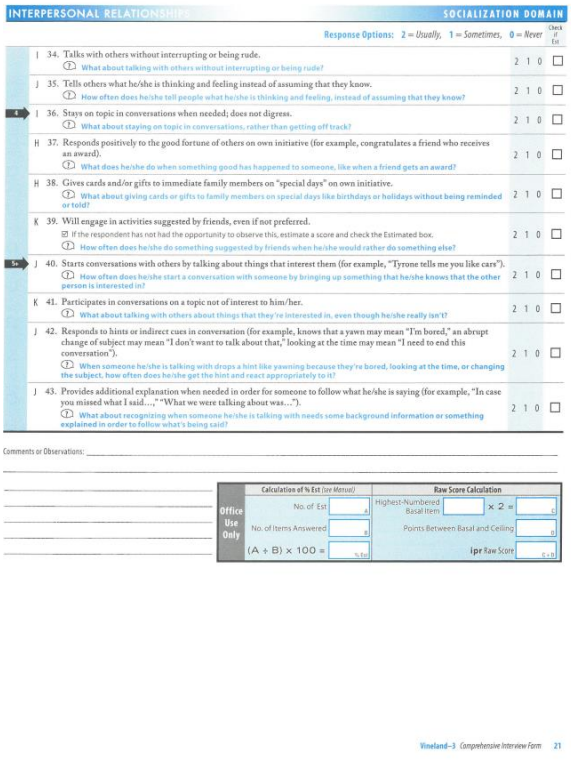
INTERPERSONAL RELATIONSHIPS — SOCIALIZATION DOMAIN

INTERVIEW TOPICS

A Beginning Social Behavior	D Identifying Self & Others	G Friendships	J Taking Others' Perspective		
B Responding to Familiar People	E Imitating Others	H Demonstrating Caring	K Interpersonal Flexibility		
C Expressing & Recognizing Emotions	F Interpersonal Appropriateness	I Conversational Skills			

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

					Check if Est
	D	18.	Says the relationship of family members to self (for example, "That's my mom," "He's my brother"); simply calling parents Mom, Dad, or equivalent doesn't count. ⓘ How does he/she describe how family members are related to him/her, things like "that's my mom" or "he's my brother"?	2 1 0	☐
	C	19.	Uses words to express own emotions (for example, "I'm happy," "I'm scared," "I don't like him"). ⓘ What words does he/she use to show his/her emotions?	2 1 0	☐
	F	20.	Maintains culturally appropriate eye contact during social interactions. ⓘ What about making proper eye contact when he/she interacts with people?	2 1 0	☐
	F	21.	Answers politely when familiar adults make small talk (for example, if asked "How are you?" says "I'm fine"; if told "You look nice," says "Thank you"). ⓘ How does he/she respond when adults who he/she knows make "chit chat" or small talk, like asking "How are you?" or saying "You look nice"?	2 1 0	☐
	E	22.	Imitates relatively complex actions several hours after watching someone else perform them (for example, shaving, putting on makeup, vacuuming, hammering nails). ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ What about copying an activity several hours after seeing you or someone else do it, like pretending to shave, put on makeup, or vacuum?	2 1 0	☐
2	F	23.	Speaks using a loudness, speed, and level of excitement that is appropriate for the conversation. ⓘ What about adjusting his/her voice to the proper loudness, talking speed, and level of excitement for the conversation?	2 1 0	☐
	H	24.	Does things to try to please others on own initiative (for example, makes someone a card or gift, helps without being asked). ⓘ How often does he/she do things to try to please others, like making them a card or gift or helping with something without being asked?	2 1 0	☐
	G	25.	Has a best friend or a few good friends. ☑ Score 2 for Yes or 0 for No. ⓘ What about a best friend or a few really good friends?	2 0	☐
	F	26.	Maintains an acceptable distance between self and others in social situations (for example, does not get too close to another person when talking). ⓘ What about not standing too close or too far away from other people in social situations?	2 1 0	☐
	G	27.	Is a good friend: Treats his/her friends fairly and with respect, is supportive, etc. ⓘ How does he/she usually treat his/her friends?	2 1 0	☐
	I	28.	Talks with others about shared interests (for example, sports, TV shows, summer plans). ⓘ What about talking with someone about things that they're both interested in?	2 1 0	☐
	G	29.	Maintains friendships over time (for example, has had the same good friend for over a year). ⓘ How long have his/her best friendships lasted?	2 1 0	☐
	J	30.	Recognizes that the likes and dislikes of others can differ from his/her own (for example, might say "Kelly likes pizza, but I don't"; "I liked that movie, but Gretchen hated it"). ⓘ How well does he/she realize that other people might not like and dislike the same things that he/she does, for example, movies or kinds of food?	2 1 0	☐
3	I	31.	Starts small talk when he/she meets people he/she knows (for example, says "How are you?"; "What's up?"). ⓘ How often does he/she start "chit chat" or small talk when seeing people who he/she knows, like asking "How are you?"	2 1 0	☐
	G	32.	Chooses friends with good qualities: Friends who treat him/her with respect, are supportive, stay out of trouble, etc. ⓘ What kinds of friends does he/she usually choose in terms of how they treat him/her, if they're troublemakers, and so on?	2 1 0	☐
	I	33.	Moves easily from one topic to another in conversation when needed; does not "get stuck" on one topic. ⓘ How flexible is he/she at moving from one topic to another in conversations?	2 1 0	☐



INTERPERSONAL RELATIONSHIPS — SOCIALIZATION DOMAIN

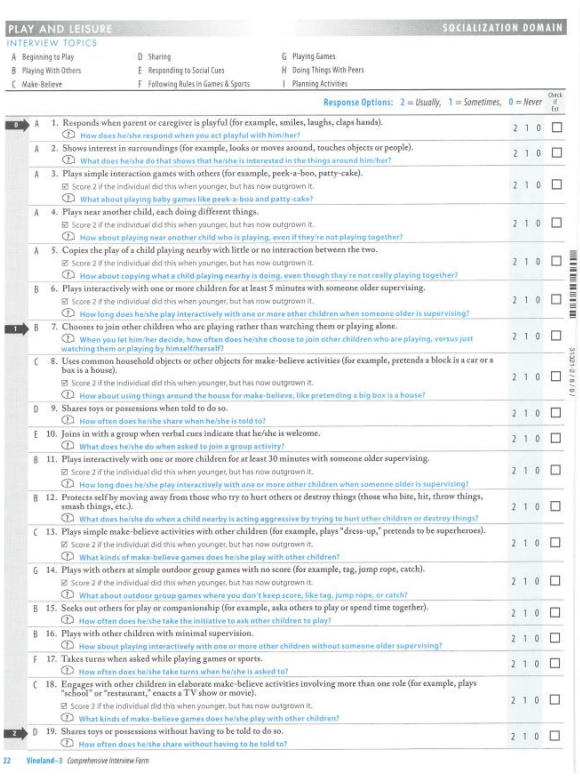
Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

I 34. Talks with others without interrupting or being rude. 2 1 0
J 35. Tells others what he/she is thinking and feeling instead of assuming that they know. 2 1 0
4 I 36. Stays on topic in conversations when needed; does not digress. 2 1 0
H 37. Responds positively to the good fortune of others on own initiative (for example, congratulates a friend who receives an award). 2 1 0
H 38. Gives cards and/or gifts to immediate family members on "special days" on own initiative. 2 1 0
K 39. Will engage in activities suggested by friends, even if not preferred. 2 1 0
5m J 40. Starts conversations with others by talking about things that interest them (for example, "Tyrone tells me you like cars"). 2 1 0
K 41. Participates in conversations on a topic not of interest to him/her. 2 1 0
J 42. Responds to hints or indirect cues in conversation (for example, knows that a yawn may mean "I'm bored," an abrupt change of subject may mean "I don't want to talk about that," looking at the time may mean "I need to end this conversation"). 2 1 0
J 43. Provides additional explanation when needed in order for someone to follow what he/she is saying (for example, "In case you missed what I said…," "What we were talking about was…"). 2 1 0

Comments or Observations:

Office Use Only — Calculation of % Est (see Manual): No. of Est (A); No. of Items Answered (B); (A ÷ B) × 100 = (% Est). Raw Score Calculation: Highest-Numbered Basal Item × 2 = (C); Points Between Basal and Ceiling (D); ipr Raw Score (C + D)

Vineland-3 Comprehensive Interview Form 21



PLAY AND LEISURE — SOCIALIZATION DOMAIN

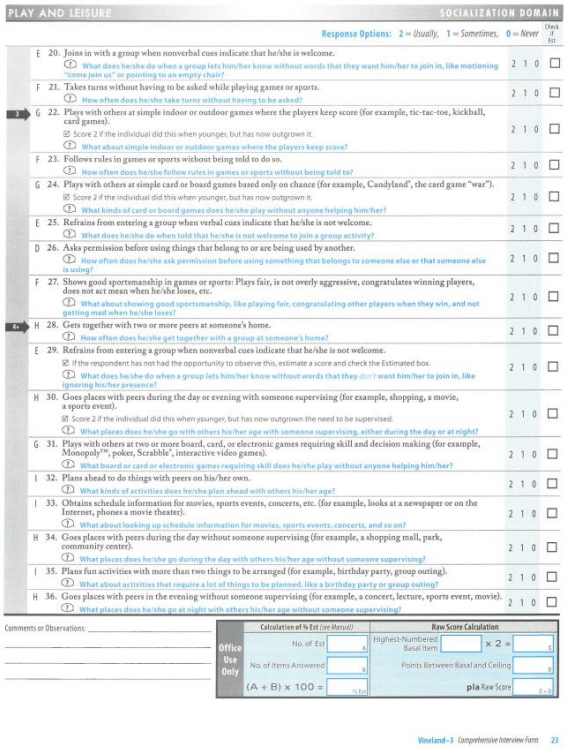
INTERVIEW TOPICS

A Beginning to Play; B Playing With Others; C Make-Believe; D Sharing; E Responding to Social Cues; F Following Rules in Games & Sports; G Playing Games; H Doing Things With Peers; I Planning Activities

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

6 A 1. Responds when parent or caregiver is playful (for example, smiles, laughs, claps hands). 2 1 0
A 2. Shows interest in surroundings (for example, looks or moves around, touches objects or people). 2 1 0
A 3. Plays simple interaction games with others (for example, peek-a-boo, patty-cake). 2 1 0
A 4. Plays near another child, each doing different things. 2 1 0
A 5. Copies the play of a child playing nearby with little or no interaction between the two. 2 1 0
B 6. Plays interactively with one or more children for at least 5 minutes with someone older supervising. 2 1 0
1 B 7. Chooses to join other children who are playing rather than watching them or playing alone. 2 1 0
C 8. Uses common household objects or other objects for make-believe activities (for example, pretends a block is a car or a box is a house). 2 1 0
D 9. Shares toys or possessions when told to do so. 2 1 0
E 10. Joins in with a group when verbal cues indicate that he/she is welcome. 2 1 0
B 11. Plays interactively with one or more children for at least 30 minutes with someone older supervising. 2 1 0
B 12. Protects self by moving away from those who try to hurt others or destroy things (those who bite, hit, throw things, smash things, etc.). 2 1 0
C 13. Plays simple make-believe activities with other children (for example, plays "dress-up," pretends to be superheroes). 2 1 0
G 14. Plays with others at simple outdoor group games with no score (for example, tag, jump rope, catch). 2 1 0
B 15. Seeks out others for play or companionship (for example, asks others to play or spend time together). 2 1 0
B 16. Plays with other children with minimal supervision. 2 1 0
F 17. Takes turns when asked while playing games or sports. 2 1 0
C 18. Engages with other children in elaborate make-believe activities involving more than one role (for example, plays "school" or "restaurant," enacts a TV show or movie). 2 1 0
2 D 19. Shares toys or possessions without having to be told to do so. 2 1 0

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PLAY AND LEISURE — SOCIALIZATION DOMAIN

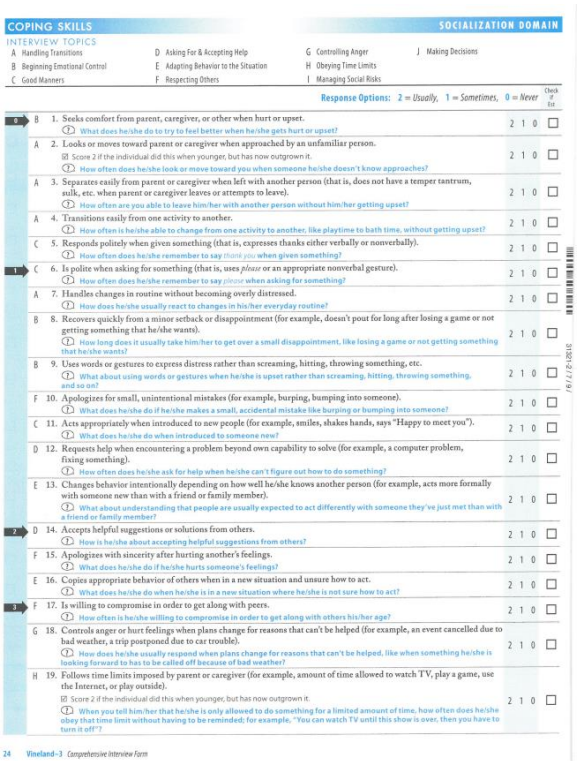
Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

E 20. Joins in with a group when nonverbal cues indicate that he/she is welcome. 2 1 0
F 21. Takes turns without having to be asked while playing games or sports. 2 1 0
3 G 22. Plays with others at simple indoor or outdoor games where the players keep score (for example, tic-tac-toe, kickball, card games). 2 1 0
F 23. Follows rules in games or sports without being told to do so. 2 1 0
G 24. Plays with others at simple card or board games based only on chance (for example, Candyland™, the card game "war"). 2 1 0
E 25. Refrains from entering a group when verbal cues indicate that he/she is not welcome. 2 1 0
D 26. Asks permission before using things that belong to or are being used by another. 2 1 0
F 27. Shows good sportsmanship in games or sports: Plays fair, is not overly aggressive, congratulates winning players, does not act mean when he/she loses, etc. 2 1 0
6m H 28. Gets together with two or more peers at someone's home. 2 1 0
E 29. Refrains from entering a group when nonverbal cues indicate that he/she is not welcome. 2 1 0
H 30. Goes places with peers during the day or evening with someone supervising (for example, shopping, a movie, a sports event). 2 1 0
G 31. Plays with others at two or more board, card, or electronic games requiring skill and decision making (for example, Monopoly™, poker, Scrabble™, interactive video games). 2 1 0
I 32. Plans ahead to do things with peers on his/her own. 2 1 0
I 33. Obtains schedule information for movies, sports events, concerts, etc. (for example, looks at a newspaper or on the Internet, phones a movie theater). 2 1 0
H 34. Goes places with peers during the day without someone supervising (for example, a shopping mall, park, community center). 2 1 0
I 35. Plans fun activities with more than two things to be arranged (for example, birthday party, group outing). 2 1 0
H 36. Goes places with peers in the evening without someone supervising (for example, a concert, lecture, sports event, movie). 2 1 0

Comments or Observations:

Office Use Only — Calculation of % Est (see Manual): No. of Est (A); No. of Items Answered (B); (A ÷ B) × 100 = (% Est). Raw Score Calculation: Highest-Numbered Basal Item × 2 = (C); Points Between Basal and Ceiling (D); pla Raw Score (C + D)

Vineland-3 Comprehensive Interview Form 23



COPING SKILLS — SOCIALIZATION DOMAIN

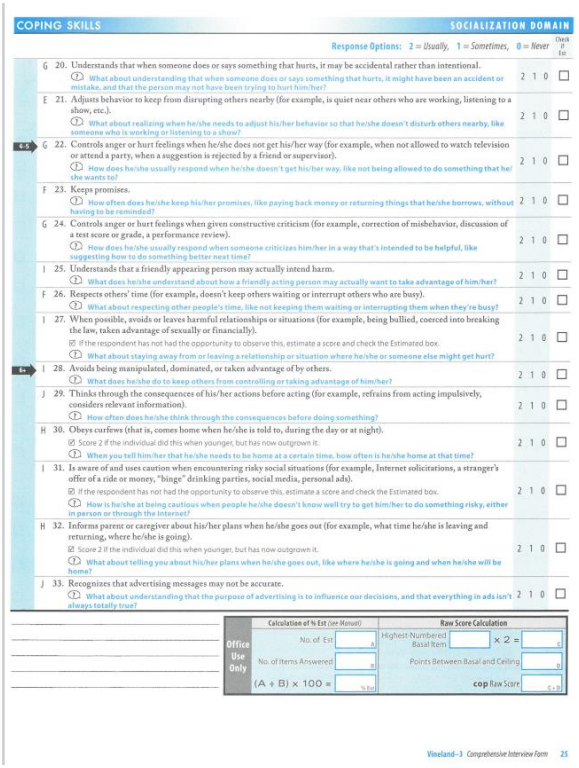
INTERVIEW TOPICS

A Handling Transitions; B Beginning Emotional Control; C Good Manners; D Asking For & Accepting Help; E Adapting Behavior to the Situation; F Respecting Others; G Controlling Anger; H Obeying Time Limits; I Managing Social Risks; J Making Decisions

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

0 B 1. Seeks comfort from parent, caregiver, or other when hurt or upset. 2 1 0
A 2. Looks or moves toward parent or caregiver when approached by an unfamiliar person. 2 1 0
A 3. Separates easily from parent or caregiver when left with another person (that is, does not have a temper tantrum, sulk, etc. when parent or caregiver leaves or attempts to leave). 2 1 0
A 4. Transitions easily from one activity to another. 2 1 0
C 5. Responds politely when given something (that is, expresses thanks either verbally or nonverbally). 2 1 0
1 C 6. Is polite when asking for something (that is, uses *please* or an appropriate nonverbal gesture). 2 1 0
A 7. Handles changes in routine without becoming overly distressed. 2 1 0
B 8. Recovers quickly from a minor setback or disappointment (for example, doesn't pout for long after losing a game or not getting something that he/she wants). 2 1 0
B 9. Uses words or gestures to express distress rather than screaming, hitting, throwing something, etc. 2 1 0
F 10. Apologizes for small, unintentional mistakes (for example, burping, bumping into someone). 2 1 0
C 11. Acts appropriately when introduced to new people (for example, smiles, shakes hands, says "Happy to meet you"). 2 1 0
D 12. Requests help when encountering a problem beyond own capability to solve (for example, a computer problem, fixing something). 2 1 0
E 13. Changes behavior intentionally depending on how well he/she knows another person (for example, acts more formally with someone new than with a friend or family member). 2 1 0
2 D 14. Accepts helpful suggestions or solutions from others. 2 1 0
F 15. Apologizes with sincerity after hurting another's feelings. 2 1 0
E 16. Copies appropriate behavior of others when in a new situation and unsure how to act. 2 1 0
3 F 17. Is willing to compromise in order to get along with peers. 2 1 0
G 18. Controls anger or hurt feelings when plans change for reasons that can't be helped (for example, an event cancelled due to bad weather, a trip postponed due to car trouble). 2 1 0
H 19. Follows time limits imposed by parent or caregiver (for example, amount of time allowed to watch TV, play a game, use the Internet, or play outside). 2 1 0

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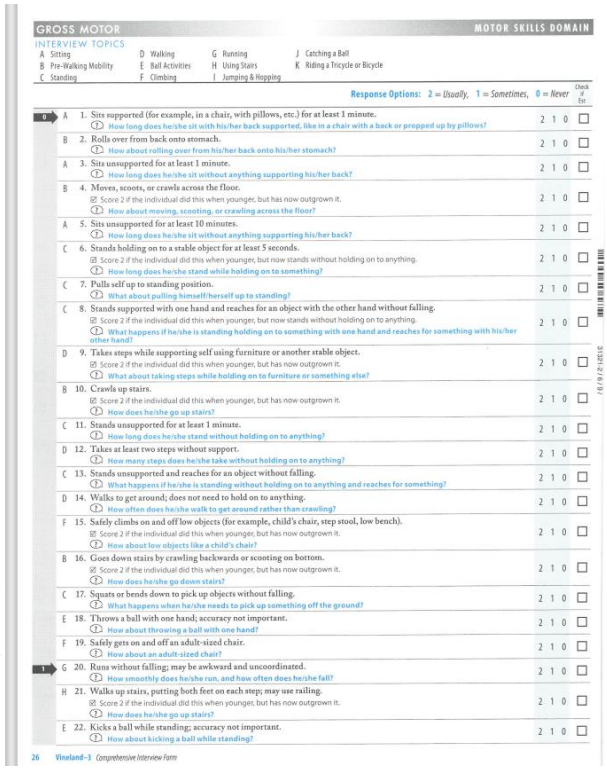
COPING SKILLS — SOCIALIZATION DOMAIN

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

					Check if Est
	G	20.	Understands that when someone does or says something that hurts, it may be accidental rather than intentional. ⓘ What about understanding that when someone does or says something that hurts, it might have been an accident or mistake, and that the person may not have been trying to hurt him/her?	2 1 0	☐
	E	21.	Adjusts behavior to keep from disrupting others nearby (for example, is quiet near others who are working, listening to a show, etc.). ⓘ What about realizing when he/she needs to adjust his/her behavior so that he/she doesn't disturb others nearby, like someone who is working or listening to a show?	2 1 0	☐
4–5	G	22.	Controls anger or hurt feelings when he/she does not get his/her way (for example, when not allowed to watch television or attend a party, when a suggestion is rejected by a friend or supervisor). ⓘ How does he/she usually respond when he/she doesn't get his/her way, like not being allowed to do something that he/she wants to?	2 1 0	☐
	F	23.	Keeps promises. ⓘ How often does he/she keep his/her promises, like paying back money or returning things that he/she borrows, without having to be reminded?	2 1 0	☐
	G	24.	Controls anger or hurt feelings when given constructive criticism (for example, correction of misbehavior, discussion of a test score or grade, a performance review). ⓘ How does he/she usually respond when someone criticizes him/her in a way that's intended to be helpful, like suggesting how to do something better next time?	2 1 0	☐
	I	25.	Understands that a friendly appearing person may actually intend harm. ⓘ What does he/she understand about how a friendly acting person may actually want to take advantage of him/her?	2 1 0	☐
	F	26.	Respects others' time (for example, doesn't keep others waiting or interrupt others who are busy). ⓘ What about respecting other people's time, like not keeping them waiting or interrupting them when they're busy?	2 1 0	☐
	I	27.	When possible, avoids or leaves harmful relationships or situations (for example, being bullied, coerced into breaking the law, taken advantage of sexually or financially). ☑ If the respondent has not had the opportunity to observe this, estimate a score and check the Estimated box. ⓘ What about staying away from or leaving a relationship or situation where he/she or someone else might get hurt?	2 1 0	☐
6+	I	28.	Avoids being manipulated, dominated, or taken advantage of by others. ⓘ What does he/she do to keep others from controlling or taking advantage of him/her?	2 1 0	☐
	J	29.	Thinks through the consequences of his/her actions before acting (for example, refrains from acting impulsively, considers relevant information). ⓘ How often does he/she think through the consequences before doing something?	2 1 0	☐
	H	30.	Obeys curfews (that is, comes home when he/she is told to, during the day or at night). ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ When you tell him/her that he/she needs to be home at a certain time, how often is he/she home at that time?	2 1 0	☐
	I	31.	Is aware of and uses caution when encountering risky social situations (for example, Internet solicitations, a stranger's offer of a ride or money, "binge" drinking parties, social media, personal ads). ☑ If the respondent has not had the opportunity to observe this, estimate a score and check the Estimated box. ⓘ How is he/she at being cautious when people he/she doesn't know well try to get him/her to do something risky, either in person or through the Internet?	2 1 0	☐
	H	32.	Informs parent or caregiver about his/her plans when he/she goes out (for example, what time he/she is leaving and returning, where he/she is going). ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ What about telling you about his/her plans when he/she goes out, like where he/she is going and when he/she will be home?	2 1 0	☐
	J	33.	Recognizes that advertising messages may not be accurate. ⓘ What about understanding that the purpose of advertising is to influence our decisions, and that everything in ads isn't always totally true?	2 1 0	☐

