A think-aloud study on the feasibility of using the ICECAP-SCM in patients with end stage organ failure and the impact of functional decline on capability wellbeing at the end of life.

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Abstract

An ageing population and the rising costs associated with end of life care have led to increased scrutiny of the economic decision-making processes at the end of life. Critics argue that the normative framework that underpins the economic evaluation of health care interventions may be inappropriate for evaluating end of life care interventions. A broader evaluative space, that goes beyond health to capture the wider benefits of end of life interventions may provide a more appropriate basis for evaluating end of life care. One option for operationalising this broader evaluative space is the use of the ICECAP-SCM which is a self-complete tool for evaluating health and wellbeing at the end of life. There is, however, little evidence about its use in patients near the end of life.

This thesis reports a PhD study that aimed to determine the feasibility of using the ICECAP-SCM alongside the more traditionally used EQ-5D-5L, and the ICECAP-A in patients with end stage organ failure. The thesis also aims to explore the impact of functional decline on health and broader aspects of wellbeing. This is the first study to investigate how the three tools measure quality of life in end stage organ failure, and explore the impact of decline on wellbeing using the three measures.

A systematic review of the literature showed that there is insufficient evidence on the feasibility of using measures suitable for economic decision making in evaluating end of life care interventions. Qualitative methods such as cognitive interviews were useful in providing a better understanding of the way measures were understood and completed by respondents, and issues that can affect their use.

The feasibility of using the ICECAP-SCM is explored through think-aloud interviews. Sixty patients with end stage heart failure, end stage renal failure and end stage COPD participated in the research. Interview transcripts were examined for errors in comprehension, retrieval, judgement and response. The constant comparative method was used to analyse qualitative

data providing an in-depth insight into measure completion. Qualitative thematic analysis further explored the impact of functional decline on health and capability wellbeing.

Error rates were low across all three measures and differences in error rates between the measures were small. The measures were acceptable, clear and easy to complete. The impact of terminal illness on health and capability wellbeing, and the extent to which participants were able to cope and adapt to the effects of their condition varied across the groups.

The ICECCAP-SCM is feasible to use with patients with end stage organ failure receiving care in different settings. Future research should consider exploring the feasibility of using the ICECAP-SCM in patients on other dying trajectories such as the frailty trajectory where patients are likely to receive care in nursing homes. The findings provide an opportunity to researchers and policy makers interested in the use of a measure suitable for economic decision making which captures the wider benefits of end of life interventions and services.

Dedication

To my parents Arinze and Ngozi Nwankwo and my six siblings Aijay, Nnamdi, Kanayo, Chinenye, Henrietta and Chioma. Thank you for all your support and for being the best family one could ask for.

To the memory of Uncle IK who died on his favourite chair with a smile on his face.

To the memory of the recently departed Uncle Obi and Aunt Uka. I wish I could have said goodbye one last time.

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Introduction

Global disease burden and cause of death have changed considerably over the last century. Infectious diseases were the leading cause of death at the beginning of the 20th century (Cohen, 2000), however, advances in medical technology and improvements in the standard of living have significantly reduced mortality from infectious diseases (Jones et al., 2012). These global trends, also evident in the United Kingdom (UK), have led to continued increases in life expectancy, with predictions that life expectancy will increase from current levels of 80 years to reach 100 years by 2064 (Office for National Statistics, 2014, Office for National Statistics, 2018c). However, as life expectancy continues to rise, many of these additional years are likely to be spent managing multiple long-term conditions for patients (Gilmour, 2017) making chronic illness the leading cause of death worldwide (Hajat and Stein, 2018).

Currently, most individuals die from chronic illnesses and multiple long-term conditions leading to increased use of health and social services before death (Hazra et al., 2018). The growing burden of chronic illness at the end of life has also contributed to rising costs of end of life care (Davis et al., 2016, Aldridge and Kelley, 2015), leading to increased public and professional scrutiny of the cost and quality of end of life services (Zhang et al., 2009). Changes in mortality, illness burden of the population and end of life care costs have implications for contemporary health policy, evident in the growing recognition of the importance of end of life care and recent policy initiatives (NHS England, 2015) aimed at addressing shortcomings in end of life care provision (Dixon et al., 2015).

A key consideration in improving end of life care may lie in measuring the impact of end of life interventions and services on patients. Measuring the outcomes of interventions can inform judgements about the effectiveness of health interventions and resource allocation decisions at various levels of a health system (Øvretveit et al., 2017).

Economic measures are currently used to evaluate the value of health interventions and assist decision makers in allocating resources in the health sector (Morris et al., 2007). Much of this work is done within an extra-welfarist framework where 'health' is considered the main outcome of interest and interventions that produce more 'health' are normatively judged to be better than those which produce less 'health'(Gafni and Birch, 2003). The EQ-5D is an economic measure recommended for use by the National Institute for Health and Care Excellence (NICE) in the UK to evaluate the impact of health interventions and services (Sculpher, 2008), the outcomes of which are defined in quality adjusted life years (QALYs). The quality adjusted life year (QALY) is 'a measure of the state of health of a person or group in which the benefits, in terms of length of life, are adjusted to reflect the quality of life' (NICE, 2019).

Measuring quality of life may be different to measuring quality at the end of life. End of life interventions are unlikely to be curative but may rather be aimed at symptom control and improving the dying experience (Hawkins et al., 2005). Hence, critics argue that the EQ-5D may be unsuitable for economic decision making at the end of life due to inherent conflicts between the goals of end of life care and principles that underpin the QALY (Normand, 2009b, Hughes, 2005).

Criticisms of the economic framework for decision making at the end of life have informed the development of new approaches for evaluating end of life care. One such approach questions the value judgements that underpin the QALY, instead arguing for a framework that broadens the 'evaluative space' to include broader aspects of wellbeing rather than just health (Coast et al., 2008b). Evaluative space refers to the identification of the objects of value relevant in economic evaluation (Sen, 2003). It essentially specifies what is considered relevant in economic evaluation. Specification of the evaluative space is essentially a discriminative exercise because of informational constraints on what is 'included' and 'excluded' in normative analysis (Brouwer et al., 2008). The ICECAP-Supportive Care

Measure (ICECAP-SCM) was developed as a self-complete economic tool to measure capability wellbeing at the end of life (Sutton and Coast, 2014b).

Measuring end of life care comes with considerable conceptual, methodological and ethical challenges such as identifying patients in need of end of life care and the feasibility of measuring outcomes in people who may be very near the end of life (Fowler Jr et al., 1999, Karnik and Kanekar, 2016). Despite these arguments, given the increased illness burden and functional decline at the end of life (Murray et al., 2017), it is important that outcome measures are appropriate and feasible to use by people near the end of life.

The feasibility of using the ICECAP-SCM in patients approaching the end of life has been considered in only one prior research which recruited patients receiving care only in one very specific context (Bailey et al., 2016). Hence, questions remain on the feasibility of using the ICECAP-SCM in patients on other dying trajectories and care setting. It is also important to explore whether the broader evaluative space the ICECAP-SCM seeks to capture, is relevant to those on different end of life trajectories.

Thesis aims and objectives

This thesis aims to investigate the feasibility of completing the ICECAP-SCM among patients on the organ failure trajectory, given the lack of any previous research in this area.

The objectives of the thesis are:

i. To investigate the evidence on feasibility of using economic measures in people near the end of life, and among people with end stage organ failure.
ii. To determine the feasibility of completing the ICECAP-SCM among patients with end stage heart failure, end stage chronic obstructive pulmonary disease and end stage renal disease.
iii. To explore the impact of function decline on health and capability wellbeing among people with end stage organ failure.

Findings from the thesis will to contribute to the existing literature on the application of the capability approach in health economics (Al-Janabi et al., 2012, Coast et al., 2008a), and the feasibility of using the ICECAP-SCM in an end of life setting (Bailey et al., 2016).

Structure of the thesis

The thesis consists of seven chapters. Chapter 1 considers conceptual issues in end of life care and end of life policy in the UK. The chapter explores the concept of end of life care and the various terms used to define end of life interventions. The chapter also considers theoretical frameworks used in end of life research and prognostic approaches in identifying people with end stage organ failure. The development of end of life policy in the UK is considered, alongside the content of recent end of life policy in the UK, and the role of economic measures in informing end of life decision making and policy.

Chapter 2 examines conceptual and methodological issues in measuring quality of life for economic decision making in health care and end of life care. The chapter critically examines theoretical framework that underpin the use of economic measures in health. The chapter considers conceptual issues in defining outcomes for economic evaluation and explores methodological challenges in conducting economic evaluation at the end of life.

Chapter 3 systematically reviews the evidence on the feasibility of using quality of life measures, and measures suitable for economic decision making in people near the end of life.

Chapter 4 specifies the methodological approach used in the thesis. The chapter considers major methodological approaches used in research. The chapter further examines the history and development of qualitative research as well as philosophical paradigms that underpin qualitative research analysis to inform the philosophical position of the researcher. The chapter outlines the study aims, data collection and analytic procedures used in the study.

Chapter 5 reports findings from the error analysis of the think-aloud transcripts. The chapter further explores the burden of completion, clarity and acceptability of the measures.

Chapter 6 examines the impact of functional decline on health and the opportunity for a good death. The chapter considers responses to the measures across the groups. Qualitative data is used to explore the impact of end stage organ failure on the capability wellbeing across the groups.

Chapter 7 discusses the principal findings of the thesis and its relationship to the wider literature. The implication for research and policy in the UK is also discussed.

Chapter 1

End of Life Care and End of Life Policy in the UK

End of life care is a multifaceted concept and its definition has evolved over time to reflect beliefs and attitudes towards death and dying (Fadul et al., 2009), and the development of palliative care as an integral aspect of medical practice (Clark and Graham, 2011). This chapter aims to explore the concept of end of life care and the development of end of life care policy in the UK. To achieve this, the chapter considers the various terminologies used to define end of life care and the settings in which end of life care is delivered. The chapter considers conceptual frameworks developed to understand the pattern of decline that occur in people as they approach the end of their lives. The chapter also discusses end of life care policy in the UK.

1.1 Definition of End of life care

End of life care is a term widely used in the literature to refer to interventions and services aimed at people approaching the end of their lives (Ahmedzai et al., 2004). The General Medical Council describes end of life care as the care given to people likely to die within the next 12 months (Bell, 2010). The National Council for Palliative Care provides a comprehensive definition of end of life care. End of life care is defined as:

> "…the care that helps all those with advanced, progressive, incurable illness to live as well as possible until they die. It enables the supportive and palliative care needs of both patient and family to be identified and met throughout the last phase of life and into bereavement. It includes management of pain and other symptoms and provision of psychological, social, spiritual and practical support" (Henry et al., 2007 p8)

Interventions and services for people approaching the end of life can be delivered in a range of settings such as in-patient wards, out-patient services and in residential homes. There is considerable variation in the terminology used to define end of life care in the literature and various clinical contexts (Pastrana et al., 2008). End of life care is often used synonymously

with palliative care, terminal care and supportive care. The definition of end of life care and the terms used to describe end of life care provision can be influenced by diagnosis, prognosis and setting of care. This apparent lack of clarity in the terms used to define end of life interventions has been identified as an issue that limits the generalisability of end of life research (George, 2002). The major terms used to describe end of life interventions are defined and discussed below.

1.1.1 Palliative care

Palliative care is a term first used by Balfour Mount to describe an approach to the care for people near the end of life (Mount, 2003). In defining palliative care, he emphasised the need for an approach that goes beyond a biomedical objective to include psychological, social and emotional support; and takes into consideration the needs of friends and family members (Mount et al., 2006). Mount is recognised as a pioneer in care of the dying, as is Elisabeth Kübler-Ross, a psychiatrist whose academic contribution on grief inspired terminal care and Dame Cicely Saunders, a nurse and then doctor who founded the hospice movement – all pioneers of palliative care as we know it today.

Attempts to differentiate between palliative care and end of life care highlight that end of life care is a subset of a broader palliative care approach. Some view palliative care as 'continuing' care that extends through the course of a life-limiting illness with curative therapy integrated into an approach that considers social and psychological wellbeing (Ferrell et al., 2018), while end of life care is viewed as the care given to people very near the end of life that does not include curative therapy (Curtis, 2008, Mularski, 2006). However, both end of life care and palliative care are used in very similar contexts and the distinction proposed above is not immediately clear in the general end of life care literature.

1.1.2 Terminal care

Terminal care is defined as the care given to people with a terminal diagnosis from which death is expected (Proot et al., 2004). Terminal care was first associated with the care given

to people dying from cancer (McCusker, 1984). However, the term has subsequently been used to describe individuals with irreversible coma (McCartney and Trau, 1990) and more recently the care given to individuals as they approach the end of their lives (Wittkowski, 2019). Prognostic eligibility for terminal care varies between three (Napolskikh et al., 2009) and twelve months (Proot et al., 2004) in the literature.

1.1.3 Supportive care

Supportive care as defined by NICE (2016) refers to the care of people with a life-limiting illness from the point of diagnosis through to treatment or therapy and into death and bereavement. Unlike terminal care, the term supportive care is often used in the literature to describe the management of cancer patients near the end of life (O'Neill, 2000, Hui, 2014) in particular, to reduce the side effects of chemotherapy and radiation on both physical and psychological wellbeing following treatment (Cherny, 2009). . The use of supportive care has however, continually evolved to include other end of life conditions (Ahmedzai et al., 2004).

1.1.4 Hospice care

Hospice care originated from the hospice movement in the UK led by Dame Cicely Saunders who founded St. Christopher's hospice to promote a multi-disciplinary community-based approach to the care of people near the end of life (Fallon and Smyth, 2008). Hospices are facilities which offer care and support to people living with terminal illness (Saunders, 1990). The definition of hospice care may vary in different cultural contexts. In the UK, hospice care has traditionally been viewed as care given in specialised palliative settings mainly through hospices, although there are recent attempts to broaden the definitional scope of hospice care to include a range of settings (National Health Service, 2019). In the US however, according to the National Institute on Ageing (NIA), hospice care is viewed as an approach to care which can be delivered in varied settings such as homes, assisted living facilities or in-patient wards (National Institute on Ageing, 2017)

Hospice care may differ from other terms used to define end of life interventions as it is often viewed in relation to the setting of care delivery i.e. hospices. Other settings of care, like nursing homes, not traditionally associated with end of life care delivery have become increasingly viewed as an important aspect of end of life care (Regulatory Quality Improvement Authority, 2013). Research shows that nursing home residents often die within two years of being admitted (Menec et al., 2009) and over a quarter die within a year of being admitted (Shah et al., 2013) highlighting the importance of end of life care in such settings.

1.1.5 Conclusion

Although the terms used to describe end of life interventions are broadly similar in the extent to which elements of end of life care are included, there are clear differences in the way these terms are used with respect to diagnosis and care setting. While 'end of life care' and 'palliative care' appear to be the most commonly used terms in the literature, there still remains, however, a lack of consensus on the appropriate term to describe end of life interventions and services (Reid et al., 2011).

It is important that end of life research clearly states and defines the approach to care used to describe end of life interventions. Hence, for the purpose of this thesis, 'end of life care' is used to refer to interventions and services aimed at people near the end of life. The definition of end of life care by the General Medical Council (Bell, 2010) and the National Council for Palliative Care (Henry et al., 2007) is the preferred working definition in this thesis. This definition is preferred because it is used in official end of life policy in the United Kingdom (UK Parliament, 2015).

Despite ongoing definitional ambiguities in end of life terminologies in the literature, there appears to be a movement towards the development of rigorous theoretical frameworks through which end of life research can be conducted. These frameworks are largely defined by different philosophical assumptions that inform the concept of dying and aim to provide

a platform through which the end of life population can be systematically organised and studied. In order to explore appropriate frameworks suitable for the thesis, common theoretical frameworks that underpin the concept of dying are examined.

1.2 Theories of dying

Death and dying has been explored extensively in much of the social science literature from various perspectives (Wein, 2008), mainly physical, psychological and social perspectives. Observational studies informed much of the early works on dying, particularly the pioneering work by Glaser and Strauss (1966) on the context-awareness theory of dying. Following observational studies involving terminally ill patients, they outlined four contexts of awareness faced by people near the end of life: closed awareness, suspicion awareness, mutual pretence awareness and open awareness. These different contexts of awareness were defined by the relationship between the patient and professionals involved in their care and distinguished by the level of transparency and communication regarding end of life preferences. Closed awareness implies that health professionals are aware that a patient is dying but do not share their judgement with the patient who remains unaware. Suspicion awareness occurs in situations where the patient begins to suspect their illness may be terminal and attempts to understand his prognosis by directly confronting those in charge of their care or looking for clues from their medical records. Mutual pretence awareness describes a situation where both the patient and health care professional are aware that death is imminent but refuse to acknowledge such in their interactions. Open awareness implies that both the patient and those in charge of their care are aware that death is imminent, and choose to openly acknowledge and discuss this in their interaction. While the context of awareness theory of dying is of little practical use in end of life research, it was influential in its methodological contribution to the development and use of grounded theory in end of life care research (Andrews and Nathaniel, 2015).

Kübler-Ross (1973) approached dying from a different perspective, defining dying as a primarily psychological process where patients go through five different emotional phases when faced with imminent death: denial, anger, bargaining, depression and acceptance. As with the context-awareness theory of dying, her theory was derived from observational studies with patients near the end of life. The denial phase is marked by disbelief and shock at the news of imminent death. Following the denial phase, the patient begins to acknowledge the risk of imminent death and reacts with anger at their prognosis. Afterwards, patients enter the bargaining phase where they contemplate abstract trade-offs in exchange for life extension. This is followed by an acknowledgement of the certainty of their condition which leads to the depression. Depression can manifest in two forms – reactive depression where the patient displays an overt emotional reaction to their condition, and a quiet depression phase where it is common to find patients withdrawn and resigned to the prospect of their condition. The final stage is acceptance during which patients come to terms with their situation and accept death as a likely outcome of their condition. The stages of grief cycle were grounded largely in reflections on practical experience. Although influential in bereavement care (Hall, 2014), it has been criticised for its apparent rigidity in the way individuals move through the various stages of grief (Stroebe et al., 2017).

Criticisms of earlier theories of dying for both their level of abstraction and a perceived inadequate focus on their implications for the care of the dying, informed Corr (1992)'s task-based notion of dying. He recommended a series of tasks based on four existential domains, the physical, psychological, social and spiritual domains, to manage the transition of patients through the end of life phase. Physical approaches should be focused on minimising pain and discomfort associated with dying; psychological approaches should be focused on achieving a sense of security and self-worth; social approaches should be focused on the relationship between dying persons and their family members; and spiritual approaches should aim to satisfy spiritual needs. Despite its seemingly practical applications, there is little evidence of its use in end of life research.

Critics of earlier theories argued that the strong value judgements implied in these theories may overlook individual differences in dying (Copp, 1998). Buckman (2001) attempts to address these criticisms through the three-stage model of dying. He defined dying as consisting of three phases: the initial stage, the chronic stage and the final stage, He lists a number of characteristics that define each stage. At each stage, individuals respond to dying in ways that are consistent with the character of an individual rather than the stage through which such individual is passing. Thus, a mixture of emotions may be present at each stage and progression from one stage to another depends on the ability of the individual to resolve the emotional elements inherent in each stage.

While these theories of dying provide a useful understanding of the care and experience of people approaching the end of life, they however lack the precision needed for the thesis. These theories of dying largely overlook key issues in end of life research such as challenges in uniquely identifying people near the end of life and a clear definition of the end of life period (Gysels et al., 2013). Prognostication at the end of life is very difficult and may differ for those with different types of diagnosis (Krawczyk and Gallagher, 2016), hence identifying people near the end of life can be challenging.

1.3 Trajectories of dying

Attempts to move beyond these problems have focused on re-defining end of life care from conceptual issues regarding its definition and scope, to one that wholly considers prognosis, the pattern of functional decline and the implications for care (Lunney, 2001). A particularly influential conceptualisation of dying was proposed by Glaser and Strauss (1980) to model the organisation of care around the needs of people near the end of life. They proposed the concept of dying trajectories, defined by two essential characteristics: duration and pattern of decline. They argued that dying takes place over time and the pattern of decline can be illustrated graphically based on the health of the patient (Glaser and Strauss, 1980). Three dying trajectories were described: sudden death; expected death (both lingering and quick

death); and a third trajectory where patients fluctuate between points of normal and poor health states until their eventual demise. These trajectories were further illustrated in case studies (Strauss and Glaser, 1970) that formed some of the early attempts to understand the trajectory of patients with acute and chronic illness.

In recent years, the trajectory framework has undergone substantial theoretical development and application in end of life research. The trajectory framework has been applied to model the pattern of decline, disability, and care needs of people near the end of life. Unlike the theories of dying examined in the previous section, the trajectory framework provides a relatively rigorous and intuitive theoretical framework through which end of life research can be conducted. Common theoretical extensions of the trajectory framework are examined below.

1.3.1 Functional trajectories

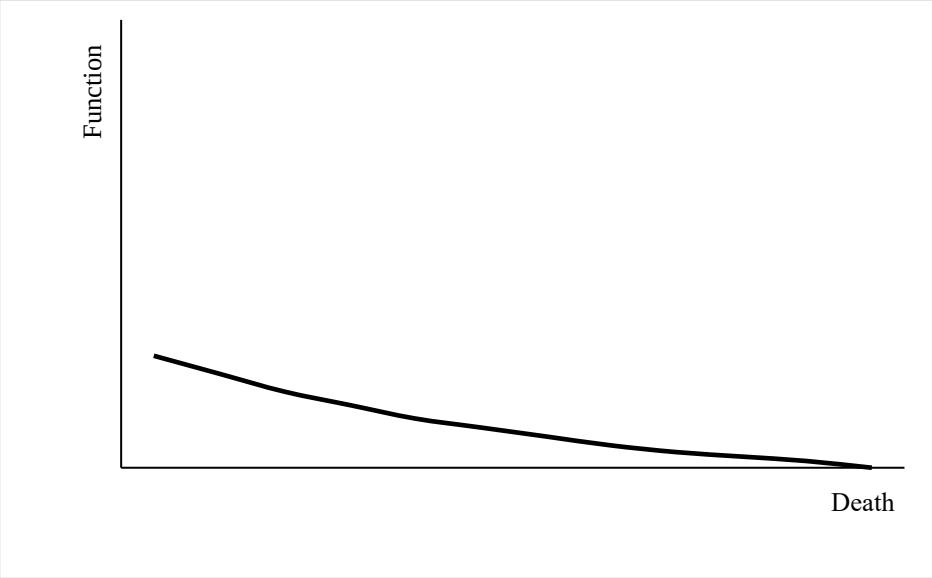
Functional trajectory is an influential application of the trajectory framework, proposed by Lunney et al. (2002) following their study of Medicare recipients in their last year of life. Functional trajectory is a term used to illustrate the pattern of functional decline in patients near the end of their lives, explored through changes in functional status over time. Functional status refers to an individual's ability to perform basic activities to maintain their health and broader wellbeing (Leidy, 1994). Functional decline refers to a reduction in functional status due to physical or cognitive impairment (Hoogerduijn et al., 2010). Four functional trajectories among people near the end of life were proposed— terminal illness trajectory, organ failure trajectory, frailty trajectory and sudden death trajectory. These trajectories are examined below.

Frailty trajectory

The frailty trajectory describes the experience of people near the end of life who have a gradual progressive functional decline leading to death (Lunney et al., 2002). Patients on this trajectory may enter the end of life phase from a low baseline of cognitive and physical

function, progressively declining to death. Patients with degenerative cognitive conditions such as dementia and Alzheimer's disease can be found in this trajectory (Lunney et al., 2003), and are likely to be found in care facilities such as nursing homes.

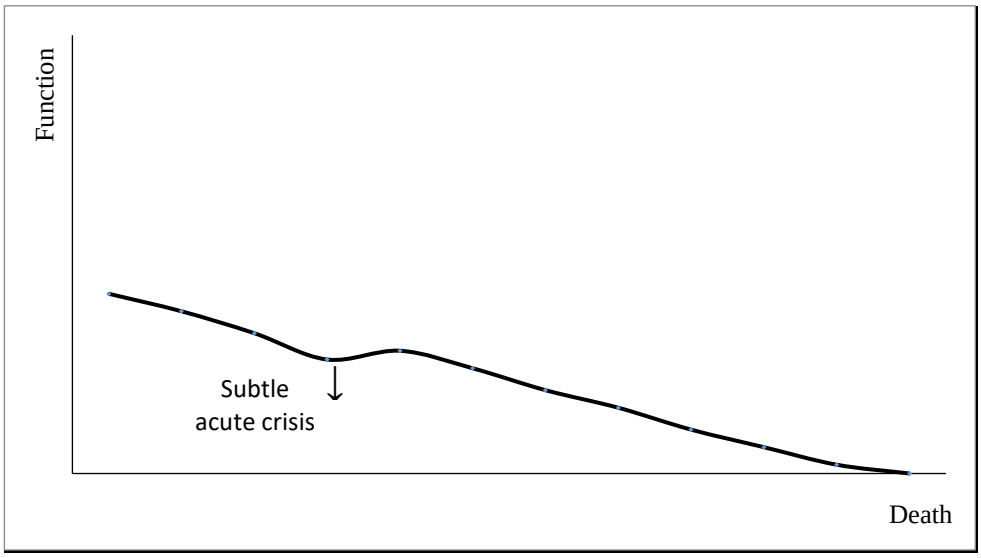
Figure 1 Death following frailty trajectory



Adapted from Lunney et al. (2003) functional trajectory

A graphical illustration of the frailty trajectory is shown in **Figure *1*** above. The patient enters the end of life phase with limited functional ability, progressively declining to death. Although the frailty trajectory may be characterised by sustained progressive decline, there may be periods of subtle acute crisis which may be missed in long-term care facilities and can result in earlier death (Cheek, 2004).

Figure 2 Death following frailty illustrating acute crisis



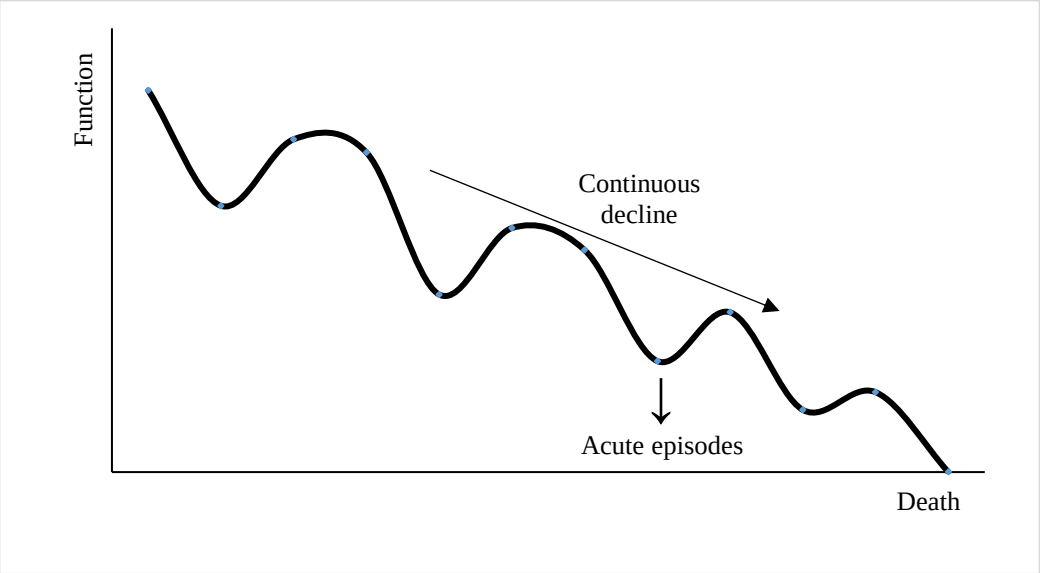
Adapted from Lunney et al. (2003) functional trajectory

In Figure 2, as patients approach the last few months and weeks of life, acute crises may worsen and hospital admissions may become more frequent in the period before death (Chamberlain et al., 2016). Age and degree of cognitive impairment can influence the rate of decline in patients on this trajectory (Nikolova et al., 2009).

Death following organ failure trajectory

The organ failure trajectory relates to patients who die from organ failure, characterised by a progressive functional decline interspersed with episodes of severe acute crisis (Murray et al., 2005). Organ failure is defined as a systemic organ dysfunction that leads to failure (Durham *et al*., 2003). Patients on the organ failure trajectory often presents with multiple co-morbidities (McClellan et al., 2002) which can influence their rate of decline (Murtagh et al., 2004). Organ failure is a leading cause of mortality, particularly among older patients (Adhikari et al., 2010, Twombly, 2005). Cardiovascular diseases and cardiomyopathies are the most common types of organ failure resulting in death (Naghavi et al., 2015). The organ failure trajectory is characterised by an unpredictable onset of acute crisis and patients on this trajectory can be found in the acute setting requiring medical care to manage illness exacerbations (Murray et al., 2002).

Figure 3 Death following organ failure trajectory



Adapted from Lunney et al. (2003) functional trajectory

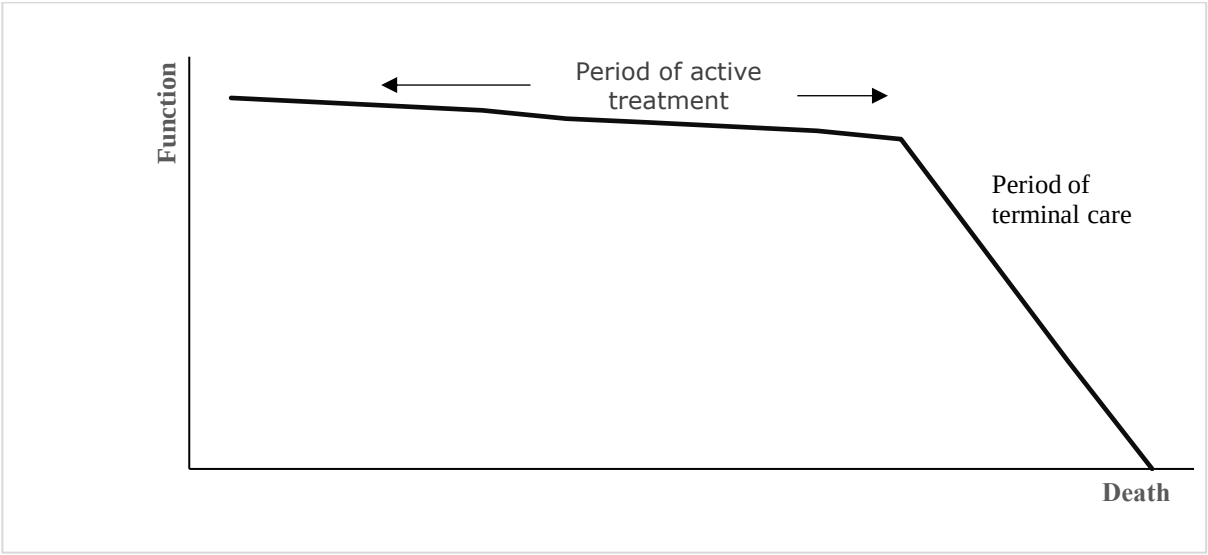
Figure 3 above presents a graphical illustration of the organ failure trajectory. Patients may enter the end of life phase from a high functional baseline but experience progressive decline that can be accelerated by acute crises. Acute crisis and medical emergencies requiring hospital admissions become more frequent as the condition worsens, and can result in death (Murtagh et al., 2004, Levenson et al., 2000b).

Organ failure is the most common cause of death in the UK (Office for National Statistics, 2018b), hence, most individuals are likely to die from conditions associated with the organ failure trajectory. Prognostication in the organ failure trajectory is difficult compared to other trajectories because of unpredictable exacerbations which can lead to death (Murray et al., 2005); and patients on this trajectory are less likely to be offered end of life services (Momen et al., 2012, Sallnow et al., 2013). Respiratory failure, heart failure and kidney failure are the most common cause of organ failure related deaths in the UK (Office For National Statistics, 2018b). The trajectory of patients with end stage heart failure and end stage chronic obstructive pulmonary disease (COPD) are typical of the organ failure trajectory (Sallnow et al., 2013).

Death following terminal illness trajectory

The terminal illness trajectory is used to describe patients who may maintain a relatively normal level of function before a rapid deterioration in health status leading to death (Murray et al., 2005). Patients with a cancer diagnosis are likely to be found in this trajectory. Individuals on the trajectory may experience a relatively normal functional level during the active treatment phase, when there is remission and a positive response to treatment. However, following recurrence of cancer or in situations where treatment becomes ineffective, patients may experience a rapid functional decline to death (Costantini et al., 2008). Parkes (1978) provides a useful guide to understanding the trajectory of patients with cancer, suggesting a distinction between the period of active treatment and the period of terminal care. Intervention before the beginning of the terminal phase of illness is referred to as the period of active treatment. The period of terminal care follows disease progression despite treatment, and the consensus opinion among clinicians that further treatment may not realistically improve prognostic survival (McCusker, 1984). The period of terminal care may be relatively short with studies suggesting a range of 2-3months following active treatment (Costantini et al., 2008). However, the distinction between the period of active treatment and terminal care may be less obvious in clinical practice where active treatment may continue alongside palliative care.

Figure 4 Death Following Terminal illness



Adapted from Lunney et al. (2003) functional trajectory

In Figure 4 above, patients may maintain a relatively high functional status before undergoing a precipitous decline to death. Patients in this trajectory are more likely to be identified for end of life care than the other trajectories and are more likely to receive specialist palliative care in settings such as hospices (Emanuel et al., 2002).

Sudden death trajectory

The sudden death trajectory describes people who die following acute illness or trauma with little or no use of medical services (Lunney et al., 2003, Lunney et al., 2002). Sudden death is regarded as death that is both unexpected and instantaneous often as a result of violence or traumatic injury such as accidents. Patients usually enter the end of life phase from a functional level before death. Excluding violence and trauma, sudden death is predominantly caused by cardiac disease (Varró and Baczkó, 2010, Ferreira et al., 2010). For example, it is common among competitive athletes and non-athletes with congenital cardiac disease (Ferreira et al., 2010).

1.3.2 Disability trajectory

The disability trajectory is an extension of the trajectory framework that considers the pattern of disability among people near the end of life (Gill et al., 2010). Using the classification scheme developed by Lunney et al. (2003), five distinct disability trajectories were described: no disability, catastrophic disability, accelerated disability, progressive

disability and persistently severe disability (Gill et al., 2010). Although terminal illnesses were shown to be poor predictors of the likely course of disability at the end of life, patients with advanced dementia were predominantly found in the persistently severe disability trajectory (Gill et al., 2010). A recent study identified frequency of hospital admission, rather than terminal diagnosis as a stronger predictor of the disability trajectory across all patient groups (Gill et al., 2015).

1.3.3 Dying trajectories in hospital settings

The trajectory framework has been applied to the management of patients in different settings such as residential care homes (Lawrence et al., 2017) and emergency departments (Bailey et al., 2011). Although associated with the sudden death trajectory, research shows that most deaths in the emergency department were of people who were known to be approaching the end of life with unexpected death occurring only in a minority of patients (Zalenski and Compton, 2004).

The trajectory in the emergency department has been further classified into spectacular and subtacular trajectories (Bailey et al., 2011). Unexpected and often traumatic loss of life was characteristic of the spectacular trajectory with dramatic life-saving measures given in response. In the subtacular trajectory, death was unsurprising and most deaths were due to complications from known medical conditions. In contrast to the spectacular trajectory, the subtacular trajectory attracted a more subtle response from the care team and care for the patients on this trajectory was not prioritised (Bailey et al., 2011).

Dying trajectories in a residential care setting have further been classified into anticipated dying, unexpected dying, uncertain dying and unpredictable dying (Lawrence et al., 2017). Lawrence et al. (2017) reported that patients on these trajectories were managed differently due to a lack of appropriate prognostic tools to identify patients in need of end of life care.

The original trajectory model developed by Glaser and Strauss has been extensively developed and applied to the management of patients near the end of life, and among those

receiving care in different settings. However, critics argue that conceptual trajectory frameworks often differ from empirical assessments (Gott et al., 2007) and factors such as cognitive decline, depression, frequency of hospital admission and duration of hospital stay are more important predictors of functional decline than terminal diagnosis (Hoogerduijn et al., 2007). Despite conceptual issues in defining and identifying patients in need of end of life care, dying trajectories provide useful frameworks that facilitate communication and organisation of care at the end of life (Sallnow et al., 2013). Furthermore, they also provide a conceptual foundation through which end of life research can be conducted (Mackintosh and Sandall, 2016).

The functional trajectory framework by Lunney et al. (2003) will be used in this thesis. This framework has been used in previous studies (Bailey et al., 2016, Fassbender et al., 2009); and it is recognised for its conceptual clarity and application in end of life research (Murtagh et al., 2008). The thesis will be focused on identifying patients with organ failure, particularly conditions that are associated with the organ failure trajectory.

1.4 Identifying patients with end stage heart failure, end stage renal failure and end stage COPD

Although the trajectory framework provides a useful theoretical foundation through which end of life research can be conducted, there remain issues in identifying patients approaching the end of life, particularly on the organ failure trajectory. Much of the uncertainty around identifying people in need of end of life care is due to inherent difficulties in predicting death. It is important however, that end of life research credibly includes patients who can be appropriately described as approaching the end of life. While there is no gold standard for clearly identifying patients in need of end of life care, prognostic classification schemes have been developed over the years to facilitate identification of patients in need of end of life care.

Given the focus of this thesis on those with organ failure at the end of life, prognostication in patients with end stage heart failure, end stage renal disease (ESRD) and end stage chronic obstructive pulmonary disease (end stage COPD) is explored. The trajectory of patients with end stage heart failure and end stage COPD are typical of the organ failure trajectory. While the trajectory of patients with ESRD differ from the organ failure trajectory (Murtagh et al., 2011a), it is a major organ failure related death and accounts for 1.3% of the total NHS expenditure (Kerr et al., 2012).

Unlike the terminal illness trajectory where research shows a relatively high prognostic accuracy in predicting death among patients with advanced cancer (Hui et al., 2019), identifying patients in need of end of life care in the organ failure trajectory remains challenging (Hogg and Jenkins, 2012, Smith et al., 2017). Identifying patients in need of end of life care is important for the initiation of end of life services, management and evaluation of end of life care. Prognostic models for classifying patients in need of end of life care is briefly considered below.

Heart Failure

Heart failure is defined as the inability of the heart to pump sufficient blood to meet the needs of the body. Heart failure can become terminal, in which case, it is referred to as end-stage HF. The Heart Failure Society of America defines and identifies patients with end stage heart failure as:

> "those patients who have advanced, persistent HF with symptoms at rest despite repeated attempts to optimize pharmacologic and non-pharmacologic therapy, as evidenced by 1 or more of the following: frequent hospitalizations (3 or more per year), chronic poor quality of life with inability to accomplish activities of daily living, need for intermittent or continuous intravenous support, or consideration of assist devices as destination therapy" (Lindenfeld, 2010 p494)

Prognostic models such as single-variable models like the 6-minute walk test (Bittner et al., 1993), and multi-variable models that consider a range of outcomes (Lee et al., 2003, Levy et al., 2006) have been used in determining prognosis among patients with end-stage HF. Some suggest that emergency hospitalisations, weight loss and history of cardiopulmonary resuscitation may serve as useful indicators of prognosis in end-stage heart failure (Adler et al., 2009). Biomedical indicators of heart failure and symptoms have been developed into severity-based classification schemes for the management of patients with heart failure. Two common classification systems exist: the American College of Cardiology (ACC)/American Heart Association (AHA) classification system (Hunt et al., 2001) and the New York Heart Association (NYHA) classification system (Criteria Committee of the New York Heart Association, 1994). The NYHA classification system divides patients into four groups based on limitations in physical activity and breathing difficulties. The classification ranges from I to IV with increasing severity of symptoms, with class IV representative of patients with end-stage heart failure. The AHA system identifies four stages of heart failure ranging from A to D, with increasing severity. Stage D denotes patients with end-stage cardiac disease requiring specialised intervention and palliative support where appropriate (Hunt et al., 2001). Patients with end-stage heart failure are defined as patients who fall into stage D of the ABCD classification of the AHA, and class III-IV of the NYHA classification system (Swedberg et al., 2005).

Both the NYHA and AHA provide a useful prognostic framework through which patients with end stage heart failure can be uniquely identified. Both classification schemes will be considered in developing suitable prognostic criteria for identifying people with end stage heart failure.

Chronic obstructive pulmonary disease

Chronic Obstructive pulmonary disease (COPD) is a term used to describe a collection of disorders of the airways characterised by an irreversible obstruction of the airway system

(Vestbo et al., 2013). Obstruction of the airway system is mainly caused by chronic exposure of the lungs to toxic gases, common among habitual smokers (Martin et al., 2005). Bronchitis and emphysema are the two most common causes of COPD (Pauwels et al., 2001). It is one of the most important causes of morbidity and the fourth leading cause of death worldwide (Grosdidier et al., 2014). COPD is classified by the Global Initiative for Chronic Lungs Diseases (GOLD) based on the degree of airway obstruction into four stages (I–IV) of increasing severity (Vestbo et al., 2013). End-stage COPD is recognised as a stage IV COPD on the GOLD framework (Habraken et al., 2008). Patients are considered as approaching the end of life if clinical indicators such as lung function, degree of breathlessness and frequency of hospital admissions are consistent with a stage IV GOLD standard classification. The GOLD framework will inform the development of prognostic criteria for identifying people with end stage COPD.

End stage Renal Disease

The kidneys serve an important function in the body by the regulation of fluid and the electrolyte balance, in particular, maintaining a stable pH and excretion of metabolic waste. (Hall, 2015). An impairment of the structural and functional integrity of the kidney can lead to renal failure. About 6-8.5% of adults in the UK have advanced renal disease (Roth et al., 2010). Renal failure can be either acute or chronic. Acute renal failure (ARF) can be defined as sudden inability of the kidney to perform its functions (Schrier et al., 2004). Persistent ARF for more than four weeks can lead to complete loss of kidney function resulting in end-stage kidney disease (Bellomo et al., 2004). Compared to ARF, chronic renal failure (CRF) is more insidious. It can be defined as the presence of kidney damage or a decrease in the functional capacity of the kidney for more than three months (Kidney Disease Improving Global Outcomes, 2013). Progressive CRF leads to end-stage renal disease (Roderick, 2002). End-stage renal failure, often referred to as end stage renal disease (ESRD), is defined as ‘an irreversible deterioration of renal function to an extent that is incompatible with life

without renal replacement therapy (RRT), either by dialysis or transplantation' (Roderick, 2002 p200). The National Kidney Foundation classifies chronic kidney disease (CKD) into 5 different classes based on Glomerular Filteration Rate (GFR). Patients with a stage 4 (GFR of 15-29 mL/min) and stage 5 CKD (GFR <15 mL/min) are classified as having ESRD and recommended for kidney transplantation (Levey et al., 2003). Renal transplant and haemodialysis have however considerably improved outcomes for people with ESRD.

The classification scheme developed by the National Kidney Foundation will inform the development of prognostic criteria for identifying patients with ESRD.

1.5 End of Life frameworks and policy in the UK

In addition to conceptual and prognostication guidance that aids in the identification and management of patients approaching the end of life, policies have been established by national governments to facilitate end of life care delivery and aid in the identification of patients in need of end of life care.

Discussions on the need for an end of life policy in the UK began at the time of the establishment of the National Health Service in 1948 (Clark, 2013). Unlike the National Health Service, which had considerable support from the government, end of life policy was largely driven by those working outside the government, many of whom came from non-profit institutions (Clark, 2013). The Marie Curie Memorial, established in 1948, was one of the first institutions devoted to the relief and welfare of cancer patients approaching the end of life. Working alongside other institutions at the time, they published a series of reports which explored the social condition of dying people in the UK (Clark, 2016). The first of these reports was the product of a collaboration with Queen's Institute of District Nursing in Belfast which outlined the needs of cancer patients receiving care at home (Joint National Cancer Survey Committee, 1952). Subsequently, the Calouste Gulbenkian Foundation published a report which critiqued deficiencies in the care of people approaching the end of life alongside recommendations for improving care (Hughes, 1960). Although

these reports had little influence on policy at the time, they provided much of the intellectual foundation that informed the early hospice movement and development of palliative care in the UK (Clark, 2013).

Official end of life policy began decades later following the establishment of the National Council for Hospice and Specialist Palliative Care Services and the publication of the Calmine-Hine report (Calman and Hine, 1995). The report identified variability in quality and access to palliative care services which was attributed to dependence on volunteer workers, underscoring the need for a national framework for the commissioning of cancer services. The Calmine-Hine report proposed a series of recommendations for the organisation and provision of palliative care services such as greater integration between palliative care and core medical specialities, provision of palliative care early in the course of disease, recognising the spiritual and psychological needs of patients and involving family members in their care, and adopting a multi-disciplinary approach to palliative care. Following the report, a framework was developed for the planning and commissioning of cancer services with a mandate to extend this to other end of life conditions. Subsequent government reviews on the quality and provision of palliative care services found that access to services remained disproportionate and existing services mainly catered to patients dying from cancer (House of Commoms Select Committee, 2004). The National Council for Hospice and Specialist Palliative Care Services later became known as the National Council for Palliative Care, reflecting the growing recognition of the broader scope of palliative care and need to extend existing services to other settings and terminal conditions (Dixon et al., 2015). Since the devolution of powers from the UK government and the establishment of local assemblies in each Scotland, Wales and Northern Ireland, health policies have differed across the four nations (McKenna and Dunn, 2015). Hence, different end of life policies has been implemented across the four nations to address issues in the quality and access to palliative care services; these are considered below.

1.5.1 End of life care policy in England

Comprehensive end of life policy in England followed the commissioning of the End of Life Care Strategy (Department of Health, 2008) in 2008 to report on the quality of end of life services delivered across the nation. The strategy identified disparities across different settings, in the quality of care, unmet patient expectations and preferences for end of life care. Identification of people in need of end of life care, greater coordination and planning of care, a particular focus on the management of patients in their last few days of life and recognition of the welfare of family members were key areas addressed by the strategy.

The strategy recommended greater collaboration with local communities and co-ordination between different end of life services to increase uptake of existing services and raise the profile of end of life care services. Regular review of care plans, needs assessments and patient preferences were also recommended. The strategy also underscored the importance of including family members and carers in needs assessment, and the provision of practical and emotional support. In addition to greater funding commitments by the government for end of life services, the strategy highlighted the need for measuring the structure, processes and outcomes of end of life care, and the need to include bereaved relatives in such assessment. The need for more research into non-cancer conditions was also emphasised.

Following the publication of the End of Life Care Strategy in 2008, end of life care became much more prominent in national health policies in England. The Mandate, a policy document outlined by the coalition government of 2010 emphasised the need for improving standards of care at the end of life (Department of Health, 2012). The NHS Commissioning Board was tasked with the responsibility of developing a long-term plan for improving the experience of people near the end of life. The Choice in End of Life care Programme Board was commissioned to provide advice to the government on improving the quality and experience of people near the end of life, their carers and family members (Henry et al., 2015). The board produced a report which identified issues with the quality of end of life care alongside a plan on how greater choice in end of life care can be achieved. They

proposed the development of an Electronic Palliative Care Coordination System where each patient can have their end of life care preferences outlined, the need for greater involvement of family and friends in decision making, and the need for local boards to ensure health care workers involved in palliative care delivery possess the right training. The report also recommended the establishment of continuous end of life support for people being cared for outside the hospital. Recommendations for suitable outcome measures for evaluating the quality of end of life services were proposed. These include care planning, access to services, quality of care, location of death, involvement and coordination of care, support for carers and bereaved people (Henry et al., 2015).

In response to the Choice in End of Life Care report, the government published a policy document which outlined a series of actions to be undertaken in response to the recommendations (Department of Health, 2016). These include a commitment to develop more personalised care for people near the end of life, implementing measures for improving the quality of services across different end of life settings, ensure training of workforce, identification and dissemination of innovation in end of life care and greater collaboration with the voluntary sector. The National End of Life Care Programme Board was established to oversee the implementation of the recommendations and report on the extent to which objectives were met (Department of Health, 2017).

Following policy initiatives on improving the quality of end of life services, assessment of end of life care services became much more emergent, informed by the need to ensure high quality service across the country. The National Institute of Health and Care Excellence (NICE) published the NICE Quality Standard for End of Life Care for adults (NICE, 2011) which outlined indicators for quality end of life care and guidance on evaluating the structure and process of care. Alongside the use of quality indicators in evaluating end of life services, the Department of Health emphasised the need for developing suitable outcome measures for evaluating end of life interventions to support decision making (Department of Health, 2009).

1.5.2 End of life policy in Wales

The All Wales Palliative Care Planning Group was set up by the government following the End of Life Care Strategy report in England. The policy review sought to establish elements of palliative care service for children and adults and demonstrate quality of palliative services (Welsh Assembly Government, 2008). Their report, published in 2008, identified disparity in the quality and access to services and the need for greater funding to enable more equitable access to palliative care.

Following the report, the Wales Palliative Care Implementation Board was established to develop quality markers for ensuring high quality end of life services and support the development of performance tools to evaluate end of life services (Wales Palliative Care Implementation Board, 2012). The Quality Standard Requirements were published to explicitly outline minimum targets end of life services were expected to meet. The subsequent policy, Together for Health: Delivering end of life care (NHS Wales, 2013), provided a framework for partnership between Local Health Boards and NHS Trusts to achieve end of life priorities of the government. The latest policy guidance, Palliative and End of Life Care Delivery Plan outlined the government's plan for palliative care delivery among different age groups and care settings, and an integrated management model of care to facilitate palliative care delivery (Steve and Finlay, 2017). The plan also prioritised the development of patient reported quality outcome measure to measure palliative care across different age groups and care setting.

1.5.3 End of life policy in Northern Ireland

End of life policy in Northern Ireland also followed the publication of the End of Life Strategy in England. The Northern Ireland end of life care strategy published in 2010 (Department of Health and Safety, 2010), aimed to develop a framework for the planning and delivery of end of life care services. The strategy considered factors influencing access and delivery of end of life services in the country, emphasising the need for continuous holistic assessment and access to end of life services. The strategy includes 25 policy

recommendations, implemented in a 5-year action plan to address key issues in the planning and delivery of end of life services. Another strategy was published in 2016 with a key focus on improving the quality of palliative care services in children (McGowan et al., 2016). End of life policy in Northern Ireland also outlines the need for identifying and measuring the outcomes of end of life care (Bradley et al., 2009).

1.5.4 End of life policy in Scotland

The Scottish end of life strategy, Living and Dying Well: A National Action Plan for End of Life Care in Scotland (NHS Scotland, 2008), sought to address issues in the quality and provision of end of life services across Scotland. Embedded within the strategy was the roll out of a national Do Not Attempt Resuscitation Order (DNACPR) across Scotland which aimed to prevent unnecessary or inappropriate cardio-pulmonary resuscitation (CPR) in people receiving end of life care (Scottish Government, 2016). The policy encouraged proactive DNACPR discussions with patients where it is clear in advance that CPR would be unsuccessful. Exceptions were made for situations where the patient lacks mental capacity, refuses to engage in the conversation or there is a clinical judgement of significant physical or psychological harm to the patient. A subsequent strategy (Scottish Government, 2011) developed to replace the previous policy, focused on reducing deaths in hospital settings, and greater integration of health and social care. The strategic Framework for Action on Palliative and End of Life Care published in 2016, acknowledges the lack of routine measurement in the process and outcomes of end of life care, and calls for the development of suitable outcome measures that can be embedded in routine care to facilitate service delivery (White et al., 2016).

Summary

The end of life policies across all four countries emphasised the need for equitable access to palliative care and consideration of the wellbeing of family members in end of life care interventions. Policies across the four nations also recognise role of measuring end of life outcomes in improving the quality of services. Recent end of life policy in England

explicitly acknowledges the need for measures suitable for economic decision making and encourages local authorities to consider the health economic impact of end of life initiatives (Public Health England, 2017).

Policies across the four nations have been supported through frameworks developed through the NHS end of life care programme that aimed to facilitate the training and delivery of end of life care services. Frameworks and guidelines that have been implemented over the years include the Liverpool Care Pathway, Gold Standard Framework, and Preferred Priorities of Care. These frameworks are considered below.

1.5.5 Liverpool Care Pathway

The Liverpool Care Pathway for the Dying Patient (LCP) is an integrated pathway developed particularly for patients in the last days to hours of life, to aid in the transfer of the hospice model of care to a broader range of settings including acute settings and nursing homes (Ellershaw et al., 1997, Curie, 2008). However, issues relating to its terminology, media and public perception (Devlin, 2009), concerns over its implementation in clinical practice (Craig, 2008), and opposition by religious groups (Pullicino, 2012) raised questions about its legitimacy in providing appropriate care for people near the end of life. These factors led to an independent review of the pathway (Neuberger et al., 2013) and subsequent recommendation for its withdrawal from routine use. The Neuberger report called for increased funding to improve access to services to support access to services. Some academics however, argued that improving funding for palliative care services may not be a sustainable long-term strategy given the little evidence base to inform resource allocation decisions (Kinghorn and Coast, 2013).

In response to criticisms of the LCP, a coalition of 21 health organisations formed the Leadership Alliance for Care of the Dying Patient to outline a new approach to replace the LCP. They proposed the gradual phasing out of the LCP and implementation of an intermediate policy which emphasised personalised development of a treatment plan and continued assessment of such plan in light of a patient's condition; a mechanism for

engaging in difficult end of life conversations with patients, their family members and carers; and participation in regular national audits and local quality assurance processes to ensure the delivery of high-quality end of life services (NHS England, 2013). Their collaboration led to the publication 'One Chance To Get It Right' (Leadership Alliance for the Care of Dying People, 2014), which identified five priorities of care for people near the end of life. These include identification of people who may be near the end of life and a clear communication of prognosis to these patients; sensitive communication between the patients, their family members and health care professionals; involving patients and family members where appropriate, in treatment decisions; recognising and meeting the needs of family members and friends; development of a comprehensive individualised care plan in collaboration with the patients.