Office Use Only

Calculation of % Est (see Manual)		Raw Score Calculation	
No. of Est	A	Highest Numbered Basal Item	× 2 = C
No. of Items Answered	B	Points Between Basal and Ceiling	D
(A ÷ B) × 100 =	% Est	cop Raw Score	C + D



GROSS MOTOR — MOTOR SKILLS DOMAIN

INTERVIEW TOPICS

A Sitting; B Pre-Walking Mobility; C Standing; D Walking; E Ball Activities; F Climbing; G Running; H Using Stairs; I Jumping & Hopping; J Catching a Ball; K Riding a Tricycle or Bicycle

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

					Check if Est
0	A	1.	Sits supported (for example, in a chair, with pillows, etc.) for at least 1 minute. ⓘ How long does he/she sit with his/her back supported, like in a chair with a back or propped up by pillows?	2 1 0	☐
	B	2.	Rolls over from back onto stomach. ⓘ How about rolling over from his/her back onto his/her stomach?	2 1 0	☐
	A	3.	Sits unsupported for at least 1 minute. ⓘ How long does he/she sit without anything supporting his/her back?	2 1 0	☐
	B	4.	Moves, scoots, or crawls across the floor. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ How about moving, scooting, or crawling across the floor?	2 1 0	☐
	A	5.	Sits unsupported for at least 10 minutes. ⓘ How long does he/she sit without anything supporting his/her back?	2 1 0	☐
	C	6.	Stands holding on to a stable object for at least 5 seconds. ☑ Score 2 if the individual did this when younger, but now stands without holding on to anything. ⓘ How long does he/she stand while holding on to something?	2 1 0	☐
	C	7.	Pulls self up to standing position. ⓘ What about pulling himself/herself up to standing?	2 1 0	☐
	C	8.	Stands supported with one hand and reaches for an object with the other hand without falling. ☑ Score 2 if the individual did this when younger, but now stands without holding on to anything. ⓘ What happens if he/she is standing holding on to something with one hand and reaches for something with his/her other hand?	2 1 0	☐
	D	9.	Takes steps while supporting self using furniture or another stable object. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ What about taking steps while holding on to furniture or something else?	2 1 0	☐
	B	10.	Crawls up stairs. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ How does he/she go up stairs?	2 1 0	☐
	C	11.	Stands unsupported for at least 1 minute. ⓘ How long does he/she stand without holding on to anything?	2 1 0	☐
	D	12.	Takes at least two steps without support. ⓘ How many steps does he/she take without holding on to anything?	2 1 0	☐
	C	13.	Stands unsupported and reaches for an object without falling. ⓘ What happens if he/she is standing without holding on to anything and reaches for something?	2 1 0	☐
	D	14.	Walks to get around; does not need to hold on to anything. ⓘ How often does he/she walk to get around rather than crawling?	2 1 0	☐
	F	15.	Safely climbs on and off low objects (for example, child's chair, step stool, low bench). ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ How about low objects like a child's chair?	2 1 0	☐
	B	16.	Goes down stairs by crawling backwards or scooting on bottom. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ How does he/she go down stairs?	2 1 0	☐
	C	17.	Squats or bends down to pick up objects without falling. ⓘ What happens when he/she needs to pick up something off the ground?	2 1 0	☐
	E	18.	Throws a ball with one hand; accuracy not important. ⓘ How about throwing a ball with one hand?	2 1 0	☐
	F	19.	Safely gets on and off an adult-sized chair. ⓘ How about an adult-sized chair?	2 1 0	☐
1	G	20.	Runs without falling; may be awkward and uncoordinated. ⓘ How smoothly does he/she run, and how often does he/she fall?	2 1 0	☐
	H	21.	Walks up stairs, putting both feet on each step; may use railing. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ How does he/she go up stairs?	2 1 0	☐
	E	22.	Kicks a ball while standing; accuracy not important. ⓘ How about kicking a ball while standing?	2 1 0	☐



GROSS MOTOR — MOTOR SKILLS DOMAIN

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

					Check if Est
	D	23.	Walks two or more blocks without having to rest or needing physical support. ⓘ What about walking two or more blocks without needing to rest or be helped?	2 1 0	☐
	H	24.	Walks down stairs, facing forward, putting both feet on each step; may use railing. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ How does he/she go down stairs?	2 1 0	☐
	I	25.	Jumps off the ground with both feet without falling. ⓘ How about jumping off the ground with both feet without falling?	2 1 0	☐
	G	26.	Runs smoothly without falling. ⓘ How smoothly does he/she run, and how often does he/she fall?	2 1 0	☐
	F	27.	Safely climbs up and down high objects (for example, jungle gym, ladder, tree). ⓘ What about high objects like a jungle gym, a ladder, or a tree?	2 1 0	☐
	D	28.	Walks carefully on a sidewalk or road that is slippery or uneven. ⓘ What does he/she do if he/she is walking outside and the sidewalk or road is slippery or uneven?	2 1 0	☐
	I	29.	Jumps forward at least three times with both feet without falling. ⓘ How many times can he/she jump forward with both feet without falling?	2 1 0	☐
	G	30.	Runs smoothly, changing speed and direction (for example, when playing tag or sports, or chasing a pet). ⓘ How about changing his/her speed and direction while running, like when playing tag or sports?	2 1 0	☐
	J	31.	Catches a beach ball–sized ball from a distance of 2 or 3 feet. ⓘ What about a beach ball–sized ball thrown from 2 or 3 feet away?	2 1 0	☐
	H	32.	Walks up stairs, alternating feet; may use railing. ⓘ How does he/she go up stairs?	2 1 0	☐
2	H	33.	Walks down stairs, alternating feet; may use railing. ⓘ How does he/she go down stairs?	2 1 0	☐
	H	34.	Climbs a flight of eight or more stairs at a normal pace; may use railing. ⓘ How fast does he/she go up a flight of eight or more stairs?	2 1 0	☐
	K	35.	Pedals a tricycle or other three-wheeled vehicle for at least 6 feet. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ How far does he/she pedal a tricycle or other three-wheeled vehicle?	2 1 0	☐
	I	36.	Hops on one foot at least once without falling; may hold on to something for balance. ⓘ How about hopping on one foot while holding on to something?	2 1 0	☐
	K	37.	Pedals a tricycle or other three-wheeled vehicle around corners. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ What about pedaling a tricycle or other three-wheeled vehicle around corners?	2 1 0	☐
3+	J	38.	Catches a beach ball–sized ball from at least 6 feet away. ⓘ What about a beach ball–sized ball thrown from 6 feet away or more?	2 1 0	☐
	I	39.	Hops forward on one foot with ease without holding on (for example, during hopscotch). ⓘ How about hopping forward on one foot multiple times without holding on?	2 1 0	☐
	J	40.	Catches a tennis- or baseball-sized ball from a distance of 2 or 3 feet, using one or both hands extended away from body. ⓘ What about a tennis- or baseball-sized ball thrown from 2 or 3 feet away?	2 1 0	☐
	K	41.	Rides a balance bike or bicycle with training wheels for at least 10 feet. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ How far does he/she ride a balance bike or a bicycle with training wheels?	2 1 0	☐
	J	42.	Catches a tennis- or baseball-sized ball from a distance of at least 10 feet, moving to catch it if necessary. ⓘ What about a tennis- or baseball-sized ball thrown from 10 feet away or more when he/she needs to move to catch it?	2 1 0	☐
	K	43.	Rides a regular bicycle without training wheels without falling. ⓘ What about riding a regular bicycle without falling?	2 1 0	☐

Office Use Only

Calculation of % Est (see Manual)		Raw Score Calculation	
No. of Est	A	Highest-Numbered Basal Item	× 2 = C
No. of Items Answered	B	Points Between Basal and Ceiling	D
(A ÷ B) × 100 =	% Est	gmo Raw Score	C + D



FINE MOTOR — MOTOR SKILLS DOMAIN

INTERVIEW TOPICS

A Beginning to Handle Objects; B Manipulating Things; C Opening Doors; D Beginning Crayon/Pen Use; E Hand Coordination; F Drawing; G Coloring; H Using Scissors; I Tying; J Precise Movements

Response Options: 2 = Usually, 1 = Sometimes, 0 = Never

					Check if Est
0	A	1.	Reaches for a toy or object. ⓘ How often does he/she reach to try to grab a toy or other object?	2 1 0	☐
	A	2.	Picks up objects; may use both hands. ⓘ How does he/she pick up things like a toy or a ball?	2 1 0	☐
	A	3.	Moves an object from one hand to the other. ⓘ What about moving an object from one hand to the other?	2 1 0	☐
	B	4.	Removes an object (for example, a block or small toy) from a box or other container with no lid. ⓘ How about taking something out of a box or other container?	2 1 0	☐
	A	5.	Picks up small objects (no larger than 2 inches on any side) with thumb and fingers (for example, raisins, beads, small blocks). ⓘ How does he/she pick up small objects?	2 1 0	☐
	A	6.	Picks up a small toy or object with one hand and hands it to someone without dropping it. ⓘ How does he/she pick up something small and hand it to someone?	2 1 0	☐
	B	7.	Puts an object (for example, a block or small toy) into a box or other container with no lid. ⓘ How about putting something into a box or other container?	2 1 0	☐
	D	8.	Marks on paper with a crayon, pen, or pencil; method of grasping the crayon, pen, or pencil is not important. ⓘ What about making marks on paper with a crayon, pen, or pencil?	2 1 0	☐
	C	9.	Opens doors that require only pushing or pulling (for example, cabinet, refrigerator, sliding, or swinging doors). ⓘ What about opening doors that only need to be pushed or pulled, like cabinet doors or sliding doors?	2 1 0	☐
1	B	10.	Stacks at least four small blocks or other small objects; alignment need not be perfect, but the stack must remain upright. ☑ Score 2 if the individual did this when younger, but has now outgrown it. ⓘ How many blocks or other small objects does he/she stack before they fall?	2 1 0	☐
	B	11.	Turns book or magazine pages one by one; books with cardboard pages don't count. ⓘ How about turning book or magazine pages one at a time?	2 1 0	☐
	E	12.	Unwraps small objects (for example, gum or candy). ⓘ How often does he/she unwrap small objects like a piece of candy or gum by himself/herself?	2 1 0	☐
	C	13.	Opens doors by turning a doorknob or handle. ⓘ What about opening doors by turning a doorknob or handle?	2 1 0	☐
2	E	14.	Uses a twisting hand–wrist motion (for example, winds up a toy or music box, screws/unscrews the lid of a jar). ⓘ What about holding something in one hand and twisting with the other, like winding up a toy or music box, or unscrewing the lid of a jar?	2 1 0	☐
	D	15.	Holds a crayon, pen, or pencil in proper position (that is, using a tripod grasp, not with fist) for writing or drawing. ⓘ How does he/she hold a crayon, pen, or pencil?	2 1 0	☐
	E	16.	Presses buttons accurately on a small keyboard or touch screen (for example, on a calculator, cell phone, or other handheld device). ⓘ What about pressing buttons on a small keyboard or touchscreen, like on a cell phone or calculator?	2 1 0	☐
	H	17.	Opens and closes scissors with one hand; does not have to cut with them. ⓘ What about opening and closing scissors with one hand?	2 1 0	☐
	F	18.	Draws a circle freehand while looking at an example. ⓘ What about copying a circle?	2 1 0	☐
	G	19.	Colors simple shapes or animals; more coloring is inside the lines than outside. ☑ Score 2 if the individual did this when younger, but has now outgrown coloring. ⓘ What kinds of things does he/she color, and how much coloring is inside the lines?	2 1 0	☐
3	F	20.	Draws more than one recognizable form (for example, a person, house, tree). ⓘ What does he/she draw well enough that you can recognize what it is?	2 1 0	☐
	E	21.	Pours liquid from one container to another with little or no spilling (for example, pouring milk or juice into a glass). ⓘ What happens when he/she tries to pour liquid, like pouring milk or juice into a glass?	2 1 0	☐
	F	22.	Draws a square freehand while looking at an example. ⓘ What about copying a square?	2 1 0	☐
	H	23.	Uses scissors to cut along a straight line across a standard sheet of paper. ⓘ How far does he/she cut with scissors on a straight line across a sheet of paper?	2 1 0	☐

FINE MOTOR — MOTOR SKILLS DOMAIN

Response Options: 2 = *Usually*, 1 = *Sometimes*, 0 = *Never*

				Check if Est
	F	24. Draws a triangle freehand while looking at an example. ? What about copying a triangle?	2 1 0	☐
	F	25. Uses an eraser without tearing the paper. ? How about using an eraser without tearing the paper?	2 1 0	☐
4	H	26. Cuts out simple shapes (circles, squares, rectangles, etc.). ? What kinds of things does he/she cut out?	2 1 0	☐
	G	27. Colors simple pictures with all coloring inside the lines. ☑ Score 2 if the individual did this when younger, but has now outgrown coloring. ? What kinds of things does he/she color, and how much coloring is inside the lines?	2 1 0	☐
	G	28. Colors a full-page drawing or scene using two or more colors; all coloring is inside the lines. ☑ Score 2 if the individual did this when younger, but has now outgrown coloring. ? What kinds of things does he/she color, and how much coloring is inside the lines?	2 1 0	☐
	F	29. Draws a straight line using a ruler or straightedge. ? What about drawing a straight line using a ruler or straightedge?	2 1 0	☐
5+	I	30. Ties a knot. ? What about tying a knot without help?	2 1 0	☐
	H	31. Cuts out complex shapes (for example, stars, animals, alphabet letters). ? What kinds of things does he/she cut out?	2 1 0	☐
	I	32. Ties a secure bow (for example, shoe laces, gift wrapping). ? What about tying a secure bow without help, for example, shoe laces or a gift wrapping bow?	2 1 0	☐
	J	33. Manipulates very small objects (for example, sets hands on a watch, threads a sewing needle, glues miniature model parts). ? What about working with very small objects, like setting the hands on a watch, threading a sewing needle, or gluing tiny model parts?	2 1 0	☐
	J	34. Assembles, builds, or creates complex building toy structures, model sets, homemade jewelry, arts and crafts, etc. ? What kinds of complex building toy structures, model sets, homemade jewelry, arts and crafts, and things like that does he/she make?	2 1 0	☐

Comments or Observations: _____

Office Use Only	Calculation of % Est *(see Manual)*		Raw Score Calculation		
	No. of Est	A	Highest-Numbered Basal Item	× 2 =	C
	No. of Items Answered	B	Points Between Basal and Ceiling		D
	(A ÷ B) × 100 =	% Est	**fmo** Raw Score		C + D

Appendix 4.4: *Self-Restraint Questionnaire*

THE SELF RESTRAINT QUESTIONNAIRE

Today's Date _____

Are you the person's teacher / nurse / keyworker / SEC instructor / parent / other *(ring as appropriate)*

Please answer all of the questions on which may or may not describe behaviours shown by the person at the present time. Read the questions carefully and then ✓ the box which is under the statement that best describes the amount of time in a day that the person engages in that behaviour if permitted to do so. If you never see the behaviour described in the question them simply ✓ the box marked **NEVER**. Two examples follow:

EXAMPLE 1. If when permitted and when wearing a long sleeved garment the person always has sleeves pulled over his or her hands then ✓ the **ALL OF THE TIME** box for question 3. It should then look like this:

3. Pull the sleeves of clothes over his or her hands if clothes have long sleeves?



ALL OF THE TIME	MOST OF THE TIME	HALF THE TIME	SOME OF THE TIME	NEVER
✓				

EXAMPLE 2. If he or she is seen to sit on his or her hands for some of the time then ✓ the **SOME OF THE TIME** box for question 15. It should then look like this:

15. Sit on his or her hands?



ALL OF THE TIME	MOST OF THE TIME	HALF THE TIME	SOME OF THE TIME	NEVER
			✓	

Please give an answer to every question. If you are unsure about an answer write a note below the question to comment on your answer and then ✓ the box you think most accurately describes the time spent by the person doing that behaviour.

QUESTIONNAIRE

If permitted does he or she . . .

1. Entwine his or her hands in the front part of the clothes that are worn *eg* t-shirt, jumper?



ALL OF THE TIME	MOST OF THE TIME	HALF THE TIME	SOME OF THE TIME	NEVER

2. Pull his or her arms out of the sleeves and push them part or all of the way into the main body of the clothes that are worn?



ALL OF THE TIME	MOST OF THE TIME	HALF THE TIME	SOME OF THE TIME	NEVER

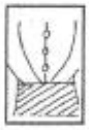
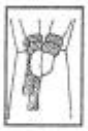
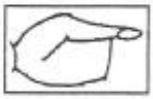
3. Pull the sleeves of his or her clothes over hands if clothes have long sleeves?



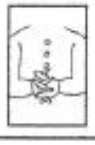
ALL OF THE TIME	MOST OF THE TIME	HALF THE TIME	SOME OF THE TIME	NEVER

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If permitted does he or she . . .

		ALL OF THE TIME	MOST OF THE TIME	HALF THE TIME	SOME OF THE TIME	NEVER
4. Push hands inside trousers, if he or she is wearing them?		☐	☐	☐	☐	☐
5. Push hands inside any pockets in his clothing if he or she has them?		☐	☐	☐	☐	☐
6. Wrap his hands/arms together with any piece of long material, *eg* sock, scarf when there is access to the material?		☐	☐	☐	☐	☐
7. Wrap a long piece of material *eg* sock, scarf, around either or both arms but not so that they are held together, when there is access to the material?		☐	☐	☐	☐	☐
8. Wrap or entwine hands in material other own clothes *eg* curtains, bed clothes, other clothing, if it is available?		☐	☐	☐	☐	☐
9. Wrap a blanket, sheet, towel or other large piece of material around himself or herself if it is available?		☐	☐	☐	☐	☐
10. Entwine arms around a chair or other piece of furniture?		☐	☐	☐	☐	☐
11. Push hands into chair/sofa cushions or other tight spaces?		☐	☐	☐	☐	☐
12. Lie on stomach with hands held behind back?		☐	☐	☐	☐	☐

If permitted does he or she . . .

		ALL OF THE TIME	MOST OF THE TIME	HALF THE TIME	SOME OF THE TIME	NEVER
13. Lie/sit in a position which restricts the movement of arms *eg* with them under the body, behind the back or pressed against a wall?		☐	☐	☐	☐	☐
14. Carry an object or objects around but not manipulate, use or play with them?		☐	☐	☐	☐	☐
15. Sit on hands?		☐	☐	☐	☐	☐
16. Hold hands together behind his back?		☐	☐	☐	☐	☐
17. Hold hands together behind the neck?		☐	☐	☐	☐	☐
18. Push hands between the legs?		☐	☐	☐	☐	☐
19. Have arms folded?		☐	☐	☐	☐	☐
20. Entwine arms with each other in any other way not described above? *(Please describe below.)*		☐	☐	☐	☐	☐
21. Interlock fingers or hands in any way. *(Please describe below.)*		☐	☐	☐	☐	☐

Does he or she . . .

22. Ask or otherwise indicate that he or she wants hands held *eg* putting hands into another's or under another's arm?



ALL OF THE TIME	MOST OF THE TIME	HALF THE TIME	SOME OF THE TIME	NEVER
☐	☐	☐	☐	☐

23. Ask or otherwise indicate that he or she wants arms held *eg* taking another's arm and putting it around him or her?



ALL OF THE TIME	MOST OF THE TIME	HALF THE TIME	SOME OF THE TIME	NEVER
☐	☐	☐	☐	☐

24. If you have answered **ALWAYS** or **MOST OF THE TIME** to any of the above questions please roughly sketch the way in which the person shows this behaviour and give a brief description below.

25. Please describe any other way not mentioned above in which the person restricts the movement of hand/arms or likes to have anything on hands or arms which is unusual *eg* gloves when indoors. Could you also describe any unusually strong preference the person may have for a particular piece of clothing (eg if they always insist on wearing a hat or will never wear garments with short sleeves) or whether they insist on carrying a particular object or objects with them most of the time. A sketch would be very useful to us. If there no other method of self restraint then please write NONE in this space.

Thank you

Appendix 4.5: *Self-Restraint Questionnaire 2*

SRQ2 v1.0 27.07.22
SIB Study
IRAS ID: 263048

SRQ2

The following questions are asked in order to get some more information about the self-restraint behaviours described in the Self-Restraint Questionnaire. Please read the questions carefully, and then indicate the statement that best describes your response.

1. How difficult is it for your child to self-injure while engaging in these behaviours?

Not at all	Slightly difficult	Moderately difficult	Extremely difficult	Impossible
1	2	3	4	5

2. When your child is engaging in these behaviours, to what extent does the self-injury decrease in frequency?

No decrease or increase in frequency	Some decrease in frequency	Moderate decrease in frequency	Significant decrease in frequency	SIB never occurs
1	2	3	4	5

3. If your child is prevented from or is unable to engage in these behaviours, to what extent do they become distressed, e.g., cry, negative vocalisations, scream or show aggression?

Never	Sometimes	Half the time	Most of the time	Always
1	2	3	4	5

4. How often does your child initiate these behaviours by themselves, without being told or asked to engage in the behaviour by another person?

Never	Sometimes	Half the time	Most of the time	Always
1	2	3	4	5

5. If your child is prevented from doing these behaviours, how often do they actively try to engage in the behaviour again?

Never	Sometimes	Half the time	Most of the time	Always
1	2	3	4	5

6. How difficult is it for your child to stop these behaviours without help?

Impossible without help	Could do so with extreme difficulty	Can do so with moderate difficulty	Can do with slight difficulty	No difficulty at all
1	2	3	4	5

Appendix 4.6: *The Childhood Executive Functioning Inventory*

CHILDHOOD EXECUTIVE FUNCTIONING INVENTORY (CHEXI) FOR PARENTS AND TEACHERS

Below, you will find a number of statements. Please read each statement carefully and thereafter indicate how well that statement is true for the child. You indicate your response by circling one of the numbers (from 1 to 5) after each statement.