A review of One Chance to Get it Right was published a year after its implementation which detailed progress made on commitments outlined in the report. The report identified progress made by professional bodies, government institutions and local palliative care networks in implementing the objectives outlined in the Priorities of Care, supporting local organisations in improving end of life care and monitoring progress made in implementing the guidance. (NHS Clinical Services Team, 2015). The Care Quality Commission and National Institute for Health and Care Excellence (NICE), Public Health England and other independent organisations within NHS England oversaw the evaluation and implementation of key aspects of the policy, and the development of policies and procedures to ensure the delivery of high-quality end of life care. The Care Quality Commission, alongside NICE and other organisations within the NHS, were tasked with the responsibility of defining what good quality end of life services should look like and evaluate end of life care services along these standards.

1.5.6 Preferred Priorities of Care

The Preferred Priorities Of Care is a document which allows patients to write down their end of life preferences and where they wish to be cared for (Storey, 2007). Originally

developed for the management of cancer patients, it is also used by patients with non-cancer conditions. The document contains a series of questions, as prompts, to enable patients write down their end of life preferences. These questions have been adapted by some hospitals, and currently used to aid end of life decision making across hospitals in the UK (Reed, 2011).

1.5.7 Gold Standard Framework

The Gold Standard Framework (GSF) is an approach to improving the care of people near the end of life with the aim of identifying people near the end of life, assessing their needs and developing a care plan (Quinn and Thomas, 2017). Identification of patients near the end of life and development of advanced care plans is supported by the GSF Proactive Identification Guidance (Gold Standard Framework, 2011). The GSF is executed alongside training and co-ordination centres that offer training for hospitals, clinicians and care professionals across a range of settings including primary care and care homes. The framework includes an accreditation process that involves a review of end of life practices for hospitals and other institutions of care (Gold Standard Framework, 2015).

End of life policy and guidelines in the UK has undergone significant changes over the last two decades. Much of these changes have focused on addressing issues relating to the quality of, and access to, palliative care services. Recent policies and guidelines for end of life care have been driven by government action supported by established non-governmental actors in palliative care. End of life care frameworks and guidelines facilitate decision making in different end of life settings and help improve the process and outcomes of care. However, there still remain questions, both on a policy and practical level, about the initiation of end of life services (Neuberger et al., 2013). In the UK, the end of life period generally follows the recommendation of the General Medical Council which defines patients approaching the end of life as those expected to die within the next 12 months (General Medical Council, 2010).

1.6 Summary

This chapter explored conceptual issues in defining and undertaking research in end of life care. The chapter also considered the development of end of life policy and key frameworks that have informed much of the current debates around the quality and provision of end of life services in the United Kingdom.

The growing importance of end of life care is informed by an ageing population and a growing burden of chronic illnesses particularly at the end of life, leading to increased demand and rising costs of end of life care. Research in end of life care has been partly undermined by a lack of consensus on the appropriate definition of end of life care and difficulties in identifying people approaching the end of life. Prognostic difficulties in identifying people in need of end of life care have been recognised by policy makers as a factor impacting on access to end of life care. This has led to initiatives aimed at improving prognostic models for identifying people approaching the end of life (People, 2014). Despite ongoing difficulties in defining and identifying people in need of end of life care, there appears to be a consensus that end of life care goes beyond biomedical objectives to include emotional and psychological needs, and the wellbeing of family members.

End of life care is delivered in a wide range of settings including in-patient wards, out-patient services and residential nursing homes, hospices and in domiciliary settings. End of life interventions vary considerably in these settings underscoring the need for appropriate theoretical framework through which research can be conducted. Various models and theoretical frameworks have been proposed to aid research and decision making at the end of life. The trajectory framework is a particularly influential approach in conceptualising the prognosis and needs of people approaching the end of life. The organ failure trajectory remains a particularly challenging trajectory due to inherent prognostic uncertainties and complex needs of people near the end of life suffering from conditions associated with the trajectory (Murray et al., 2005).

In addition to theoretical frameworks which seek to aid the organisational and delivery of end of life services, various policies have been developed and implemented by national governments in to facilitate end of life decision making in various settings of end of life care delivery. These policies and framework are targeted at improving the structure, processes and outcomes of care. All strategies explicitly acknowledge the lack of suitable outcome measures to evaluate end of life services, and advocate for the development of evaluative measures to support their long-term goals of improving the quality of palliative care services.

Measuring the outcomes of end of life care is particularly important in establishing the effectiveness of end of life care interventions and measuring quality in end of life services. The functional trajectory model and prognostic criteria examined in the chapter provides a conceptual framework for the thesis and offers a means for identifying people with organ failure approaching the end of life.

Developing appropriate measures for use in evaluating end of life care interventions to inform decision making has been identified by the Department of Health and Care Quality Commission as an important aspect of end of life delivery (Department of Health, 2017). However, there is little policy and guidance on how such measurement should proceed, and there is considerable debate regarding the appropriate normative framework through which economic evaluation at the end of life should occur and how such outcomes should be valued (Kinghorn and Coast, 2019). The next chapter considers conceptual and methodological issues in measuring outcomes for economic evaluation of health care interventions and decision making at the end of life.

Chapter 2

Measuring Outcomes for Economic Evaluation of Health Interventions and End of Life Decision Making

2.1 Introduction

Rising costs of health care and concerns over equity in the distribution of health resources, has led to an increasing reliance on explicit normative criteria to inform resource allocation decision making. In some countries with single payer health systems, for example the United Kingdom, economic analysis of health interventions has been recognised as a process that aids decision makers in the allocation of scarce resources in the health sector (Maynard and Bloor, 1995). For a number of years, health-related quality of life has been an important (even dominant) outcome of interest for economic decision making in health (Bodenheimer and Fernandez, 2005, Crosby et al., 2003).

Nevertheless, there is considerable debate in the literature regarding the use of health-related quality of life as the principal outcome of interest in economic decision making. Much of this debate relates to the theoretical foundations through which analytical techniques were derived (Birch and Donaldson, 2003). Critics also argue that health-related quality of life may be insufficient, and in some cases, inappropriate in measuring the outcomes of interventions whose benefits may go beyond 'health'(Brazier and Tsuchiya, 2015). This may be particularly relevant at the end of life where treatment goals may go beyond biomedical objectives to include aspects of wellbeing other than 'health'(Hughes, 2005, Normand, 2009b, Coast, 2014).

Regardless of the normative framework through which economic analysis is conducted, measuring end of life care outcomes involves considerable methodological and conceptual challenges, most of which relates to general challenges in defining relevant domains of

measurement and sensitivities of undertaking research with people approaching the end of life.

This chapter begins by considering the frameworks that govern economic decision making in health and the role of the evaluative space in economic decision making. Section 2.3 considers conceptual issues in economic evaluation of health interventions. Section 2.4 examines conceptual issues in economic evaluation of end of life care, and discusses economic measures used in evaluating end of life interventions. Section 2.5 considers methodological challenges in measuring end of life care outcomes. Section 2.6 summarises the chapter

2.2 Theoretical frameworks that govern economic evaluation in health

The techniques used for economic evaluation in health care originate from its parent discipline, economics; thus, the principles and conceptual frameworks used in analysis are derived from traditional and contemporary economic theories (Folland et al., 2007). Three major frameworks underpin much of the normative work in economic evaluation of health interventions: the classical welfare theory, extra-welfare theory and the capability approach. These are considered below.

2.2.1 Welfare economics in health care

The application of the principles of welfare economics in the economic analysis of health interventions, an approach referred to as welfarism, is derived from classical and neo-classical economic theory (Petratos, 2018). The welfarist approach is underpinned by a set of basic assumption in relation to utility derived from the consumption of goods and services.

i ‘Individuals are rational and consistently act to maximise their utility

ii Individuals are the best and only judges of what constitutes their utility

iii Utility is derived from the outcomes of a process rather than the process itself

iv The value of a situation is judged solely on the basis of the utility derived from individuals in the state.’ (Brouwer et al., 2008 p327).

Utility is a term that denotes an individual's preference ordering over a bundle of goods or social states (Hurley, 2000). In welfarism, individuals are regarded as the best and only acceptable source of utility. Processes and characteristics of individuals are only considered relevant to the extent to which they affect utility. Non-utility information is considered irrelevant in the evaluation of social states (Pigou, 2017).

In classical welfare theory, utility is cardinally measured and added across individuals and a socially desirable state is one that yields the most utility. The notion of utility as defined in welfare economics, creates problems due to the inherent trade-offs involved in deriving utility. A simple summation of individual utilities creates a problem in aggregation due to the strong value judgements implied in the interpersonal preference orderings from which the utilities from two different individuals are derived (Hammond, 1990). In neo-classical Paretian welfare economics, utility is ordinally measured and interpersonal comparison is considered invalid. Socially desirable states are judged using the pareto principle (Backhaus, 1980). A Pareto optimal state is one in which there can be no further increase in utility of one individual without a corresponding loss in utility for another individual (Baujard, 2015). The Pareto principle was criticised for its disregard for the distribution of resources and its lack of guidance in ranking numerous pareto optimal states (Sen, 1979b). Furthermore, the pareto principle offers no mechanism for decision making in states where the only possible outcomes involve winners and losers particularly in the health sector where decisions may involve decommissioning and reintroduction of new services (Morris et al., 2007). The Kaldor-Hicks compensation principle was introduced due to these criticisms, which is based on the notion of hypothetical compensation for any corresponding loss in utility (Chipman, 2008).

The arbitrary nature of the concept of utility as a preference ordering makes it difficult to directly estimate and measure, particularly in the health sector; hence, most analytical approaches measure utility in monetary terms. Cost-benefit analysis is an analytic technique based on welfarist principles, used in evaluating health care interventions. Cost benefit

analysis is the comparative analysis of two or more interventions based on their costs and benefits. Benefits are estimated in monetary terms using willingness-to-pay through contingent valuation methods. Contingent valuation methods and choice experiments are approaches used for the evaluation of non-market goods and currently used by analyses that adopt a welfarist perspective to economic evaluation of health interventions (Ryan et al., 2001). Willingness-to-pay (WTP) is the maximum amount a respondent is willing to give to enjoy the benefits of an intervention. Benefits can also be estimated through 'willingness to accept', the minimum amount accepted in compensation for the deprivation of a service or intervention (Zweifel and Frech, 2012).

Critics of the welfarist approach question the suitability of market-based approaches in estimating the benefits of goods and services that may violate assumptions that underpin a perfect market. Information asymmetry, supplier-induced demand and extensive regulatory barriers in the health sector have been identified as factors that may undermine the applicability of market-based approaches in the health sector (Culyer, 1989). Criticism of the welfarist approach also relates to the means by which benefits are estimated. Critics argue that monetary valuation of benefits ignores wealth and income distribution (Javan-Noughabi et al., 2017), fails to sufficiently address equity concerns (Dong et al., 2005), and may bias resource allocation to the wealthy (Smith, 2005, Aizuddin et al., 2012). Furthermore, questions remain on the ethics of valuing health in monetary terms (Tsuchiya and Williams, 2001) with some studies reporting that patients refused to engage in monetary valuation of health interventions (Protière et al., 2004). Critics also argue that an exclusive focus on utility ignores non-utility information such as individual characteristics and the nature of utility itself which may be relevant in resource allocation (Brouwer et al., 2008).

Some academics, however, argue that the cost benefit approach has the potential to include a richer informational space in its valuation of benefits of an intervention because it places no restriction on what individuals are allowed to value (Birch and Donaldson, 2003,

Robertson et al., 2019). Hence, both health and broader outcomes, processes, and other factors deemed relevant to individual utility are included in evaluation.

However, within this approach, questions still remain about the valuation of benefits in monetary terms, the apparent lack of consideration for equity and the characteristics of individuals which may influence the values obtained. Furthermore, the extent to which processes and other broader outcomes are actually considered during WTP tasks is unclear (Olsen and Smith, 2001). These criticisms have informed the development and use of new frameworks in the evaluation of health interventions. The most common are the extra-welfarist framework and more recently the capability framework.

2.2.2 Extra-welfarist framework

The extra-welfarist approach emerged following conceptual criticism of the welfarist utilitarian approach, including its apparent inability to address equity concerns in resource allocation (Seixas, 2017). Critique relates to its exclusive focus on utility and disregard for non-utility information in the evaluation of a state (Sen, 1979a). The extra welfarist approach differs from welfarism in four major ways:

i 'It allows the use of outcomes other than 'utility'

ii It allows the use of sources of valuation other than the affected individual

iii It allows the weighting of outcomes according to principles that may not necessarily be preference-based.

iv It permits interpersonal comparison of utility beyond that currently allowed under welfarism' (Brouwer et al., 2008 p327)

Culyer (1991) extended Sen's critique of welfarism (Sen, 1979a), arguing that the notion of capabilities can be broadened to include non-good characteristics of individuals, such as 'health', in economic evaluation. Although the theoretical framework proposed by Culyer sought to include health as an additional outcome of interest alongside utility, 'health' has largely been replaced with 'utility' and treated as the sole outcome of interest which should be maximised (Coast, 2009).

The application of extra-welfarism to economic evaluation of health interventions generally entails outlining sets of characteristics relevant to health; a description of health states to distinguish between 'more' or 'less' health; and assigning utility values to such states (Brouwer et al., 2008). Cost utility analysis (CUA) is an economic analytic technique based on extra-welfarist principles. CUA can be defined as the comparative analysis of two or more competing interventions based on their costs and utility (Morris et al., 2007).

In CUA, the outcomes of interventions are measured directly from patients through health-related quality of life measures such as the EQ-5D (Brooks, 1996) and SF-6D (Brazier et al., 2002). Utility weights are attached to the health states derived from health-related quality of life valuations. Utility weights may be derived from the public using preference elicitation methods such as standard gamble (Gafni, 1994), a technique based on von-Neuman Morgenstern utility theory of decision making under uncertainty (Von Neumann and Morgenstern, 2007). Preference elicitation techniques such as time-trade off (Dolan et al., 1996) and discrete choice experiments (Ryan et al., 2007) are also used to value health states. Utility weights for each health state are treated as values which lie on a scale of 0 to 1 to reflect the best and worst possible health state, with 0 being dead and 1 being full health. However, states deemed worse than death are also allowed. Quality adjusted life years (QALYs) are then calculated by multiplying utility weights by the length of time spent in that health state (Morris et al., 2007). The QALY is thus the main definitive outcome of CUA and measures the length of life adjusted for quality (Torrance, 1986).

Despite arguments that the extra-welfarist framework does not support an exclusive focus on 'health' (Brouwer et al., 2008), empirical application of the framework has largely embraced the QALY as the outcome of interest for valuing the benefits of health interventions. Hence the evaluative space has been largely restricted to 'health'.

Criticisms of the extra-welfarist evaluative space mainly relate to the 'health' and 'life years' focus of the QALY. An exclusive focus on health ignores other non-health benefits

of health interventions. The benefits of health interventions often go beyond health and may fall under different service areas, for example, social care or the criminal justice system. The production of health simultaneously follows its allocation; hence, a lack of distinction between the production and allocation of health undermines the normative rule of treating health as a maximand which makes it difficult to practically incorporate efficiency and equity concerns unless equity concerns are dealt with specifically and concurrently (Coast, 2009). QALY maximisation is also critiqued as being inherently ageist because older individuals with fewer life years are likely to have a shorter life expectancy than younger individuals, hence resource allocation may be biased against older people (Tsuchiya, 2000). The criticisms of the QALY framework on its principal focus on 'health gain' and 'survival' may be particularly relevant in end of life care where interventions may not necessarily improve health-related quality of life and survival but may be targeted at improving outcomes associated with a good death (Normand, 2009b, Ellershaw et al., 2010).

2.2.3 Capability framework

The capability framework was proposed by Amartya Sen in his seminal lecture, 'equality of what' (Sen, 1979a), where the welfarist principle of utilitarianism was critiqued on the basis of its exclusive focus on utility, ignoring non-utility information. Sen proposed the notion of 'basic capability equality' with two normative claims that underpin the framework: freedom to achieve wellbeing is of prime importance in the evaluation of states of the world, and the freedom to achieve wellbeing is conceptualised in terms of an individual's capabilities (Sen, 2005). The capability approach makes a distinction between two essential concepts, functionings and capabilities. Functionings are defined as 'beings' and 'doings' which are characteristics of individuals. For example 'beings' can include being in good health, being well nourished, being hungry, and 'doings' can range from activities like walking, sleeping, eating to exercising certain basic rights, freedom of expression (Sen, 1999). Capabilities refer to the freedom of an individual to 'do' and 'be' what they have reason to value and judge to be important. The concept of choice and freedom in the

evaluation of social states is relevant to the capability framework. For instance, being well nourished may be an important objective in the evaluation of a social state but a focus on nutritional status ignores other potentially relevant information that may affect nutritional status; an individual may have unfettered access to food but may choose to fast. Thus, while being well nourished is important as an end in itself, the opportunity of access to food may be potentially more relevant in this scenario. Sen advocates for a shift in the evaluative space from specified functionings to a richer and more informative evaluative space which accounts for the real opportunities an individual has to do and be what they have reason to value (Anand et al., 2004).

The capability approach has been applied to a range of settings, including in the evaluation of health interventions. The Oxford Capability instruments (OxCap) (Lorgelly et al., 2008), Adult Social Care Outcomes Tool (ASCOT) (Netten et al., 2012) and the ICECAP measures (Coast et al., 2008a) are examples of capability-based measures developed for use in evaluating the outcomes of health interventions. The ICECAP family of measures include the ICECAP-A (developed for the general adult population) (Al-Janabi et al., 2012), ICECAP-O (developed to measure capability in older people) (Grewal et al., 2006) and ICECAP-SCM (developed to measure capability at the end of life) (Sutton and Coast, 2014b).

Critics of the capability approach argue that despite its conceptual appeal and potential to inherently address equity concerns, questions remain about the operationalisation of the framework due to its deliberately underspecified nature and multidimensionality in relation to wellbeing (Robeyns, 2000, Comim et al., 2008, Sugden, 1993). Unlike other economic frameworks, appropriate methods for measuring and valuing capabilities for use in economic decision making remain largely unspecified (Alkire, 2005, Coast et al., 2015). Using the capability approach at the end of life also faces considerable challenges in determining what capabilities are important at the end of life and how such should be measured and valued (Coast, 2014). Furthermore, unlike the explicit maximisation principle

of welfarism and extra-welfarism, there is no clear decision-making principle in relation to end of life capabilities (Round, 2016).

2.3 Defining outcomes for economic evaluation

Theoretical frameworks that govern economic evaluation of health interventions are mostly focused on the nature of the evaluative space and defining the 'informational space' of analysis, i.e. either 'utility', 'health' or 'capability'. This should not be confused with questions that seek to answer what precise aspect of health or capability is to be measured. Identifying aspects of health for economic evaluation of health interventions may lie in a subjective view of what health-related quality of life entails. The term health-related quality of life was reportedly introduced by the Medical Literature Analysis and Retrieval System Online (MEDLINE) in 1975 to describe comprehensive assessments of health status (Walters, 2009). Health-related quality of life has been defined in many different ways in the literature (Karimi and Brazier, 2016), mostly in relation to its parent concept of health. Historically, health has been viewed from a functional standpoint of being free from bodily harm and disease. There appears to be a consensus however, that health is a multi-dimensional concept encompassing physical, emotional, social and psychological domains of wellbeing (Huber et al., 2011). This multi-dimensional view of health is recognised in the definition of health by the World Health Organisation which defined health as a 'state of complete physical, mental and social wellbeing and not merely the absence of disease' (Grad, 2002 p981).

2.3.1 Types of health-related quality of life measures

The multi-dimensional nature of the concept of health poses challenges in defining outcomes for evaluating health interventions (Cano and Hobart, 2011). Despite conceptual difficulties in aggregating seemingly diverse aspects of health, a number of outcome measures have been developed and used in evaluating health-related quality of life. These can be broadly classified into generic and specific measures. Generic measures are not specific to any disease area and can be used both in a healthy population and a wide range

of disease conditions (Walter, 2009). These measures may contain multiple domains of interest to a broad group of patients and the general population. Specific measures on the other hand, are developed for a particular disease (Saab et al., 2011), patient population (Williams et al., 2009), dimension or problem (Melzack and Katz, 2001), and functional ability (Morley et al., 2013). Unlike generic measures, specific measures may not be used to make inferences beyond the patient population or condition the instrument was designed to evaluate.

Generic health-related quality of life measures may be profile or index measures. Profile measures of health such as the SF-36 and Nottingham Health Profile generate separate scores for different domains of health. These domains are not necessarily aggregated to generate a single score but rather scores from the different domains of health are independently summarised to reflect a particular area of interest (McDowell, 2006). In index measures, different domains of health are aggregated to generate a summary score for the measure (Streiner et al., 2015).

2.3.2 Economic measures used in evaluating health interventions

Index measures of health such as the EQ-5D (Brooks, 1996), Health Utility Index (Furlong et al., 2001) and SF-6D (Brazier et al., 2002) are used in economic decision making in the health sector. Some of these measures to a large extent, are defined in relation to 'physical' aspects of health. For example, the EQ-5D, widely used in the UK for economic evaluation of health interventions, mostly measures physical health and to a lesser extent psychological aspect of health, defined in the question on 'anxiety and depression'(Brooks, 1996). However, other less widely used measures such as the Assessment of Quality of Life instrument, includes attributes associated with psychological wellbeing and social roles (Hawthorne et al., 1999).

Health interventions may have different purposes and thus outcomes of interest may differ. For example, outcomes of public health interventions to influence behaviour such as interventions to reduce smoking and obesity are likely to differ from outcomes of health

interventions such treatment for respiratory infections. Thus, measuring outcomes for the economic evaluation of a health intervention involves a deliberate understanding of the goals of treatment and the appropriate aspect(s) of health or wellbeing to be measured (Gold et al., 2002).

2.4 Economic evaluation of end of life interventions

Critics have long argued that the use of outcome measures such as the EQ-5D, and the normative QALY framework with which it is associated, may be inappropriate in capturing the wider benefits of interventions (Ventegodt et al., 2003), particularly in situations where the benefits of health interventions may fall under a different service area (Wilkinson et al., 1990). This argument is particularly relevant in end of life care where the most definitions agree on the importance of aspects of wellbeing that go beyond health, such social and psychological wellbeing, and the importance of friends and family members (Henry et al., 2007).

Critics have also called into question the use of the QALY for economic decision making at the end of life. In addition to the general critique the of the QALY framework outlined in section 2.2.2, critics also argue that the strong value judgements implied in the additivity of time (that is the value of time is constant across an individual's life) ignores differences in the marginal benefit of 'time' which may be valued greater as individuals approach the end of life (Normand, 2009b).

At the end of life, interventions may not necessarily be aimed at improving health but may be geared towards providing care, managing symptoms and achieving a good death (Ellershaw et al., 2003). Furthermore, terminal illnesses are incurable and interventions may not necessarily increase prognostic survival. Due to functional decline at the end of life, a significant aspect of end of life interventions involves social care particularly for patients who die at home (Bardsley et al., 2010).

However, questions remain about the essential constituents of a good death and the appropriate framework through which the benefits of end of life care can be captured (Johnston, 2016).

2.4.1 Defining outcomes for evaluating end of life care interventions

Outcome measures used in evaluating end of life interventions and services should be sensitive to aspects of wellbeing relevant to people near the end of life. Relevant outcomes for evaluating end of life interventions are often associated with outcomes associated with a good death. End of life policies in the UK emphasise the importance of achieving a good death and the evaluation of end of life services and interventions based on the achievement of a good death (Ellershaw et al., 2010). A good death is defined as a death that is free of avoidable suffering for both the patient and their close persons; and respects their wishes in accordance with clinical, ethical and cultural standards. A bad death, on the other hand, is defined as one that is characterised by needless suffering, neglect, unnecessary and unwanted medical treatment; and does not consider the wishes of patients and their close persons (Field and Cassel, 1997).

There are however, varied notions to what constitutes a good death in the literature. These range from expert-led approaches (Clarke et al., 2003) to more deliberative and participatory approaches involving the views of patients near the end of life and their close persons (Heyland et al., 2006). Emanuel and Emanuel (1998) developed a model that defines a good death as one which considers a multi-dimensional notion of dying consisting of fixed characteristics of the patient, modifiable characteristics of the patient, care system interventions and the final outcome, reflected in the overall dying experience. Conceptual elements of a good death have also been derived through exploring the views of both patients near the end of life and their family and friends. Following qualitative interviews of patients near the end of life in the United States, Singer et al. (1999) described five indicators of quality in end of life care: adequate symptom management, avoiding inappropriate

prolongation of dying, achieving a sense of control, relieving burden, and strengthening relationships with close persons.

Following in-depth, qualitative focus groups with patients and their relatives in the United States, Steinhauser et al. (2000) reported six components of a good death. They are: pain and symptom management; clear communication and decision making between patients and health care professionals; a better understanding of the changes that occur during the course of their illness and how they could prepare for it (termed 'preparation for death'); resolution with family and friends; being able to participate in spiritual rituals (termed 'completion'); contributing their time and experiences to others; and being valued and treated with respect (termed 'affirmation of the whole person'). Indeed, most end of life researchers tend to agree on the relevance of aspects of wellbeing that go beyond physical health, such as the social support, spiritual support and bereavement care in end of life care (Kelley and Morrison, 2015, Schenker and Arnold, 2015).

2.4.2 Economic measures developed for evaluating end of life care

Despite inherent difficulties in defining constituent outcomes for evaluating end of life care, a number of measures have been recently developed and used over the years to evaluate end of life interventions (Mularski et al., 2007a). However, most of these measures are unsuitable for use in economic decision making.

There have however, been efforts in recent years to develop outcome measures for use in economic decision making at the end of life. These measures differ from each other, in relation to the nature of their respective evaluative space and outcomes considered relevant in evaluation. Recently developed economic measures used in evaluating end of life care include the Palliative Care Outcome Scale (POS-E) (Dzingina et al., 2017) and the ICECAP-Supportive Care Measure (ICECAP-SCM) (Coast et al., 2018a).

The POS-E is an end-of-life specific preference-based measure of health developed from the ten item Palliative Care Outcome Scale and reduced to seven items containing 2-3 levels of increasing severity. The items assessed in the POS-E include: family anxiety, other

symptoms, pain, depression, anxiety, practical matters, and feeling good (Dzingina et al., 2017). The POS-E remains within the existing QALY paradigm with its focus on 'health', but considers the health proprieties of people receiving palliative care. The POS-E is a relatively new economic measure and as yet, there is no evidence on its valuation.

The ICECAP-Supportive Care Measure (ICECAP-SCM) is a capability-based instrument developed to measure capability associated with the opportunity for a good death. The ICECAP-SCM consists of seven domains with four levels of severity. The domains assessed by the measure include autonomy, love, physical suffering, emotional suffering, dignity, support and preparation (Sutton and Coast, 2014b). Value sets for the ICECAP-SCM was derived using discrete choice experiments and best worst scaling techniques in a survey of the general population (Huynh et al., 2017). Pilot work has also been conducted showing that it is feasible to obtain values for the ICECAP-Supportive Care Measure (SCM) from patients receiving hospice care (Bailey et al., 2018).

The ICECAP-SCM and the POS-E are the only self-complete economic measures to date, developed for evaluating end of life care. Both measures differ from each other in both the outcomes considered relevant in evaluation and the normative framework through which end of life care is evaluated.

The ICECAP-Close Person Measure (ICECAP-CPM) is a proxy-completed economic measure developed for use by close persons (that is family, friends, and carers) in evaluating end of life care outcomes. The ICECAP-CPM is a capability-based measure developed from qualitative interviews with recently bereaved relatives and relatives of patients near the end of life. The ICECAP-CPM consists of six domains. They are: communication with those providing care services; practical support; emotional support; privacy and space; emotional distress; preparing and coping (Canaway et al., 2017).

2.5 Methodological challenges in the economic evaluation of interventions at the end of life

Measuring outcomes for economic evaluation at the end of life is conceptually and methodologically challenging. The challenges include general concerns regarding the precise focus of measurement – either the structure, process or outcomes of care; defining the outcomes to be measured, the source and timing of assessment, the method of data collection, and identifying patients for end of life research (Streiner et al., 2015). Methodological challenges specific to economic measures include theoretical consideration about the appropriate framework through which outcomes should be evaluated and the value judgements that inform economic decision making at the end of life. There are also practical concerns regarding the feasibility of using outcome measures in people near the end of life and the ability of patients to complete such measures (Theofilou, 2012). These issues are considered below.

What aspects of end of life care should be measured?

End of life policies emphasise the importance of evaluating both the structure, process and outcomes of care (Henry et al., 2007). Structural elements include material, organisational and human resources that influence the quality of care. The process of care includes activities that occur between the patient and the health care professional during the delivery of care while the outcomes of care are the effect of care on the patient themselves (Donabedian, 1988). The delivery of end of life care is marked by inherent complexities (Ding et al., 2018) which may undermine efforts to identify relevant aspects of the structure and process of care. End of life care is delivered in settings such as long-term care facilities, homes and acute settings which have different structures and processes. Hence, it may be difficult to link positive outcomes to a particular characteristic of the process of care (Fowler Jr et al., 1999).

Outcomes currently used to evaluate end of life care include measures based on satisfaction with care (Morita et al., 2002), quality of dying, quality of life and wellbeing measures.

Evaluating the outcomes of care based on satisfaction with care may lead to bias as patients may have low expectations of care and may be unwilling to criticise the quality of care received (Aspinal et al., 2003). Furthermore, as discussed in the previous section, there is little agreement on relevant outcomes in quality of life, dying or wellbeing measures. Although end of life policies and frameworks highlight important aspects of care that should be measured, translating these into measures that are psychometrically rigorous and can be easily used in evaluating the quality of care remains an issue.

Timing of assessment

A key methodological concern in quality of life measurement is the appropriate timing of assessment to overcome recall bias and reduce non-response. There is, however, little guidance in the literature on the ideal time to administer outcome measures. Some suggest measures should be administered immediately after an event to overcome recall bias and reduce non-responses (Casarett et al., 2003a). The feasibility of undertaking such an exercise is questionable given that symptoms may be unpredictable and fluctuate among patients near the end of life. Evaluating outcomes when health states fluctuate has been shown to lead to inaccurate estimation of quality of life (Giesinger et al., 2014).

Source and method of data collection

More general methodologic concerns regarding quality of life measurement relates to the method of data collection and the source of assessment. Outcome measurement can utilise various means of data collection such as postal surveys, telephone or in-person interviews, self-administration or online surveys. There is little evidence, however, on how these methods compare against each other in an end of life population. There may also be the need to consider the physical burden any proposed method given that patients are likely to enter the end of life phase from a low baseline of cognition and function which may affect their ability to complete evaluative measures (Thompson and Chochinov, 2006).

Patients very near the end of life may be unable to complete quality of life measures due to their terminal illness, hence responses from proxies such as health care professionals, family

members and friends may be useful in evaluating end of life interventions. However, the role of proxies in providing valid and reliable responses on behalf of the patient has been questioned in the literature with research showing that proxies tend to rate quality of life lower than patients (Rand and Caiels, 2015). Responses may also differ based on the relationship between the patient and the proxy with studies showing differences in family members and health care professional quality of life rating (Crespo et al., 2013). Research suggests that while proxies can reliably report quality of care and observable symptoms, they may be unsuitable for evaluating subjective experiences such as anxiety or pain (McPherson and Addington-Hall, 2003). There are also questions about the degree to which proxy responses reflect the memory of respondents or their own emotions. The end of life phase can be emotionally intense for close persons who may not be immune to complex emotions and burden that may arise from providing support and care to people near the end of life (Forbes et al., 2000).

Identifying patients approaching the end of life

Identifying patients for end of life research is a crucial first step and major methodologic challenge in evaluating end of life care. Difficulties in predicting death and identifying patients approaching the end of life in recognised in both research and practice (Kennedy et al., 2014). However, prognostic criteria are widely used in both research and clinical practice to aid in the identification of patients near the end of life. These include the use of the 'surprise' question 'would you be surprised if this patient died in the next year' (Weeks et al., 1998), clinical indicators (Boyd and Murray, 2010), single (Kohl et al., 2012) and multi-variable prognostic model (Gold Standard Framework, 2011). The extent to which prognostic criteria can predict patients in need of end of life care varies according to terminal condition (Brar and Tangri, 2015, Ayesta et al., 2018). These prognostic criteria, however, provide a useful framework through which end of life patients can be systematically identified. Thus, the use of clear prognostic criteria can ensure that patients recruited for end

of life research share similar prognostic characteristics that can reasonably be deemed to be indicative of approaching the end of life, informing the generalisability of findings.

Ability to complete measures

Patients near the end of life are likely to struggle with progressive functional decline and multiple long-term conditions which may affect their ability to complete quality of life measures (Chochinov and Breitbart, 2009). Hence, recruiting patients into psychosocial research at the end of life often faces extensive gate-keeping barriers due to ethical concerns relating to the burden of research (Emanuel et al., 2004). End of life research using evaluative measures should be designed to minimise the risk to patients and reduce the burden of completion (Casarett et al., 2003b). There needs to be a greater consideration of the burden of completion and the practical issues faced by patients near the end of life when asking them to complete questionnaires.

Response shifts and Adaptation

Another important consideration in evaluating end of life care interventions relates to the nature of response shifts particularly among people near the end of life (Preston et al., 2013). Response shift can be defined as a change in internal standards and re-conceptualisation of quality of life due to illness (Sprangers and Schwartz, 1999). Patients near the end of life with long-term conditions may adapt to the effects of their condition on their wellbeing which may be reflected in their quality of life scores raising questions on the ability of quality of life scores to truly reflect health states (Nagl and Farin, 2012). Research by Coast et al. (2018b) however suggests that while there is evidence of adaption among those approaching the end of life, this is not reflected in self-assessments of health and capabilities in some groups of patients.

2.6 Summary

This chapter examined conceptual and practical issues in measuring outcomes for economic evaluation of end of life interventions. Different theoretical frameworks underpin economic

evaluation of health interventions and these differ in terms of the nature of the evaluative space and outcomes considered relevant in evaluation. These include the welfarist, extra-welfarist and capability approach. The ICECAP-SCM is a condition-specific preference-based measure based on the capability approach, developed to measure capabilities associated with the opportunity for a good death.

The use of economic measures in end of life care faces key methodological challenges such as identifying people in need of end of life care, defining outcomes to be measured and the feasibility of using quality of life measures in patients approaching the end of life. Given that people near the end of life are more likely to manage multiple long-term condition, it is important that measures used for economic decision making are practically feasible to complete by people near the end of life. The next chapter systematically examines the evidence on the feasibility of using quality of life measures and measures suitable for economic decision making among patients receiving end of life care

Chapter 3

Feasibility of using quality of life measures in patients receiving end of life care: a systematic review

3.1 Introduction

The salience of quality of life measures to contemporary health research and service evaluation at the end of life is well established in the literature (Eichler et al., 2004). However, questions remain about the feasibility of using quality of life measures for assessing interventions at the end of life given the significant practical and methodological challenges faced when conducting research in people near the end of life.

Methodological challenges in evaluating quality of life among people near the end of life has been explored in the previous chapter. In particular, patients near the end of life are likely to have complex conditions associated with frailty, older age and chronic illness, which may cause significant functional and cognitive decline that can influence the effective use of quality of life measures (Harada et al., 2013). These challenges may be particularly important in patients on the organ failure trajectory where in addition to functional and cognitive decline, periodic exacerbation of illness (Van den Heuvel et al., 2016) and fluctuating conditions (Sanghera and Coast, 2018) may influence the use of quality of life measures. Lack of evidence on the feasibility of using existing quality of life measures presents a significant barrier to their widespread use (Higginson and Carr, 2001).

There has been a number of reviews on the use of quality of life measures in patients receiving palliative care. Jordhoy et al. (2007) reviewed measures developed to assess physical functioning in palliative patients, comparing aspects of physical functioning assessed across all measures. Their review identified inconsistencies in domains of physical functioning assessed and further highlighted inconsistencies in words and phrases used that are likely to affect response.

Similar reviews have also focused on measures used to evaluate symptoms in patients with cancer. Kirkova et al. (2006) evaluated the psychometric properties, content validity, method of administration and practicality of the measures used to evaluate symptoms of cancer patients. They reported that completion rates decreased as patients became more ill, with less than 5% completion rates reported by a study with critically ill patients near the end of life (Bruera et al., 1991).

Other reviews have focused on the use quality of life measures generally within patients near the end of life. Most of these reviews focused on the use of generic quality of life measures and measures developed to assess palliative care interventions, providing a description of their content, method of administration, time required to complete the measures, and their psychometric properties (Hearn and Higginson, 1997, Mularski et al., 2007b, Albers et al., 2010, Bruley, 1999, Kaasa and Loge, 2002, Massaro and McMillan, 2000, McMillan, 1996). These reviews reported that the way questionnaires were administered affected the ability of participants to complete them (Hearn and Higginson, 1997), and most measures did not have satisfactory content validity (Hearn and Higginson, 1997, Mularski et al., 2007b).

Despite a clear intention to address questions on feasibility and practicality of using the measures in patients near the end of life (Albers et al., 2010), most of these reviews largely focused on assessing the clinimetric properties of the measures used with little or no information on the practicality, acceptability, burden of completion and the way questions are understood and interpreted. A focus on psychometric properties of a measure may undermine differences in the way questions are understood and interpreted, and the willingness of participants to answer such questions (Collins, 2003).

Given significant challenges in measuring outcomes with patients with end stage organ failure, no review has focused on identifying evidence on the feasibility of using quality of life measures in patients on the organ failure trajectory. Therefore, this review aims to assess

the feasibility of using quality of life measures in patients receiving palliative care and in patients with end stage organ failure.

3.2 Definition of feasibility

A key consideration in determining the feasibility of using a measuring instrument is the definition of the term 'feasibility'. Feasibility is a term commonly used in the literature to denote a proposed plan or action to test key aspects of a main study (Jairath et al., 2000). Feasibility studies are regarded as integral aspects of research, common in randomised controlled trials to assess the validity of a prospective study (National Institute of Health Research, 2017). Feasibility studies are also used in qualitative research to explore socio-cultural and political barriers to patient recruitment (Hundley and van Teijlingen, 2002, Kelly, 2007, Kim, 2011).

However, it is important to distinguish between feasibility studies and feasibility of using a measuring instrument. Feasibility studies typically have many of the hallmarks of a prospective major study albeit on a smaller scale (Arain et al., 2010), while the feasibility of using a measuring instrument focuses on the acceptability of the instrument, degree of effort, burden to patients, and the disruption to normal care that arises from the use of such an instrument (Fitzpatrick et al., 1998). Thus, feasibility in the context of quality of life measures, is circumscribed to a narrower range of issues such as burden, practicality, acceptability, cognitive response problems, time required to complete, non-completion and missing responses; which can affect the way an instrument is used (Bouwmans et al., 2013). Acceptability can be defined as the ease with which a measure is completed, and the proportion of patients who find it difficult, impossible or unacceptable to complete (Williams, 2003). Practicality can be defined as the burden of an instrument to respondents, which can be explored through refusal rates, administration time and missing responses (Hwang et al., 2003). Both acceptability and practicality can be explored qualitatively through interviews (Sprangers et al., 1993) and quantitatively through information on measured response rates (Fitzpatrick et al., 1998). A mixed methods approach to obtain both

data types may be an appropraite framework through which the feasibility of using quality of life measures can be determined (Collins, 2003). For the purpose of this review, feasibility is defined in terms of the acceptability, practicality, burden of completion and cognitive response issues that affect the use of a maeasure; and the ability of patients to complete a measure

3.3 Aims and objectives

This chapter aims to systematically review evidence on the feasibility of using quality of life measures in evaluating health and wellbeing in patients receiving palliative care and in patients with end-stage organ failure. The review objectives were:

i. To systematically review and summarise the evidence on the ability of people near the end of life to complete quality of life measures and measures suitable for economic evaluation.
ii. To systematically review and summarise the evidence on the problems faced by people near the end of life while completing quality of life measure and measures suitable for economic evaluation.
iii. To systematically review and summarise the evidence on the acceptability and burden of completing quality of life measures and measures suitable for economic evaluation by people near the end of life.

3.4 Method

This systematic review was conducted in line with the Preferred Reporting Items for Systematic Review and Meta-analysis (PRISMA) guidelines (Liberati et al., 2009). The review question and search terms were developed using the PEO criteria (Pollock and Berge, 2018). The search terms and strategy, selection criteria, and quality assessment of eligible studies are described in the following sections.

3.4.1 Information sources

A comprehensive literature search was carried out to identify all relevant articles that addressed the aim and objectives of the review.

Electronic searches

The following databases were searched for the review:

i. Medline
ii. Web of Science
iii. Social policy and practice
iv. Embase
v. HMIC
vi. PsycINFO
vii. EconLit (EBSCO)

In addition to electronic databases, the reference lists of papers included were hand searched to identify potentially relevant articles. The searches were conducted between March 2017 and June 2019. The search was restricted to articles published between January 1990 and May 2019. This time frame was expected to capture relevant studies and hence meet the research objectives. The time frame selected was expected to be sufficient for the review as the use of quality of life measures in evaluating palliative care interventions became more prominent in the 1990's (Clark, 2007). Previous reviews on quality of life measures in end of life care, used 1990 as a starting point for selecting studies (Albers et al., 2010, Mularski et al., 2007b).

Four phrases from the review aim was used as keywords in the search: 'feasibility', 'quality of life measures', 'palliative care', and 'end stage organ failure'. Free text variation of these key words, including truncation, were used in the search. In addition to key text variations, feasibility was substituted for qualitative methods such as cognitive interviews and think-aloud interviews. Similarly, common generic and end-of-life specific economic measures were substituted for "quality of life measures" to ensure that all literature associated with

known relevant measures was identified. Alternative words and phrases used in the search are shown in *Table 1* below:

Table 1 Free text variations of search terms

Feasibility	**Quality of life measures**	**Palliative care**	**End stage organ failure**
Feasibility	Quality of life measure$	Palliative care	End stage organ failure
Acceptability	Patient outcome measure$	End of life care	End stage heart failure
Burden	Questionnaire$	Terminal care	End stage COPD
Cognitive interview	Palliative care Outcome Scale	Supportive care	End stage chronic obstructive pulmonary disease
Think-aloud	POS-E	Hospice care	End stage renal disease
Practical$	ICECAP-O		End stage kidney disease
	ICECAP-SCM		End stage renal failure
	Health Utility index		End stage kidney failure
	EQ-5D$		End stage liver disease
	SF-6D		

A free text search of each key word with their alternative was undertaken, separated by 'OR'. Search results from free-text search of 'feasibility', 'quality of life measures' and

'palliative care' were combined using 'AND'. Search results from free-text search of 'feasibility', 'quality of life measures' and 'end stage organ failure' were also combined using 'AND'. The results obtained were screened for their title and abstracts according to the inclusion and exclusion criteria. Medical Subject Headings (MeSH) were also searched for each keyword and their alternatives. Alternative MeSH terms were separated by 'OR' and individual search results were combined using 'AND'. Search results were screened based on their titles and abstracts, against the inclusion and exclusion criteria developed.

Other sources of information

The key words above were searched in both Google scholar and Google to identify relevant articles. Searches on Google were restricted to the first ten pages to capture grey literature and unpublished works.

3.4.2 Inclusion Criteria

Articles were included in the review if they met the following criteria

i Published in English language (due to lack of expertise in non-English languages)

ii Used quality of life or wellbeing measures (patient satisfaction questionnaires were excluded)

iii Reported the feasibility of using quality of life/outcome measures

iv Focused on people near the end of life and those with end stage organ failure

v Addressed at least an aspect of feasibility (as defined in section 3.2)

vi Questionnaires were self-completed

vii Participants were over 18 years old

Studies were included if they met the above criteria irrespective of research design.

Exclusion criteria

Articles were excluded from the review if they met the following criteria:

i. Published in a language other than English

ii. Failed to address aspects of feasibility as defined in section 3.2

iii. Articles are conference abstracts

iv. Articles are protocols for systematic reviews

v. All questionnaires were proxy-completed

vi. Used non-quality of life measures such as service evaluation measures and adherence measures.

vii. Participants were under 18 years old

Articles that satisfied the selection criteria were included in the review, irrespective of research design. Articles that only assessed psychometric properties of the quality of life measures without addressing their feasibility of use were excluded from the review.

3.4.3 Data collection and analysis

Study selection

Selection of papers for review involved two main steps. The first involved screening of titles and abstracts only, against the inclusion criteria. Articles were only excluded if they clearly failed to meet the selection criteria outlined. Articles that appeared to meet the inclusion criteria were marked for full-text screening. In cases where decisions on eligibility could not be made based on information contained in the title and abstract, the article was marked for full-text screening. Conference abstracts that appeared to meet the inclusion criteria were included in this stage to enable identification of full papers from the same study. Selected articles were exported to EndNote referencing software where duplicates were removed.

The second stage involved full text screening of articles marked from the first stage of the screening process. Each article was screened against the eligibility criteria. Articles meeting the selection criteria were marked for inclusion in the study. A Google and Google Scholar search was undertaken to identify the main article from conference abstracts selected from the first stage of screening. If the main article of conference abstracts included from the first stage of the review could not be found, it was excluded from the review. If full-text articles from conference abstracts were found, they were screened against the inclusion criteria. Articles that did not meet the inclusion criteria were excluded in the review.

Articles that satisfied both stages of the inclusion process were included in the review. All stages of the selection process are shown in the Prisma flow diagram in *Figure 5*

Data extraction and management

A standardised data extraction spreadsheet was adapted from the Cochrane data extraction form template (Cochrane, 2011) enable extraction of the relevant information from eligible articles. Eligible articles were summarised under the following sections: General Information, Eligibility, Population and Setting, Methods, Participants, Questionnaire, and Results. Each section was managed in a different work sheet with additional information relevant to each criterion. For example, under General information the information extracted included: date form completed, name of person extracting information, study title, publication type, possible conflict of interest, author and year of publication. An example of the data extraction form developed for all sections can be found in **Error! Reference source not found.**

Quality assessment

All included articles were assessed using the CASP checklist for qualitative studies (Critical Appraisal Skills Programme, 2017). While the CASP tool is recommended as a guide for evaluating the quality of studies rather than a scoring system, each question in the tool was scored and summarily reported. Each study was evaluated against each question in the tool and reported as either 'yes', 'can't tell' or 'no'. Articles that had a greater number of 'yes' responses were deemed of higher quality than those with a lower number. Articles with a higher number of 'no' and 'can't tell' responses were deemed of lower quality than those with a lower number. A summary of the quality scores for each study in each category are presented in the results. A sample quality assessment form for included studies can be found in **Appendix 2**

The CASP tool provides guidance for assessing the quality of studies and does not contain rules for decision making regarding which studies should be included in a review. Given

that any decision to exclude studies based on the CASP tool would be arbitrary, all studies that met the inclusion criteria were included in the review.

3.4.4 Data synthesis

Articles were summarised based on the methodological approach used, their use of generic or end of life specific quality of life measures, the palliative care population and study setting, and the aspect of feasibility addressed. A narrative review was undertaken to identify and summarise the evidence (Popay et al., 2006) on the ability to complete quality of life measures, the difficulties faced while completing quality of life measures, and the acceptability, clarity and burden of completion. Qualitative data relating to these key themes were identified and summarised in a data extraction form (presented in **Appendix 1**).

The strengths and weaknesses of each study were summarised with the aid of the CASP checklist and a score assigned based on the number of 'yes', 'no' and 'can't tell' responses to the checklist. Quality assessment of the included studies using the CASP checklist is presented in **Appendix 3**.

3.5 Results

3.5.1 Search results

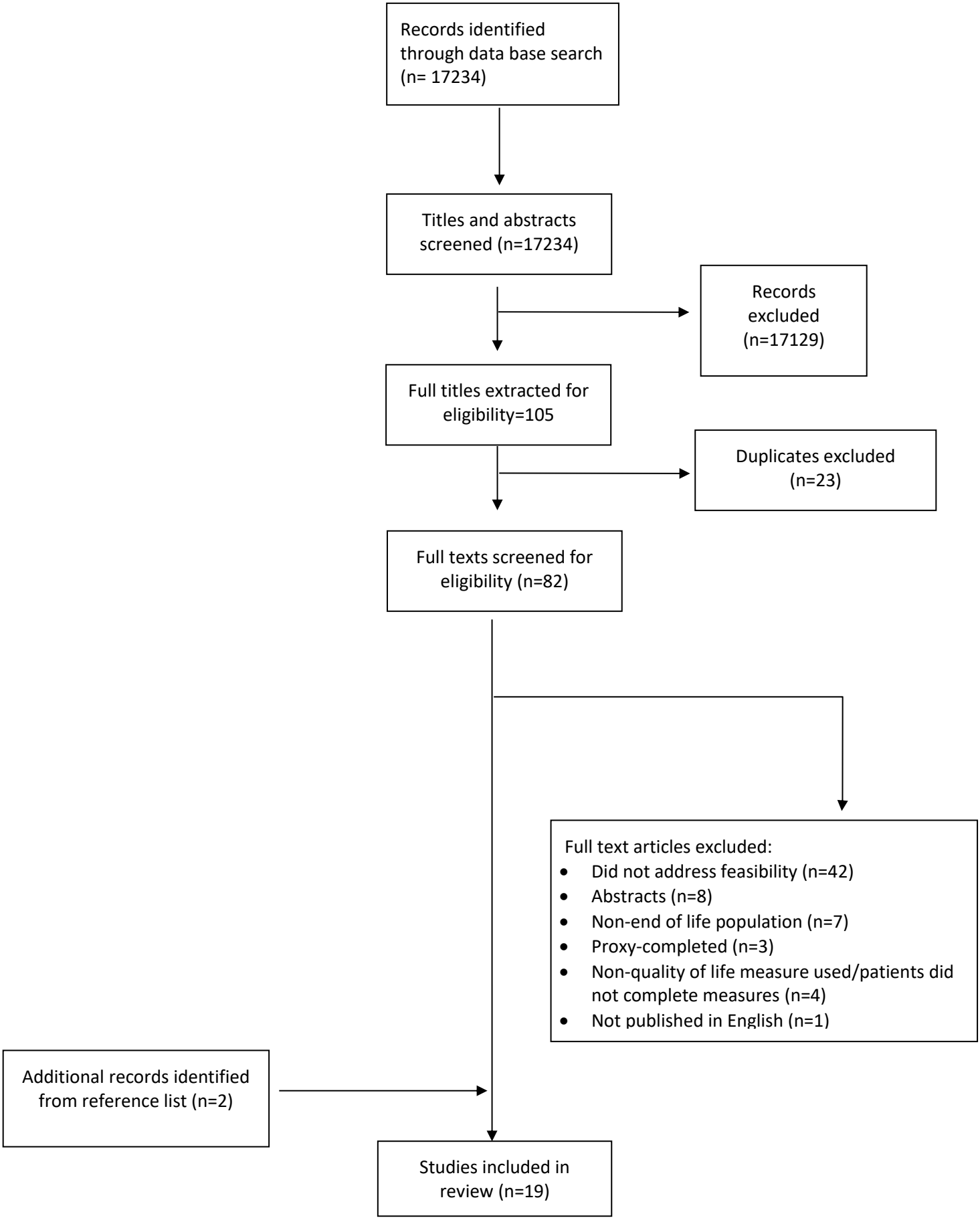
The initial data base search using the search strategy generated 17,234 articles across all databases. Medline generated 12,318 articles, Web of Science generated 2,709 articles, PsychInfo, Embase and HMIC generated 2,203 articles and EconLit generated 4 articles. These records were screened by title and abstracts, and 17,129 articles were excluded following the first round of screening. 105 articles were then exported to EndNote and screened for duplicates. Twenty-three duplicates were removed and 82 full text articles were screened against the selection criteria. Following full-text screening of the articles, 41 articles were excluded because they failed to address any aspect of the feasibility of using quality of life measures. Eight articles were excluded because they were conference abstracts and the full texts of the articles were unavailable. Seven articles were excluded

because the patient population was apparently non-end of life. Patients were deemed to be non-end of life if the population description did not explicitly state that patients were approaching the end of life or receiving supportive, terminal, hospice and palliative care. Articles were included if the population was described as having 'advanced' disease and were receiving palliative care.

Three articles were excluded because they were proxy-completed. Four articles were also excluded because feasibility assessment did not involve the use of a quality of life measure or patients completing quality of life questionnaires. One article was excluded because it was published in German and an English-translated version could not be found. Despite explicit *a priori* feasibility criteria defined in terms of completion and response rate, one study was excluded as it provided no information on the way participants understand and respond to questions in the measure (Kane et al., 2017).

Seventeen studies met the systematic review criteria and two studies were identified from the reference list of included studies and retrieved through hand search. In total, 19 studies were included in the review. A summary of the study selection process is shown in the figure below.

Figure 5 Study selection process



3.5.2 General Study characteristics

Of the 19 studies retrieved, most were cross-sectional studies with cognitive and semi-structured interviews. Twenty measures were evaluated by the included studies including generic measures, disease-specific measures, condition-specific and end-of-life specific measures. A brief summary of the general study characteristics can be found in ***Table 2*** below. This is to show a simple description of the study population and setting of care. The measures assessed in the included studies and their target population can be found in **Appendix 4**

Most patients received care exclusively in a hospice setting. However, some patients were recruited from out-patient palliative care departments of hospitals, acute settings, dialysis centres, rehabilitation centres and oncology outpatient units. Patients mainly had a cancer diagnosis with some studies reporting a mixture of both malignant and non-malignant conditions such as end stage heart failure, advanced respiratory illnesses, and palliative patients with HIV. Three studies evaluated the use of quality of life measures exclusively in patients with ESRD.