Definitely not true	Not true	Partially true	True	Definitely true
1	2	3	4	5

1.	Has difficulty remembering lengthy instructions	1	2	3	4	5
2.	Seldom seems to be able to motivate him-/herself to do something that he/she doesn't want to do	1	2	3	4	5
3.	Has difficulty remembering what he/she is doing, in the middle of an activity	1	2	3	4	5
4.	Has difficulty following through on less appealing tasks unless he/she is promised some type of reward for doing so	1	2	3	4	5
5.	Has a tendency to do things without first thinking about what could happen	1	2	3	4	5
6.	When asked to do several things, he/she only remembers the first or last	1	2	3	4	5
7.	Has difficulty coming up with a different way of solving a problem when he/she gets stuck	1	2	3	4	5
8.	When something needs to be done, he/she is often distracted by something more appealing	1	2	3	4	5
9.	Easily forgets what he/she is asked to fetch	1	2	3	4	5
10.	Gets overly excited when something special is going to happen (e.g., going on a field trip, going to a party)	1	2	3	4	5
11.	Has clear difficulties doing things he/she finds boring	1	2	3	4	5
12.	Has difficulty planning for an activity (e.g., remembering to bring everything necessary for a field trip or things needed for school)	1	2	3	4	5
13.	Has difficulty holding back his/her activity despite being told to do so	1	2	3	4	5
14.	Has difficulty carrying out activities that require several steps (e.g., for younger children, getting completely dressed without reminders; for older children, doing all homework independently)	1	2	3	4	5

www.chexi.se

Definitely not true	Not true	Partially true	True	Definitely true
1	2	3	4	5

15. In order to be able to concentrate, he/she must find the task appealing	1	2	3	4	5
16. Has difficulty refraining from smiling or laughing in situations where it is inappropriate	1	2	3	4	5
17. Has difficulty telling a story about something that has happened so that others may easily understand	1	2	3	4	5
18. Has difficulty stopping an activity immediately upon being told to do so. For example, he/she needs to jump a couple of extra times or play on the computer a little bit longer after being asked to stop	1	2	3	4	5
19. Has difficulty understanding verbal instructions unless he/she is also shown ***how*** to do something	1	2	3	4	5
20. Has difficulty with tasks or activities that involve several steps	1	2	3	4	5
21. Has difficulty thinking ahead or learning from experience	1	2	3	4	5
22. Acts in a wilder way compared to other children in a group (e.g., at a birthday party or during a group activity)	1	2	3	4	5
23. Has difficulty doing things that require mental effort, such as counting backwards	1	2	3	4	5
24. Has difficulty keeping things in mind while he/she is doing something else	1	2	3	4	5

Appendix 4.7: *SIB study remote executive function researcher manual*

Sleep - Impulsivity - Behaviour (SIB) Study

General Instructions

During the VABS
Go through "VABS Checklist" document with parent (task, room and device checks).

Prior to the testing session:

- Work out which EF tasks to conduct according to the child's overall age equivalent on the VABS.
    - If <2 years overall age equivalent start with Game 3 'Time To Sort!'.
        - If participant passes, move on to complete Game 4.
    - If 2-4 years overall age equivalent start with Game 4 'Ball Sorting'.
        - If participant fails the first rule (PT 2), go back and attempt Game 3.
        - If participant passes the Silly Game (PT 4), attempt DCCS.
        - If participant passes the first rule and fails the Silly Game, use judgement to determine if DCCS should be attempted.
- Work out where to start on the BPVS based on the child's receptive language age equivalent:

If ≤ 3 years 11 months	Start at Set 1 Item 1
If 4 years	Start at Set 2 Item 13
If 5-6 years	Start at Set 4 Item 37
If 7 years	Start at Set 5 Item 49
If 8 years	Start at Set 6 Item 61
If 9-11 years	Start at Set 7 Item 73
If 12-13 years	Start at Set 9 Item 97
If 14+ years	Start at Set 10 Item 109

- Open up and fill in Participant Scoring Sheet
- Remind parent to charge device(s) and headphones in advance (or charge during session)

At the start of the testing session:

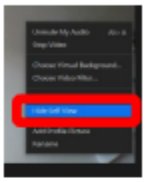
- Introduce yourself to the parent and child
- Check it's still a good time / okay to proceed today; check if need toilet break

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Setting up

- Check yours and parent's device(s) are charging/charged
- Guide parent to set up device(s), check WIFI connection is stable
  - If 2 devices, disconnect audio from one (preferably the device you won't use for Game 7)
    - → "Mute > leave computer audio" on laptop/PC
    - → "More > disconnect audio" on tablet/phone
- Guide parent to set up headphones - reminder to only have one in so they can hear the child!
  - Advise turning off Bluetooth of other device(s) nearby
  - Check parent can hear you through the one headphone
  - Warn not to touch the right earphone – button disconnects call
- Check you have the best view of the child and parent, then ask parent to set Zoom to "Gallery View" and "Hide Self View":
  - Laptop/PC: right click over face and select "Hide Self View"
  - iPad/tablet: click "...More" > "Meeting Settings" > untick "Show Self View"



Set parent's expectations

- There will be times where I will guide you through games more generally, and other times where I'll ask you to repeat specific wording and actions after me.
- Try not to prompt or give your child any feedback until I say you can (so if child gets something right, I'll let you know when you can say "yes well done", likewise if they answer in a way that you don't think seems 'right', I'll let you know if you can correct them.
- Reminder of trying to not laugh if child responds in a way you don't think they 'should'

Statements

- Safeguarding statement
  - *"For the safety of your child, we ask that you do not leave your child alone during the course of the Zoom call. If you need to leave the room at any point, for example to go to the toilet or attend to other family members, please take your child with you. Are you happy to continue?"*
- Zoom recording disclaimer
  - *"We will be recording this play session via Zoom today, and will be storing it on the University's secure server. Are you happy to continue?"*
- Say *"Ok, I am going to start the recording now. [Start recording]. I just wanted to formally check that you have read the information about the study and are happy to continue? If you or your child need to stop at any time that's fine, just let us know. And just to note, everything that happens within this game session is confidential and will only be shared with members of the research team. However if we see anything that makes me feel concerned about you, your child or anyone's safety then I may have to pass that on - but I would try and come back to you before I did that. Is that all okay?"* [We will need to note this formal re-consenting procedure on the participant's consent form]
- **Pin family device(s)**

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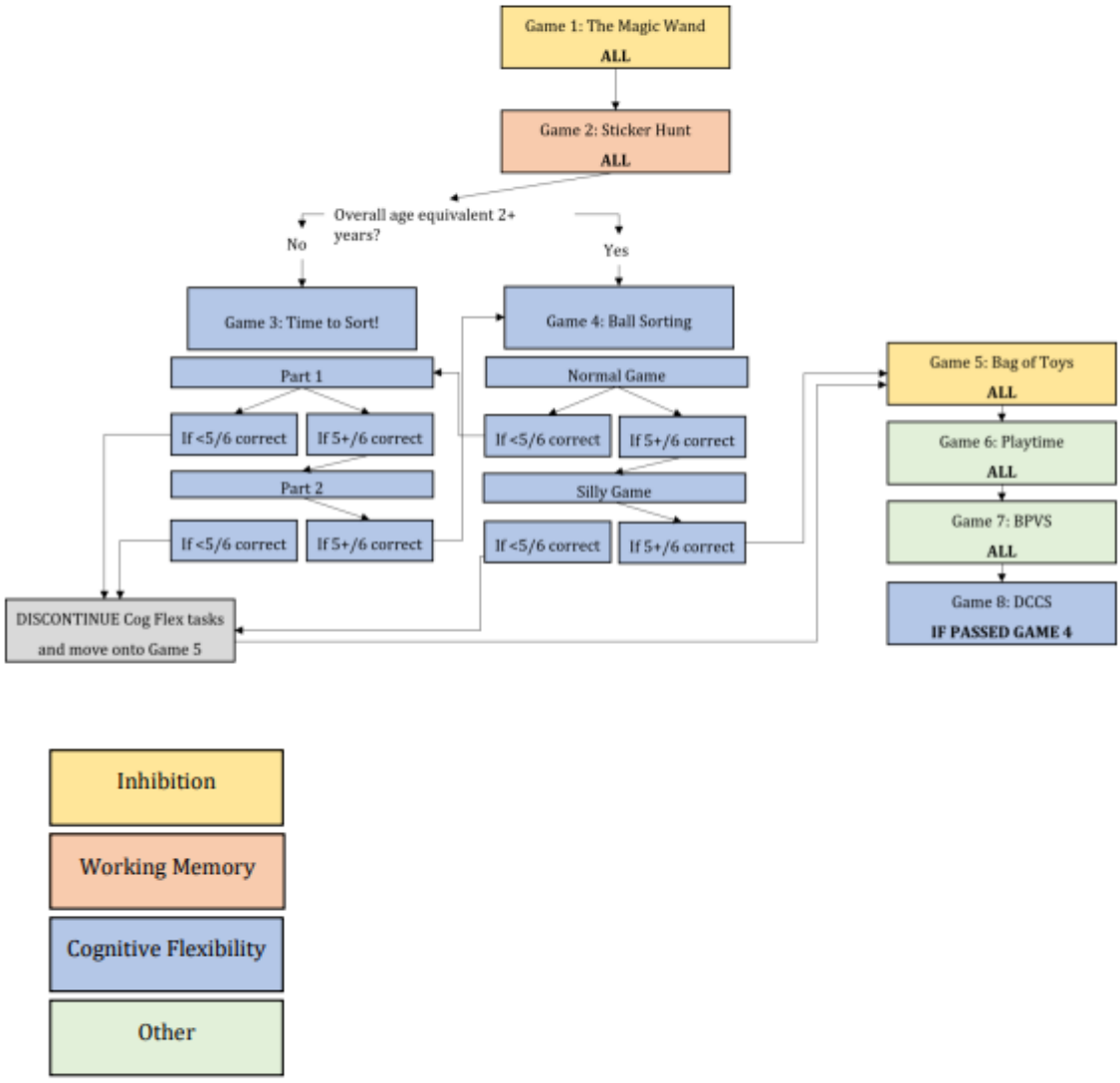
3 Researcher Manual

- Ask to see autism diagnosis letter
- Let parent know that if their call disconnects, re-click the Zoom link and we will be waiting on the call for their return

<u>During the testing session</u>:

- Turn off your camera (except for Game 7 – BPVS/DCCS)

<u>Games flow chart</u>



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Game 1: The Magic Wand

Direct parent towards page 2 of 'Games Instruction' page (front and back).

What parent will need:

- Glitter Wand

Found in: package labelled "Game 1"



Package



Wand

What you will need:

- Timer ready to countdown from 30 seconds





Task set up

Child should not see the wand before starting the game.

If possible, parent should sit opposite child, facing them.

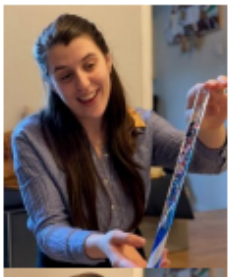
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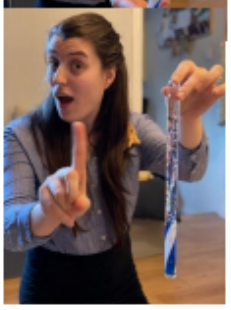
How to play

- Check parent and child are comfortable, and ready to play
- Ask parent to attract child's attention by telling them they're going to play a game together
- Ask parent to open the package to reveal the magic wand toy
- Remind parent that it is important child sees their instructions during this game
- **N.B. Don't let child touch the wand before Step 3**

1. Ask parent to show child the glitter wand for a few seconds, drawing attention to the wand by saying **'Oh look [child name]'.**
   Say they can twist the wand upside down so that the glitter in the wand moves.
   Make sure the child is paying attention to the wand.
   - If the child isn't interested in the wand, encourage the parent to keep playing with it (twist upside down, repeat phrase, get closer)

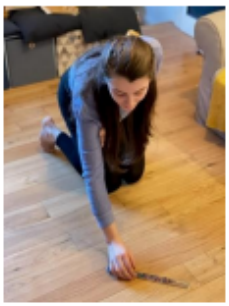


2. Ask parent to say **'Look [child name]. No, don't touch'** using their finger to emphasise 'no'.
   Make sure child attends to this instruction.
   - If child does not seem to attend to this instruction, ask parent to repeat it before placing the wand down (Step 3).



3. Ask parent to place the wand down in front of child, where they can clearly see it.

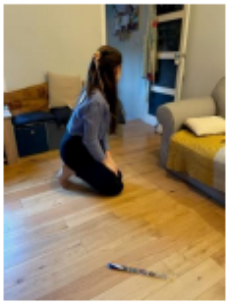
   Start the timer to count down from 30 seconds immediately after the wand has been placed down.



4. Ask parent to turn around, away from child.

5. **Stop the timer immediately after the child touches the wand**, and make a note of the time in the "Participant Scoring Sheet" (this marks the end of the trial/game). Tell the parent that the game has ended and that they can turn back around towards the child. If the child doesn't touch the toy for the 30 seconds, tell the parent they can now turn back around and say **"It's okay, you can touch it now"**

   Praise child for playing the game and say you'll now start the next game.



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Game 2: Sticker Hunt

Direct parent towards page 3 of 'Games Instruction' pack (front and back).

What parent will need:

- 5 stickers
- 5 cups
- Laminated card
- Certificate

Found in: package labelled "Game 2" / tucked in postage box.



Package | Stickers | Cups | Laminated card

Task setup

Ask parent to place the 5 cups in a row on the table/floor in the following order: (**from child's POV**, left to right): orange, blue, purple, green, red.

Ask parent to place stickers behind the cups (from child's POV).



If possible, have parent sit opposite child. If there's a second caregiver present, ask them to sit with the child and gently encourage them to focus on the game.

Ask parent to keep toys out of child's reach for this game.

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How to play

- Check parent and child are comfortable, and ready to play
- Ask parent to attract child's attention by telling them they're going to play a game together
- If child tries to direct parent's attention away during the task, tell the parent to give a short reply but then direct focus back to the game.
- At yours and the parent's discretion, end the game at any time if the child seems distressed or unable to play.
- **N.B. Emphasise stickers are to be found one by one**

Praise if correct? YES. Correct if wrong? YES (don't laugh)

1. Ask parent to pick up any one cup and put a sticker down underneath it. Tell parent stickers and cups are matched on colour, so the red sticker goes under the red cup, the purple sticker under the purple cup, and so on. Do this for all 5 cups.



2. Ask parent to cover the cups with the laminated card, ensuring the child's view of the cups is completely blocked. **Make sure parent has child's attention**, then ask parent to **count out loud for 5 seconds** then remove the card.



3. Ask parent to say to child: **"Can you point to a sticker?"**. A sticker is 'found' by any gesture or action that shows the child is indicating a sticker's location (e.g., point, grab, kick, lurch towards etc). Remind only one sticker should be found.



4. The child will find a sticker in any of the five cups on the first go.
When they do, ask parent to lift the cup, and pick up the sticker. Ask parent to praise their child by saying phrases like **"Well done!" / "Good job!" / "You got the sticker!"**.

5. **Make sure child is watching parent**, then ask parent to put the sticker out of sight say **"Look! Let's put this one here"**.



6. **Make sure child is watching parent,** then ask parent to lift up the empty cup and say: **"Look! This cup is now empty!"** (it is now the only cup without a sticker underneath). All of the cups should now be stacked upright.

7. **Make sure child is watching parent,** then ask parent to cover the cups with card, **count out loud for 5 seconds** then remove the card.



8. Ask parent to say **"Can you get another sticker?"**

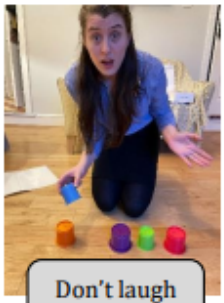
Do not use the word 'AGAIN'

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9. CORRECT RETRIEVAL: If child points towards a <u>different</u> cup this time (for example they chose a red cup the first time and a purple cup this time), do the following:
    - Ask parent to lift up the cup, and pick up the sticker.
    - Praise child with phrases like **"Well done!"/ "Good job!"/ "You got the sticker!"**.
    - **<u>Make sure child is attending to parent</u>**, then ask parent to put the sticker out of sight and say: **"Look! Let's put this one here"**. There should now be two stickers on the chart.
    - **<u>Make sure child is attending to parent</u>**, then ask parent to lift up the empty cup and say: **"Look! This cup is now empty!"**.
    - Go back to Step 2 (cover cups with card)

INCORRECT RETRIEVAL: If child points towards the <u>same</u> cup for a second time (for example they chose a red cup the first *and* second time), do the following:

- Ask parent to lift the cup to show it's empty, and say: **"It's not there. Let's have another go."** Remind parent to not smile or laugh.
- **<u>Make sure child is attending to parent</u>**, then ask parent to lift up the empty cup and say: **"Look! This cup is now empty!"**.
- Go back to Step 2 (cover cups with card)



10. Keep following procedure until either
    - Child finds all 5 stickers, *or*
    - Child makes an incorrect choice 3 times consecutively.

Tell the parent when the game has finished. Be encouraging – say well done and invite parent and child to stick the stickers on their certificates.

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Game 3: Time To Sort!

Direct parent towards page 4 of 'Games Instruction' pack (front and back).

What parents will need

- 2 boxes (1 big, 1 small) **lids off**
- 1 large ball (from the bag of 6)
- 1 small ball (from the bag of 6)

Found in: package labelled "Game 3/4"



Boxes



Balls

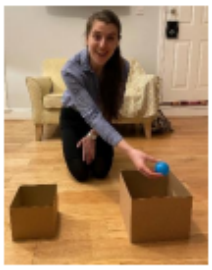
Task setup

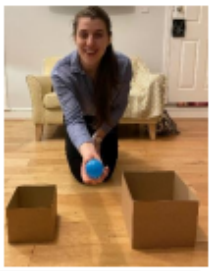
Ask the parent to set up the two boxes **without lids**, about 30cm (shoulder-width) apart, ideally opposite the child.

Ask the parent to have one big and one small ball ready.

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How to play





N.B. Note down on Participant Score Sheet whether the child places the ball in the boxes themselves, or whether they point/gesture towards the boxes and the parent places the ball in. Either is fine.

Part I: Big ball in big box

1. Ask the parent to get the child's attention and say **"Oh [child name] look at this!"** as they demonstrate putting (one) big ball in the larger box, then picking it back up. If you are not certain that child was looking at the parent, ask them to repeat the demonstration.

2. Ask the parent to say **"Now you try"** and either pass the child the ball or indicate where the child would like the ball to go. Remind the parent to offer the ball centrally, so that they do not cue the child to place the ball in either box.

   If the child places the ball in the big box, this is correct. Encourage the parent to say **"Well done [child name]"** and pick the ball back up.
   If the child places the ball in the smaller box ask the parent to say **"Oh no, the ball doesn't go in there!"** pick up the ball, ***point*** towards the bigger box and say it goes in there. Remind the parent not to laugh or praise the child if wrong.

   Praise if right
   Say + point if wrong
   Don't demonstrate

3. Ask the parent to repeat this **6** times without cuing child. Each time they should offer the ball centrally and say **"Where does this go?"**
   (The ball should go in the larger box). Do not offer any feedback. Tell the parent when they have done 6 turns and ask them to gather all the balls.

   NO FEEDBACK –
   "Let's do another one"

Encourage parent to congratulate child for playing the game, and either:

Next part: If the participant correctly sorts 5/6 balls, proceed to the next stage and tell the parent to start the second part of the game.
New game: If they correctly sort 4/6 or less, instead ask the parent if they/the child would like a break then start a new game (5). Do not proceed to Part II and do not administer Game 4.

Part II: Small ball in small box

In the second part of Game 3, the rule is reversed so that children need to put the small ball into the smaller box. Instruct the parent to teach them this new rule:



4. Ask the parent to get the child's attention and say **"Oh [child name] look at this!"** as they demonstrate putting (one) small ball in the smaller box and then removing it. If you are not certain that the child was looking at the parent, ask them to repeat the demonstration.

5. Ask the parent to say **"Now you try"** and either pass the child the ball or indicate where the child would like the ball to go. Remind the parent to offer the ball centrally, so that they do not cue the child to place the ball in either box.

   If the child places the ball in the smaller box, this is correct. Encourage the parent to say **"Well done [child name]"** and pick the ball back up.
   If the child places the ball in the smaller box ask the parent to say **"Oh no, the ball doesn't go in there!"** pick up the ball, ***point*** towards the smaller box and say it goes in there. Remind the parent not to laugh or praise the child if wrong.

   Praise if right
   Say + point if wrong
   Don't demonstrate

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6. Ask the parent to repeat this **6** times without cuing child. Each time they should offer the ball centrally and say "**Where does this go?**" (The ball should go in the smaller box). Do not offer any feedback. Tell the parent when they have done 6 turns and ask them to gather all of the balls.

NO FEEDBACK –
"Let's do another one"

END. Encourage parent to congratulate child ("you're doing so well!") and say you'll now start a new game:

If the participant correctly sorts 5/6 balls proceed to Game 4.

If they do not pass Game 3, do not proceed to Game 4. Instead ask the parent if they/the child would like a break, then proceed to Game 5.

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Game 4: Ball Sorting

Direct parent towards page 5 of 'Games Instruction' pack.

What parents will need

- 2 boxes (1 big, 1 small) **lids on**
- 6 large balls
- 6 small balls

Found in: package labelled "Game 3/4"



Boxes Balls

Task setup – if starting here

Ask the parent to set up the two boxes **with the lids on**, about 30cm (shoulder-width) apart, ideally opposite the child.



Task setup – if continuing from Game 3

Ask parent to put the lids on the boxes and get both bags of balls. Tell the parent that now is a good time to empty both boxes and make sure that you have all the balls back in the bags they came in.

Tell the parent that in this game, the size of the balls and the boxes are important. We are interested to see whether child will sort according to size.

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How to play

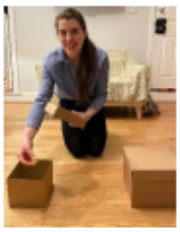
Part I: Demonstrate big in big & small in small



1. Ask the parent to have the bags of big balls and little balls ready next to them. Ask the parent to get the child's attention and say **"Oh [child name] look at this! Here I have a Big Box, and here I have a Little Box. In this game I'm going to put the Big Balls in the Big Box and I'm going to put the Little Balls in the Little Box"** as they demonstrate putting one large ball in the larger box, and one small ball in the smaller box. You can leave the balls in the boxes. If you are not certain that child was looking at the parent, ask them to repeat the demonstration.
   Make sure it's the parent that demonstrates, even if the child wants to place it themselves.

2. Ask the parent to tell the child, **"Now it's your turn to put the Big Balls in the Big Box and the Little Balls in the Little Box"**. Ask the parent to pick up a big ball and show/pass it to the child as they say, **"Now here is a Big Ball. Where does this one go?"** Allow child to point or place the ball in the box.
   - ✓ If child correctly places the ball in the big box this is correct. Encourage the parent to say, **"Yes, very good. The Big Balls go in the Big Box"** and close the lid.
   - ✗ If child tries to place the ball in the little box ask the parent to say **"No, this one's a Big Ball. It goes over here in the Big Box."** Remind the parent not to laugh or praise the child here.

Praise & correct



3. Ask the parent to pick up a Little Ball and show/pass it to child, as they say **"Now here is a Little Ball. Where does this one go?"** (Child point or place.)
   - ✓ If child places the ball in the little box this is correct. Encourage the parent to say, **"Yes, very good. The Little Balls go in the Little Box"** and close the lid.
   - ✗ If child tries to place the ball in the big box ask the parent to say **"No, this one's a Little Ball. It goes over here in the Little Box."** Remind the parent not to laugh or praise the child.