The studies differed in their approaches and the methods used for evaluating the feasibility of using quality of life measures. Ten studies used cognitive interviewing methods with both concurrent and retrospective verbal probing. Half of these studies primarily used cognitive interviewing techniques to identify problems faced during completion of the measures. Problematic or unclear questions were modified and re-tested using cognitive interview to ensure completion difficulties had been resolved. The rest of the studies used cognitive interviewing techniques to investigate the acceptability and ability of patients to complete a measure.

Table 2 Summary of included studies

Study number	Author (Year)	Population description	Setting
1	Addington-Hall et al. (2014)	Terminal diagnosis not clear but patients were recruited in a hospice	Hospice
2	Bailey et al. (2018)	Hospice patients	Hospice
3	Bailey et al. (2016)	Hospice patients	Hospice
4	Bausewein et al. (2005)	hospice patients and referrals to Palliative care units	Hospice
5	Cohen et al. (1995)	Patients with advanced cancer receiving care in a hospice or at home	Home and hospice
6	Flythe et al. (2019)	ESRD patients receiving dialysis	in-centre dialysis
7	Harding et al. (2010)	Palliative care patients with advanced disease	Hospice
8	Hearn and Higginson (1999)	Hospice patients	Hospice and in-patient wards
9	Iris et al. (2016)	Palliative patients	Palliative care unit in hospital
10	Bergh et al. (2011)	Patients in a palliative care unit	Palliative care unit in hospital
11	Lua et al. (2005)	In patient wards, out-patient palliative care clinic and hospice	Both in-patient and out-patient clinic and hospice
12	Murtagh et al. (2007)	patients with ESRD	receiving care from renal units
13	Pratheepawanit et al. (1999)	Out-patient palliative care patients	out-patient
14	Renovanz et al. (2018)	Cancer patients	Oncology outpatient
15	Shah et al. (2019)	ESRD patients receiving dialysis ad managed through conservative care	Rena units
16	Wang et al. (2010)	Patients with GI cancer	Oncology clinic
17	Watanabe et al. (2008)	Patients with advanced cancer	Outpatient oncology
18	Weis et al. (2013)	Patients with cancer	Acute care, rehab and palliative settings
19	Wilkinson et al. (2014)	Patients receiving palliative care at acute settings	acute settings

Cognitive interviewing techniques were exclusively used in 5 studies. The rest of the included studies used cognitive and semi-structured interviews in combination with other methods of assessment, particularly tests of validity and reliability. Across these studies, the extent to which findings from cognitive and qualitative interviews were reported varied with one study providing only a brief summary of findings from interview (Harding et al., 2010). Most of the studies had multi-faceted objectives which included psychometric assessment alongside cognitive interviewing and greater emphasis was placed on reporting the psychometric properties of the measure (Addington-Hall et al., 2014, Harding et al., 2010). One study required participants to provide written comments to questions they found difficult or confusing (Weis et al., 2013).

The nature of comprehension difficulties encountered by participants while completing the measures, acceptability, clarity and burden of completion were reported by all studies. The extent to which these were reported varied across the studies. These are discussed below.

3.5.3 Ability to complete the measures

Given the increased burdens of terminal illness and general frailty that occur at the end of life, the ability to complete quality of life measures is an important consideration in evaluating the use of such measures.

Inability to participate in the research

The number of eligible participants, approached for the study but who declined participation, was reported by only 8 studies. These ranged from as low as 26% (Watanabe et al., 2008) of those approached to as high as 60% (Bailey et al., 2016, Harding et al., 2010). Although the reasons for non-participation varied across the studies, health-related reasons were identified as a major cause of non-participation. Harding et al. (2010) reported that 8% of patients felt too unwell to participate in a study to complete the African Palliative Outcome Scale. Bailey et al. (2016) reported that 21% patients felt too unwell to participate in a study to complete three measures (the ICECAP-SCM, ICECAP-A and EQ-5D). Cohen

et al. (1995) reported that 20% of patient could not participate in a study to complete the McGill Quality of Life measure due to health-related reasons. These figures could be much higher given that many eligible patients do not disclose their reasons for non-participation. Flythe et al. (2019) reported that 59 of 128 patients approached declined participation for undisclosed reasons.

Participating but unable to complete the measures without help

The extent to which participants were able to independently complete the measures without any help was often not clearly reported by the studies. However, one study reported that up to 67% of participants required some form of help in reading out the questions and explaining unclear terms upon first assessment although the number was reduced to 57% by the third assessment (Bausewein et al., 2005).Watanabe et al. (2008) reported that most participants asked for clarification of certain terms in the ESAS, and about 60% of participants indicated that the presence of a health care professional would be needed to complete the measure. Some participants however were either unable to rate their quality of life or found rating their quality of life difficult. Addington-Hall et al. (2014) reported that 3 of 13 participants were unable to rate their quality of life while a considerable number of those who were able to do so found it difficult.

3.5.4 Issues observed during completion of the measures

Cognitive and semi-structured interviews were useful in providing greater clarity on the problems faced by patients completing the measures. These include comprehension difficulties with some words and phrases used in the measures, problems with response formats and question sensitivity.

Comprehension difficulties were most common among participants who completed the measures evaluated in the studies (Bailey et al., 2019, Bailey et al., 2016, Bergh et al., 2011, Cohen et al., 1995, Flythe et al., 2019, Harding et al., 2010, Iris et al., 2016, Murtagh et al., 2007, Watanabe et al., 2008, Weis et al., 2013, Wilkinson et al., 2014, Renovanz et al., 2018). Response errors and errors in judgement were also reported by some studies

(Murtagh et al., 2007, Jordhoy et al., 2007, Bailey et al., 2016). Most comprehension difficulties were due to a misunderstanding of certain words and phrases. Watanabe et al. (2008) reported that some participants struggled with words like 'tiredness' and 'drowsiness' in the Edmonton Symptom Assessment Scale (ESAS). Some participants also requested clarification on 'wellbeing', 'anxiety' and 'depression'. Bausewein et al. (2005) reported that some patients had difficulties understanding 'self-esteem' in the Palliative Care outcome Scale (POS). Murtagh et al. (2007) reported that participants had difficulties understanding words such as 'nausea' 'urination' and 'soreness' in the Memorial Symptom Assessment Scale (MSAS-SF), which were later modified with explanatory phrases eliminating such errors in subsequent interviews. Weis et al. (2013) evaluated the use of the EORTC QLQ-FA13 and its translated versions in participants from Italy, Spain, Germany and Sweden, reporting that some participants found questions about 'vulnerability' and 'frustration' difficult and confusing to answer. These comprehension issues differed by countries with Swedish patients finding questions about 'frustration' difficult to answer due to the word being rarely used in general speech. Indeed, Iris et al. (2016) also reported differences in comprehension errors among German and English participants which were due to the translation of the measure to German.

Differences in the way some words and phrases were understood and interpreted by participants were reported by some studies. Murtagh et al. (2007) reported that questions in the Geriatric Depression Scale and (GDS-10) were interpreted differently by participants. For example, in response to the question 'do you feel something bad is going to happen to you?', one participants answered yes because she viewed death as "something bad" while three participants answered no because they were not afraid of dying Iris et al. (2016) reported that the interpretation of 'uneasiness' in the POS varied across participants. Similarly, the word 'wellbeing' was interpreted differently by participants completing the ESAS (Watanabe et al., 2008).

There were some comprehension difficulties that were unrelated to words or phrases in the measures but rather directly linked to the legibility and format of the measure itself. Murtagh et al. (2007) reported comprehension issues with the MSAS-SF among patients with ESRD which were related to the legibility of the measures. Using a larger font for those with visual problems and modifying the appearance of the measure were recommended by patients completing the measure which subsequently reduced completion errors.

Response errors and difficulties identifying suitable responses were reported by many studies. Some of these studies reported that response options based on disease severity were difficult to answer due to the fluctuating nature of symptoms which varied daily. Bergh et al. (2011) reported that some participants found it difficult to answer questions based on severity due to the fluctuating nature of their symptoms. Wilkinson et al. (2014) reported that some participants found it difficult to quantify their quality of life due to the fluctuating nature of their symptoms. Indeed, some studies reported that participants had difficulty choosing a response option from the list of available options, rather choosing a range of options to reflect the uncertainty associated with their symptoms (Watanabe et al., 2008). Other studies also reported response errors due to multiple response options being selected, although it is unclear the extent to which this relates to uncertainty about their fluctuating symptoms (Renovanz et al., 2018).

Lua et al. (2005) reported that patients completing the McGill Quality of Life questionnaire preferred a wider recall period to capture the fluctuating nature of their symptoms. Iris et al. (2016) also reported errors in judgement due to a 3-day recall period, which participants felt was unable to sufficiently capture their fluctuating symptoms. Another study reported that participants requested that they complete the questionnaires repeatedly over time to capture day to day changes in their condition (Flythe et al., 2019).

Response errors were also identified as a result of the nature of the response expected of participants completing a measure. Some studies reported that participants struggled to identify suitable responses to open ended question with such questions having a significantly

higher number of missing values than questions with response options (Bausewein et al., 2005, Wilkinson et al., 2014, Pratheepawanit et al., 1999).

3.5.5 Acceptability, burden of completion and sensitivity of the measures

The acceptability of the measures to patient groups, burden of completion and sensitivity of certain items were often reported by participants during cognitive debriefing and semi-structured interviews. Most studies identified a number of questions that patients reported as being sensitive and the reasons that underpin their sensitivity. The burden of illness on family members was a sensitive issue for some participants. Addington-Hall et al. (2014) reported an unusually high number of missing items in the Palliative Care Outcome Scale (POS) relating to questions on family anxiety; further probing revealed concerns by patients about the impact of their illness on family members. Studies using the POS and its adapted versions such as the African Palliative Care Outcome Scale, reported that questions about family members were upsetting to some participants (Harding et al., 2010, Hearn and Higginson, 1999). Bergh et al. (2011) reported that some participants found questions relating to burden in the ESAS difficult to answer. Another study by Watanabe et al. (2008) on the ESAS found that participants found questions on anxiety and depression particularly sensitive. Wilkinson et al. (2014) reported that participants found questions in the QUAL-E related to the burden placed on family members difficult to answer and upsetting, often becoming emotional while reflecting on the burden of their illness on their family members. Iris et al. (2016) also reported that participants became upset while reflecting on the burden placed on family members due to their illness suggesting a particularly pervasive feeling of self-perceived burden among people approaching the end of life.

Questions relating to sexual intimacy were often deemed sensitive and inappropriate by participants completing measures containing such questions (Weis et al., 2013, Murtagh et al., 2007). These questions were often described as intrusive, annoying and upsetting (Weis et al., 2013). However, sensitivity may not be entirely based on the question asked but may be influenced by cultural norms with a study reporting that Italian and Spanish participants

reacted strongly to questions on sexual interest while German participants were more receptive (Weis et al., 2013).

Participants across a number of studies had difficulties answering questions related to advanced care planning and end of life decision making. Bailey et al. (2016) reported that the question on 'having a say' in decision-making at end of life in the ICECAP-SCM had a relatively high number of completion errors which may suggest difficulties among participants in addressing the issue. Harding et al. (2010) reported that questions on advanced care planning were particularly difficult for participants completing the African Palliative care Outcome Scale leading to errors in completion. Wilkinson et al. (2014) also reported that participants struggled to engage with questions in advanced care planning in the QUAL-E.

Some studies reported that while some participants found completing the measure they were given upsetting burdensome and intrusive, they preferred its comprehensiveness (Pratheepawanit et al., 1999, Murtagh et al., 2007). Wilkinson et al. (2014) reported that some participants did become distressed while completing measures about their wellbeing, they however finished answering the questions and ultimately found reflecting on their wellbeing a positive experience.

3.5.6 Quality assessment

A summary of the CASP quality scores for included studies is found in Table 3 below. Most studies had an average 'yes' score of 7 out of 9 criteria evaluated. The quality assessment form contains a series of questions regarding the quality of included studies which were judged under three response options: 'yes', 'no' and 'can't tell'. A sample of the quality assessment form with a detailed list of the criteria adapted from the CASP qualitative checklist can be found in **Appendix 3**

Table 3 Summary scores for all included studies

Study number	Yes	Can't tell	No
1	7	2	0
2	8	1	0
3	8	1	0
4	8	1	0
5	5	3	1
6	7	2	0
7	7	1	1
8	7	2	0
9	8	1	0
10	8	1	0
11	7	1	1
12	8	1	0
13	6	2	1
14	8	1	0
15	5	0	4
16	6	0	3
17	9	0	0
18	7	2	0
19	8	1	0

3.6 Discussion

This systematic review identified 19 articles which evaluated the use of 20 measures. Most of the measures were developed specifically for people near the end of life with a few studies evaluating the use of generic measures. End of life specific measures evaluated in the studies included both generic and disease-specific end of life measures. Six measures for economic decision making (ICECAP-SCM, ICECAP-A, EQ-5D-5L, QUAL-E, ICECAP-O, SF-6D) were assessed in only 3 of 19 studies. Of the 3 studies that evaluated the use of measures suitable for economic decision making, only one study investigated the use of economic measures (ICECAP-O and SF-6D) exclusively in patients with organ failure (End stage renal disease). The number of questions contained in the measures and the type of response formats varied across the measures. While most of the studies had satisfactory quality, the extent to which feasibility findings from interviews were reported varied.

The relatively high number of patients that declined participation and required further assistance from health care professionals during completion raises important questions about the use of quality of life measures in evaluating health and wellbeing at the end of life, and the practicality of patients independently completing such measures. As patients approach the end of life, they are likely to deteriorate and may be either unwilling or unable to complete quality of life measures (Thompson and Chochinov, 2006). Hence, patients very near the end of life may be systematically excluded from quality of life measurement which can lead to bias.

Understanding the various issues that may affect the ability of patients to complete quality of life measures can inform the development of appropriate steps to improve the content and clarity of questionnaires particularly for use with patients near the end of life. However, most research on the development of quality of life measures for use in evaluating end of life care are rather focuses on the psychometric properties of the measures, and tends to overlook issues relating to the interpretability of the items and the practical issues faced by patients during completion (Albers et al., 2010).

Some response problems were due to difficulties in ranking health states on quality of life measures due to fluctuating symptoms. This was particularly evident among patients with end stage organ failure who felt that day to day changes in their conditions made it difficult to 'quantify' their quality of life (Flythe et al., 2019). Measuring quality of life when health states fluctuate has been identified as a problem in research particularly in conditions with episodic symptoms (Norquist et al., 2012). Solutions to address this issue in the studies reviewed were focused on two main approaches—increasing the recall period (Lua et al., 2005) and repeated measurements over time (Flythe et al., 2019). Some suggest increasing the recall period in the measures may help reduce the impact of fluctuating symptoms on quality of life measurement (Iris et al., 2016), and in some cases, patients themselves suggested repeated measurements overtime to capture day to day fluctuations in their conditions (Flythe et al., 2019). There is however considerable debate regarding the extent to which increasing the recall period of measures can capture fluctuation in symptoms, with a systematic review identifying considerable challenges inherent in different recall periods (Stull et al., 2009). Besides, increasing the frequency of quality of life measurements to capture day to day fluctuations may not be economically feasible. However, a focus on frequency of symptoms rather than severity may be a way to evaluate the impact of symptoms that fluctuate over time (Bailey et al., 2016). Furthermore, given that most of these studies were conducted in a hospice setting with a diagnosis of cancer, further research is needed in different settings and trajectories such as the end stage organ failure trajectory where patients are more likely to struggle with fluctuating symptoms (Murray et al., 2005). Indeed, despite there being only three studies that investigated the use of measures exclusively in patients on with organ failure, response problems due to fluctuating symptoms was identified (Flythe et al., 2019). Fluctuating symptoms may be particularly prominent in conditions typically associated with the organ failure trajectory such as end stage heart failure and end stage COPD. No study has investigated the use of quality of life

measures, particularly measures suitable for economic decision making, exclusively in patients with these conditions.

This review has minor limitations that need consideration. First, the review was restricted to English language which could exclude potentially relevant research from different cultures and setting. A second limitation of the review is the arbitrary definition of feasibility used in the study. This is due to the fact that there is no consensus on relevant characteristics of a measure or a recommended set of tests that comprehensively addresses the feasibility of using a measure. However, a scoping review of the literature was undertaken to identify relevant aspects of feasibility which were used in the search strategy. Although the review instituted a comprehensive search strategy in different databases, there is a possibility that some articles may be missed. This is unlikely to be the case as the references of included studies were comprehensively searched to identify articles relevant to the review.

In conclusion, this review has underscored the value of the cognitive and semi-structured interview in providing greater clarity on the nature of problems faced by patients while completing quality of life measures. Questions remain about the ability of patients very near the end of life to complete quality of life measures given the very high rates of decline reported by most studies. Given that most patients were recruited in a hospice, the majority of which had a cancer diagnosis, questions remain on the impact of fluctuating symptoms on the patients receiving care in settings where symptoms may be less well controlled and on the organ failure trajectory characterised by periodic exacerbation of symptoms.

Only three studies evaluated the use of measures suitable for economic decision making among patients near the end of life. No research to date has explored the use of quality of life measures, particularly measures useful for economic decision making exclusively in patients with patients associated with the organ failure trajectory. Hence, this research aims to address this gap, and investigate the feasibility of using the ICECAP-SCM alongside other quality of life and wellbeing measures in patients on the organ failure trajectory.

Chapter 4

Methodology

This thesis aimed to determine the feasibility of completing the ICECAP-Supportive Care Measure (ICECAP-SCM), ICECAP-Adult (ICECAP-A) and the EQ5D-5L in patients with end stage organ failure. Think-aloud interviews alongside semi-structured probes were applied to patients with end stage heart failure, end stage COPD and ESRD completing the measures.

This chapter discusses the research methods and frameworks used in the thesis. The concept of cognitive interviewing techniques and its role in social science research is examined. The use of a retrospective think-aloud protocol is justified given alternative methodological approaches. The choice of questionnaires used in the research are justified given the research questions.

The chapter also considers the research setting, recruitment sites and population used for the study. These are justified given the research objectives. Appropriate sample sizes for think-aloud studies are explored in the literature and the patient population and sample size used in the research is justified. The study set-up and data collection procedures are discussed alongside methods developed to test the research design and data collection procedure.

The chapter critically examines the data analytical procedures used, justifying the suitability of the analytic methods used to address the research question. Ethical considerations in planning, conducting and evaluating the research are discussed, and ethical approvals from regulatory bodies are outlined. Lastly, issues of internal and external validity are explored given the proposed research design and data analytic procedure.

4.1 Research Design

This thesis used a mixed methods research design in data collection and analysis. A mixed methods paradigm combines elements of both qualitative and quantitative methodologies in

addressing a research question. Qualitative and quantitative research methods are the two major study designs used in research. A third approach, a mixed methods design, which incorporates elements common to both qualitative and quantitative research is has evolved into a distinct methodological framework widely used in research. Research evaluating the use of questionnaires in evaluating health and wellbeing have used qualitative and quantitative approaches, either separately or in combination (Oberguggenberger et al., 2018, Murtagh et al., 2007).

4.1.1 Research Methods

To provide greater clarity on the philosophical position of a mixed methods approach, this section considers the approach to research and philosophical position of quantitative research methodology. The history and development of qualitative research alongside its underpinning philosophies is considered. This section also considers the philosophical position of a mixed methods approach in light of qualitative and quantitative research paradigms.

Quantitative research methodology

Quantitative methodology emphasises the systematic assessment of phenomenon through a reliance on objective reasoning using mathematical and statistical approaches in data analysis (Kaplan, 2004). Quantitative research is often positivist and deductive in nature, and largely defined by the type of data set used in analysis (Bahari, 2010). Numerical data are used to obtain information about the world and explain relationships among variables to support or reject a pre-determined hypothesis (Moghaddam and Moballeghi, 2008).

Qualitative research methodology

Qualitative research on the other hand assumes an inductive, interpretivist approach in the collection and analysis of data (Denzin and Lincoln, 2011), although qualitative research using a positivist framework is not uncommon in the literature (Benbasat et al., 1987, Shek et al., 2005). Qualitative research can be traced to the 17th century in the ethnographic works of British colonialists on the indigenes of colonial states (Vidich and Lyman, 2003).

Historical development of qualitative research

Modern qualitative research evolved through five distinct phases each characterised by different intellectual and philosophical schools of thought (Denzin and Lincoln, 2011). These include the traditional period, the modernist phase, blurred genres, crisis of representation, and the fifth moment (Denzin and Lincoln, 2011).

The traditional period is represented in the works of classic ethnographers undertaking research in different cultures, most of which were published around 1900-1950s. The period is characterised by the appropriation of naturalism in social research with the aim of producing a social science version of literary naturalism, represented by the works of the Chicago School in the 1920's (Denzin and Lincoln, 2011). Qualitative research was marked by a focus on reliability, validity and objectivity in the interpretation of findings. The traditional period led to the modernist phase (1950s - 1970s), characterised by elements of the traditional period such as naturalism and social realism. Post-positivism emerged as a dominant theme during this period, and elements of quantitative research were appropriated to intellectually situate qualitative research in already established quantitative research methods (Becker, 2002). The period was marked by a push towards standardisation of qualitative data and quasi-statistical analysis of qualitative data (Becker, 1958). Rigidity in research was a core theme of the traditional and modernist era with a view that knowledge should be subject to verification. Reliability was interpreted as consistency and research was considered reliable if findings could be replicated by another researcher (Schwandt, 2014). Member checking, triangulation, peer debriefing and other methods were developed to enhance the validity of qualitative research (Cho and Trent, 2006).

The period of blurred genres, a successor to the modernist phase was characterised by a movement away from a post-positivist, quasi-quantitative approach, to a more pluralistic, interpretive paradigm (Denzin and Lincoln, 2011). While some research remained firmly within the positivist and post-positivist paradigm, there were elements of intellectual diffusion in published works, evident in the rejection of the validity and reliability checks

by constructivists and naturalists in favour of credibility and an emphasis of transferability of findings rather than generalisability (Lewis, 2009).

The fourth phase, crisis of representation, represents a period of emerging reflexivity in research and the erosion of objectivism as researchers sought new models of truth. The phase was marked by a lack of consensus between the different paradigms that inhibit qualitative research (positivists, post-positivists, constructivists, oriental inquirists and naturalists) on standards of reliability or validity in qualitative research with research being dispersed in various interpretive forms (Lewis, 2009). The final stage, the fifth moment defines the state of modern-day qualitative research still mired in the crisis of representation, with an emerging consensus that qualitative research cannot be removed from intrinsic characteristics of the researcher such as race, gender, and lived experiences. Thus previous issues settled in earlier phases of qualitative research such as reliability and validity of qualitative results became much more emergent and continues to generate considerable debate (Denzin and Lincoln, 2011).

The representation of the historical development of qualitative research is not distinctly chronological but rather reflect the dominant philosophical and intellectual viewpoint that underpinned the development of qualitative research and elements of each phase are seen in contemporary qualitative research (Silverman, 2016).

Qualitative research designs

Research designs used in qualitative research include ethnography, case studies, phenomenological studies and grounded theory. These different methods differ in the manner by which data are collected and analysed. Ethnography is mainly concerned with the study of people within a particular culture. Researchers using an ethnography approach usually immerse themselves into the culture for a defined period of time and findings are usually circumscribed to particular cultures and settings (Hammersley and Atkinson, 2007). Phenomenology is concerned with the study of an individual's lived experience within a particular setting. Truths are defined and explored within the confines of human experience

(Teherani et al., 2015). Phenomenological studies can adopt a transcendental and hermeneutic approach. Transcendental or descriptive phenomenology assumes that the lived experience of a phenomena contains characteristics that are commonly experienced by all who experience such phenomena. Hence, the goal of the researcher is to identify the universal essence of a phenomenon, requiring continuous reflexivity to ensure elimination of all forms of individual bias (Neubauer et al., 2019). Hermeneutic or interpretive phenomenology argues that individual experience is inextricably bound to the world within which they exist and a distinction between a person's experience and their background is impossible. The researcher's subjective experience cannot be separated from the interpretation of a phenomenon but can be a useful guide to inquiry (Neubauer et al., 2019).

A case study is a research method that involves a systemic and in-depth exploration of an individual, event or group of people in a natural setting (Baxter and Jack, 2008). Case studies can be classified according to time dimensions e.g. retrospective case studies, snapshot case studies and diachronic case studies (Starman, 2013). Cases studies can also be classified based on whether they lead to theory formation, e.g. atheoretical case studies, disciplined case studies, heuristic case studies, theory testing, building blocks and plausibility probe studies (Starman, 2013).

Grounded theory is an analytic framework concerned with the generation of theory to explain observed data (Noble and Mitchell, 2016). Theories are generated through methodological collection and analysis of data, which evolve over time as observation, analysis, and reflection on data continues (Glaser, 2017). In grounded theory, observations are first summarised into categories and explored within a theoretical sample. Relationships between categories are developed into themes and explored reflexively. As new data emerges, categories are further refined into themes and theories that help explain a given phenomenon (Charmaz and Belgrave, 2007). Theoretical sampling and constant comparison inform the methodological approach to data collection and analysis using grounded theory. Theoretical sampling is concerned with identifying where data will be collected based on

provisional theoretical concepts. Constant comparison is a method of analysis in which each interpretation of a finding is compared with existing interpretations to identify conceptual similarities, refine distinctive patterns among categories and further develop existing themes and patterns (Boeije, 2002). Theoretical sampling occurs alongside constant comparative analysis facilitating comparison with existing data. Constant comparison contributes to the internal and external validity of grounded theory research, informing reasonable generalisation of theories beyond subjects not studied but who experience the same phenomena under investigation (Boeije, 2002).

Application of qualitative research methods generally involve documentation, categorisation and exploration of data to illustrate relationships between identified concepts (Adams et al., 2007). Documentation refers to procedures through which qualitative data are collected which may be interviews or field notes from observations (Polkinghorne, 2005). Categorisation refers to the iterative conceptualisation and coding of data to generate meaning. The relationships between concepts developed are critically examined in a reflexive manner to form conclusions about the subject of inquiry (Dey, 2003).