4. Repeat steps 2 and 3 so that the child has an opportunity to sort each size of ball twice. Continue onto Part 2 regardless of whether sorting is correct, unless child is clearly upset or distressed. If child does not want to play, say well done and move onto Game 5.

Part II: Child's go

In the second part of this game, you will prompt the parent to ask the child to sort some more balls.
N.B. Explain that it is important that at this stage of the game the parent does not give the child feedback after each turn. Instead they should say ***"Let's do another one!"***

5. Ask the parent to say to the child, **"Now let's try some more!"** Ask the parent to present the balls in the following order and ask the child to sort them. As the parent holds up each ball each time say **"If it is a Big Ball, it goes here *[point to big box]* and if it is a Little Ball it goes here *[point to little box]*. Here is a ___ ball. Where does it go?"** Remind the parent not to give feedback here. They can say **"Let's do another one"** to encourage the child to stay engaged.
   a) Little ball
   b) Big ball
   c) Little ball
   d) Big ball
   e) Big ball
   f) Little ball

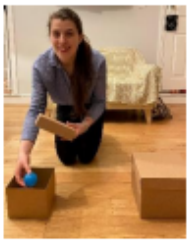
NO FEEDBACK – "Let's do another one"

Ball goes wherever child chooses – even if wrong.

Record how many balls the child is able to sort correctly in the Participant Scoring Sheet.

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If they correctly sort 5/6 balls, proceed to the next stage of the game.
If they do not correctly sort 5/6 balls, instead ask the parent if they/the child would like a break. Do not proceed to the next part of the game, move onto Game 5.



Part III: Demonstrate Silly Game

Explain to the parent that in the final part of the game, we are interested to see how child can switch between rules. The set-up for this part is the same, but this time the Big Balls will be sorted into the Little Box and the Little Balls into the Big Box. We call this the Silly Game!

6. Ask the parent to tell the child, **"Now we're going to play a Silly Game! In this game, we put the Big Balls in the Little Box and the Little Balls in the Big Box"** as they demonstrate putting a small ball in the larger box, and a large ball in the smaller box.
   If you are not certain that child was looking at the parent, ask them to repeat the demonstration.

7. Ask the parent to tell the child, **"Now it's your turn to play the Silly Game!"**. Ask the parent to pick up a big ball and show/pass it to the child as they say, **"Now here is a Big Ball. Where does this one go in the Silly Game?"** Allow child to point or place the ball in the box.

   ✓ If child places the ball in the small box this is correct. Encourage the parent to say, **"Yes, very good. The Big Balls go in the Little Box"** and close the lid.
   ✗ If child tries to place the ball in the big box ask the parent to say **"No, this one's a Big Ball. It goes over here in the Little Box."** Remind the parent not to laugh or praise the child.

Praise & correct

8. Ask the parent to pick up a Little Ball and show/pass it to child, as they say **"Now here is a Little Ball. Where does this one go in the Silly Game?"** Allow child to point or place the ball in the box.



   ✓ If child places the ball in the big box this is correct. Encourage the parent to say, **"Yes, very good. The Little Balls go in the Big Box"** and close the lid.
   ✗ If child tries to place the ball in the little box ask the parent to say **"No, this one's a Little Ball. It goes over here in the Big Box."** Remind the parent not to laugh or praise the child.

9. Repeat steps 7 and 8 so that the child has an opportunity to sort each size of ball twice. Continue onto Part 4 regardless of whether sorting is correct, unless child is clearly upset or distressed. If child does not want to play, say well done and move onto Game 5.

Part IV: Child's go – Silly Game

10. Ask the parent to tell the child, **"Now let's try some more!"**. Ask the parent to present the balls in the following order and ask the child to sort them. As the parent holds up each ball each time they should say **"If it is a Big Ball, it goes here *[point to little box]* and if it is a Little Ball it goes here *[point to big box]*. Here is a ___ ball. Where does it go?"**
    a) Little ball
    b) Big ball
    c) Little ball
    d) Big ball
    e) Big ball
    f) Little ball

NO FEEDBACK – "Let's do another one"

Ball goes wherever child chooses – even if wrong.

Record how many balls the child is able to sort correctly – if 5/6 the continue to DCCS in Game 7.
Ask the parent if they/the child would like a break. Then move on to Game 5.

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Game 5: Bag of Toys

Direct parent towards page 6 of 'Games Instruction' pack.

What parent will need:

- Stuffed animal toy
- Toy car
- Squeezy ball toy

Found in: package labelled "Game 5"



What you (researcher) will need:

- Timer ready to countdown from one minute



Task set up

Child should not see the toys before starting the game.

If possible, parent should sit opposite child, facing them. Make sure there is space between the parent and child, so that the parent can lay out the toys in front of them.



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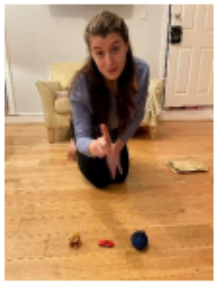
How to play

- Check parent and child are comfortable, and ready to play
- Ask parent to attract child's attention by telling them they're going to play a game together
- Ask parent to open the package containing the toys
- If child reaches up to grab package, ask parent try keep it out of their reach
- Remind parent that it is important child sees their instructions during this game
- **Remind parent that they should not hand child any toys until the end (but don't say "don't touch", just hold them out of reach)**

1. Ask parent to take the toys out of the package one by one in the following order: **cuddly toy → car → squeezy ball**, drawing attention to each toy by saying **'Oh look [child name]'**. Do not let parent place toys down.
   Make sure the child is paying attention to the toys.



   - **N.B. It is crucial the child isn't offered/allowed to touch any of the toys before Step 3. If child wants to touch, do not tell parent to *say* "don't touch", but instead encourage parent to hold toys away and say "hold on" / "wait a second" / "just a moment" / "not yet"**
   - If the child isn't interested in the toys, encourage the parent to keep playing with them
   - If there are two caregivers present, ask the second person to gently hold the child to discourage them touching the toys



2. Make sure child is paying attention to parent, then ask parent to place all three toys down at the same time, and say **'Look child. No, don't touch'** using their finger to emphasise 'no'.

   Start the timer to count down from 60 seconds immediately after this instruction has been given.

3. Ask parent to turn around, away from child.



4. **Stop the timer immediately after the child touches any of the toys**, and make a note of the time (this marks the end of the trial/game).
   Tell the parent that the game has ended and that they can turn back around towards the child.
   If the child doesn't touch the toys for the 30 seconds, tell the parent they can now turn back around and say **"It's okay, you can touch it now"**.

   As soon as child touches a toy, let parent know playtime has begun.

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Game 6: Playtime

Direct parent towards page 7 of 'Games Instruction' pack.

1. Tell parent it's now playtime.

   "Now, we would like you and your child to play in any way you want. You can use the toys we've sent you, or any other toys you may already have. We will let you know when playtime is over."

   Make sure parent / child stays in frame of camera; if they move out of frame, let the parent know and direct play back towards the camera shot.

2. After 10 minutes, tell parent playtime is over and it's time for our final game.

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Game 7: Point To A Picture

Direct parent towards page 8 of 'Games Instruction' pack.

What you (researcher) will need:

- BPVS manual
- Digital visualiser

Task set up

Tell parent that for this final game, you would like them and child to sit in front of the laptop, with a clear view of Zoom. This may involve moving the device to a more suitable place (e.g., desk/table).

How to play

1. Tell parent to disconnect their headphones, so that their sound plays through the device speakers and the child can hear you. Test this by interacting with the child.

2. If using 2 devices, tell parent to end the call on the second device. Keep the call going on the device more suitable for the testing (e.g., laptop > phone).

    - Ensure the device that is kept running has audio connected.

3. Check family's device(s) are pinned

4. Check if parent has hidden Self View if this will distract child (and it wasn't selected at the beginning of the visit)
    - Laptop/PC: right click over face and select "Hide Self View"
    - iPad/tablet: click "...More" > "Meeting Settings" > untick "Show Self View"

5. Check parent has selected "Speaker View" – advise they select "View" then "Speaker View"

6. Use digital visualizer to project BPVS onto Zoom as a screen share via camera app. Turn the brightness up to around 50%.

    Explain how to play this game to parent and child, then follow the instructions in the BPVS manual. Instruct parent to call out the number of the image the child points to. Make a note of the number the parent calls out.

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DCCS – Boats and Rabbits

ONLY play if child passes Game 4: Ball Sorting

What you (researcher) will need:

- DCCS PowerPoint Presentation

Task set up

Open Participant Scoring Sheet to keep track of child's score.

Tell parent that you're going to play another pointing game. This time it's a card sorting game.

Zoom Setup
Ask parent and child to sit in front of the laptop, with a clear view of Zoom.

Tell parent to disconnect their headphones, so that their sound plays through the device speakers and the child can hear you. Test this by interacting with the child.

Pin family's device(s)

Check parent has selected "Speaker View" – advise they click "View" then "Speaker View"

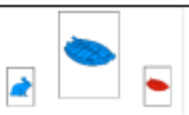
Move onto pages 20 & 21 for DCCS instructions.

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20 *Researcher Manual*

How to play

Note: during pre- and post-switch trials a slide will appear showing empty target cards – this is to differentiate trial cards.

Slide #	Script	Screenshot
1 *Demo Phase*	Tell the parent you're going to play a card sorting game with them and their child, where you'll ask the child to point towards cards on the screen. Direct the parent's attention towards the numbers in the bottom right/left corners, and ask them to call out the **number** of the card pile that the child points to (1 or 2).	
2	Ask parent to help engage child's attention, and say: "Now we're going to play a game using these cards"	
3	Q. "What is this?" Q. "What colour is it?" A. "Yes, it's a blue rabbit".	
4	Q. "What is this?" Q. "What colour is it?" A. "Yes, it's a red boat".	
5 *Colour Game*	"This is the colour game. In the colour game, all the blue ones go here *[click x2]* and all the red ones go there *[click x2]*."	
6	"Here is a **blue** one, so it goes here *[click]*"	
7	"So if it's a blue one it goes here *[click x2]*, and if it's a red one it goes there *[click x2]*.	
8	"Now here is a **red** one. Where does this one go?" Correct: "Yes, very good. You know how to play the colour game." Incorrect: "No, this one's red. It goes over here in the colour game *[click]*	
10 *Pre-Switch Phase*	"Okay, now it's your turn. Remember, if it's blue it goes here *[click x2]* and if it's red it goes there *[click x2]*. Here's a **red** one *[click]*. Where does it go?" Whether or not child responds correctly, say "Let's do another one". DO NOT GIVE ANY FEEDBACK AT ANY POINT; DO NOT SAY OKAY.	
12	"Remember, if it's blue it goes here *[click x2]* and if it's red it goes there *[click x2]*. Here's a **blue** one *[click]*. Where does it go?" … "Let's do another one"	

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21 *Researcher Manual*

14	"Remember, if it's blue it goes here *[click x2]* and if it's red it goes there *[click x2]*. Here's a **blue** one *[click]*. Where does it go?" … "Let's do another one"	
16	"Remember, if it's blue it goes here *[click x2]* and if it's red it goes there *[click x2]*. Here's a **red** one *[click]*. Where does it go?" … "Let's do another one"	
18	"Remember, if it's blue it goes here *[click x2]* and if it's red it goes there *[click x2]*. Here's a **blue** one *[click]*. Where does it go?" … "Let's do another one"	
20	"Remember, if it's blue it goes here *[click x2]* and if it's red it goes there *[click x2]*. Here's a **red** one *[click]*. Where does it go?" … "Let's do another one"	
21	END OF PRE-SWITCH PHASE: ○ If child scored 5/6 correct, move onto next phase. ○ If child scored 4 or less correct, end game here.	
22 *Post-Switch Phase*	"Now we're going to play a new game. We're not going to play the colour game anymore. We're going to play the shape game. In the shape game, all the rabbits go here *[click x2]* and all the boats go there *[click x2]*."	
23	"Remember, if it's a rabbit it goes here *[click x2]* and if it's a boat it goes there *[click x2]*. Here's a **rabbit** *[click]*. Where does it go?" … "Let's do another one".	
25	"Remember, if it's a rabbit it goes here *[click x2]* and if it's a boat it goes there *[click x2]*. Here's a **boat** *[click]*. Where does it go?" … "Let's do another one".	
27	"Remember, if it's a rabbit it goes here *[click x2]* and if it's a boat it goes there *[click x2]*. Here's a **rabbit** *[click]*. Where does it go?" … "Let's do another one".	
29	"Remember, if it's a rabbit it goes here *[click x2]* and if it's a boat it goes there *[click x2]*. Here's a **boat** *[click]*. Where does it go?" … "Let's do another one".	
31	"Remember, if it's a rabbit it goes here *[click x2]* and if it's a boat it goes there *[click x2]*. Here's a **boat** *[click]*. Where does it go?" … "Let's do another one".	
33	"Remember, if it's a rabbit it goes here *[click x2]* and if it's a boat it goes there *[click x2]*. Here's a **rabbit** *[click]*. Where does it go?" … "Let's do another one".	

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End of Games

- Thank parent and child, check in/listen to feedback
- Remind family can keep toys and we will send feedback report

After call has ended

- Save local recording to participant's folder
- Fill in your codes for the session and work out BPVS score
- Ensure kit (visualiser, BPVS) is accessible for the next visit/researchers

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Appendix 4.8: *SIB study coding crib sheet*

SIB Study Executive Function Tests – Coding Crib Sheet

Written by Katherine Marlow

Coding

Coding in SPSS

- 888 = task was purposefully not administered
  - E.g., child was not eligible to play Game 4, so Game 4 was not attempted
- 999 = true missing data
  - E.g., a big ball is lost during Game 4, so 6/6 balls could not be sorted
  - E.g., any materials are missing or lost preventing full game play
  - E.g., task discontinued

How to Score

- Open a new Participant Scoring Sheet (excel) and save against SIB ID number
- Transfer codes to Master SPSS file once coded / scores checked if needed

Game 1/5

- Time latency to touch toys using available timer e.g., iPhone timer (time twice to ensure you have the most accurate latency)

Games 3, 4 and 8

- Note the predetermined starting point (based on child's overall age equivalent - <2 years = Game 3, >2 years = Game 4)

Validity Checks – Scoring

- 0 = No / fail (indicates poorer validity)
- 1 = Yes / pass (indicates stronger validity)
- A total validity score can be calculated based on items 1-8, item 9 allows specification of any other validity concerns
- N.B. be careful of the wording – 0 is always a fail, and 1 is always a pass (e.g., V5 is worded as 'did child ***not*** hold item in hand' to keep the coding consistent across checks)
- Scores of **less than 4** (i.e., 3 or less) are deemed too low to be 'valid'. In the first instance such examples should be considered by the broader SIB study team (e.g., PI and/or collaborator(s). Scores of three or below should not be included in SIB Study analyses.

Validity Checks

V1: Was the child in the room when the instruction was given?
Examples of where V1 would be scored as 'yes' (1):

- Researcher can see that child was in the room when task instruction was given

Example of where V1 would be scored as 'no' (0):

- Child cannot be seen on camera to be in the room during instruction delivery
- Child is not in/leaves the room when instruction is given

V2: Did the child attend to the instruction?
Examples of where V2 would be scored as 'yes' (1):

- Child looks to the caregiver and/or looks at the task materials when instruction is delivered
    - e.g., looks at wand reveal/stickers going under cups/ball and box demonstrations/array reveal
- Child may provide verbal or non-verbal feedback to indicate they heard/understood task instructions
    - E.g., nods
- *Note – child does not need to have followed instructions to show they were listening.*
- *Note that children's listening may not be best represented by neurotypical eye contact*

Example of where V2 would be scored as 'no' (0):

- Child was not facing caregiver when instruction given
- Child was walking around room when instruction given
- Child was talking, signing, singing or doing something else which indicates they were not attending to caregiver's voice during instruction delivery
- Caregiver verbally indicates that they do not think the child was attending to the instruction delivery

V3: Did the researcher deliver the instructions correctly?
N.B. "Correctly" means everything that must have been administered was, and nothing that shouldn't be administered wasn't. Note that there is some allowance – words need not be copied verbatim from manual, acknowledging the need to adapt and 'keep' family on side (e.g., not repeating rules if child asks to stop repeating them), however key task instructions must have still been delivered in order to pass.

Examples of where V3 would be scored as 'yes' (1):

Wording:

- Researcher delivered task instructions following Researcher Manual, using the approved wording or similar – so long as key task instructions were communicated, and correct wording was given when imperative
  E.g., ...
  - Game 1/5: researchers asked parent to say "don't touch"
  - Game 2: researcher asked "can you find **ANOTHER** sticker" on all but the first search
  - Game 3/4: rules clearly communicated
- Researcher correctly told caregiver when/when not to deliver praise.
  E.g., ...
  - Game 2: praise throughout, both when child is correct or incorrect
  - Game 3/4: praise during Game 3 Pt 1, Game 4 Pt 1, Game 4 Pt 3 (demonstrations), no praise during Game 3 Pt 2, Game 4 Pt 2, Game 4 Pt 4
- Researcher delivered praise / withheld praise during appropriate times of DCCS (i.e., praised and corrected training cards, did not praise during pre- and post-switch trials)
  - A note on 'praise' – comments on effort allowed, comments on performance are not

Order:

- Researcher delivered task instructions following Researcher Manual, in the correct order. E.g., ...
  - Game 3/4: balls sorted in the standardised order
  - Game 5: toys revealed in standardised order

Set-up:

- Researcher directed caregiver to set up task appropriately
  E.g.,
  - Game 1/5: told caregiver not to let child touch toy(s) during reveal, mentioned they should hold toy(s) close to chest if child does reach out
  - Game 2: told caregiver to set up the cups in the correct order, ensure all cups were adequately hidden from child's view when A3 card is used
  - Game 3/4: told caregiver to set up boxes shoulder width apart *(exception – when camera angle is too narrow to spread boxes out without losing sight of boxes/child)*, told caregiver to hand balls centrally

Example of where V3 would be scored as 'no' (0):

Wording:

- Researcher did not deliver task instructions following Researcher Manual, using the approved wording or similar – key task instructions were not communicated, correct wording was not given when imperative, and incorrect wording was given
  E.g.,
  - Game 2: researcher did not ask parent to say **"another"** on all but the first search
  - Game 3/4: rules were clearly communicated
- Researcher did not correctly advise when/when not to deliver praise. E.g., …
  - Game 2: did not advise to give praise throughout
  - Game 3/4: did not advise praise during Game 3 Pt 1, Game 4 Pt 1, Game 4 Pt 3 (demonstrations), did not say **no** praise during Game 3 Pt 2, Game 4 Pt 2, Game 4 Pt 4
- Researcher did not deliver praise / withhold praise during appropriate times of DCCS (i.e., did not praise and correct during training cards, did praise/correct/said 'okay' during pre- and post-switch trials)

Order:

- Researcher did not deliver task instructions following Researcher Manual; instructions or materials in the wrong order, e.g., …
  - Game 3/4: researcher gives wrong order to sort balls in (i.e., non-standardised)
  - Game 5: researcher gives wrong order to reveal toys in (i.e., non-standardised)

Set-up:

- Researcher did not direct caregiver to set up task appropriately, e.g., …
  - Game 1/5: did not tell caregiver to not let child touch toy(s) during reveal
  - Game 2: did not tell caregiver to set up the cups in the correct order, did not instruct caregiver to adequately hide cups with A3
  - Game 3/4: did not tell caregiver to set up boxes shoulder-width apart *(exception – when camera angle is too narrow to spread boxes out without losing sight of boxes/child, so long as caregiver does not obviously prime or cue child to correct answer)*, did not tell caregiver to hand balls centrally

V4: Did the caregiver deliver the instructions correctly?

Examples of where V4 would be scored as 'yes' (1):

- Caregiver repeats what researcher asks them to, does not laugh or add their own interpretation of instructions
- **N.B. V4 is always scored as a pass for DCCS, even if parent prompts/intervenes (in this case, fail V7)**

Example of where V4 would be scored as 'no' (0):

Note: when to score zeros for V3 and V4

* If researcher delivered instruction correctly, and caregiver did not, score V3 = 1 and V4 = 0 *(e.g., if researcher told caregiver not to praise during Game 3 Pt 2, but the caregiver gave praise anyway, you may score V3 = 1, V4 = 0).*

* If researcher delivered instruction incorrectly, and caregiver also delivered instruction incorrectly, score 0 for both V3 and V4 *(e.g., if researcher did not tell the caregiver not to praise during Game 3 Pt 2, and caregiver gave praise during this trial, score a zero for both V3 and V4).*

* (Less likely) – if researcher delivered instruction incorrectly, and caregiver delivered task correctly, score V3 = 0, V4 = 1 *(e.g., if researcher forgets to tell caregiver not to praise, but caregiver does not praise, V3 = 0, V4 = 1).*

- Caregiver does not repeat what researcher asks them to, laughs or adds their own interpretation of instructions

Caregiver Praise

- Caregiver praises child at any inappropriate time (i.e., Game 3 Pt 2, Game 4 Pt 2, Game 4 Pt 4, DCCS trials)
- Caregiver does not praise when they should (i.e., Game 3 Pt 1, Game 4 Pt 1, Game 4 Pt 3)

Set up

- Tasks set up inappropriately
    - Game 1/5: child touched toy(s) during reveal
    - Game 2: cups not set up in the correct order, move around during the duration of the trial, cups not hidden completely from child view when obscured by A3 card
    - Game 3/4: boxes not shoulder-width apart *(exception – when camera angle is too narrow to spread boxes out without losing sight of boxes/child, so long as caregiver does not obviously prime or cue child to correct answer)*, balls not handed centrally

Caregiver gives any hints, cues or tips, e.g., …

- Game 2 – points towards a particular cup / names cup to search *("Can you find the orange sticker?", "Can you find the last sticker?")*
- Game 3/4 – points to, touches, hovers over correct box
- Games 1/5 - caregiver turns towards child during the 30/60 second waiting period and engages with child (talks, signs, sings, gestures, touches toy[s], touches child, re-delivers 'don't touch' instruction)
    - *Note – exception for children who show pica and are at risk of swallowing toys. In this instance, caregiver should sit with child in peripheral vision during waiting period for safety, but not interact with child or toys (e.g., talk, play, touch toys). This highlights the importance of safety and taking an adaptive/individualised approach to administering the tasks.*

- DCCS – any general context references to boats/rabbits/blue/red

Specific Game Examples

- Games 1/5:
  - Caregiver says "don't touch" or similar BEFORE the official 'don't touch' instruction is given (i.e., Step 2 of Games 1/5);
  - caregiver lets child touch toy(s) before Step 2
- Game 2:
  - Caregiver retrieves more than one sticker per search (e.g., countdown happens, child finds orange sticker which is removed, then child immediately finds blue sticker, and caregiver removes it rather than putting back and re-setting)
- Game 3/4:
  - Caregiver sorts balls in an order different to that stated by researcher (including when balls are lost during trial, e.g., roll under sofa, and so standardised order cannot be followed);
  - lids were open while child was sorting balls in Game 4

V5: Was the child not holding another object during the task?

Note – 'toy' used to represent any item, material, toy, device or object the child may play with or want to hold. Examples may include stuffed animal, tablet, cutlery, stationary, pets, etc.