Mixed methods research methodology

There is considerable debate among advocates of qualitative and quantitative research methods regarding issues of internal and external validity of findings, driven by existing tensions between the distinct philosophical traditions that underpin each paradigm. Qualitative purists thoroughly reject positivism, objectivism and the notion of time and context-free generalisations advocated by quantitative researchers (Lincoln et al., 2011). Quantitative purists, on the other hand, stress the need for the elimination of bias in social science research, and the desirability of time and context-free generalisations (Nagel, 2001). However, research methods are increasingly nuanced on the philosophical underpinnings of both qualitative and quantitative research; hence critics on both sides of the debate either implicitly or explicitly validate the notion of the intractability of a paradigm that combines qualitative and quantitative philosophies and their methods (Onwuegbuzie and Leech,

2005). While it may be impossible to reconcile the distinct philosophies that underlie qualitative and quantitative methodologies, various scholars have written extensively about the 'commonalities' that exist between both paradigms and the need to focus on pragmatism rather than adopting a rigid stance on research design (Johnson and Onwuegbuzie, 2004).

Research that combines elements of qualitative and quantitative paradigms in research design is referred to as a mixed methods approach. It is regarded as the 'third wave' in research and can be formally defined as "the type of research where the researcher mixes or combines qualitative and quantitative research techniques, methods, approaches, concepts or languages into a single study" (Johnson and Onwuegbuzie, 2004 p17).

Mixed methods research has been extensively used in a diverse range of research fields including in the development of outcome measures (Bailey et al., 2016, Eckard et al., 2017, Fielding, 2010). A mixed methods approach has the potential to combine both deductive and inductive reasoning providing researchers with a greater scope and theoretical lens through which research questions can be addressed (Almalki, 2016). A mixed methods approach can also provide greater understanding to research findings and contribute to methodological rigour (Chow et al., 2010). Research using both qualitative and quantitative attributes should clearly articulate how both paradigms are integrated and their respective contribution to the research question (Lingard et al., 2008).

Philosophical positions

This section explores the philosophical positions that underpin the major research designs and their application in research in order to inform the philosophical position taken in this thesis.

Qualitative and quantitative approaches are underpinned by distinct ontological and epistemological assumptions. These assumptions are linked to philosophical and often rhetorical questions in research, on the nature of reality and the basis of knowledge. Metaphysical and epistemological theories in philosophy provide support for methodological approaches used in research.

Ontology is a branch of metaphysics concerned with the nature of reality and how reality can be studied (Heidegger, 2014). Realism and idealism are two branches of ontology that shape the debate on the nature of reality. Realism is a school of thought that asserts that there is an objective reality that can be studied which is independent of a person's belief or understanding. Idealism on the other hand argues that reality is intrinsically linked to the human mind and socially constructed truths (Ritchie et al., 2013). Hence, the notion of an objective reality independent of the individual is impossible.

Epistemology is a branch of philosophy primarily concerned about the theory, nature and basis of knowledge (Ritchie et al., 2013). Epistemology examines the relationship between the researcher and knowledge (Carson et al., 2001), and explores the means by which knowledge is best acquired. A major epistemological school of thought holds that knowledge is best acquired through an inductive, 'bottom-up' process where observations are made of the natural world from which patterns and trends are established (Denzin and Lincoln, 2011). An alternative school of thought argues that knowledge is best acquired through a deductive, 'top-down' approach by testing hypothesis against observations (Ritchie et al., 2013). In an inductive approach, a hypothesis is first generated and tested against observations in the natural world. The hypothesis is either confirmed or rejected based on the observations (Ritchie et al., 2013).

Positivism, interpretivism, realism and pragmatism are major ideologies that reflect particular epistemological and ontological assumptions. Positivism and positivist research argues that reality is objective and can be independently studied through a controlled, structural approach by testing a hypothesis using rational and logical approaches (Carson et al., 2001). Positivism emphasises the distinction between positive and normative facts, and a rational and logical approach to research, often relying on mathematical and statistical means for data analysis. Interpretivism on the other hand makes less of a distinction between objective and subjective truths. The researcher is seen as an inextricable aspect of the research process. Truths are believed to be socially constructed and human interactions and

perception of reality are regarded as being fundamental to understanding a social phenomenon (Myers, 2013). Reflexivity is an important theme in interpretivism and truths are regarded as being multiple and socially dependent. Interpretivism is philosophically aligned to idealism and relies on situating and theorising truths in the social world within which they exist (Goldkuhl, 2012b).

Pragmatism emphasises the use of both interpretivist and positivist philosophy in research, with a particular view that both philosophies exist in a continuum rather than being distinct (Goldkuhl, 2012a). Pragmatism to some extent, avoids the contentions the debate around 'truth' and 'knowledge' embedded in positivism and interpretivism, and rather focuses on 'action' and 'change' in a social world (Kilpinen, 2008). Pragmatists argue that the meaning of an idea or phenomena is inextricably bound to its practical consequences (Goldkuhl, 2012b).

Realism reflects the ontological position of the independence of reality, both observable and non-observable, from the human mind (Kukla, 1998). Philosophic realism is 'the view that entities exist independently of being perceived, or independently of our theories about them' (Phillips, 1987 p 205). Critical realism has emerged as a dominant application of the realist perspective in social science research. Critical realist accepts the existence of an objective reality but argue that knowledge is historically, culturally and socially situated; hence, positivist reasoning alone cannot be exclusively relied on (Archer et al., 2016). Bhaskar (2013) outlines three inter-related ontological domains that constitutes critical realism: the real, actual and empirical domain. The real domain consists of structures, mechanisms, events, actions and experiences. The actual domain consists of events, actions and experiences while the empirical domain consists only of what is experienced. For critical realists, claims to knowledge rests on all three domains (Banfield, 2004). Subtle realism is another emergent philosophy from the field of realism. Subtle realism acknowledges the subjective nature of research but does not preclude the existence of an independent knowable phenomena (Hammersley, 2002). Claims of knowledge are ultimately constrained

by cultural assumptions and different methods and approaches may produce a different understanding of a concept (Duncan and Nicol, 2004). In articulating his approach to analysis, Hammersley defines subtle realism as an approach that rests on "the idea that research investigates independent, knowable phenomena. But it breaks with [realism] in denying that we have direct access to those phenomena, in accepting that we must always rely on cultural assumptions, and in denying that our aim is to reproduce social phenomena in some way that is uniquely appropriate to them" (Hammersley, 2013 p 52).

Reflecting on these philosophical positions, and the epistemological and ontological theories that underpin them, the researcher identifies as a subtle realist.

4.1.3 Study design

Research that aims to investigate the use of quality of life measures has relied on quantitative methods such as psychometric tests of validity and reliability (Coombes et al., 2016). However, as discussed in Chapter 3, these tests do not provide sufficient information on how the measures are understood and the problems faced by patients during completion. Qualitative methods such as semi-structured interviews have also been used to investigate the feasibility of using quality of life measures. However, concerns over interviewer bias remain. Cognitive interviewing techniques such as think-aloud interviews can be used to eliminate interviewer bias and accurately reflect issues faced by respondents completing a questionnaire (Howlett et al., 2018).

Cognitive interviewing techniques have their roots in information processing theories of psychology (Jobe and Mingay, 1989) and have been widely used in health research for developing, refining, evaluating or pre-testing questionnaires and quality of life instruments (May and Warren, 2001, Johnson-Kozlow et al., 2006). Cognitive interviewing methods are based on the theory of survey response which conceptualises response to questions as consisting of four cognitive phases (Tourangeau and Bradburn, 2010). The first stage is comprehension in which respondents attempt to make sense of the question. The next stage is retrieval in which the respondent undertakes a memory search for appropriate information

that would be used to answer the question as it is understood by the respondent (Tourangeau et al., 2000). The third stage is judgement in which the respondent estimates the relevance of the recalled information to the question as understood. The final stage is response in which respondents provides an answer to the question as understood (Tourangeau and Bradburn, 2010). Tourangeau and Yan (2007) suggests that errors can arise from any of these four distinct phases and responses can be influenced by question sensitivity and social bias.

There are generally two types of cognitive interviewing techniques — think-aloud interviewing and verbal probing. In think-aloud interviewing respondents are asked to verbalise their thoughts as they complete the questionnaires. The interviewer does not 'interfere' with the completion process and only interjects after a prolonged silence to encourage the respondent to continue thinking aloud (Fonteyn et al., 1993). The process through which the subjects arrives at an answer to a question is captured through notes or a recording device. Thus, the hallmark of a think-aloud interviewing process is the minimal or non-interference of the interview process by the interviewee. In verbal probing, responses to questions are probed as the respondents complete the survey question (Willis, 1999). In essence, as the interviewee answers the questionnaire, their responses are probed to elicit the basis for their response and any relevant information of interest to the researcher. There are two general approaches to verbal probing: concurrent and retrospective verbal probing techniques. In concurrent verbal probing techniques, the responses are probed during the course of the interview whereas in retrospective verbal probing, responses are probed after the interview (Willis, 1999). Probes used during the interview may either be scripted or unscripted.

Feasibility of completing quality of life measures can be determined by evaluating their acceptability, burden, and cognitive response issues faced by respondents during completion. The consensus-based standard for the selection of health status measurement instruments (COSMIN) identifies reliability, validity, responsiveness and interpretability as

four key measurement properties and characteristics of health-related outcome measures (Mokkink et al., 2010b). The COSMIN guidelines recommends the assessment of content validity in terms of the relevance and comprehensiveness of items in a questionnaire to the target population and to the purpose of measurement; and that such assessments should be judged by patients themselves (Mokkink et al., 2010a). The extent to which the health domains assessed in a questionnaire are relevant to the patient population, the description of the domains and the theoretical foundation through which the domains are derived should also be assessed (Mokkink et al., 2016). The guidelines also recommend the evaluation of the interpretability of items—the degree to which qualitative meanings can be appropriately assigned to an instrument's scores or change in scores (Mokkink et al., 2010c).

Think-aloud interviewing techniques provides an appropriate framework through which (i) important concepts of content validity such as relevance and comprehensiveness of domains of a questionnaire, and (ii) interpretability of items can be explored among patients. Think-aloud interview alongside retrospective verbal probing and a short semi-structured interview were used to evaluate the feasibility of completing the ICECAP-SCM, ICECAP-A and EQ-5D-5L in recruited patients. Think-aloud interviews alongside retrospective verbal probing were used to understand the nature of problems faced by participants completing the measures and explore these problems in a way that minimised interviewer interference. Semi-structured interviews consisting of scripted questions were used following the think-aloud interviews to further explore the acceptability of the measures and burden of completion.

Think-aloud transcripts were analysed systematically for errors in comprehension, retrieval, judgement and response. A mixed form of analysis was used in analysing the data. Although the nature of data was primarily qualitative, errors were systematically analysed and quantitatively reported. Responses were examined for errors in completion, based on four categories: comprehension, recall, judgement and response (Tourangeau and Bradburn,

2010). A fifth category 'struggle' was added to identify areas where the participants did not make an error but had difficulties reaching an appropriate response.

The use of a think-aloud technique in combination with a retrospective verbal probing technique in the study aimed to minimise interviewee bias and to understand participants' completion of the questionnaires unaided. The use of scripted probes following the think-aloud interview assessed issues of interest to the research such as acceptability of the questionnaires, burden of completion and comprehensibility of the questionnaire and its instructions. Unscripted probes were used to explore observed difficulties during completion of the questionnaires. Verbatims transcripts of both the think-aloud and semi-structured segments of the interview were thematically analysed using constant comparative methods.

Previous research on the use of quality of life measures in patients near the end of life has employed a mixture of qualitative and quantitative methods (Bailey et al., 2016, Hearn and Higginson, 1999, Murtagh et al., 2007). Hearn and Higginson used standard piloting techniques in assessing the psychometric properties of the Palliative Care Outcome Scale in patients receiving palliative care (Hearn and Higginson, 1999). In-depth face to face interviews were subsequently carried out in a small sample following completion of the measure, to explore acceptability and the views of people of the measure. Murtagh and colleagues used think-aloud interviewing alongside concurrent verbal probing techniques to refine a symptom questionnaire in patients with end stage renal disease (Murtagh et al., 2007). Bailey et al. (2016) used cognitive interviewing techniques in evaluating the feasibility of using an end-of-life-specific capability instrument with patients receiving care in a hospice. Think-aloud interviews alongside retrospective verbal probing, were used to explore patient and close person completion of quality of life and capability-based instruments. This thesis builds on the work done in a hospice setting by Bailey et al. (2016) and extends the ICECAP-SCM to patients with organ failure who received care in different settings.

4.2 Research aims and objectives

This research aims to determine the feasibility of completing the ICECAP-SCM, ICECAP-A and EQ5D-5L in patients with end stage heart failure, end stage COPD and end stage renal disease and to explore the impact of functional decline on health and capability wellbeing.

In order to achieve this aim, the research was designed to achieve the following objectives:

I. To explore difficulties in completing the ICECAP-SCM, ICECAP-A and EQ-5D-5L in terms of errors in comprehension, retrieval, judgement and response
II. To understand the views of participants about the ICECAP-SCM, ICECAP-A and EQ-5D-5L
III. To explore the impact of functional decline (in relation to terminal illness) on capability wellbeing and capabilities associated with the opportunity for a good death as captured by the measures.

The first objective was achieved through systematic analysis of errors in completion by independent raters. Comprehension, retrieval, judgement and response errors were reported for each participant group completing each measure. The pattern of errors across the measures provided greater clarity on the difficulties faced by participants completing the measures and the reason for such difficulties. Findings are reported in Chapter 5.

Through seeking the views of participants about the measures, the second objective aimed to specifically examine the acceptability, burden, clarity and interpretability of the measures across participant groups. Acceptability, burden, interpretability and clarity of items in a questionnaire are important aspects of both content and face validity (Mokkink et al., 2010c). Acceptability denotes the relevance of the items in a questionnaire to aspects of wellbeing deemed important by the patient. Burden of completion is the physical and emotional difficulties experienced by patients while completing the questionnaires. Examination of clarity and interpretability of a questionnaire focuses on the extent to which items in a questions are clearly understood and quantitative scores or changes in scores are

given appropriate qualitative connotations (Mokkink et al., 2010b). These aspects of content validity were examined by analysis of responses to individual items in the questionnaires, using retrospective verbal probes, in semi-structured interview following completion of the questionnaire. Findings are reported in Chapter 5.

The third objective was to understand the impact of symptoms associated with end stage organ failure on capability wellbeing and capabilities associated with the opportunity for a good death. Previous research have largely explored the impact of symptoms on physical wellbeing, emotional wellbeing (Murtagh et al., 2011b), and help seeking behaviour (Habraken et al., 2008) in patients with end stage organ failure. These studies report an increased symptom distress and health-related concerns as patients approach the end of life. Patients were also more likely to view their conditions as a normal part of day to day living rather than as a disability; and as such were less likely to request help.

There is however little or no research on how these findings and other symptoms related to their terminal condition affects capabilities associated with the opportunity for a good death, particularly among patients on the organ failure trajectory in different care settings.

Furthermore, questions remain about whether the ICECAP-SCM, alongside other measures, captures the effects of patient illness on broader aspects of wellbeing; and the extent to which the effects of symptoms related to terminal illness impact capabilities associated with day to day living. This element of the research involved thematic analysis of both the think-aloud and semi-structured segments of the interview to explore the effects of terminal illness on capabilities associated with day to day living, and the opportunity for a good death. Findings are reported in Chapter 6.

4.3 Research Setting and participants

The research was conducted in a tertiary hospital in England and patients with end stage heart failure, end stage COPD and end stage renal failure were recruited for the study. To assist in the identification of suitable patients, recruitment teams consisting of cardiac, respiratory and renal consultants, alongside nurse specialist and junior doctors were set up

in the heart failure, renal failure and respiratory unit. Each team assisted in recruiting patients from their respective units.

The research aimed to purposefully recruit 60 patients with end stage organ failure split equally across patients with end stage heart failure, end stage COPD and end stage renal disease. Although there is no specific guidance on an appropriate sample sizes for a think-aloud study, the sample size was deemed sufficient for the study as previous think-aloud studies had used much smaller sample sizes.

Think-aloud interviewing techniques alongside retrospective verbal probing techniques were used with all sixty patients.

Recruited patients were the following generic eligibility criteria:

- Participants were able to provide informed consent.
- Participants were willing and able to directly participate
- Participants able to communicate in the English language
- Participants were over 18 years of age.
- Patients who are not be able to give consent or directly participate in the study will be excluded from the study.

Due to uncertainty regarding prognosis and survival, common among people with end stage organ failure (Murray et al., 2005), prognostic criteria were developed in collaboration with consultants and nurses in the various units from which the patients were recruited. Over a three-month period, numerous meetings were held with consultants, patient groups, nurse specialists, and junior doctors in the relevant units to agree feasible prognostic criteria that could be used to identify potential participants and to set up a system to use them. The prognostic criteria were chosen using established clinical guidelines and advice from the clinical specialists involved in the study. The aim was that the use of the prognostic criteria would ensure patients recruited were in the last 12 months of life.

The following prognostic criteria were developed:

4.3.1 End-stage heart failure

1. History of severe heart failure at baseline (New York Heart Association Class III or IV) despite being treated medically with appropriate drugs as prescribed by their clinician;
2. A definite history of New York Heart Association Class IV CHF at hospital admission or ICU transfer as manifested by baseline dyspnoea at rest related to primary cardiac failure, systolic blood pressure 100 mm Hg or less related to primary cardiac failure, or a history of hypotension despite the use of recommended medications;
3. Chart documentation of CHF and a left ventricular ejection fraction less than or equal to 35%;
4. QRS with left bundle branch block configuration, and duration >120 ms.

The selected criteria were developed following consultations with consultant cardiologists and nurse specialists in heart failure. Prognostic classification schemes such as the American Heart Association classification scheme (Hunt et al., 2001) and the New York Heart Association classification scheme (Association, 1994) were used in developing the prognostic criteria. The selected criteria were also compared with current guidelines on the diagnosis and management of patients with end stage heart failure to ensure similarity (Ponikowski et al., 2016). In the Study to Understand Prognosis and Preferences for Outcomes and Risks of Treatment (SUPPORT) patients meeting one of the first three criteria were found to have a 12-month mortality of 38% (Levenson et al., 2000a). Another study reported a 50% mortality rate in patients with the above criteria (Cleland et al., 1999).

The selected criteria have been used in various studies involving patients with end stage heart failure (Wong et al., 2016, Aissaoui et al., 2017). Patients were required to meet all four criteria and were expected to die within the next 12 months.

4.3.2 End Stage Renal Disease

For patients with ESRD, one of the following criteria had to be met:

1. Glomerular filtration rate of < 15mL/min and/or
2. Diagnosed as ESRD* (i.e. eligible for dialysis in the anticipation that they will need renal replacement therapy or an indefinite haemodialysis) and
3. Managed by conservative care i.e. patients who decline renal replacement therapy or dialysis.

Patients may or may not have had dialysis in the past but patients participating in the study were expected to be on conservative therapy.

The first two criteria were previously used in studies of end-stage renal failure (London et al. 2001) and the UK registry definition of end stage renal failure (Cullen et al., 2013). However, evidence suggests that among patients diagnosed with ESRD, those who undergo renal replacement therapy or haemodialysis have greatly increased odds of survival that can last for years (Locatelli et al., 2004). Hence, the third criterion ensured that patients participating in the study could be appropriately described as being in an 'end of life' phase.

4.3.3 End-stage COPD

Patients suspected of having end stage COPD were expected to have at least two of the indicators below:

1. Disease assessed to be severe (e.g. FEV1 <30% predicted)
2. Recurrent hospital admissions (at least 3 in last 12 months due to COPD)
3. Fulfils long term oxygen therapy criteria
4. MRC grade 4/5 – shortness of breath after 100 metres on the level of confined to house
5. Signs and symptoms of right heart failure

6. Combination of other factors i.e anorexia, previous Intensive Therapy Unit/Non-Invasive Ventilation resistant organisms.
7. More than 6 weeks of systemic steroids for COPD in preceding 6 months

These criteria are based on the Gold Standard Framework Prognostic Indicator Guidance (Thomas, 2010) and have been used in prospective studies on end-stage COPD in the UK (Gore et al., 2000) and other countries (Habraken et al., 2008).

In addition to meeting at least two of the above criteria, patients were expected to die within the next 12 months.

4.4 Measurement instruments

The ICECAP-SCM, ICECAP-A and EQ-5D-5L were administered to all participants. The ICECAP SCM and ICECAP-A were developed from Sen's philosophical notion of capabilities (Sen, 1993). The EQ-5D is underpinned by the extra-welfarist framework (Culyer, 1989). A summary of the key attributes of each measure can be found in **Table *4*** below.

The ICECAP Supportive Care Measure (ICECAP-SCM) was developed as a self-complete economic tool for patients receiving supportive and palliative care (Sutton and Coast, 2014a). The ICECAP-SCM measures patients' wellbeing in terms of their capabilities when they are receiving supportive care at end of life. The conceptual attributes of ICECAP-SCM are the result of in-depth interviews conducted with older people at different points along the trajectory towards death. A detailed evaluation of the completion of the measure and how it compares to other widely used measures for economic evaluation has been successfully carried out in a hospice setting on people near the end of their lives, mostly with a cancer diagnosis (Bailey et al., 2016).

The descriptive measure covers seven attributes:

- Autonomy (Having a Say – your ability to influence where you would like to live or be cared for, the kind of treatment you receive, the people who care for you);
- Love (Being with People who Care about You – being with family, friends or caring professionals);
- Physical suffering (Physical Suffering – experiencing pain or physical discomfort which interferes with your daily activities);
- Emotional suffering (Emotional Suffering – experiencing worry or distress, feeling like a burden);
- Dignity (Dignity – being yourself, being clean, having privacy, being treated with respect, having your religious or spiritual beliefs respected);
- Support (Being Supported – having help and support);
- Preparation (Being prepared – having financial affairs in order, resolving things that are important to you, having treatment preferences in writing or making a living will).

The descriptive system has four levels for each attribute and attributes are expressed in terms of capabilities and frequency of symptoms. Scores from the ICECAP-SCM are converted to an index value which lie on a scale of 0 to 1 with 0 representing 'no capability' and 1 representing 'full capability'. Capability scores are derived using value-sets derived from the general population using preference-based elicitation methods (Huynh et al., 2017). A version of the ICECAP-SCM can be found in Appendix 5

The ICECAP-A was developed as a self-complete economic measure of capability for the general adult population. It comprises five attributes: Attachment (ability to have love, friendship and support), Stability (ability to feel settled and secure), Achievement (ability to achieve and progress in life), Enjoyment (ability to experience enjoyment and pleasure) Autonomy (ability to be independent) (Al-Janabi et al., 2012). Attributes are described in terms of capabilities. For example, 'I am able to be completely independent' 'I am unable

to feel settled and secure in any areas of my life'. Scores from the ICECAP-A are converted to an index value which lie on a scale between 0 and 1 with 0 representing 'no capability' and 1 representing 'full capability'. Capability scores were also derived using value-sets obtained from the general population using preference-based elicitation techniques (Flynn et al., 2015). A version of the ICECAP-A used in the study can be found in Appendix 6.

The EQ5D-5L is a self-complete economic tool developed as a preference-based measure of health. It consists of five attributes: mobility, self-care, usual activities, pain and discomfort, anxiety and depression (Devlin et al., 2016). The levels of attributes are expressed in terms of functionings and disease severity. For example: 'I have no problems in walking about' 'I have moderate problems doing my usual activities'; 'I have severe pain or discomfort'. The version of the EQ-5D-5L used in the research can be found in Appendix 7. The EQ-5D-5L is based on some elements of the extra-welfarist framework where health rather than utility is the focus of evaluation (Brouwer et al., 2008). Health gain is expressed through the Quality Adjusted Life Years (QALY). The EQ5D-5L is used to measure QALYs gained through application of a value set developed using preference elicitation methods (Oppe et al., 2007).

Table 4 Summary of key attributes of the ICECAP-SCM. ICECAP-A and EQ-5D-5L questionnaire

ICECAP-SCM	**ICECAP-A**	**EQ-5D-5L**
Having a say	Feeling settled and secure	Mobility
Being with people who care about you	Love, friendship and support	Self-care
Physical suffering	Being independent	Usual activities
Emotional suffering	Achievement and progress	Pain and discomfort

Dignity	Enjoyment and pleasure	Anxiety and depression
Help and support		
Being prepared		

4.5 Study set-up and data collection

All the research was conducted in a single acute hospital in the UK's West Midlands. During the development of the topic guide, information sheet and protocol, consultations were held with various stakeholders, including representatives from the Patient Advisory and Liaison Service (PALS), clinical consultants and nurse specialists. Revisions were made to the information sheet and protocol based on recommendations from these consultations. The information sheet and topic guide can be found in Appendix 8 and Appendix 9 respectively. The patient consent form can be found in Appendix 10.

The data collection procedure for the heart failure unit was discussed with a heart failure nurse specialist, following which a meeting was held with other health care professionals working in the heart failure unit. The aims and objectives of the research were presented at a scheduled Multidisciplinary Team meeting (MDT) involving heart failure consultants, heart failure nurse specialists and heart failure palliative nurse specialists. Following the presentation, the heart failure team agreed to support the research and a bi-weekly time and place for recruiting prospective participants was agreed.

A similar exercise was conducted with the staff in the Chronic Kidney Disease Community Support Team in charge of patients with ESRD under conservative (palliative) management. To obtain familiarity with the setting, the researcher was invited to attend scheduled clinics pending ethical approval from the Foundation Trust for the research. Notes were taken during each session and the nature of patients attending each session was observed. This

was done while awaiting formal approval of the Trust and involved the explicit consent of both the palliative nurse specialist in charge of the clinic and the patients that were attending the session. Relationships were developed with other health professionals and administrative staff responsible for running the clinics. From observing the clinics, it was observed that most of the patients attending the clinic had substantial mobility problems, hence a room situated close to the consultation room used by the nurse specialists was made available for the research by the administrative team. It was also observed that most patients attending the clinic had significant hearing difficulties and the nurses had to speak louder than normal when speaking to patients. Hence, during data collection deliberate attempts were made to speak slowly and carefully to ensure patients understood the task.

The data collection procedure for the COPD unit was developed in collaboration with a respiratory consultant who also served as the Principal Investigator (PI) for the research in line with the policy of the recruitment site. A meeting was held with other consultants, nurses and junior doctors within the unit where the research was introduced to the wider team and a weekly recruitment schedule was agreed. To facilitate collaboration with the supervisory team, a meeting was held between the PI and the academic team where data recruitment and other issues related to the research were discussed.

Prior to commencing the actual think-aloud interview, mock think-aloud exercises were held to appraise key aspects of the data collection procedures. An audio-recorded think-aloud interview with retrospective verbal probing was undertaken with a colleague, a fellow PhD student. The interview was encrypted, played back and assessed for clarity and coherence of the interview session. Feedback was received from the PhD student and recommended improvements were implemented. Another practice interview session was conducted with a nurse specialist in the heart failure unit with experience of caring for patients with end stage heart failure, sensitive interviewing and qualitative research. Notes were taken on the feedback from both sessions and the recording was listened to for clarity and coherence.

Following both think-aloud sessions, it was observed that answers to the preliminary think-aloud task were inadequate. The preliminary think-aloud task asks a participant to think out loud while counting how many windows were in their homes. In both sessions, the volunteers only reported the actual number of windows rather than a verbal description of their thought process. Hence, during the study, the think-aloud task was clearly explained to the participant before the preliminary task was undertaken.