Examples of where V5 would be scored as 'yes' (1):

- Child was not holding any toys unrelated to the present game during task
- Child picked up toy(s), but caregiver removed from child, then repeated instruction/continued with task

Example of where V5 would be scored as 'no' (0):

- Child held toys that were unrelated to the present game during task, and rejected any attempts to remove toy from hands (this may include when children help safety/comfort items)

V6: Were the instructions delivered without interference from additional caregivers?

Examples of where V6 would be scored as 'yes' (1):

- No other caregiver(s) or sibling(s) were present during task delivery
- Additional caregiver(s) or sibling(s) were present, but did not interfere verbally or physically during task

Example of where V6 would be scored as 'no' (0):

- Additional caregiver(s) or sibling(s) were present, and interfered verbally (e.g., reminded child of 'don't touch' instruction during the inhibition tasks; hinted verbally where to find stickers; hinted verbally where to put balls)
- Additional caregiver(s) or sibling(s) interfered physically (e.g., held child during Game 1/5, pointed towards 'correct' cup/box response)

V7: Did the parent/caregiver (i.e., person administering Games 1-5) not interfere when child was responding to an item?

Examples of where V7 would be scored as 'yes' (1):

- After instructions had been presented, parent/caregiver (i.e., person in the room with the child, administering task) did not interrupt child's response in anyway
- Allowed child to answer however they chose

Example of where V7 would be scored as 'no' (0):

- Caregiver interrupted or interfered with child's response in any way after instructions had been administered, e.g., …
    - Stopped child from responding how they wanted to, and re-directed them to answer differently *(e.g., prevented child from picking orange cup and told them to pick blue instead)*
- DCCS: parent comments on child's performance/offers feedback during pre- and post-switch trials of DCCS (*e.g., "that's right, well done!"*)

V8: Was the task delivered without any other validity concerns? (If no, please describe in next variable).

Examples of where V8 would be scored as 'yes' (1):

- No other validity concerns

Example of where V8 would be scored as 'no' (0):

- Other validity concern not better captured by V1 – V6, e.g.,
    - Parent did not wear headphones during task, so researcher's voices playing through device (i.e., child could hear researchers)

V9: Please specify any other validity concerns.

Free-text to describe why V8 was answered as "no". V9 is not included in the total validity score, rather it is an open space to record why V8 failed.

Appendix 4.9: *SIB study caregiver consent form*

Parent Guardian Consent Form Study 1 v3.0 24.03.21_clean
SIB Study
IRAS ID: 263048

Richards Lab | Medical Research Foundation | University of Birmingham | MRC Medical Research Council | CEREBRA Working wonders for children with brain conditions

Sleep - Impulsivity - Behaviour (SIB) Study

Site name: _____ Principal Investigator: _____

If you are reading this information on behalf of someone you care for who is a child under the age of 16 years, then we would like to ask you to decide whether or not you think that it is in your child's / person you care for's best interests for them to participate in the study. If you would like your child / person you care for to participate in this study, please complete this consent form.

Please initial box...

1. I confirm that I have read and understood the information sheet dated v3.1 12.04.21 for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐
2. I understand that the participation of my child / person I care for is voluntary and that we are free to withdraw at any time without giving any reason, without my or that of my child's / person I care for's medical care or legal rights being affected. I agree that data collected prior to withdrawal can still be used in the research analysis. ☐
3. I understand that all information collected during the study will be confidential. Participant names will not be published in any research reports. Information will be treated as strictly confidential and handled in accordance with the provisions of the General Data Protection Regulation 2018 (GDPR) and the Data Protection Act 2018. ☐
4. I understand that as part of the above study, a video recording will be made of me, my child and members of my family who are present at the time of the recording may also be captured on the video and/or audio. This footage will be captured via Zoom and the recording will be stored on the University's secure Research Data Store. I consent for this recording to be made and stored for further review. ☐
5. I understand that the University of Birmingham will hold the copyright of any video/voice recordings collected during the study but that this does not entitle the University of Birmingham to edit, copy or use the videos for teaching purposes without my written permission. ☐
6. I understand that relevant sections of my child's / person I care for medical notes and data collected during the study may be looked at by individuals from regulatory authorities, Sponsors and/or NHS bodies, where it is relevant to my child / person I care for taking part in this research. I give permission for these individuals to have access to my records. ☐
7. I understand that anonymised data collected during the study will be made available on an open science platform and that it will not be possible to identify any participant in the study from this data. I give permission for my child's anonymised data to be made available. ☐
8. I agree to my child's GP being informed of my child's participation in this study. ☐

Page **1** of **2**

OFFICE USE To be removed following return of consent form. **ID Number:**

9. I agree to the University of Birmingham research team sharing my research data with any health professionals or clinicians working with the person I care for. ☐

10. I agree that my child can take part in the above study. ☐

11. I am willing to administer the tasks as required in the above study. ☐

Optional clauses: The statements below are optional. Please delete Yes/No as appropriate:

1. I agree to being contacted with information about participating in future parts of the above study using the contact details I provide. Yes/No

2. I agree for my contact information to be kept so that study newsletters and feedback can be sent to me. Yes/No

3. I agree to the University of Birmingham research team using images and video recordings taken as part of the study for non-research purposes e.g. teaching, presentations, websites Yes/No

Please complete the details below

Your Name: _____

Address: _____

_____

Telephone Number 1: _____ Telephone Number 2: _____

Email: _____

Full Name of Your Child / Person You Care for: _____

Relationship to Participant: _____

Signature: _____Date: _____

To be completed at the play session:

Researcher responsible for confirming consent: _____

Signature: _____ Date:_____

Completed Consent Form Filing
Original completed consent forms will be filed in a secure and safe location at the University of Birmingham. One copy will also be stored in the Investigator Site File and one copy will be given to the parent/carer of the participant.

OFFICE USE To be removed following return of consent form. **ID Number:**

Appendix 4.10*: SIB study parent general information sheet*

Sleep - Impulsivity - Behaviour (SIB) Study

Games Session: General Information

Welcome to our study!

We know that the Covid-19 pandemic has been really tough for families, and so we are more grateful than ever for your support in our research. The SIB study gives you the opportunity to play a few games with your child and a researcher over Zoom. The researcher will guide you through each game using Bluetooth earphones, telling you how to play and when to move on to the next game. The researcher will help you to play the games step by step, but please make sure you are familiar with these instructions before starting the games session. If your child becomes upset whilst playing one of the games, the researcher will check whether you are both comfortable to go ahead. If not, the researcher will help you to stop the game or take a break. We hope that you and your child enjoy participating in this study, and thank you for your time!

Setting up

We know that each family will have their own preferences, and every device and room will be different. So, whilst we offer suggestions for setting up for the games session, please do things in a way that you are comfortable with, and suits you.

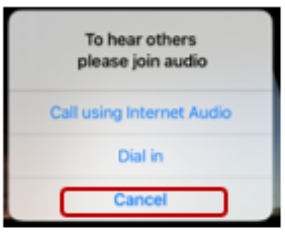
Setting up your device(s)

We ask that you use any device you own that connects to WiFi, has Bluetooth capability and supports Zoom. This could be a laptop, tablet or mobile phone (so long as the device has a webcam and speakers). You only need one device for this study, but if you have more than one suitable device (say a tablet and phone) a second device could be set up elsewhere in the room to give us the best view of the games.

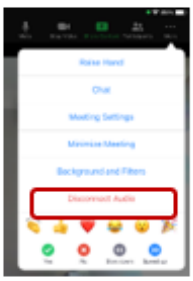
If you can use two devices, one will need to be muted and disconnected from audio (this avoids getting high-pitched feedback between devices).

You can easily do this one of two ways:

1. When joining the Zoom call, instead of connecting to audio, click 'Cancel'



2. When on the Zoom call, click 'More' then 'Disconnect Audio'



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Accessing Zoom

If you're using a laptop or computer, you don't need to install any software. All you will need to do is click on the link we send you nearer the time, which will take you directly to the Zoom call.

If you're using a tablet or mobile phone, we ask that you download the free Zoom App. Just type "Zoom" into the App or Google Play Store, and tap "Get" on the app called "**ZOOM Cloud Meetings**" (see screenshots to the right). You don't need to create a Zoom account, you just need to click on the link we will send you before the session.



Zoom link

A member of our team will email you with a link to the Zoom meeting close to the planned date. Don't worry if you don't receive this link until the day of the games session!
Once received, all you will need to do is click or tap on the link.

Earphones

The researcher will talk you through each game through wireless earphones. You may already have your own and would prefer to use those – if you do, that's great! Please check they connect to the device you plan to use. Otherwise, we can send you a pair of Bluetooth earphones. You will only need to wear **one** headphone during the games session.

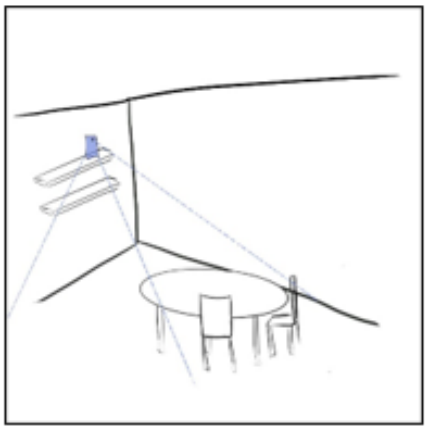
Suggestions for setting up the room

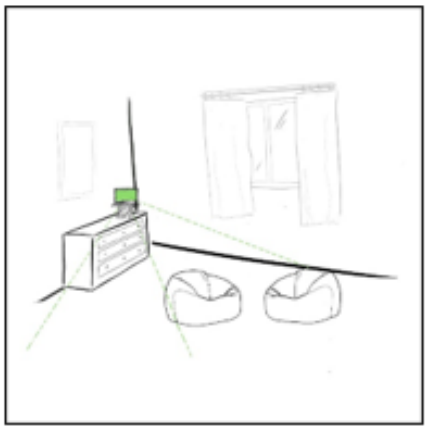
You can use any room for the games session. Please choose a room that will work best for you and your child. This may be a living room or play room. If you are playing the games at school, please use a room that is unoccupied by other children. You and the researcher will have the chance to discuss how to set up the device(s) in the room before the day of the games session.

We know no two rooms look the same, but when deciding where to put the device the most important thing is the camera(s) shoot an angle wide enough to capture you and your child play. If possible, this would be **in a corner, by the wall, propped up high and angled downwards** towards the games area. For example, you could safely prop the device on a wall shelf, on top of a table or on a bookshelf.

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Here are a couple of suggestions:





Your role

So that we can continue conducting essential research during Covid times, we are asking that you take on a 'researcher' role. This means we're asking you to play games directly with your child. While we hope you and your child find the games fun, there are some rules that we must follow to make sure we play the games properly:

- We ask that you follow the instructions given over the earphones. These will include physical actions (for example, the researcher may say to you *"Now pick up the magic wand toy"*) and verbal instructions for you to say to your child (for example, the researcher may say *"Now say to your child 'Can you find a car?'"*).
- Don't smile or laugh if your child doesn't respond how you think they 'should'

Please read on to the next page to learn more about the games…

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Overview of Games

During the games session, we will ask you to play some games with your child and the researcher. Not all of the games are appropriate for every child so the researcher will tell you which games to play. **It is important that you don't let your child see the toys before we start the play session.**

Most of the games can be played on the floor or table, with your child facing you. If helpful, another caregiver can help you to keep your child comfortable and engaged during the session. For Game 7, the researcher will lead the game and ask your child to point at pictures on a screen. We will need your help to tell us where your child is pointing.

When playing the games, please don't prompt your child unless asked to by the researcher – we are interested in seeing what they can do alone. Also, please don't smile or laugh if your child doesn't respond how you think they 'should'. Thank you for your help with our research!

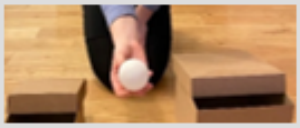
Game 1 – The Magic Wand
In this game, we are interested in how children respond to being asked not to do something.



Game 2 – Sticker Hunt
In this game, we are interested in how children remember things.



Game 3 – Time To Sort!
In this game, we are interested in how well children can follow the rules of a game.



Game 4 – Ball Sorting
In this game, we are interested in how well children can follow the rules of a game, and then adapt when the rules of the game change.



Game 5 – Bag of Toys
In this game, we are interested in how children respond to being asked not to do something.



Game 6 - Playtime
In this game, we are interested in how your child plays without our prompting!



Game 7 – Point To A Picture
In this game, we are interested in your child's understanding of words. We will ask them to point to some pictures on a screen.





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Appendix 4.11: *SIB study pre-visit checklist and risk assessment*

Ppt ID:	Examiner:	Date:	Status (Y/N)	Notes
Task Checks				
Have you read General Info & Overview of Games sheets?				
Any questions / obvious issues?				
Run through the games using kit to demonstrate				
Will there be 1 or 2 caregivers present during the assessment?				
Explain role and set expectations (i.e., we will feed instructions through headphones, parent should not to laugh/giggle)				
Risk Assessment				
Fill in Risk Assessment Qs on next sheet				
Room Checks				
Do you have / have you chosen a quiet room for play session?				
Can you show me the room?				
Devices				
What device(s) will you use? Where will you set these up?				
Will you be able to use device for the duration of the play session (i.e., if another family member would usually use iPad at this time, ensuring family				
Will you be able to charge the device(s) and headphones before or during the assessment?				
Do you have wireless Bluetooth headphones?				
If yes, check they pair with device				
If no, we will send you some!				
Other				
Was Wi-Fi stable during VABS call?				
Tips for boosting connection: moving closer towards router, closing down other internet pages, disconnecting from unnecessary VPNs				
Mention if possible to bring autism diagnosis letter to Games Session				
Booked in a time for Games Session / scoped what times may suit best?				
Ask if parent will be able to receive kit on the suggested postage date (if date hasn't been arranged, alert parent the kit will need signing for)				

Turn this box green once senior member of team has approved risk assessment prior to EF session.					NB: This must be completed following the VABS (PRIOR to the research assessment day). If any of these behaviours are present, please discuss with the team prior to further planning of the research assessment day.	Please use information from the CBQ, QABF and SRQ to fill in the shaded boxes prior to completing this risk assessment. Unshaded boxes need to be completed during the risk assessment, using the prompts provided.
Ppt ID:	**RISK ASSESSMENT (STUDY 1)**					
	Y/N	Longest burst of behaviour	Worst outcome of behaviour ever	Likely function(s)	Typical management of this behaviour (*ask parent what they do to stop behaviour e.g. distract child, change activity, use of restraint, protective clothing)*	Health and safety materials to be present *(e.g. if child shows head banging, protective padding)*
SELF-INJURY						
AGGRESSION						
DESTRUCTION						
STEREOTYPY						
OTHER (*normalise this for the parent e.g. "sometimes children remove their clothing or smear in public, it's just really helpful if we know about this so we can plan for it")*						
Does participant show **SELF-RESTRAINT** according to SRQ?						
If yes, ask parent how they manage this behaviour and what happens if child is prevented from restraining?						
*Please asterisk any behaviours which are managed by the use of restrictive/protective clothing or restraints.						

Appendix 4.12: *Overview of SIB study games parent booklet*

Sleep - Impulsivity - Behaviour (SIB) Study

Overview of Games

In this play session, we will ask you to play some games with your child and the researcher. Not all of the games are appropriate for every child so the researcher will tell you which games to play.

It is important that you don't let your child see the toys before we start the play session.

Most of the games can be played on the floor or table, with your child facing you. If helpful, another caregiver can help you to keep your child comfortable and engaged during the session.

For Game 7, the researcher will lead the game and ask your child to point at pictures on a screen. We will need your help to tell us where your child is pointing.

When playing the games, please don't prompt your child unless asked to by the researcher – we are interested in seeing what they can do alone. Also, please don't smile or laugh if your child doesn't respond how you think they 'should'.

Thank you for your help with our research!

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Game 1 – The Magic Wand
In this game, we are interested in how children respond to being asked not to do something.



Game 2 – Sticker Hunt
In this game, we are interested in how children remember things.



Game 3 – Time To Sort!
In this game, we are interested in how well children can follow the rules of a game.



Game 4 – Ball Sorting
In this game, we are interested in how well children can follow the rules of a game, and then adapt when the rules of the game change.



Game 5 – Bag of Toys
In this game, we are interested in how children respond to being asked not to do something.



Game 6 - Playtime
In this game, we are interested in how your child plays without our prompting!



Game 7 – Point To A Picture
In this game, we are interested in your child's understanding of words. We will ask them to point to some pictures on a screen.



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Game 1: The Magic Wand

What is it about?

In this game, we're interested in how children respond to being asked *not* to do something.

Let your child respond however they want – there are no right or wrong responses.

What you'll need...

- Glitter Wand } Found in: package labelled "Game 1"



Package

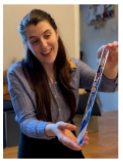
Magic wand

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How to play

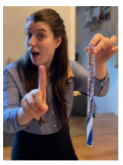
1. Attract your child's attention, then show them the glitter wand.

   Say: **"Oh look [say child's name]"**.



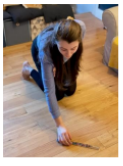
2. Say: **"Look [say child's name]. No, don't touch"**.

   Use your finger to emphasise 'no'.

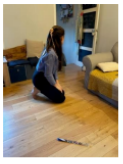


3. Put the wand down within your child's reach.

4. Turn around, away from your child.



5. The researcher will tell you when the game has finished and you can turn back towards your child.



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Game 2: Sticker Hunt

What is it about?

This is a memory game, where we're interested in learning about how children remember things.

What you'll need...

- 5 stickers
- 5 cups
- Laminated card
- Certificate

Found in: bag labelled "Game 2"

Found in: loose in between large and postage box


Package


Stickers


Cups


Laminated card

Getting started

From your child's perspective, line up the cups in the following order (left to right): orange, blue, purple, green, red.


Cup order

Have the stickers and sticker chart to hand.

Keep the toys out of your child's reach for this game.

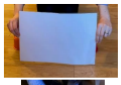
Copyright © 2021 Marlow, Agar, Richards & Devine

How to play

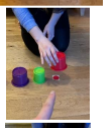
Make sure your child sees you do all of these steps – it is important they see your actions!



1. Attract your child's attention, then pick up any cup and put a matching-coloured sticker underneath it. Do this for all five cups.
   Put the red sticker under the red cup, the purple sticker under the purple cup, and so on.



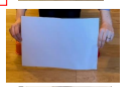
2. Cover the cups with the laminated card, **count out loud for 5 seconds,** then remove the card.
3. Say: **"Can you point to a sticker?"** to encourage your child to look for a sticker.



4. When your child finds any sticker (e.g., by gesturing towards or grabbing a cup), lift the cup and pick up the sticker. Praise them with phrases like: **"Well done!", "Good job!", "You got the sticker!"**.
5. Say: **"Look! Let's put this one over here"** and put it out of your child's sight.



6. Say: **"Look! Let's put the cup back!"** then stack the cup back upright (it is now the only cup without a sticker underneath).
7. Cover the cups with the card, **count out loud for 5 seconds,** then remove the card.
8. Say: **"Can you get another sticker?"** Do not use the word 'again'.



9. If your child finds another sticker, keep repeating steps 2 – 8.
   If your child gestures towards the same cup for a second time, do not smile or laugh. Instead:
   - Lift the cup to show it's empty.
   - Say: **"It's not there. Let's have another go."**
   - Stand the cup upright and say: **"Look! Let's put the cup back!".** Try again (repeat steps 2 – 8).



The researcher will tell you what to do, and when to finish the game.

You can then help your child stick the stickers on their certificate.

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Game 3: Time To Sort!

What is it about?

In this game, we are interested in how well children can follow the rules of a game, and then adapt when the rules of the game change. We will guide you through each stage of the game.

What you'll need...

- 2 boxes (1 big, 1 small)
- 1 large ball (from the bag of 6)
- 1 small ball (from the bag of 6)

Found in: bag labelled "Game 3/4"


Boxes

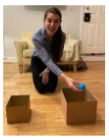

Balls

Place the two boxes in front of your child, with the **lids off**.

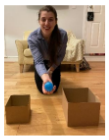
Have one large and one small ball ready.

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How to play



1. Attract your child's attention and say "**Oh [say child's name] look at this!**" as you put the big ball in the larger box. Then pick it back up.



2. Pass your child the big ball. Pass it in the middle of the two boxes so that you're not suggesting which box they should put it in. Say "**Now you try**".

   If your child places the ball in the larger box, this is correct. You can say "**Well done [say child's name]**"

   If they try to place the ball in the smaller box, say "**Oh no, the ball doesn't go in there!**" and remove it.

3. You'll now repeat this 6 times – each time pass your child a big ball and say "**Where does this go?**" without giving them a hint.

You don't need to keep count of how many times you've done this game, the researcher will prompt you in your headphones.

After 6 goes, the researcher will either stop the game to give your child a break, or ask that you start the second part of this game.

In the second part of the game, we will introduce a new rule, so that your child has to place a small ball in the smaller box. We will guide you through these steps, in the same order as above.

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Game 4: Ball Sorting

What is it about?

In this game, we are interested in how well children can follow the rules of a game, and then adapt when the rules of the game change.

If you played Game 3, it is very similar. We will guide you through each stage of the game.

What you'll need...

- 2 boxes (1 big, 1 small)
- 6 large balls
- 6 small balls

Found in: bags labelled "Game 3/4"



Boxes



Balls

Set up the two boxes in front of your child, with the **lids on**.

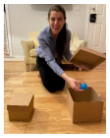
Have **both bags** of balls ready next to you.

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How to play

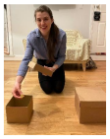
1. Attract your child's attention and say "**Oh [say child's name] look at this! Here I have a Big Box, and here I have a Little Box".**

   Demonstrate placing the balls in each box as you say **"In this game, the Big Balls go in the Big Box and the Little Balls go in the Little Box**" and close the lids.



2. Tell your child "**Now it's your turn to put the Big Balls in the Big Box and the Little Balls in the Little Box**".

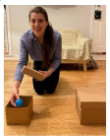
   The researcher will prompt you to ask your child to sort big and little balls, and will tell you when the game is finished. The researcher will let you know when you can give them feedback.



In the final part of the game, the Big Balls will be sorted into the Little Box and the Little Balls into the Big Box. We call this the Silly Game!

3. Tell your child "**Now we're going to play a Silly Game!"** Demonstrate placing the balls in each box as you say **"In this game, we put the Big Balls in the Little Box and the Little Balls in the Big Box"** and close the lids. Then remove the balls.

   The researcher will prompt you to ask your child to sort big and little balls, and will tell you when the game is finished. The researcher will let you know when you can give them feedback.





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Game 5: Bag of Toys

What is it about?

In this game, we're interested in how children respond to being asked not to do something.

Let your child respond however they want – there are no right or wrong responses.

What you'll need...

- Stuffed toy
- Toy car
- Squeezy ball toy

Found in: package labelled "Game 5"



Package



Toys

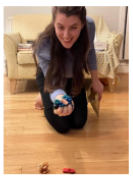
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How to play

1. Attract your child's attention by saying **"Oh look [say child's name]"** as you playfully reveal the cuddly toy, then the toy car, then the squeezy ball. Keep hold of all the toys.

   Once all three toys have been revealed and are in your hands, place them all down in front of your child.

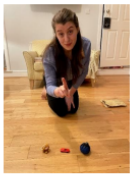
   If your child reaches for a toy before they are placed down, try to hold them out of their reach and say "**Not yet!**" / "**Hold on!**"



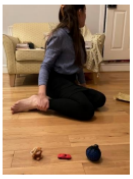
2. Say: **"Look [say child's name]. No, don't touch"**.

   Use your finger to emphasise 'no'.

3. Turn around, away from your child.



4. The researcher will tell you when the game has finished and you can turn back towards your child.



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Game 6: Playtime!

Now, we would like you and your child to take a break together and play in any way you want. You can use the toys we've sent you, or any other toys you may already have.

We will let you know when playtime is over.

Have fun! 😊

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Game 7: Point To A Picture

What is it about?

This game is all about children's understanding of language.

Getting ready…

Tell your child that you will both play one last game with the researcher. For this game, you won't need any toys – just yourselves!

You'll play this game with you and your child sat facing the laptop. You may need to move your device somewhere more suitable, like a table or desk.

Here is some guidance for getting Zoom ready for this game:

- **Disconnect your headphones** so that your device's sound plays through the speakers, and your child can hear the researcher.
- Make sure you have **expanded the Zoom call** to cover the whole screen, so that you have the best view of the game.

If you're using a tablet, this should be automatic (see screenshot)



If you're using a laptop, you can use the "expand" icons circled (see screenshot)



- **Switch to "Speaker View"** to make the researcher's screen appear much bigger, and give you a really clear view of the game.

On a tablet, click "Switch to Active Speaker" (usually in the top left corner, see screenshot)



On a laptop click "View" then select "Speaker View" (usually in the top right corner, see screenshot)



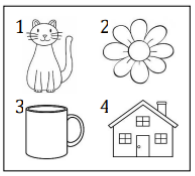
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How to play

1. The researcher will talk you both through the game, which involves the researcher saying a word and your child choosing a picture that best relates to that word from a selection of pictures.

   Please **tell us the <u>number</u>** of the picture that your child points to.

   For example, if your child pointed to the flower, you would say "two".



2. The researcher might continue onto the second part of this game, which is very similar. This part of the game involves your child choosing a picture based on its colour or shape.

   Again, please **tell us the <u>number</u>** of the picture you're your child points to.

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Appendix 4.13: *Live games session scoring sheet*

Participant ID		Overall Age Equivalent (VABS)	
Test Date		RL Age Equivalent (VABS)	
Name of scorer		Tick when seen autism diagnosis letter:	

	Name researcher administering	Start at	Attempted (Y/N/A)	Completed (Y/N)
Game 1: Wand		/		
Game 2: Cups & Stickers		/		
Game 3: Balls & Boxes				
Game 4: Ball Sort				
Game 5: Array		/		
Game 6: Free Play		/		
Game 7: BPVS				
Game 7: DCCS				

General Notes		
Time	Behaviour (e.g., SIB, self-restraint)	Notes

	Time to touch wand/toys (00:00.00)	Touch (Y/N)	Note any potential threats to validity	General Notes
Game 1				
Game 5				

Game 2	Number search	Note colour of cup tried (in order):	Was sticker retrieved? (Y/N)	Notes
	1			
Total number of stickers retrieved:	2			
	3			
	4			
	5			
Total number of searches:	6			
	7			
	8			
	9			
	10			

Game 3	Pass/Fail	Who places in ball?	Notes
Big			
Big			
Big			
Big			
Big			
Big			
Small			
Small			
Small			
Small			
Small			
Small			

	Game 4	Pass/Fail	Who places in ball?	Notes
Demonstration	Big			
	Small			
	Big			
	Small			
Normal game	Small			
	Big			
	Small			
	Big			
	Big			
	Small			
Demonstration	Big			
	Small			
	Big			
	Small			
Silly game	Small			
	Big			
	Small			
	Big			
	Big			
	Small			

Game 6 *(code FLACC later)*	
Notes	

Game 7: BPVS	Number of sets	Raw score, standardised score	Age equivalent	Top 3 words
(Use separate score sheet)				
Notes				

			Game 7: DCCS	
	Stimulus		Response	Pass/Fail
Pre-switch	Red	2		
	Blue	1		
	Blue	1		
	Red	2		
	Blue	1		
	Red	2		
If 5/6 correct…				
Post-switch	Rabbit	1		
	Boat	2		
	Rabbit	1		
	Boat	2		
	Boat	2		
	Rabbit	1		

Appendix 4.14: *SIB study feedback report*



ID_REF: [0000]

Report on [Child's Full Name]

DOB: 00/00/0000

Compiled by:

Catherine Laverty
Research Associate
Katherine Marlow
PhD Student
Dr Caroline Richards
Senior Lecturer in Neurodevelopmental Disorders
Clinical Psychologist

From
The School of Psychology
University of Birmingham

This report is based on the information collected about [Child's name] by [Researcher names] and Dr Caroline Richards as part of the research project 'Sleep-Impulsivity-Behaviour (SIB) Study'. The information included in the report does not comprise a comprehensive psychological evaluation nor has it necessarily focused on specific problems that might be experienced by the person. It is hoped that the report will be of use to carers and professionals involved with the individual by providing the results of these assessments.

Any questions arising from this report can be directed to Dr Caroline Richards*.

* Contact address and telephone number: 52 Pritchatts Road, School of Psychology, University of Birmingham, Edgbaston, Birmingham, B15 2TT - 0121 415 8098. E-mail: c.r.richards@bham.ac.uk

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REPORT SUMMARY

Name:
DOB: 00/00/0000

This report outlines the results of a number of assessments carried out with [Child's name] and his/her [insert relationship of informant]. Direct assessments of executive functioning, language and behaviour were completed with [Child's name] over video conferencing. An interview and questionnaires about a range of behaviours and skills were also completed by his/her [insert relationship of informant].

For some assessments, expected age equivalent scores are reported alongside [Child's name]'s scores, which can be used as a guide to aid understanding of [Child's name]'s scores. For example, if a person is reported to have an age equivalent score of 7 years old, this is not to suggest that they are the same as a 7 year old, only that we would expect the majority of typically developing 7 year olds to achieve a similar score.

Assessment Summary

The key information derived from these assessments is summarised below. For more information, please see the relevant sections.

- **Language and Adaptive Ability:** Assessments of adaptive functioning revealed that [Statement of any areas of relative strength and/or weakness.]

- **Executive Functioning:** [Child's name] [statement of any areas of relative strength and/or weakness.]

- **Behaviour and Social Communication:** [Qualitative description of the participant's overall behaviour, highlighting any challenging behaviour, self-restraint behaviour, adaptive behaviours of particular interest. Highlight any areas of relative strength and/or weakness.]

1. Language and Adaptive Ability

1.1 The British Picture Vocabulary Scales, Third Edition (BPVS-III; Dunn & Dunn, 2009)

Assessment Date: 00/00/0000

The BPVS assesses children's understanding of spoken words. We used the BPVS to assess [child's name]'s receptive language.

[Qualitative summary of BPVS results. Include an explanation of any areas of relative strength and/or weakness.]

1.2 Vineland Adaptive Behaviour Scales, Third Edition (VABS-III; Sparrow, Cicchetti & Saulnier, 2016)

Assessment Date: 00/00/0000

The VABS is used to obtain an overall indication of a person's level of ability on a day-to-day basis. The assessment takes into consideration an individual's communication, daily living, socialisation, and motor skills. This assessment was used to obtain an indication of [child's name]'s adaptive ability.

Table 1: [Child's name]'s developmental age on the VABS-III

Domain (*sub-domains*)	**Example of types of skills included in each subdomain.**	**[Child's name]'s developmental age**
Communication		
Receptive language	Understanding of what others are saying.	Xy Xm
Expressive language	The ability to verbally express wants, needs and feelings.	Xy Xm
Written language	A person's writing abilities	Xy Xm
Daily living skills		
Personal	Adaptive skills such as feeding, washing, dressing.	Xy Xm
Domestic	Domestic skills such as helping with household chores.	Xy Xm
Community	Skills such as using the telephone, carrying money in a safe place, telling the time, staying safe.	Xy Xm
Socialisation		

Interpersonal relationships	Includes understanding the dislikes of others, understanding personal space and non-verbal cues.	Xy Xm
Play and leisure time	Includes skills such as turn taking and game playing.	Xy Xm
Coping skills	Abilities such as repairing relationships, saying please and thank you.	Xy Xm
Motor skills		
Gross motor skills	Abilities such as climbing stairs or riding a bicycle	Xy Xm or *
Fine motor skills	Includes abilities such as unwrapping small sweets and manipulating small objects.	Xy Xm or *

*Motor skills age equivalents cannot be calculated for individuals over the age of 10

[Qualitative summary of VABS results: are domain scores low/high.as expected for age group. Include explanation of any areas of relative strength and/or weakness.]

2. Executive Functioning

Executive functioning refers to a set of brain processes that control and regulate our behaviour. These processes allow us to: a) start and stop behaviours appropriately (*for example*- turn taking in a back and forth game), b) move from one task to another and deal with changes in routine (*for example*- planning and changing your normal selection of clothes based on the weather), and c) hold things in our mind so we can update information for the current task (*for example*, remembering a list of instructions and carrying these out in the correct order). These processes are important for us to be able to learn and integrate new information.

2.1 The Prohibition Task (Friedman, 2011)

Assessment Date: 00/00/0000

The Prohibition Task measures a child's ability to inhibit or stop a natural or impulsive behaviour.

[Qualitative description of the child's performance on the task]

2.2 The Three Boxes Task (Miller & Marcovitch, 2015)

Assessment Date: 00/00/0000

The Three Boxes Task is used to assess a child's ability to hold information in mind, and then update it with new information. We used a variation where [child's name] was asked to find stickers hidden underneath cups.

[Qualitative description of the child's performance on the task.]

2.3 Cognitive Flexibility Tasks (Marlow, Agar, Richards & Devine, 2021)

Assessment Date: 00/00/0000

The cognitive flexibility tasks measure set-shifting behaviour, which is the ability to shift attention between one task and another. In this task, [child's name] was asked to learn a rule about sorting balls and then switch this rule.

[Qualitative description of the child's performance on the task]

2.4 The Dimensional Change Card Sort Task (Zelazo, 2006)

Assessment Date: 00/00/0000

The Dimensional Change Card Sort Task measures set-shifting behaviour by asking children to learn one rule for sorting cards, and then switch to playing the game with another rule.

[Qualitative description of the child's performance on the task]

2.5 The Activity Questionnaire (TAQ; Burbidge *et al.*, 2010)

Assessment Date: 00/00/0000

The TAQ is an informant-report questionnaire used to assess an individual's activity level. The measure has three subscales: overactivity, impulsivity and impulsive speech. A higher score on this subscale indicates a greater degree of overactivity, impulsivity and impulsive speech. The table below presents [child's name]'s scores and the mean scores for a group of individuals with autism.

Table 2: [Child's name]'s scores on the TAQ

TAQ Subscales	[Child's name]'s score	Average score of 214 individuals with autism aged 4-15 years
Overactivity	X	18 (0-36)
Impulsivity	X	16 (2-24)
Impulsive Speech*	X	6 (0-12)
Total Score	X	39 (4-72)

* A score on the impulsive speech subscale was not calculated for those individuals who were not verbal or for whom information regarding verbal ability was missing.

3. Behaviour and Social Communication

3.1 The Social Communication Questionnaire Lifetime Version (SCQ; Berument, Rutter, Lord, Pickles & Bailey, 1999)

Assessment Date: 00/00/0000

[Child's name]'s social communication skills were assessed using the SCQ, which is an informant measure suitable for use in individuals over the age of four years, with a range of intellectual ability. The table below presents [child's name]'s scores and the mean scores for a group of individuals with autism. A higher score on each subscale indicates greater difficulties in social interaction and social communication skills.

Table 3: [Child's name]'s scores on the SCQ

SCQ subscales	[Child's name]'s score	Average score of 215 individuals with autism aged 4-15 years
Social Interaction	X	6 (1-15)
Communication*	X	9 (1-13)
Repetitive behaviour	X	6 (0-8)
Total score	X	25 (7-38)

*A score on the communication subscale was not calculated for those individuals who were not verbal or for whom information regarding verbal ability was missing.

3.2 The Challenging Behaviour Questionnaire (CBQ; Hyman, Oliver & Hall, 2002)

Assessment Date: 00/00/0000

The CBQ is used to assess the presence of four types of challenging behaviour in people with intellectual disability. The table below indicates whether or not [child's name] was reported to show self-injurious behaviour, physical aggression, destruction of property or stereotyped behaviour in the past month and the proportion of individuals with autism who also showed these behaviours.

Table 4: [Child's name]'s scores on the CBQ

CBQ Subscales	[Child's name]'s behaviour YES/NO	Percentage of 215 individuals with autism aged 4-15 years who show behaviour
Self- Injurious Behaviour	X	40%

Physical Aggression	X	68%
Destruction of Property	X	58%
Stereotyped Behaviour	X	64%

3.3 The Self-Restraint Questionnaire (Oliver, 2009)

Assessment Date: 00/00/0000

The Self-Restraint Questionnaire is a questionnaire completed by an informant that asks about the presence and frequency of self-restraint behaviours such as sitting on hands and pulling items of clothing (e.g. jumpers) over the hands.

The Self-Restraint Questionnaire was completed by [child's name]'s [parent/carer] in order to identify any self-restraint behaviours that [child's name] engages in. These may occur in order to control or manage other behaviours.

[Qualitative description of any self-restraint behaviours that the participant was reported to show and the frequency of these behaviours.]

3.4 Face, Legs, Activity, Crying and Consolability (FLACC) Scale (Merkel, Voepel-Lewis, Shayevitz & Malviya, 1997)

Assessment Date: 00/00/0000

The FLACC scale allows a trained observer to code for potential indicators of pain based on a child's face and leg movements, general activity, crying and level of consolability. Each category is scored 0-2, with higher scores suggesting greater discomfort. This was coded by the research team during a time when [child's name] was playing freely, not engaging in a task. [Child's name]'s FLACC score was X, which indicates [qualitative description of pain indicators, suggestion of seeking appointment with health professional where appropriate].

Many thanks for participating in this study. We are very grateful to you for your time and effort and hope that you have found this report helpful. Please do not hesitate to contact us with any queries or if you need assistance interpreting the report (contact Dr Caroline Richards on c.r.richards@bham.ac.uk, 0121 415 8098 or Catherine Laverty on c.laverty@bham.ac.uk, 0121 414 7209).

For further information on aspects of behaviour relating to children with autism and/or intellectual disability, please see:

- Cerebra website https://www.cerebra.org.uk/
- FIND website http://www.findresources.co.uk/
- Challenging behaviour Foundation website https://www.challengingbehaviour.org.uk/
- Research Autism website http://www.researchautism.net/

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Appendix 4.15: *SIB study ethical approval*

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Dr Caroline Richards
52 Pritchatts Road, School of Psychology
University of Birmingham, Edgbaston
Birmingham
B15 2TT

Email: hra.approval@nhs.net

19 August 2019

Dear Dr Richards

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**Sleep-Impulsivity-Behaviour (SIB) Study: examining pathways to self-harm in children with autism and intellectual disability**
IRAS project ID:	**263048**
Protocol number:	**RG_19-087**
REC reference:	**19/SW/0141**
Sponsor	**University of Birmingham**

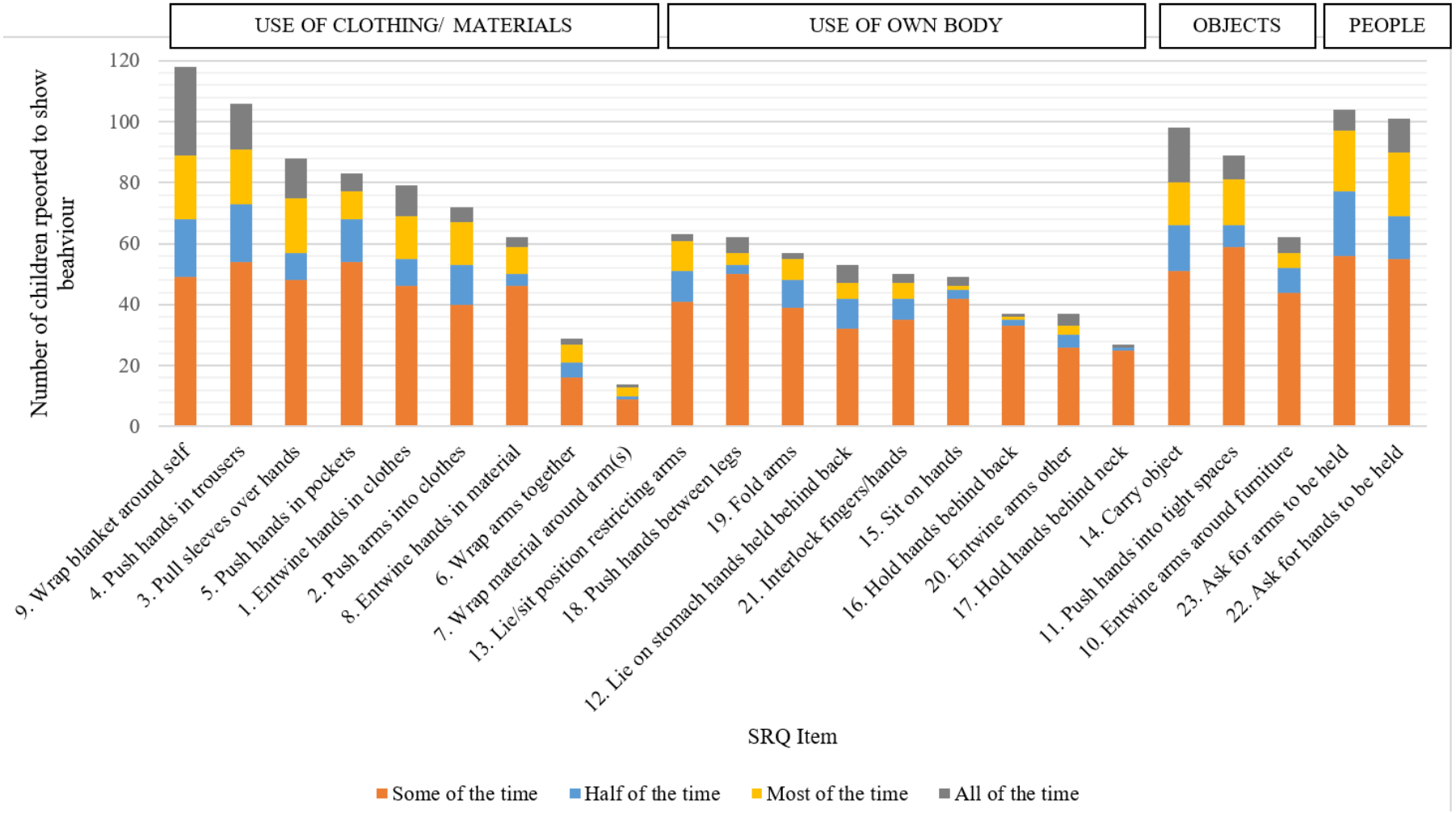
I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Appendix 4.16: *Distribution of data across SRQ items and subscales*



Appendix 5.1: *Naturalistic observation coding manual*

Coding Manual

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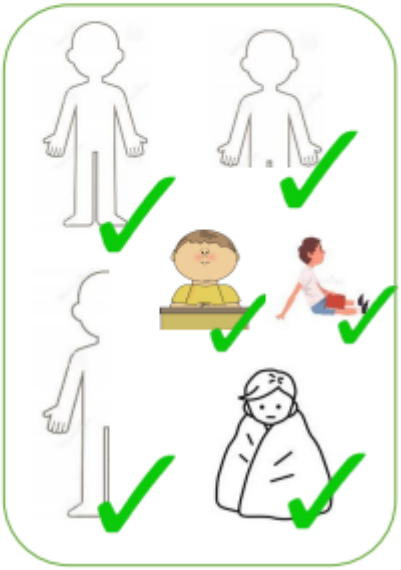
Stage 1: Coding duration child is visible on screen

Coding duration child is visible on screen

Set onset as the first second child is visible. Set offset as the last second before child is no longer visible. Repeat for full duration of video clip (note some participants have more than one video clip saved as individual files – code all clips).

Child is visible ('onset') when using all camera angles available you can see:

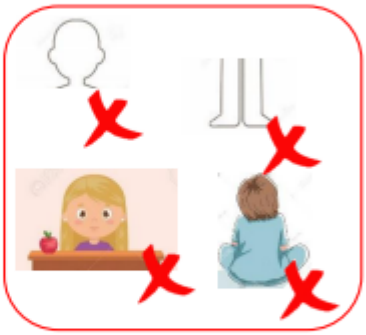
- One ***or*** both hands from any angle where you can tell what child is doing with their hands
  - E.g., sat at table with hands above table
  - E.g., can see child's fingertips and confident you know what child is doing with their hand
- Can't see hands because child is restraining them
  - E.g., sat on hands, folding arms, push hands into clothing, holding items, holding other people, pushing hands in-between legs, holding child's hand(s), holding hand(s) behind head
- Child is in full camera shot and wrapped in a blanket/material
- Child is seeking hugs/physical affection from caregiver but you can't see their hand(s) / child hugs caregiver and cannot see hands
- You can see item that child is holding (even if can't see child's hand)



Essentially, if self-restraint was happening, would you be able to see it from this angle?
Don't have to see: Face, Head, Legs, Feet

Child is *not* visible ('offset') when:

- You cannot see child's hands / what child is doing with their hands (and child is not self-restraining)
  - E.g., child is off screen, caregiver or objects are blocking view of child, child is facing away from camera restricting view of arms/hands, child is sat at table with arms beneath table, child has hand inside box
- Video footage freezes
- Lighting of video is too dark to tell what child is doing with their hands
- Clip is too short to be meaningful
  - e.g., you see a flash of a hand/fingers for less than one second

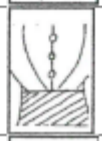
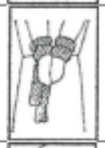
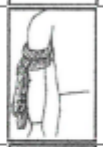


2

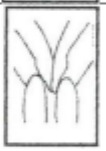
Stage 2: Coding self-restraint

Behaviour

N.B. If behaviour observed does not fall under items listed below but appears to be self-restraining, add as an additional item and label with following number

1	Entwines hand(s) in the front part of the clothes that are worn (e.g., t-shirt, jumper)	
2	Pulls arm(s) out of sleeves and pushes them part or all of the way into the main body of the clothes that are worn	
3	Pulls sleeve(s) of their clothes over hands	
4	Pushes hand(s) inside trousers, skirt, shorts or similar (i.e., clothing on lower half), push hands inside/up trouser legs	
5	Pushes hand(s) inside pockets in their clothing	
6	Wraps their hands or arms <u>together</u> using any piece of long material (e.g., sock, scarf)	
7	Wraps a long piece of material (e.g., sock, scarf) around either or both arms but not so that they are held together	
8	Wraps or entwines hand(s) in material other than own clothes e.g., curtains, bed clothes, other clothing, comfort/safety blanket	
9	Wraps a blanket, sheet, towel or other large piece of material around themselves	

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10	Entwines arms around a chair or other piece of furniture	
11	Pushes hands into chair or sofa cushions or other tight spaces, pushes part of or whole body under tight space/furniture e.g., floor rug	
12	Lies on stomach with hands held behind back	
13	Lies or sits in a position which restricts the movement of arm(s) e.g., with arms under the body, behind the back or pressed against a wall except when lying on front and looking at device screen (leaning)	
14	Carries an object(s) around but doesn't manipulate, use or play with them	
15	Sits on hand(s)	
16	Holds hands together behind back	
17	Holds hands together behind neck	
18	Pushes hands between own legs with legs closed (indicating effort to 'trap' hands in-between legs) or sat crossed-leg with hand(s) pushed in tight space behind knee. Excluding when pushing on floor to stand up.	
19	Folds arms excluding when folding arms and resting/leaning on table.	