4.5.1 Participant recruitment

Once ethics approvals and appropriate governances were achieved, participants were identified through relevant out-patient palliative clinics and wards in their different units. The recruitment process differed across the three units. Differences were due to administrative and structural factors peculiar to these units. The study set-up and pre-data collection procedures in the different units are described below.

End stage renal failure

All participants with end stage renal failure were recruited from a specialised out-patient clinic for patients with end-stage renal disease. The out-patient clinic is run by a dedicated out-patient support team consisting of renal nurse specialists and doctors. All patients attending the clinic were diagnosed as having end-stage renal disease and had voluntarily forgone curative options including dialysis and renal replacement therapy, opting instead for conservative management. Before each clinic, the list of prospective participants and their addresses was accessed from the online Portal of the recruitment site. The information sheet was sent to the patients before their scheduled appointment. On attending the clinic, the nurse specialist inquired if the patient had received the information sheet and were willing to participate in the research. The nurse specialist recorded those who declined to participate and their reasons for decline. Patients consenting to participate in the study were referred to the researcher. The researcher was located in a room close to the consultation

room used by the renal specialist nurses and consultants. Interviews were either conducted following consultation by the nurse specialist or in the patient's home at a later date.

End stage heart failure

Participants were recruited from both the outpatient heart failure palliative clinic and in-patient wards. Most participants were recruited from the wards because most of the patients attending the specialised heart failure clinic did not meet the specified prognostic criteria. Discussions with the nurses and consultants who ran the clinic revealed an administrative decision to expand the clinic to accommodate those who may have less severe heart failure. Interviews with patients from the heart failure palliative clinic took place in a separate room situated close to the consultation room used by the specialist nurse or consultant attending to the patient. In the wards, eligible patients were identified by the consultants and their team. Initial approach to the patient was be done by the consultant or health professional looking after the patient. Patients who agreed to learn more about the study were referred to the researcher. The information sheet was read to the patient and any questions they had were answered. Interviews were arranged with the patient at time of their choosing, giving them sufficient time to think about participating in the study.

End stage COPD

Participants were recruited from the respiratory unit of the hospital. All participants were recruited from the wards. Prospective patients meeting the recruitment criteria were identified and approached by the consultant or members of their team. During the initial approach, the participants were informed of their eligibility for a study and asked if they were willing to participate in the study. Participants who expressed a willingness to learn more about the study were referred to the researcher. Interested participants were approached by the researcher and the information sheet was given to them. The information sheet was read to the patient and any questions they had were answered. Interviews were

arranged with the patient at a time of their choosing, giving them sufficient time to think about participating in the study.

4.5.2 Interview Process

Participants were interviewed at their preferred time and location. Before each interview, the information sheet was re-read to them and any questions the participant had about the study were clarified. The patient was then asked to sign the consent form and verbal acknowledgement was also sought before each interview began as outlined in the topic guide (see **Appendix *9***).

The interview began with a recap of the study aims and an explanation of the format of the interview. Participants were asked some basic questions about their age, marital status and family circumstances. As a 'warm up' task, the participant was asked how many windows were in their houses, and were encouraged to verbalise their thoughts as they answered the question. In the event, the think-aloud warm up process was unsatisfactory, the researcher performed the task using his own home as an example. Subsequently, the participant was asked to think-aloud as they count how many doors were in their homes.

Following the warm-up task, participants were given the questionnaires and asked to concurrently verbalise their thoughts as they completed the measures. The interviewer sat quietly during the process. The order in which participants were presented the three questionnaires was varied to minimise bias. Following completion of the questionnaires by participants, difficulties encountered over the course of the interview were probed. The probes used a combination of scripted and unscripted questions. A guide to the probes can be found in the **topic guide** in Appendix 9. The interviews were transcribed verbatim and examined for errors in comprehension, recall, judgement and response. Transcripts were also analysed to identify any areas of 'struggle' (i.e. difficulty in answering that is not so severe as to constitute a response problem). Error scoring and analysis were used to examine the nature of difficulties encountered while completing the respective questionnaires.

4.5.3 Data processing and storage

Immediately after the interview, a record of the session was made on the participant's file using standardised notes developed for the research. When the interview took place in an out-patient clinic or a participant's home where their files were not readily available, a note was made on their electronic record through the Prescribing Information and Communication System at the earliest opportunity. All patients participating in the study were also registered on the Research Facilitation Group database following consent and conclusion of the interview in line with existing Research and Development Department policy of the recruiting site. Consent forms were triplicated and one copy given to the participant, another stored in the main site file and the third kept at the site file at the University of Birmingham.

Audio-recorded interviews were encrypted and transcribed verbatim by the researcher and by an independent transcription company. The quality of the transcripts was reviewed to ensure rigor and minimize errors (Poland, 1995). Confidentiality of all information was maintained in line with the Data Protection Act (Primary Care Trusts, 1998). Names and addresses of informants were removed and individuals were identified on transcripts by means of a serial number only. Reporting of data occurred in the form of anonymised quotes. Recordings were uploaded to a secured drive on a server and will be destroyed after 10 years, in line with University of Birmingham policy.

An encrypted audio device was used for all interviews. Interviews were encrypted immediately after each session and transferred to the university server at the earliest opportunity.

4.6 Ethical issues

The research involved administering sensitive questionnaires to individuals near the end of their lives using cognitive interviewing techniques. It is possible that during the course of the interview, there might have been incidental disclosures of malpractice or abuse by

participants. Had this happened, a protocol was in place to facilitate reporting of such to the relevant authority. In the event of such a disclosure, this was to be brought to the immediate attention of the nurse in charge of the ward, or where relevant, the responsible clinician in charge of the care of the patient. Members of the academic supervisory team were also to be notified of such a disclosure. Before the interview, written consent was sought from patients regarding the disclosure of malpractice or abuse that may come up over the course of the interview. If there were any fears regarding the immediate safety of the patient, plans were made to contact the Police using 999. However, no such disclosure of abuse or malpractice occurred during the interviews.

Possible challenges identified prior to commencement of the study included gate-keeping issues in relation to accessing patients, expected emotional distress from patients due to the sensitive nature of the questions asked and appropriately handling sensitive interviewing materials from patients in compliance with ethical requirements. A protocol was developed with the help of the recruitment team to deal with these issues and other forms of emotional distress in patients during the interview. Patients were also provided with phone numbers for third party agencies, to call if they needed further help following the interview.

The research was sponsored by the University of Birmingham after undergoing a favourable university-based ethical review process (ERN_16-1308). The research further obtained ethical approval from the North Wales Research Ethics Commission and the Health Research Authority (REC reference: 17/WA/0022). Approval was obtained from the Research Facilitation Group and Research and Development Department of University Hospitals Birmingham NHS Foundation Trust (Project reference: RRK 5927). These ethical approval documents can be found in Appendix 11.

4.7 Data analysis

All interviews were transcribed verbatim to reflect the appropriate context of words used, and provide sufficient clarity. Transcription is usually the first step in the analysis of

qualitative data and involves close observations and repeated listening to data. Transcription requires judgements about the level of detail to include in the transcription, interpretation and representation of data (Bailey, 2008). Selectivity, the level of detail to include in transcription, is an important aspect of the transcription process, and largely depends on the type of data and proposed analytic framework (Davidson, 2009). For example, audio-visual data may include some visual description of surroundings and non-verbal reactions to questions in order to provide sufficient context to data. Ideally, the level of detail to include in a transcript should be informed by the aims of the research and considerations about readability and accuracy (Bailey, 2008). Interpretation and representation of data is largely informed by the person undertaking transcription. Considerable knowledge about the subject area may be necessary for interpretation and accurate representation of data (Pope and Mays, 2013). However, the use of hired transcribers, who may have little knowledge of the subject area, is often inevitable in qualitative research. In cases where transcripts are delegated to an external authority, attempts should be made to provide sufficient context to the transcribers and careful consideration should be given to ethical and data protection issues (Tilley and Powick, 2002). Transcripts should be reviewed throughout the analytic process by the researcher to ensure accurate interpretation and representation of data.

Transcription of the qualitative data was done by the researcher and with the help of a recognised transcription company with a history of transcribing audio recordings for the Health Economics Unit at the University of Birmingham. Audio recordings were completely anonymised and ethical and data protection requirements were contractually considered. All transcripts were reviewed by the researcher for content and clarity.

Transcripts were segmented to material relating to each of the attributes on the three measures, and were distributed with instructions for analysis by five independent raters (HN and his four academic supervisors, AH, CB, JC,PK) for errors in comprehension, retrieval, judgement and response (Tourangeau and Bradburn, 2010). Transcripts were also analysed

to identify questions that participants struggled with in situations where there were no errors. A struggle was defined as instances where the participant had difficulties answering the final question even when their response was not judged to be associated with any errors.

A standardised classification scheme was employed to consistently identify the aforementioned types of response problems. The classification scheme was developed following an initial review of six transcripts (two from each of the organ failure group) and a consensus reached amongst the raters on responses that constituted an error. The classification scheme defined constituents of a comprehension error, retrieval error, judgement error, response error and struggle. It further outlined the process by which a consensus was reached in attributing an error, including the process for dealing with disagreement about the nature and classification of an error. A detailed outline of the standardised classification scheme developed here can be found in Appendix 12. The remaining transcripts were then independently examined for errors by each rater. The researcher then collated these independent judgements to identify those segments of the transcripts that would require further discussion in a consensus meeting. In situations where there was disagreement over the error classification, a consensus was reached amongst the raters in a meeting led by the researcher. Errors were reported in terms of the percentage error rate by group and measure completed. Qualitative quotes were used to illustrate the reasons the errors across the groups and to highlight issues around the acceptability of the measures and burden of completion. Think-aloud segments of the interview and responses to the probes and semi-structured interview were qualitatively analysed to explore the impact of functional decline on capability wellbeing and the opportunity for a good death.

The next section describes in greater detail the qualitative analytical procedure that led to the development of the qualitative section in Chapter 5 and Chapter 6.

4.7.1 Development of the qualitative analytical process

Qualitative accounts were used to illustrate the reasons for errors made by participants when completing the measures, and the acceptability and clarity of the measures, reported in Chapter 5. Qualitative accounts were also used to explore the impact of functional decline on capability wellbeing and capabilities associated with the opportunity for a good death, reported in Chapter 6. The analytic process that led to the development of both qualitative accounts differed.

Qualitative accounts used to illustrate the reasons for errors across participant groups were developed following a review of verbatim transcripts for errors, and a consensus among all raters on the error made by a participant while completing an item. Following consensus on the specific error (i.e. either comprehension, retrieval, judgement or response [C, T, J, R] made by a participant in an item, segments of the transcript that formed the basis of the error classification were highlighted and coded under the specific error type. Codes were classified by item, measure and participant group. These were managed using NVivo 12.1 software.

Errors made in each item were analysed and coded to illustrate the reasons for the errors. These reasons were compared across all participants making a specific error type in an item to identify similarities. Analytic accounts (Coast, 2017a)were then developed to illustrate the reasons for the errors across each item in the measure. The reasons for errors were also compared across items within each measure and analytic accounts developed to illustrate similar reasons across multiple items in the measure. All stages of the analytical process were reviewed by members of the supervisory team.

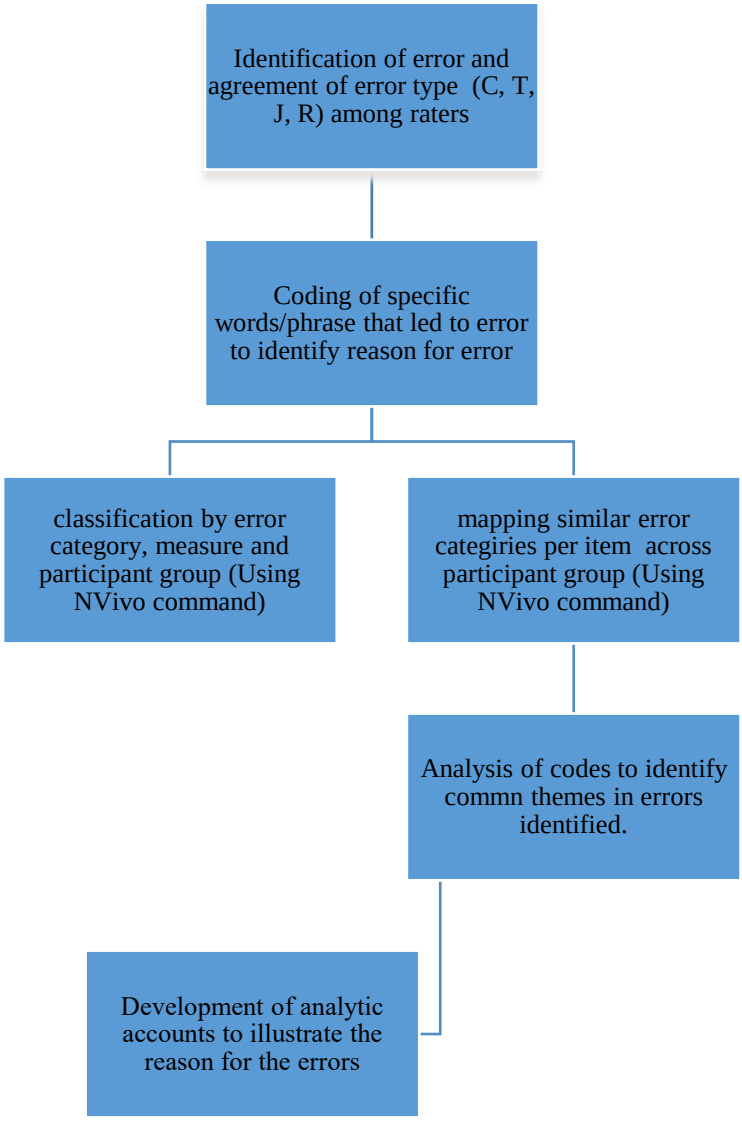
A similar process was used to develop the qualitative accounts that explored the acceptability of the measures and burden of completion. Specific probes were used to explore the acceptability, clarity and burden of completing the measures among all participants in the semi-structured interview that followed completion of measure.

Responses to each probe were coded to reflect a participant's view on the clarity of the measure and burden of completion. Codes were assigned to identify similarities in analytic accounts which were subsequently grouped to develop emerging themes which reflected the acceptability, clarity and completion of the measure. For example, in response to the question on achievement and progress a response error from the following quote was coded as 'difficulties linking achievement to wellbeing' which was then compared across similar responses to more clearly illustrate completion errors due to apparent difficulties in identifying the relevance of capabilities associated with Achievement to their wellbeing.

> *"I don't think I can progress anymore, I couldn't get anything extra that I do now anyway. I don't think I could so I don't know about that question so I don't know what to put for that so do you mind if I leave that one?"* [PT-28R]

Each stage of the analytic process was also reviewed by members of the supervisory team. The figure below provides a graphical illustration of the analysis of the qualitative account which identified themes in the errors reported in Chapter 5.

Figure 6 Flow chart to illustrate the development of qualitative accounts on the reasons for errors across the measures

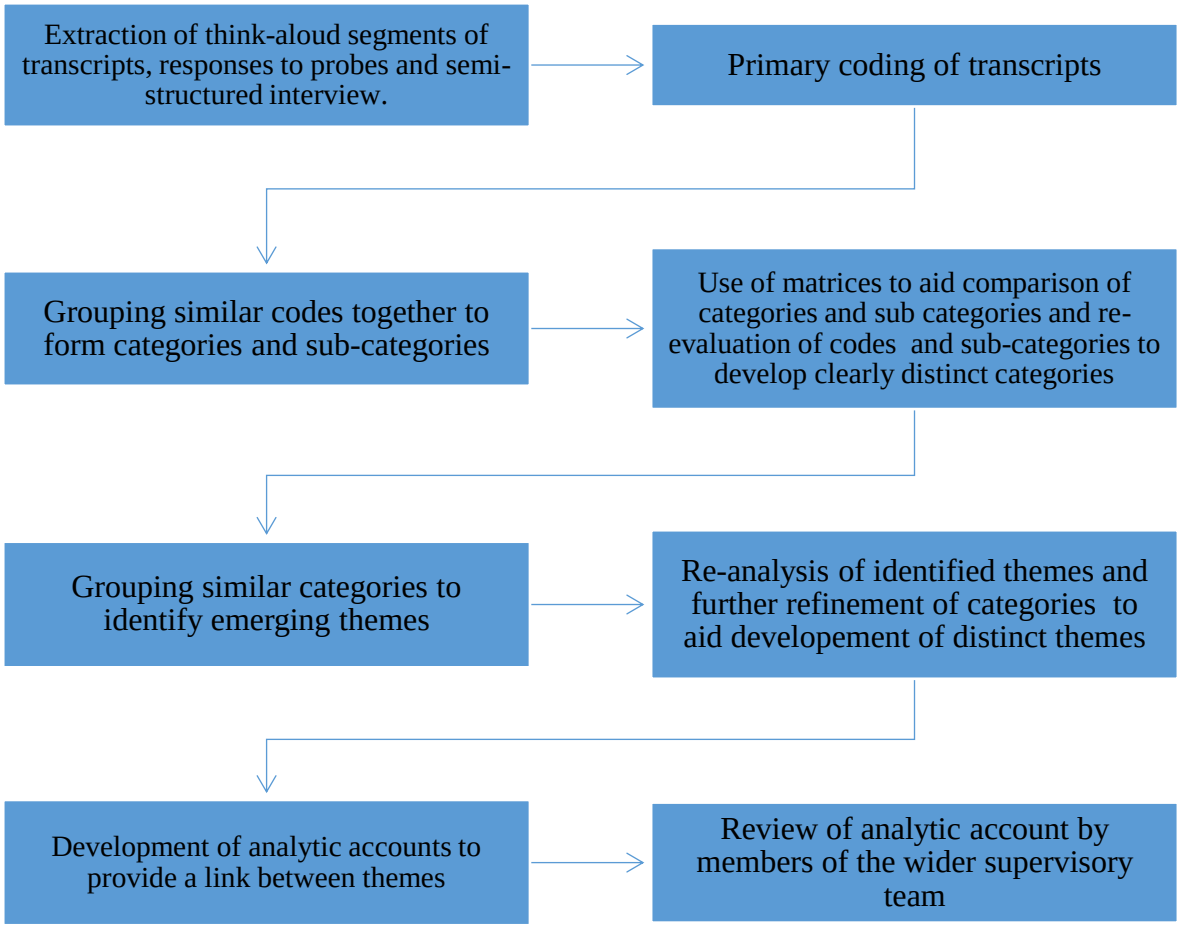


Think-aloud responses to items across the measures, responses to retrospective verbal probes and the semi-structured interview were analysed using constant comparative methodology (Coast, 2017b). Analysis focused on exploring the impact of functional decline on capability wellbeing, and capabilities associated with the opportunity for a good death.

Transcripts were read and re-read, and categories and sub-categories were developed to identify emerging themes. Matrices were used to aid comparison of codes and categories among the various groups. Analysis was carried out using the NVivo 12.1 software. Analytic accounts were developed by the researcher which were critiqued and reviewed by members

of the supervisory team who have different professional backgrounds and experience in conducting qualitative research with palliative patients. The diverse experience of members of the supervisor team provided a more comprehensive analysis that informed the development of the analytic account. As more data emerged, themes were examined in a reflexive process, analytic accounts were further developed and externally reviewed. Analytic accounts explored the nature of response problems during completion of the measures as well as the impact of functional decline on health and wellbeing. Themes were explored across participant groups and between participants on different phases of their dying trajectory. For example, the code ‘assistance’ contained sub-codes ‘family’ ‘friends’ and ‘carers’, which was further developed and integrated into a theme that explored the role of relatives and carers in coping with loss of capabilities. The figure below provides a graphical illustration of the analytical process that informed the development of the qualitative accounts that explored the impact of functional decline on capability wellbeing and the opportunity for a good death, reported in Chapter 6.

Figure 7 Flow chart to illustrate development of the qualitative account to explore the impact of functional decline on capability wellbeing



The flow chart above provides an overview of the analytic process that led to the development of the qualitative content of Chapter 6 and does not entirely reflect the sequential order of events. The development of the qualitative account involved an iterative process that involved all phases in a non-sequential order and multiple analytical accounts were written and reviewed by all members of the supervisory team.

4.8 Presentation of findings

The findings are reported in the following two chapters. The first details findings from the error analysis of think-aloud transcripts and the second reports the health profile of the measures across participant groups. Thematic analysis of the think-aloud aspects of the interview is used to further explore differences in health and capability profiles and the impact of functional decline on capability wellbeing.

In Chapter 5 the recruitment rate across the groups including the reasons for decline and non-participation is reported. The sociodemographic characteristics of participants, including recruitment location are outlined. Completion rate is reported for the whole sample and by condition group. Percentage error rates are reported for the whole sample group across each measure, and by error type across each measure. Percentage error rates are reported by condition group – on an aggregate level across each condition group and on a more disaggregate level for each measure by condition group. Errors are also reported across items of each measure. The reason for the errors is explored qualitatively alongside issues regarding acceptability and burden of completion.

Chapter 6 presents the responses to each attribute of the measures, and their actual utility and capability score. The health profiles of the measures and their corresponding utility and capability scores are compared across participant groups. Differences observed within the groups are further examined through thematic analysis of the qualitative data to explore the impact of functional decline on capability wellbeing and capabilities associated with the opportunity for a good death.

Chapter 5

Feasibility of completing the ICECAP-SCM, EQ-5D-5L and ICECAP-A

One means of assessing the feasibility of the different tools is to examine the degree of accuracy with which they were completed by the participants. This chapter reports findings from the error analysis of think-aloud transcripts. Errors and percentage error rates relating to the participants' completion of the three measures are presented, by measure and by group.

The chapter further explores the nature of completion difficulties faced by participants completing across items in the ICECAP-SCM, EQ-5D-5L and ICECAP-A. Analysis of the qualitative interview data is used to explore reasons for errors and illustrate issues relating to acceptability, clarity of the measures and burden of completion.

5.1 Recruitment rate

One hundred and fifteen patients were identified as being potentially suitable for inclusion in the study. Those approached included 46 patients with end stage renal disease, 41 patients with end stage COPD and 28 patients with end stage heart failure. Consent rates for the three groups are presented in Table 5

Table 5 Recruitment rate across participant groups

	ESRD	**End stage HF**	**End stage COPD**
Identified (n)	47	28	41
Excluded	12	4	3
Invited	35	24	38
Declined	17	3	17
Accepted	18 (38%)	21 (75%)	21 (51%)

Nineteen patients were excluded from the research by the recruitment team. Three patients were considered too ill to be interviewed and were excluded based on the advice of the

consultant and nurse specialist. One patient was discharged and another died before an interview could be arranged. Fourteen patients were excluded on the basis of the inclusion criteria. Of these, six had cognitive difficulties and were unable to understand the information sheet; eight patients could not communicate in the English language. Ninety-seven patients were invited to take part in the research. Thirty-seven patients declined participation and the reasons are presented in Table 6.

Table 6 Reasons for decline across participant groups

Reasons for decline	ESRD	COPD	HF	Total
Breathing difficulties	0	5	0	5
Poor eyesight	1	1	0	2
Upsetting speaking about quality of life.	1	2	0	3
Transport	4	0	0	4
Non-consent to interview/audio record	1	4	1	6
Pain	0	1	0	1
Bereavement	0	1	0	1
Fatigue	0	0	1	1
Unease with research	1	0	0	1
No reason given	9	3	1	12
Total	17	17	3	37

5.1.1 Reasons for decline across participant groups

Reasons for declining varied across the participant groups. Some were specific to prognosis while others were due to organisational arrangements of the clinics they were recruited from. Reasons for declining across the groups are discussed below.

End stage COPD

Patients with end stage COPD had a relatively higher rate of decline compared to the end stage heart failure group. Seventeen of 38 patients invited to participate in the research declined (44.7%). A considerable number of patients with end stage COPD declined to

participate due to breathing difficulties. Five patients declined due to significant breathing difficulties which limited their ability to perform the think-aloud task. These patients were willing to answer the questions but did not feel able to think-aloud about their answers. Four patients declined to be audio recorded and offered to complete the questionnaires without talking through their answers. Two patients declined to participate because they did not wish to speak about their quality of life or reflect on the impact of their illness on their wellbeing. One patient declined to participate due to pain and another declined due to a recent bereavement. One patient declined to participate due to a poor eye sight. Three patients did not give a reason for decline.

End stage renal disease

Patients with ESRD had the highest rate of decline among the three condition groups. Seventeen of 35 patients invited to participate in the research declined (48.6%). A considerable number of patients declined due to logistical issues specific to the recruitment site. An outpatient clinic was provided for patients with ESRD who were being managed conservatively. Consequently, most patients attending the clinic were reliant on the patient transport service or a family member to get them home. Hence, participation in the study was dependent on the agreement of family members or close persons assisting the patient by waiting whilst the interview was conducted. Four patients declined due to logistical issues with transportation. One patient declined due to poor eye sight. One patient expressed reservations about participating in research. One patient declined to be audio-recorded. Nine patients offered no reason for decline.

End stage heart failure

Patients with end stage heart failure had the lowest rate of decline across the groups. Three of 24 patients invited to participate in the research declined (12.5%). One reason for the higher rate of consent observed in the heart failure group may be the ability of patients to perform the think-aloud task without breathing difficulties. One patient declined due to fatigue and another declined to be audio recorded. One patient offered no reason for decline.

5.2 Basic socio-demographic information and health characteristics of participants

Interviews were conducted between June 2017 and November 2017. Eighteen patients with end stage renal disease (represented as PT-*n*K – kidney - in quotes), 21 patients with end stage heart failure (represented as PT-*n*H – heart - in quotes) and 21 patients with end stage COPD (represented as PT-*n*R – respiratory - in quotes) participated in the study. Patients included in the study were aged between 35 and 95 years old. Forty-three out of 60 participants were over 75 years old.

Participants were recruited at various stages of their terminal illness including very near the end of life with 23 of the 60 participants who were interviewed having died within 8 months of the interview and 19 of these participants having died within four months of being interviewed. In the remainder of this thesis, participants who died within four months of being interviewed will be referred to as being in the late end of life phase.