20	Entwines arms with each other in any other way not described above – e.g., as shown in picture, crossing/wrapping arms behind one another	
21	Interlocks fingers or hands including grasping own hand or wrist and hand-over-hand grasp, excluding when interlocking fingers while squeezing squeezy ball (or other toys)	
22	Asks or otherwise indicates to others that they want their hands to be held, e.g., puts hands into another's or under another's arm, excluding when trying to retrieve item from others' hand, excluding when others initiate (record from first indication of desire to end of hand-hold)	
23	Asks or otherwise indicates to others that they want their arms to be held e.g., takes another's arms and puts it around themselves	
24	Restraint device worn (e.g., headgear, gloves)	
25	Hold hands together above head, must grab wrists or interlock fingers with arms resting on top of head	
26	Entwine self (any body part) in others' clothing e.g., grabs others' t-shirt	
27	Grabs/holds another's body part e.g., holds others' hair or body, clasps hands behind others' neck, grabs others wrist with both hands, entwines arms around others' arm	

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Stage 3: Coding behaviours before and after self-restraint

CAVEAT – when jumping back and forward in video clip child might not be visible – account for this by using an N/A code when child is completely out of shot for full 30 seconds

- Open Stage 2 DataVyu file and re-save as SIB000_3
- Add two more columns each with three codes –
  - a (S, A, D, R) (antecedent – SIB, aggression, destruction of property, self-restraint)
  - c (S, A, D, R) (consequence – SIB, aggression, destruction of property, self-restraint)



- Jump back 30 seconds and watch for SIB, aggression, destruction of property and self-restraint, then…
- Jump forward 30 seconds from off-set of behaviour and watch for SIB, aggression, destruction of property and self-restraint

If child is visible for some or full 30 seconds:

- If SIB is observed, code S as Y ('yes') followed by the number item observed
  - *E.g., if child bites hard object, code S as YS1*
- If SIB is not observed, code S as N ('no')
- If two or more SIB behaviours shown during those 30 seconds preceding SR, code all
  - *E.g., if child bits hard object twice, code S as YS1, YS1*
- Repeat for aggression, destruction of property and self-restraint

If child is not at all visible 30 seconds prior to SR onset / there is no existing video footage prior to or after the self-restraint (i.e., child restrains for the full duration of the clip):

- Code S ('SIB'), A ('aggression'), D ('destruction of property') and R ('other self-restriant') as N/A – this indicates behaviours were not codable

Before self-restraint

Did any of the following behaviours occur 30 seconds before the first onset of self-restraint?

SIB

Item	Code	Definition
S1	Bite hard object	Child clenches jaw and teeth around a solid object (e.g., wood, chair, table). Must use top and bottom teeth and definite biting movement.
S2	Bite self	Child clenches jaw and teeth around own body part
S3	Cheek press	Child forcefully pushes thumbs into cheek
S4	Eye press	Child pushes fingers on eye, pulls or pokes eyelashes
S5	Finger bite	Child clenches jaw and teeth on side of finger
S6	Hand bite	Child clenches jaw and teeth on hands or arms
S7	Hand to hand hit	Child forcefully brings hands together, palm to palm
S8	Hand to object hit	Child forcefully makes contact with hard surface with palm of hand
S9	Head bang	Child forcefully makes contact with head against hard surface
S10	Head hit	Child forcefully makes contact with fist and/or open palm on their head
S11	Hit self	Child forcefully makes contact with fist and/or open palm on own body part
S12	Leg slap	Child forcefully makes contact on leg with palm of hand
S13	Mouth hit	Child forcefully makes contact on mouth/around mouth with palm of hand
S14	Scratch self	Child drags nails across or pinches their own body part
S15	Skin pick	Child scrapes skin using forefinger at right angles to the surface of skin
S16	Teeth hit	Child forcefully makes contact with top teeth against object
S17	Wrist hit	Child forcefully makes wrist to wrist contact
S18	Pulls	Pulls hair or skin
S19	Choke self	Child wraps hand(s) around neck and squeezes, and/or constricts neck using clothing worn or other material

Aggression

Item	Code	Definition
A1	Hit other with body part	Child forcefully hits other using a body part e.g., hit or slap by quickly striking other with hand, punching, head-butting other anywhere on body
A2	Kick other	Child strikes foot out, making contact with other
A3	Bite other	Child clenches jaw and teeth around other's body part
A5	Pull other	Child holds onto other's body or clothes, moving them towards themselves, excluding when pull directs caregiver towards object
A6	Push other	Child reaches for other's body or clothes, makes contact and moves them away from themselves
A7	Rub, pinch or scratch other	Child forcefully rubs hands against other's skin, pinches other's body part, or drags nails across other's body part
A8	Spit	Child spits at or on other
A9	Hit other with object	Child throws object at other or uses object as a tool directed at other
A10	Verbal aggression	Child verbalises towards caregiver/researcher using threatening language, verbal swearing, swearing with hands

Destruction of Property

Item	Code	Definition
D1	Bite or chew item	Biting or chewing small items (e.g. books, clothing, testing materials)
D2	Tear or rip item	Tearing or ripping small items (e.g. books, clothing)
D3	Throw, kick or smash item	Throwing, kicking, or smashing small items (e.g. cups, mobile phones) excluding ball when engaging in game-play
D4	Hit object	Slamming, hitting, or kicking doors, walls or windows
D5	Smash or throw item	Tipping, smashing or throwing large items (e.g. furniture, televisions), hitting or punching device
D6	Pull item	Pulling items from walls or shelves

Other self-restraint

Refer to codes listed in Stage 2

After self-restraint

Did any of the following behaviours occur 30 seconds after the final offset of self-restraint?

SIB

Item	Code	Definition
S1	Bite hard object	Child clenches jaw and teeth around a solid object (e.g., wood, chair, table). Must use top and bottom teeth and definite biting movement.
S2	Bite self	Child clenches jaw and teeth around own body part
S3	Cheek press	Child forcefully pushes thumbs into cheek
S4	Eye press	Child pushes fingers on eye, pulls or pokes eyelashes
S5	Finger bite	Child clenches jaw and teeth on side of finger
S6	Hand bite	Child clenches jaw and teeth on hands or arms
S7	Hand to hand hit	Child forcefully brings hands together, palm to palm
S8	Hand to object hit	Child forcefully makes contact with hard surface with palm of hand
S9	Head bang	Child forcefully makes contact with head against hard surface
S10	Head hit	Child forcefully makes contact with fist and/or open palm on their head
S11	Hit self	Child forcefully makes contact with fist and/or open palm on own body part
S12	Leg slap	Child forcefully makes contact on leg with palm of hand
S13	Mouth hit	Child forcefully makes contact on mouth/around mouth with palm of hand
S14	Scratch self	Child drags nails across or pinches their own body part
S15	Skin pick	Child scrapes skin using forefinger at right angles to the surface of skin
S16	Teeth hit	Child forcefully makes contact with top teeth against object
S17	Wrist hit	Child forcefully makes wrist to wrist contact
S18	Pulls	Pulls hair or skin
S19	Choke self	Child wraps hand(s) around neck and squeezes, and/or constricts neck using clothing worn or other material

Aggression

Item	Code	Definition
A1	Hit other with body part	Child forcefully hits other using a body part e.g., hit or slap by quickly striking other with hand, punching, head-butting other anywhere on body
A2	Kick other	Child strikes foot out, making contact with other
A3	Bite other	Child clenches jaw and teeth around other's body part
A5	Pull other	Child holds onto other's body or clothes, moving them towards themselves, excluding when pull directs caregiver towards object
A6	Push other	Child reaches for other's body or clothes, makes contact and moves them away from themselves
A7	Rub, pinch or scratch other	Child forcefully rubs hands against other's skin, pinches other's body part, or drags nails across other's body part
A8	Spit	Child spits at or on other
A9	Hit other with object	Child throws object at other or uses object as a tool directed at other
A10	Verbal aggression	Child verbalises towards caregiver/researcher using threatening language, verbal swearing, swearing with hands

Destruction of Property

Item	Code	Definition
D1	Bite or chew item	Biting or chewing small items (e.g. books, clothing, testing materials)
D2	Tear or rip item	Tearing or ripping small items (e.g. books, clothing)
D3	Throw, kick or smash item	Throwing, kicking, or smashing small items (e.g. cups, mobile phones) excluding ball when engaging in game-play
D4	Hit object	Slamming, hitting, or kicking doors, walls or windows
D5	Smash or throw item	Tipping, smashing or throwing large items (e.g. furniture, televisions), hitting or punching device
D6	Pull item	Pulling items from walls or shelves

Other self-restraint

Refer to codes listed in Stage 2

Appendix 5.2: *Intensity of each topography of self-restraint observed (%)*

	Percentage of time visible spent engaging in self-restraint topography																
ID	**1**	**3**	**4**	**5**	**9**	**13**	**14**	**15**	**16**	**17**	**18**	**19**	**21**	**22**	**23**	**26**	**27**
001													0.10				
002	0.10							2.80			1.47	0.30					
003	0.80										1.20	0.35	3.40				
004														14.40	0.30	0.41	
005					16.80	2.20				0.04	0.15	0.59				0.53	0.06
006																	
007													0.20				
008						7.10		0.30									1.04
009							4.30			0.10		23.76	0.43				
010												0.40	0.90	1.02			
011	0.40												0.50	4.36		0.46	
012	1.30																1.40
013						1.00					1.80	3.21					
014	1.50					0.40								1.07			
015												0.50	0.20				0.10
016				0.10				2.80		0.18	0.45	1.52	3.84	0.32			0.14
017	0.80		3.40			0.18			0.15	0.46	0.60	2.86	1.05				
018														8.40			3.00
019											11.30		0.10	0.13			
020													12.80				
021		3.30											9.60				
022													0.20				
023																	
024												0.05	0.70				

Note. Red = very low intensity (<1%), orange = low intensity (1 – 24.99%), yellow = moderate intensity (25 – 49.99%), green = high intensity (50 – 97.99%), blue = very high intensity (98 – 100%) and white = topography not observed. Codes 2, 6, 7, 8, 10, 11, 12, 20, 24 and 25 excluded from table section as topographies were not observed.

Appendix 5.2 cont.

	Percentage of time visible spent engaging in self-restraint topography																		
ID	**1**	**4**	**5**	**8**	**9**	**13**	**14**	**15**	**16**	**17**	**18**	**19**	**20**	**21**	**22**	**23**	**25**	**26**	**27**
025						0.50						0.50							
026				0.20						2.20				0.87					0.07
027																			
028						0.50						0.10		6.95					
029		0.50												0.50			0.14		
030						0.20	0.50							1.25					
031														3.20					
032						0.50						0.10	4.14	3.83					
033				2.90	4.60			0.22				0.60							
034	0.10											0.70		1.61	1.40	1.00		0.30	0.57
035						1.00								0.10					
036			0.04		3.50	1.84						1.43							
037																			
038						7.40						1.30							
039						4.70													
040	0.80														0.60				
041						4.00						7.60		0.90					
042						0.40					1.80					0.12			
043						2.00													
044				0.90	10.60	2.64													
045														1.10					
046															0.20				0.30
047															0.30				0.50
048									0.20	1.00				0.16					1.24
049						0.20												1.80	
050														0.80					
051																			
052						2.00					1.20	1.03		0.59	0.25				

Note. Red = very low intensity (<1%), orange = low intensity (1 – 24.99%), yellow = moderate intensity (25 – 49.99%), green = high intensity (50 – 97.99%), blue = very high intensity (98 – 100%) and white = topography not observed. Codes 2, 3, 6, 7, 10, 11, 12 and 24 excluded from table section as topographies were not observed.

Appendix 5.2 cont.

	Percentage of time visible spent engaging in self-restraint topography																		
ID	**1**	**3**	**4**	**8**	**9**	**11**	**13**	**14**	**15**	**16**	**17**	**18**	**19**	**21**	**22**	**23**	**24**	**25**	**26**	**27**
053	0.50	0.40					1.19		0.56				0.10	0.93						
054				0.20			0.20											0.08		0.23
055	1.00								0.20					0.27	20.00					
056														0.30						
057															0.80				0.20	
058														1.30						0.60
059				0.20										0.30						
060						0.30	1.80		2.05		0.21	0.81		0.28						
061							2.40							8.20						
062							0.80													
063														2.00						
064													8.30	0.80			99.68			
065			2.50	18.60										0.52	1.24					
066													0.60							
067					94.50						0.30		2.46	0.08		14.89				
068														2.00						
069																				
070					13.70										0.70					
071													5.30	2.50						
072	0.30							7.30					0.30	4.71						
073										3.00				2.00	0.32					
074	3.10																			
075						0.50	0.50							0.38						4.29
076			0.30											0.02						0.40
077															0.90					1.60

Note. Red = very low intensity (<1%), orange = low intensity (1 – 24.99%), yellow = moderate intensity (25 – 49.99%), green = high intensity (50 – 97.99%), blue = very high intensity (98 – 100%) and white = topography not observed. Codes 2, 5, 6, 7, 10, 12 and 20 excluded from table section as topographies were not observed.

Appendix 5.2 cont.

	Percentage of time visible spent engaging in self-restraint topography																		
ID	**1**	**3**	**5**	**8**	**9**	**11**	**13**	**14**	**16**	**17**	**18**	**19**	**21**	**22**	**23**	**24**	**25**	**26**	**27**
078																			
079										4.20	0.40								
080		0.40									0.50								
081	1.90		1.50						0.29			0.22	0.96						
082							100									100			
083					62.00														
084													1.10				0.50		
085												1.70							
086						7.90	7.20							0.87	1.82				2.33
087	0.40						1.20				7.73	0.54	3.13						
088						1.20	5.00			0.36									
089																			
090													0.20						
091					97.60		20.90					0.18	2.83						
092																			
093													0.30						4.00
094				47.60	1.50								0.08	0.91					
095										0.60			0.30				0.84		
096							0.40							3.50	0.30			6.41	13.65
097	0.30											0.30							
098							0.20					1.20							
099										0.10									
100				2.90				43.10						5.07				1.01	15.76
101						1.20		0.80						1.50					
102													0.60	0.10					0.38
103				17.50			0.20							0.11					0.25
104																			

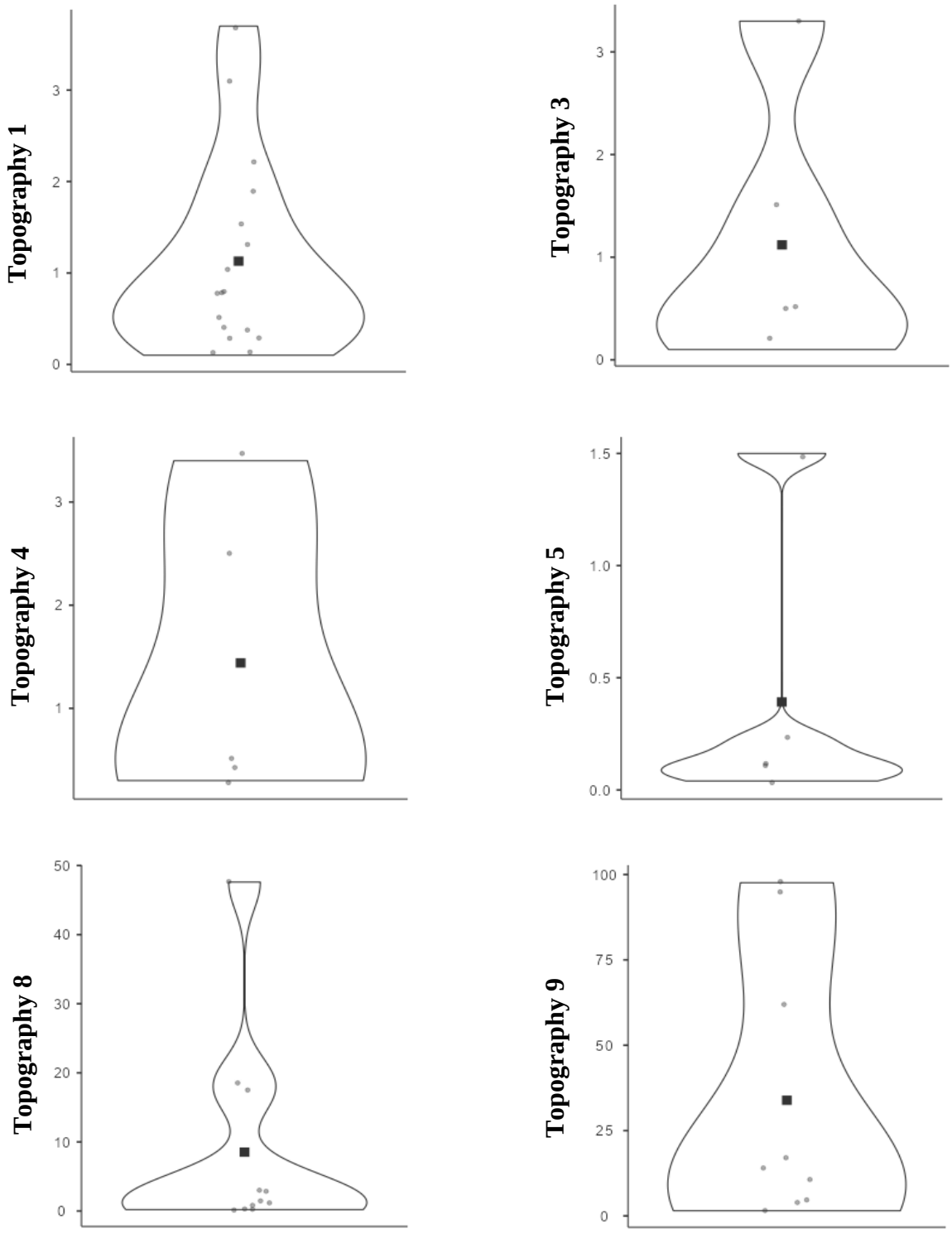
Note. Red = very low intensity (<1%), orange = low intensity (1 – 24.99%), yellow = moderate intensity (25 – 49.99%), green = high intensity (50 – 97.99%), blue = very high intensity (98 – 100%) and white = topography not observed. Codes 2, 4, 6, 7, 10, 12, 15 and 20 excluded from table section as topographies were not observed.

Appendix 5.2 cont.

	Percentage of time visible spent engaging in self-restraint topography																	
ID	**1**	**2**	**3**	**4**	**5**	**8**	**10**	**11**	**13**	**16**	**17**	**18**	**19**	**21**	**22**	**25**	**26**	**27**
105												1.10	2.30	0.48				
106														0.50				
107													0.10					
108	2.20													2.20	0.20			
109								2.70	0.10				0.35		10.78			
110											0.02	2.00	0.18	5.05				
111						1.50			2.00			0.08	0.64		0.21	0.32		0.43
112				0.50											0.60			2.00
113									6.30									
114					0.10		3.10		0.70	0.73			0.78					
115																		
116			0.10						0.10					0.13				
117									0.60									
118														0.70				
119																		
120																		
121		3.50	1.40		0.22									1.46				
122						1.20												
123																		
124	3.70								0.70								0.47	
125										0.20								
126														0.10				

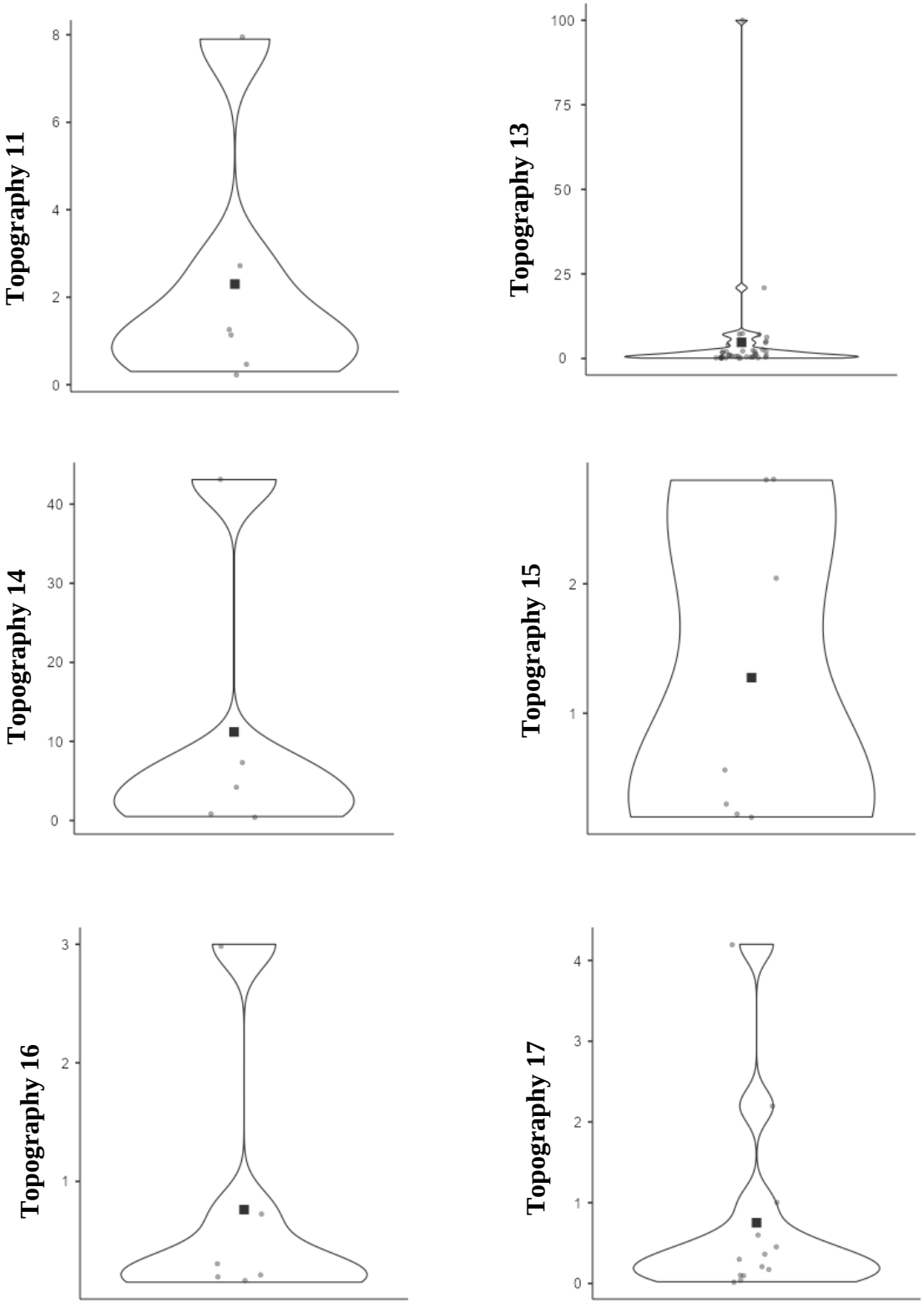
Note. Red = very low intensity (<1%), orange = low intensity (1 – 24.99%), yellow = moderate intensity (25 – 49.99%), green = high intensity (50 – 97.99%), blue = very high intensity (98 – 100%) and white = topography not observed. Codes 6, 7, 9, 12, 14, 15, 20, 23 and 24 excluded from table section as topographies were not observed.

Appendix 5.3: *Violin plots of observed self-restraint intensity proportions*

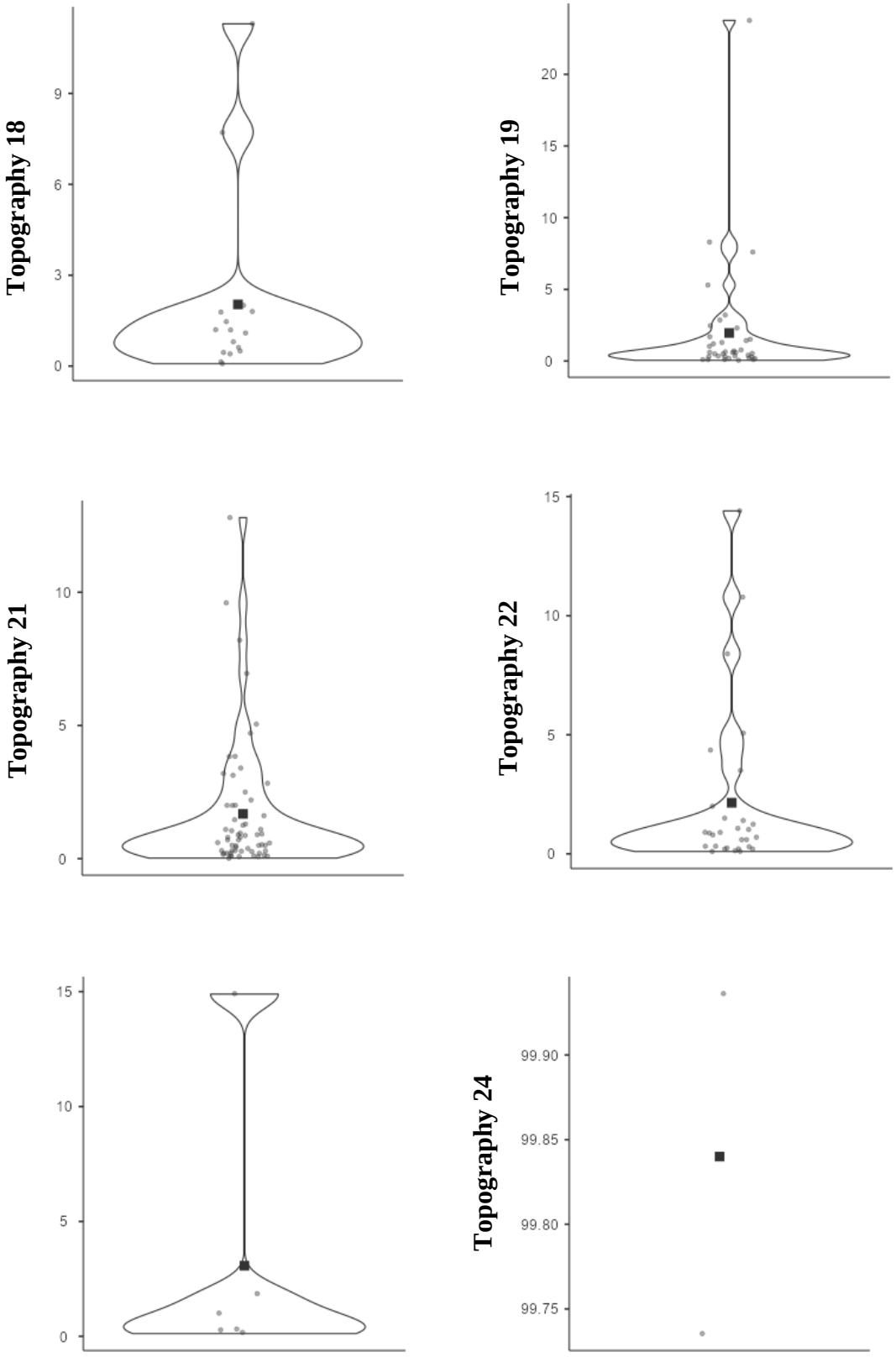


Note. Black squares indicate mean intensity score, grey dots represent individual data points and line shape represents density.

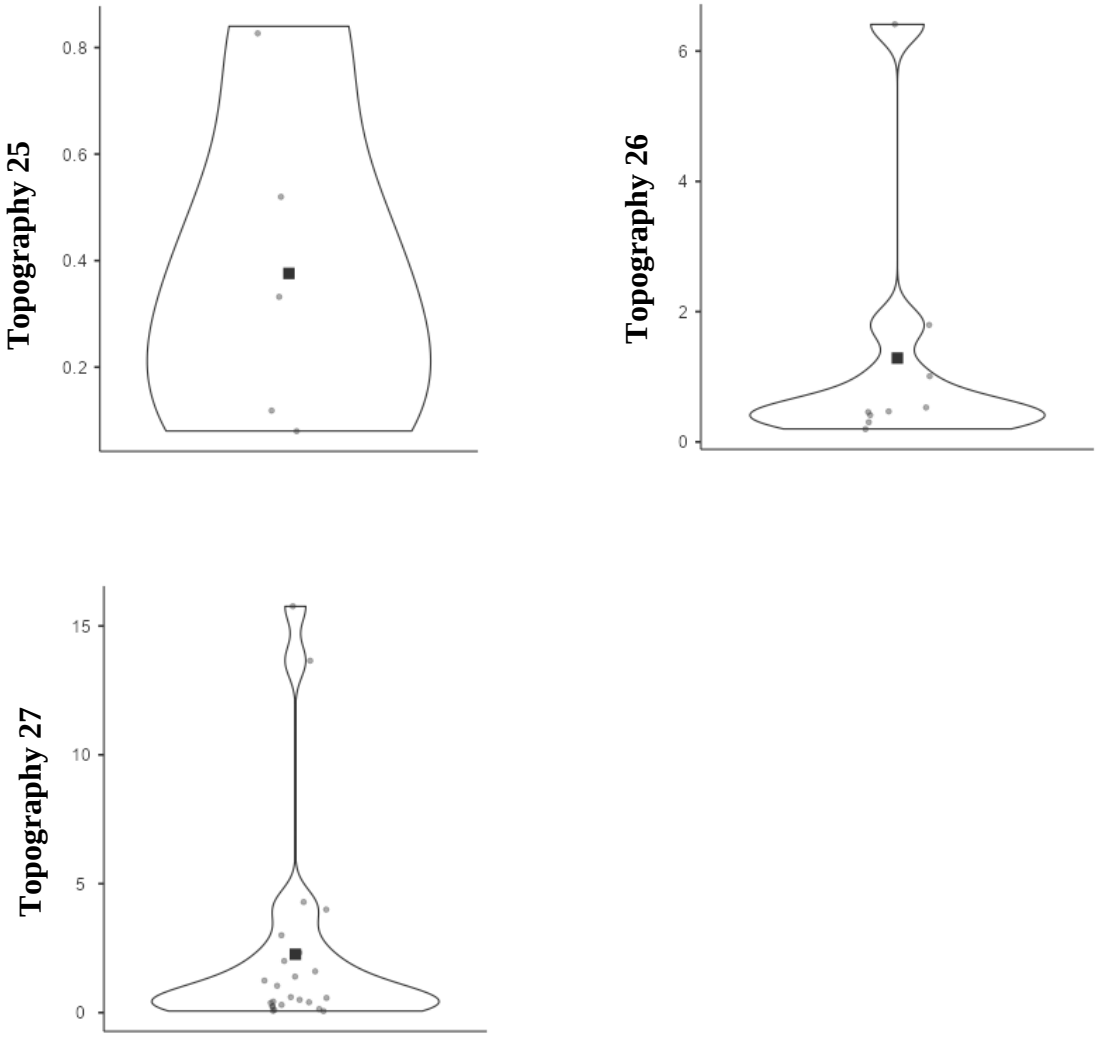
Appendix 5.3 cont.



Appendix 5.3 cont.



Appendix 5.3 cont.



Note. Plots for topographies 2, 6, 7, 10, 12, and 20 not produced due to lack of variance (n=1 or n=0).

Appendix 5.4: *Observation intensity internal consistency values*

Item Reliability Statistics

	if item dropped
	McDonald's ω
Item1	0.364
Item2	0.359
Item3	0.379
Item4	0.373
Item5	0.308
Item8	0.372
Item9	0.376
Item10	0.350
Item11	0.368
Item13	0.350
Item14	0.376
Item15	0.346
Item16	0.361
Item17	0.335
Item18	0.340
Item19	0.323
Item20	0.382
Item21	0.364
Item22	0.355
Item23	0.353
Item24	0.385
Item25	0.375
Item26	0.363
Item27	0.351

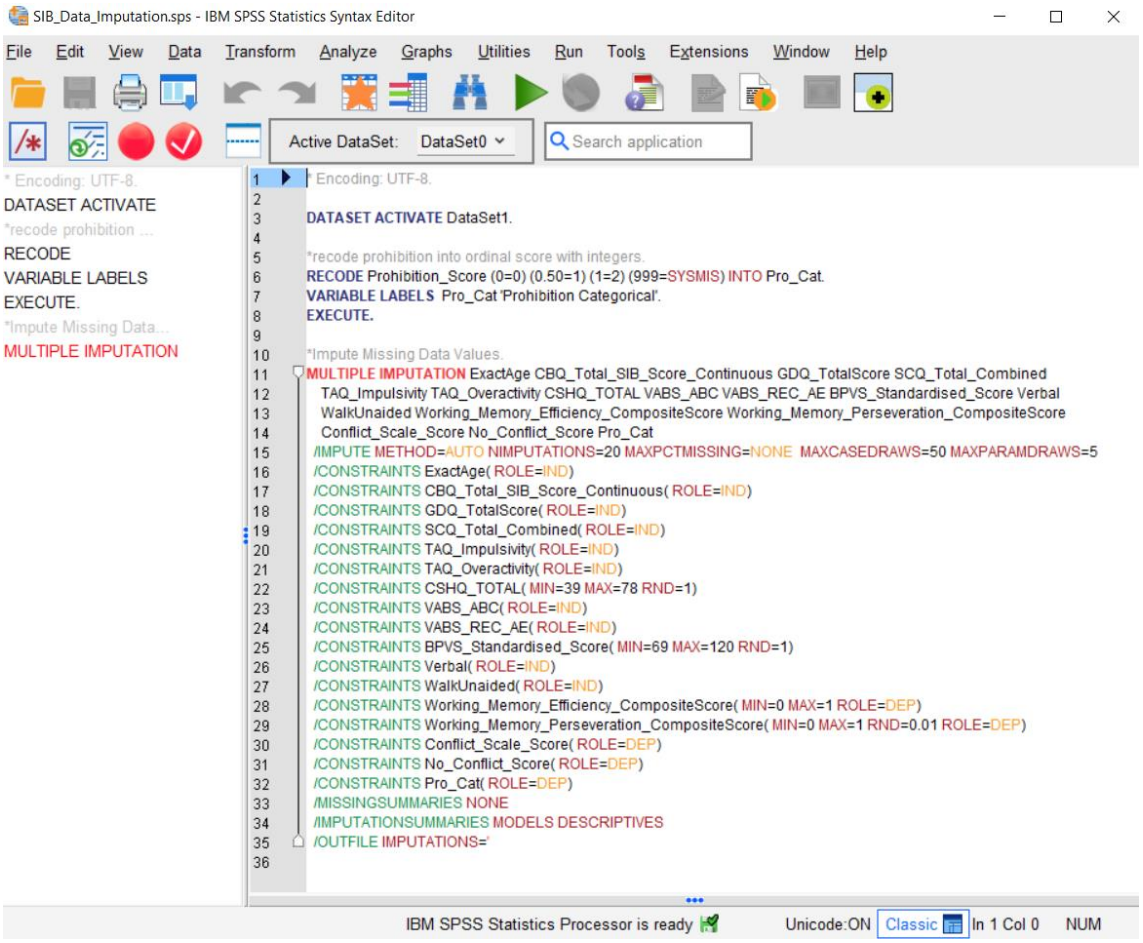
Note. Items refer to self-restraint codes. Codes 6, 7 and 12 excluded due to no item variance (i.e., behaviours not observed)

Appendix 6.1: *Demographic characteristics of the SRQ2 subsample*

Developmental variables	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Chronological age (years)	69		10.01 (9.70)	5.09 – 15.49 (2.96)
Sex (female/male)	69	16/53 (23/77)		
Mobility (walk unaided/unable)	69	65/4 (94/6)		
SCQ total (all participants)	69		26.48 (26)	16 – 35 (4.47)
SCQ total verbal		38 (55)	26.58 (26)	16 – 35 (5.40)
SCQ total non-verbal		31 (45)	26.35 (27)	20 – 32 (3.06)
Autism description	68			
Autism spectrum disorder		35 (51)		
Autism		30 (44)		
High functioning autism		2 (3)		
Autistic spectrum		1 (1)		
VABS-III ABC	67		44.25 (40)	20 – 87 (15.87)
VABS-III receptive language age equivalent (months)	67		20.70 (19)	0 – 67 (11.65)
ID severity	69			
Mild		5 (7)		
Moderate		21 (30)		
Severe		28 (41)		
Profound		3 (4)		
Unknown		11 (16)		
Other		1 (1)		
BPVS raw score [a]	35		69.46 (69)	2 – 142 (39.71)
BPVS standardised score [b]	35		74.46 (69)	69 – 118 (11.58)
BPVS age equivalent (months) [c]	21		81.71 (74)	45 – 137 (26.68)

Note. SCQ = Social Communication Questionnaire; VABS-III ABC = Vineland Adaptive Behavior Scale, Third Edition Adaptive Behavior Composite.
[a] BPVS discontinued for n = 34; [b] Includes n = 24 that scored lower than the average of a normed sample (70), indicated as '69'; [c] Age equivalents incalculable for n = 16 as raw scores fell below normed sample scores.

Appendix 6.2: *SPSS imputation syntax for performance-based executive function composite scores*



Appendix 6.3: *Behavioural characteristics of SRQ2 subsample*

Behavioural variables	**Possible score range**	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Self-restraint - SRQ		69			
Monthly self-restraint/no SR			62/7 (90/10)		
Total SRQ intensity score	0 – 92			14.75 (13)	0 – 84 (13.30)
Behaviours of concern – CBQ					
Monthly SIB/no current SIB		69	47/22 (68/32)		
Total SIB severity score	0 – 14			5.72 (7)	0 – 14 (4.51)
Monthly AGG/no current AGG		69	46/23 (67/33)		
Total AGG severity score	0 – 14			5.07 (5)	0 – 14 (4.44)
Monthly DoP/no current DoP		69	51/18 (74/26)		
Total DoP severity score	0 – 14			5.58 (6)	0 – 14 (4.27)
Monthly STPY/no current STPY		65	58/7 (89/11)		
Total STPY severity score	0 – 14			7.54 (8)	0 – 14 (3.68)

Note. SRQ = Self-Restraint Questionnaire; SR = self-restraint; CBQ = Challenging Behaviour Questionnaire; SIB = self-injurious behaviour; AGG = aggression; DoP = destruction of property; STPY = stereotypy.

Appendix 6.3 cont.

Behavioural variables	**Possible score range**	***N***	***n* (%)**	**Mean (median)**	**Range (SD)**
Activity – TAQ					
Impulsivity subscale	0 – 24	69		20.27 (22)	0 – 24 (4.69)
Overactivity subscale	0 – 36	69		24.58 (26)	4 – 36 (8.26)
Impulsive speech subscale	0 – 12	39		6.03 (6)	0 – 12 (4.08)
Total score	0 – 72	69		48.27 (50)	12 – 72 (12.95)
Executive functions – CHEXI					
Inhibition factor	0 – 55	68		46.21 (47)	11 – 55 (2.82)
Working memory factor	0 – 65	68		53.57 (55)	13 – 65 (9.86)

Note. TAQ = The Activity Questionnaire; CHEXI = Childhood Executive Functioning Inventory.

Appendix 6.4: *Chi-square tests comparing SIB and no SIB groups on presence of self-restraint topographies*

SRQ item	SIB group	Topography *N*		Test statistic	*p*
		No	Yes		
1. Entwine hands in clothes	1	24	15	1.815	.178
	2	62	64		
2. Push arms into clothes	1	24	15	0.556	.456
	2	69	57		
3. Pull sleeves over hands	1	21	18	1.058	.304
	2	56	70		
4. Push hands in trousers	1	14	25	0.000	.983
	2	45	81		
5. Push hands in pockets	1	23	16	1.758	.185
	2	59	67		
6. Wrap arms together	1	34	5	0.797	.372
	2	102	24		
7. Wrap material around arm(s) [a]	1	38	1	-	.191
	2	113	14		
8. Entwine hands in material	1	22	8	0.139	.710
	2	27	12		
9. Wrap blanket around self	1	12	18	2.294	.130
	2	9	30		
10. Entwine arms around furniture	1	21	9	0.005	.945
	2	27	12		
11. Push hands into tight spaces	1	17	13	0.429	.512
	2	19	20		

Note. Group 1 = No SIB, Group 2 = SIB; SRQ = Self-Restraint Questionnaire.
[a] Fisher's exact test employed due to behaviour containing group n<5, no test statistic available (indicated by dash).

Appendix 6.4 cont.

SRQ item	SIB group	Topography *N*		Test statistic	*p*
		No	Yes		
12. Lie on stomach hands behind back	1	25	5	1.267	.260
	2	28	11		
13. Lie/sit restricting arms	1	19	11	0.626	.429
	2	21	18		
14. Carry object	1	17	13	0.750	.387
	2	18	21		
15. Sit on hands	1	24	6	1.020	.313
	2	27	12		
16. Hold hands behind back [a]	1	26	4	-	.430
	2	32	7		
17. Hold hands behind neck [a]	1	29	1	-	.105
	2	33	6		
18. Push hands between legs	1	20	10	0.049	.825
	2	25	14		
19. Fold arms	1	25	5	2.443	.118
	2	26	13		
20. Entwine arms other [a]	1	26	4	-	.169
	2	29	10		
21. Interlock fingers/hands	1	22	8	0.020	.887
	2	28	11		
22. Ask for hands to be held	1	14	16	0.145	.704
	2	20	19		
23. Ask for arms to be held	1	9	21	3.152	.076
	2	20	19		

Note. Group 1 = No SIB, Group 2 = SIB; SRQ = Self-Restraint Questionnaire.
[a] Fisher's exact test employed due to behaviour containing group n<5, no test statistic available (indicated by dash).

Appendix 6.5: *Fisher-Freeman-Halton exact tests comparing presence of self-restraint topographies across four SRQ2 groups*

SRQ item	SRQ2 group	Topography *N*		Fisher-Freeman-Halton exact test statistic	*p*
		No	Yes		
1. Entwine hands in clothes	1	4	1	7.240	.060
	2	14	11		
	3	13	4		
	4	8	14		
2. Push arms into clothes	1	4	1	2.382	.495
	2	15	10		
	3	12	5		
	4	11	11		
3. Pull sleeves over hands	1	4	1	5.788	.122
	2	13	12		
	3	12	5		
	4	8	14		
4. Push hands in trousers	1	1	4	3.746	.298
	2	14	11		
	3	7	10		
	4	7	15		
5. Push hands in pockets	1	2	3	1.031	.830
	2	13	12		
	3	10	7		
	4	10	12		
6. Wrap arms together	1	5	0	.825	.922
	2	21	4		
	3	15	2		
	4	18	4		
7. Wrap material around arm(s)	1	5	0	2.883	.430
	2	22	3		
	3	16	1		
	4	22	0		

Note. SRQ = Self-Restraint Questionnaire; SRQ2 = Self-Restraint Questionaire 2. Group 1 = ‘no self-restraint/no SIB’; Group 2 = ‘no self-restraint/yes SIB’; Group 3 = ‘yes self-restraint/no SIB’; Group 4 = ‘yes self-restraint/yes SIB’.

Appendix 6.5 cont.

SRQ item	SRQ2 group	Topography *N*		Fisher-Freeman-Halton exact test statistic	*p*
		No	Yes		
8. Entwine hands in material	1	4	1	1.003	.833
	2	18	7		
	3	13	4		
	4	14	8		
9. Wrap blanket around self	1	2	3	3.037	.374
	2	10	15		
	3	5	12		
	4	4	18		
10. Entwine arms around furniture	1	5	0	7.254	.053
	2	16	9		
	3	15	2		
	4	12	10		
11. Push hands into tight spaces	1	5	0	6.051	.111
	2	12	13		
	3	10	7		
	4	9	13		
12. Lie on stomach hands behind back	1	5	0	5.698	.109
	2	20	5		
	3	15	2		
	4	13	9		
13. Lie/sit restricting arms	1	5	0	9.909	.015
	2	14	11		
	3	13	4		
	4	8	14		
14. Carry object	1	2	3	8.569	.028
	2	15	10		
	3	12	5		
	4	6	16		
15. Sit on hands	1	4	1	6.113	.095
	2	20	5		
	3	15	2		
	4	12	10		

Note. SRQ = Self-Restraint Questionnaire; SRQ2 = Self-Restraint Questionaire 2. Group 1 = ‘no self-restraint/no SIB’; Group 2 = ‘no self-restraint/yes SIB’; Group 3 = ‘yes self-restraint/no SIB’; Group 4 = ‘yes self-restraint/yes SIB’.

Appendix 6.5 cont.

SRQ item	SRQ2 group	Topography *N*		Fisher-Freeman-Halton exact test statistic	*p*
		No	Yes		
16. Hold hands behind back	1	5	0	3.464	.309
	2	21	4		
	3	16	1		
	4	16	6		
17. Hold hands behind neck	1	5	0	4.290	.195
	2	24	1		
	3	16	1		
	4	17	5		
18. Push hands between legs	1	4	1	2.395	.508
	2	16	9		
	3	13	4		
	4	12	10		
19. Fold arms	1	4	1	3.783	.257
	2	21	4		
	3	13	4		
	4	13	9		
20. Entwine arms other	1	5	0	4.710	.178
	2	21	4		
	3	15	2		
	4	14	8		
21. Interlock fingers/hands	1	4	1	.521	.976
	2	18	7		
	3	13	4		
	4	15	7		
22. Ask for hands to be held	1	2	3	.410	1.000
	2	12	13		
	3	9	8		
	4	11	11		
23. Ask for arms to be held	1	1	4	5.342	.146
	2	8	17		
	3	11	6		
	4	9	13		

Note. SRQ = Self-Restraint Questionnaire; SRQ2 = Self-Restraint Questionaire 2. Group 1 = ‘no self-restraint/no SIB’; Group 2 = ‘no self-restraint/yes SIB’; Group 3 = ‘yes self-restraint/no SIB’; Group 4 = ‘yes self-restraint/yes SIB’.