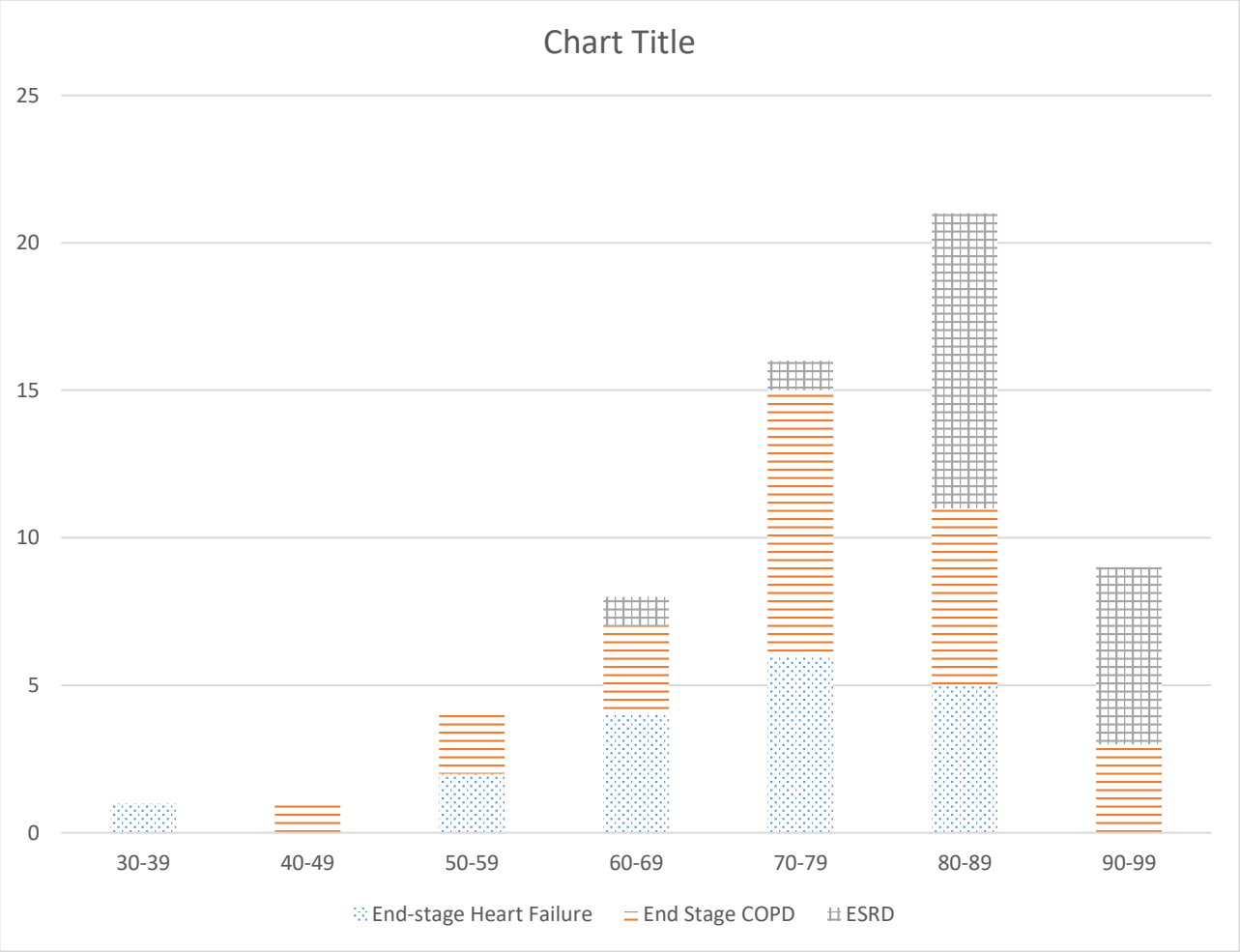
Demographic characteristics of participants included in the study are presented in Table 7.

Table 7 Basic socio-demographic information of participants

CHARACTERISTICS	ESRD [n=18]	COPD [n=21]	HF [n=21]	Total [n=60]
Age Group (years)				
30-39	0	0	1 (5%)	1 (2%)
40-49	0	1 (5%)	0	1 (2%)
50-59	0	2 (9.52%)	2 (10%)	4 (2%)
60-60	1 (6%)	3 (14%)	4 (19%)	8 (13%)
70-79	1 (6%)	9 (43%)	6 (29%)	16 (27%)
80-89	10 (56%)	6 (29%)	5 (24%)	21 (35%)
90-99	6 (33%)	0	3 (14%)	9 (15%)
Gender				
Male	11 (61%)	6 (29%)	15 (71%)	32 (53%)
Female	7 (39%)	15 (71%)	6 (29%)	28 (47%)
Race				
White	14	20	20	54 (90%)
Black	3	1	1	4 (8%)
Asian	1	0	0	2%
Recruitment location				
Outpatient clinic	18	0	2	20 (33%)
Ward	0	21	19	40 (67%)

Participants with ESRD were generally older than participants with end stage COPD and end stage heart failure. The mean age of participants with ESRD was 85 years while the mean age of participants with end stage heart failure and end stage COPD was 74 and 73 years respectively. Overall, the mean age of the participants was 77 years and the median age was 80 years. The distribution of participants across age groups is shown in Figure 8.

Figure 8 Age distribution of participants



5.3 Reporting for the whole sample group

5.3.1 Completion of the measures

The mean completion time for the think-aloud task was 25 minutes. Completion time varied between 14 and 49 minutes. Mean completion times for each of the three groups were very similar, and are shown in

Table 8.

Table 8 Completion time across the groups

	ESRD	End stage COPD	End stage HF
Mean completion time (mins)	27	23	23
Range (mins)	15—39	16—40	14—49

5.3.2 How many participants were able to complete the measures without errors?

Absolute errors in completion across the measures

All participants completed all three measures. Thirty-four of the 60 participants (56.7%) completed all three measures without any errors, although six of these 34 participants did experience struggle. The ICECAP-A had the highest number of participants completing the measure without an error (52 out of 60) but two experienced struggle. One participant struggled with 'achievement' and the other struggled with the 'autonomy' attribute of the ICECAP-A. Participants struggled to identify capabilities associated with achievement and progress:

> "*I really don't know what to make of it to be honest…this is what I'm thinking, I can make what I like out of what I want to…I'm thinking, if I want something, if I work hard then I'll probably get it. I'm trying so hard…I don't know what to say*" [PT-17R]

The ICECAP SCM and the EQ-5D-5L were each completed by 45 of the 60 participants without an error. Six participants without errors in the EQ-5D-5L did struggle. Two participants struggled with 'mobility', two participants struggled with 'usual activities', one participant struggled with 'self-care', and one participant struggled with the 'anxiety' attribute of the EQ-5D-5L. Some of these participants appeared to struggle with the recall period of the EQ-5D-5L (which asked about 'today'), which they felt did not reflect fluctuations in their condition that affected how they managed from day to day. Participants struggled with the recall period of the attributes 'self-care' (PT-38K) and 'usual activities' (PT-28R)

> *"I should say, I don't really know because I don't know how they'd class it myself to be honest…I should say they're moderate, I mean they can be severe I suppose but it just, depends how you feel yourself"* [PT-38K]

> *"I mean I could do both of those, I have moderate problems doing my usual activity. And I have severe problems doing my usual activity. It's all according how I am on the day I mean so I don't know which one to tick on that one... I'll put the moderate one. But again like it's all according to what they talk about usual activities, what's a usual activity? I don't do work and I don't do housework, I don't do much leisure activities now because of the shortness of breath"* [PT-28R]

Two participants struggled with the mobility attribute on EQ-5D-5L as they reflected on their own state.

> *"If I don't agree with what's there, what do I put?... I have no problems in walking about but I do, so what would I put there? ...I can't walk at all. So just do what?"* [PT-14R]

Although these participants struggled in responding to the questions while reflecting on their state, they were able to provide an appropriate response.

Two participants who completed the ICECAP-SCM without error struggled with the 'love and affection' and 'preparation' attributes. Participants appeared to struggle with understanding the descriptive level of the 'love and affection' attribute, expressed in terms of capabilities (PT-10K)

> *"I don't understand that last question. If I want to, I am able to be with people who care about me most of the time, which is what I would like. At least they've got my interests at heart"* [PT-10K]

Table 9 summarises the struggles experienced and errors made by participants while completing the ICECAP-SCM, ICECAP-A and EQ-5D-5L.

Table 9 Errors, struggle and percentage error rates in completion of the measures

	ESRD [n=18]	**COPD [n=21]**	**HF [n=21]**	**Total [n=60]**
EQ-5D-5L (a=5)	**b=90**	**b=105**	**b=105**	**b=300**
Errors (P)	2 (11%)	6 (29%)	6 (29%)	14 (23%)
Struggle (P)	1 (6%)	3 (14%)	2 (10%)	6 (10%)
No error/struggle (P)	15 (83)	12 (57%)	13 (62%)	39 (65%)
Error (n)	2 (2.2%)	7 (6.7%)	10 (9.5%)	19(6.3%)
Struggle (n)	1 (1.1%)	3 (2.9%)	2(1.9%)	6(1.7%)
ICECAP-A (a=5)	**b=90**	**b=105**	**b=105**	**b=300**
Errors (P)	1 (6%)	2 (10%)	5 (24%)	8 (13%)
struggle (P)	1 (6%)	1 (5%)	0	2 (3%)
No error/struggle (P)	16 (89%)	18 (86%)	16 (76%)	50 (83%)

Error (n)	1(1.1%)	3 (2.9%)	5 (4.8%)	9(3%)
Struggle (n)	1 (1.1%)	1 (1%)	1 (1%)	3(1%)
ICECAP-SCM (a=7)	**b=126**	**b=147**	**b=147**	**b=420**
Errors (P)	5 (28%)	6 (29%)	4 (19%)	15 (25%)
Struggle (P)	2 (11%)	0	0	2 (3%)
No error/struggle (P)	11 (61%)	15 (71%)	17 (81%)	43 (72%)
Error (n)	11 (8.7%)	7 (4.8%)	6 (4.1%)	24 (5.7%)
Struggle (n)	2 (1.6%)	1 (0.7%)	0	3 (0.7%)
Total (a=17)	**b=306**	**b=357**	**b=357**	**b=1020**
Error (P)	7 (39%)	10 (47.6%)	9 (42.9%)	26 (43.3%)
Struggle (P)	2 (11%)	2 (10%)	2 (10%)	6 (10%)
No error/struggle (P)	9 (50%)	9 (43%)	10 (48%)	28 (47%)
Error (n)	14 (4.6%)	17(4.8%)	21 (5.9%)	52(5.1%)
Struggle (n)	4 (1.3%)	5 (1.4%)	2 (0.6%)	12(1.2%)

***Error(P)**: Number of persons with errors*
***Struggle (P)**: Number of persons with struggle without error*
***No error/struggle (P)**: Number of persons without error or struggle*
***Error (n):** Number of errors*
***Struggle (n)**: Number of struggles*
***n**: number of persons in a group*
***a**: Number of attributes*
***b**: Number of opportunities for error [a*n]*

Participants completing the ICECAP-A had the lowest error rate while error rates in the EQ-5D-5L and ICECAP-SCM were similar. With 60 participants responding to the seven attributes or questions on ICECAP-SCM, there were 420 opportunities for either error or struggle; 24 errors (5.7%) and 3 struggles (0.7%) were identified. There were 300 opportunities for error or struggle for the EQ-5D-5L and 19 errors (6.3%) and 6 struggles (1.7%) were identified. In the case of ICECAP-A, there were nine errors (3%) and three struggles (1%) (as with EQ-5D-5L, there were 300 opportunities for error/struggle).

5.3.3 What problems did participants have while completing the measures?

Nature of errors across the measures

Response and comprehension errors were the most common error types. Average response error rates across groups were low (3.5%) but varied slightly across the measures. The EQ-5D-5L had the highest response error rates across the measures (6%). The ICECAP-SCM and ICECAP-A had the lowest response error rates (3.3% and 1.3% respectively). The nature of errors across the measures is shown in Table 10.

Comprehension error rates across the groups were low. The EQ-5D-5L was entirely free of comprehension errors. The ICECAP-SCM and ICECAP-A had comprehension error rates of 2.4% and 1.3% respectively. Retrieval and judgement errors were rare across the groups. There was one instance of retrieval error in the ICECAP-A and two instances of judgement error in the ICECAP-A and EQ-5D-5L respectively.

Table 10 Nature of errors and percentage error rates across the groups

	ESRD [n=18]	**COPD [n=21]**	**HF [n=21]**	**Total [n=60]**
EQ-5D-5L (a=5)	**b=90**	**b=105**	**b=105**	**b=300**
Comprehension	0	0	0	0
Retrieval	0	0	0	0
Judgement	0	0	1 (0.1%)	1 (0.3%)
Response	2 (2.2%)	6 (5.7%)	10 (9.5%)	18 (6%)
ICECAP-A (a=5)	**b=90**	**b=105**	**b=105**	**b=300**
Comprehension	1 (1.1%)	1 (1.0%)	1 (1.0%)	3 (1%)
Retrieval	0	0	1 (1.0%)	1 (0.3%)
Judgement	0	0	1 (1.0%)	1 (0.3%)
Response	0	2 (1.9%)	2 (1.9%)	4 (1.3%)
ICECAP-SCM (a=7)	**b=126**	**b=147**	**b=147**	**b=420**
Comprehension	5 (4.0%)	4 (2.7%)	1 (0.7%)	10 (2.4%)
Retrieval	0	0	0	0
Judgement	0	0	0	0
Response	6 (4.8%)	3 (2.0%)	5 (3.4%)	14 (3.3%)
Total (a=17)	**b=306**	**b=357**	**b=357**	**b=1020**
Comprehension	6 (2.8%)	5(1.4%)	2 (0.6%)	13 (1.3%)
Retrieval	0	0	1 (0.3%)	1(0.1%)
Judgement	0	0	2 (0.6%)	2 (0.1%)
Response	8 (3.7%)	12 (3.4%)	16 (4.5%)	36 (3.5%)

5.4 Do participants with organ failure make errors when reporting outcome measures?

5.4.1 Aggregate error and struggle across participant groups

One participant with end stage heart failure and another with end stage COPD made errors while completing all three measures. No participant with ESRD made errors across all three measures.

As shown in *Table 9,* 11 of 18 (61.1%) participants with ESRD completed all three measures without error. One participant who made no errors struggled with the ‘preparation’ attribute

of the ICECAP-SCM and another struggled with the 'self-care' attribute of the EQ-5D-5L. Eleven of 21 (52.4%) participants with end-stage COPD completed all three measures without error. Two participants without an error struggled with the 'usual activities' attribute of the EQ-5D-5L. Twelve of 21 (57.1%) participants with end stage heart failure completed all three measures without error. One participant without error struggled with completing the 'mobility' attribute and another struggled with the 'anxiety' attribute of the EQ-5D-5L.

5.4.2 Errors and percentage error rates by condition

ESRD

More participants (n=5, 27.8%) in the ESRD group made an error when completing ICECAP-SCM than made an error on EQ-5D-5L (n=2, 11.1%) or ICECAP-A (n=1, 5.6%). Error rates across the measures (rates allow for the fact that some individual participants made errors across multiple attributes within the same measure) were similar for EQ-5D-5L (2.2%) and ICECAP-A (1.1%), but higher for ICECAP-SCM (8.8%).

End Stage COPD

EQ-5D-5L and ICECAP-SCM were both completed without errors by 15 of the 21 participants with end stage COPD (71.4%). However, error rates varied slightly across the measures. The EQ-5D-5L had a slightly higher error rate (6.7%) than the ICECAP-SCM (4.8%). The ICECAP-A was completed without error by the greatest number of patients (19 of 21) and had the lowest error rate (2.9%).

End Stage HF

Of the 21 participants with end stage HF, 15 (71.4%) completed the EQ-5D-5L without errors, 16 (76.2%) completed ICECAP-A without errors and 17 (81%) completed ICECAP-SCM without errors. Although the error rates for the ICECAP-A (4.9%) and ICECAP-SCM (4.1%) were similar, the error rate for the EQ5D-5L (9.6%) was higher.

5.4.3 What types of errors were made by participants for each questionnaire and with each condition?

While the section above looked at the total number of participants in each group who made errors across each measure, this section looks at type of errors made by participants across each measure.

EQ-5D-5L

There were no comprehension or retrieval errors for the EQ-5D-5L and across the three participant groups, only one participant made a judgement error (from HF group). However, there was a markedly higher number of participants from the end stage heart failure group (10) who made a response error, compared to two ESRD and six end stage COPD participants. Similarly, error rates were highest in the end stage heart failure group (9.6%) and lowest in the ESRD group (2.2%).

ICECAP-A

There was a low rate of errors made by participants when completing the ICECAP-A. Comprehension and response errors were the most common. Participants with ESRD and end stage COPD made no retrieval and judgement errors. Comprehension errors were made by participants from all groups while response errors were found only in the end stage COPD and end stage heart failure group.

ICECAP-SCM

Comprehension and response errors were the only common errors across the three participant groups (there were no retrieval and judgement errors) completing the ICECAP-SCM. ESRD and end stage COPD group each had four participants with comprehension errors while completing the ICECAP-SCM compared to just one participant with end stage heart failure. However, participants with ESRD had a slightly higher comprehension error rate than participants with end stage COPD (5 (4.0%) versus 4 (2.7%)); one participant with ESRD made two comprehension errors while completing the ICECAP-SCM. The number of participants who made response errors when completing the ICECAP-SCM across the

groups was quite similar. The end stage heart failure and ESRD groups each had three participants with response errors while two participants with end stage COPD made response errors while completing the ICECAP-SCM. However, the response error rates varied slightly across the groups. Participants with ESRD had higher response error rates (6 (4.0%)) than participants with end stage heart failure (5(3.4%)). Participants with end stage COPD had the lowest response error rates (3(2.0%)).

5.5 What difficulties did participants face while completing the ICECAP-SCM?

While the section above examined the types of errors made across the three measures, this section specifically looks at the type of error made across each attribute of the ICECAP-SCM and how participants across the groups differed in their ability to complete the ICECAP-SCM. Qualitative data were used to explore the reasons for the errors made by participants across the attributes.

As presented in Table 9, 45 of 60 participants completed the ICECAP-SCM without errors. Whilst the number of participants making errors when completing ICECAP-SCM was similar across the three patient groups, the error rates did vary. The ESRD group had the highest error rate (8.7%) across the groups. End stage heart failure (4.1%) and end stage COPD (4.8%) had a similar error rate. Response and comprehension errors were the only errors in the ICECAP-SCM (Table 10) and participants with ESRD had the highest response and comprehension error rates (4.8% and 4% respectively).

Table 11 Nature of errors across attributes of the ICECAP-SCM among participant groups

Error	Choice	Love	Physical Suffering	Emotional suffering	Dignity	Support	Preparation	total
Comprehension								
ESRD	0	1	0	1	0	0	3	**5**
COPD	0	0	1	0	0	0	2	**3**
HF	0	0	0	1	0	0	1	**2**
Retrieval								
ESRD	0	0	0	0	0	0	0	**0**
COPD	0	0	0	0	0	0	0	**0**
HF	0	0	0	0	0	0	0	**0**
Judgement								
ESRD	0	0	0	0	0	0	0	**0**

COPD	0	0	0	0	0	0	0	**0**
HF	0	0	0	0	0	0	0	**0**
Response								
ESRD	0	0	2	3	0	1	0	**6**
COPD	0	0	0	1	0	1	2	**4**
HF	1	0	0	0	1	1	1	**4**
Total	**1**	**1**	**3**	**6**	**1**	**3**	**9**	**24**

Participants had more errors relating to two attributes (preparation and emotional suffering) compared to the other attributes, as can be seen in Table 11. Participants had nine errors in 'preparation' and six errors in 'emotional suffering' while completing the ICECAP-SCM. Together, both attributes were responsible for 15 of 24 (62.5%) errors observed in the ICECAP-SCM. The attributes 'choice', 'affection', and 'dignity' had the fewest errors. Participants with ESRD made the most errors while completing the ICECAP-SCM and made the most errors in 'emotional suffering' and 'preparation'. Seventeen of 15 errors in both attributes were made by participants with ESRD.

Most of the errors identified in 'preparation' were comprehension errors. Participants who made errors on this attribute often associated 'preparation' with the ability to get things done, rather than advanced care planning, financial and legal preparations at the end of life, as in the following two quotes.

> *"Some of the work I can do. I can dress myself, I can wash myself, I don't need anybody to give me baths so far I'm okay and on the food side, when she [the carer] come in the morning, she cook my food and wash my clothes"* [PT-18K]

> *"If I wanted something, there's always somebody in my family that I could depend on and they'd come and help me out"* [PT-69R]

Six of ten comprehension errors in the ICECAP-SCM were found in 'preparation', and half of these comprehension errors were found in participants with ESRD. Participants appeared

to conflate ‘preparation’ with day to day decision making rather than decision making at the end of life.

> *“I deal with my own. I look after myself and make up my own mind. Well some things I could say no to and yes if you know what I mean, the jobs that I wouldn’t do I don’t want to do. I can do the jobs they're asking but I don’t want to that's all”* [PT-42H]

> *“I make a decision myself. I have to ask somebody if I want to make a decision very rare now. I make my own decisions”* [PT-63K]

‘Emotional suffering’ has the second highest error rate in the ICECAP-SCM and again, most of these errors were made by participants with ESRD (4 of 6 errors). Participants seemed unable to understand the description of ‘emotional suffering’ and in some instances appeared to conflate the attribute with aspects of their lives that appeared unrelated to the question.

> *I can’t really understand that question. I don’t have anything to argue about and nobody to argue with. I don’t get emotional about things like that, no. I can’t get emotional about anything.* [PT-30K]

> *I don’t know what to say, I’d say number two or is it number three but I don’t know the answer. It’s… I don’t know what you mean. I mean I suffer most proper pain in my head or what, I don’t know what you mean I don’t understand that question, I will come back to it* [PT-23R]

Some comprehension errors appeared to be due to difficulties in understanding the description of the levels of attributes expressed in terms of capabilities.

> *“I want to be able to - people who care about me some of the time. Well, I know my son cares about me. I want to be able to be with people who care about me only a little of the time. No, I don't want - I want the answer to be able to be with people who care about me. I don't understand half of them, really”* [PT-11K]

There were fourteen response errors among participants who completed the ICECAP-SCM and half of these errors were also found in ‘emotional suffering’ (4 errors) and ‘preparation’ (3 errors). Participants with ESRD had the most response errors while completing the ICECAP-SCM, and most of these errors were found in ‘emotional suffering’. Most response errors were due to participants choosing more than one response option, suggesting that participants may not have understood the instructions in the questionnaires. Furthermore, response errors were due to participants being unable to distinguish between levels of capabilities expressed in the measure. For instance, one participant’s stated

preference suggested a lack of capabilities in end of life decision making and advanced care planning while her experience appeared to suggest otherwise.

> *"I've not had the opportunity, the right opportunity to make preparations in my life. Just my son. I have, I've made arrangements to give things to my son."* [PT-14R (Ticked I have not had the opportunity to make any of the preparations I want to make]

Response errors were also due to some participants refusing to choose a response option. Although a relevant experience relating to the question was recalled, they felt their experience did not reflect the question being asked.

> *"I: You didn't tick any box for physical suffering or on the emotional suffering or being supported. Why is that?*
> *IV: I often experience physical discomfort but it's just my legs and my back and if I have a paracetamol and then it eases off, you see, and I can walk… it's not discomfort, it isn't. I think it's just an everyday sort of thing"* [PT-11K]

> *"I: What about emotional suffering? You didn't tick any of the boxes there. Why is that?*
> *IV: Emotional suffering, no. I don't have emotional suffering at all. I just carry on with daily life as I go. If I can't do anything I don't do it…I don't worry about anything really. I lose my temper if I can't do anything. I fall out with myself… I don't think it's emotional suffering really, no"* [PT-11K]

Some participants refused to consider questions that appeared sensitive. For example, one participant refused to answer a question on her emotional wellbeing and when probed further during the interview appeared unwilling to discuss the question further.

> *"I: …this one about emotional suffering while experiencing worry or distress like a burden, you didn't really, so, you didn't really feel like you could answer the question?*
> *IV: I don't know, I don't know. That one was the hardest.*
> *I: This is the hardest for you?*
> *IV: Hmm.*
> *I: And why, and is there any particular reason why it was the hardest one?*
> *IV: I don't want to say, I don't want to say"*
> [PT-33R]

In some instances, although a response option was chosen, participants suggested that their desired response was missing from the available response options

> *"I'm going to put number four which is I am able to maintain my dignity and self-respect most of the time, all the time to be honest but that's not on there"* [PT-24H]

> *"I am able to have the help and support that I need most of the time, should be all of the time but we'll have most of the time"* [PT-24H]

5.6 What difficulties did participants face while completing the ICECAP-A?

This section examines the specific errors made across each attribute of the ICECAP-A. Qualitative data is used to explore the reasons for the errors made. Fifty-two of 60 participants completed the ICECAP-A without any errors although 2 respondents experienced struggles (see Table 9). The ICECAP-A had the lowest number of persons with errors and the lowest error rate across all three measures. Participants with end stage HF had higher error rates and number of persons with errors compared to those with ESRD and end stage COPD.

Table 12 Nature of errors across attributes of the ICECAP-A among participant groups

Error	Stability	Attachment	Autonomy	Achievement	Enjoyment	Total
Comprehension						
ESRD	0	0	0	1	0	**1**
COPD	0	0	0	1	0	**1**
HF	0	0	0	1	0	**1**
Retrieval						
ESRD	0	0	0	0	0	**0**
COPD	0	0	0	0	0	**0**
HF	0	0	0	1	0	**1**
Judgement						
ESRD	0	0	0	0	0	**0**
COPD	0	0	0	0	0	**0**
HF	0	0	0	1	0	**1**
Response						
ESRD	0	0	0	0	0	**0**
COPD	1	0	1	0	0	**2**
HF	0	1	1	0	0	**2**
Total	**1**	**1**	**2**	**5**	**0**	**9**

Table 12 indicates comprehension, retrieval, response and judgement errors were made by participants while completing the ICECAP-A. Response and comprehension errors were the most common error types. Most errors were made while answering the question about achievement and participants struggled to understand the item and questioned its relevance to their day to day lives.

> *"Progressing in what, I don't know what I'm supposed to progress with, I don't know"* [PT-28R]
> *"I really don't know what to make of it to be honest. I've no idea… I'm trying so hard. I don't know what to say"* [PT-17R]

When probed further during the interview, age was often cited as the reason they felt achievement was irrelevant to their wellbeing.

> *"I'm 93… I'm not going to progress. Some really don't apply (to) what I can do anymore. There's nothing left for me to achieve 'cause of my age"* [PT-20H]

> *"if you want to achieve something it's like you being young and learning and going from here and doing your job… at 75 what can I want to achieve? It's such a hard word that is, it's… I don't know why but it is to me"* [PT-56R]

Thus, difficulties faced by participants completing the ICECAP-A seem to be related at least as much to the generally older sample, with a median age of 80 years, as to issues associated with end of life.

5.7 What difficulties did participants face while completing the EQ-5D-5L?

This section examines the type of errors made by participants across each attribute of the EQ-5D-5L. Qualitative data is used to explore the reasons for the errors made during completion. Forty-five of 60 participants completed the EQ-5D-5L without any errors. Among these, 6 participants did struggle with some questions (Table 9). Participants with end stage HF and end stage COPD had much higher error rates and the number of participants with errors, wad also higher compared to participants with ESRD which had very low error rates.

Table 13 Nature of errors across attributes of the EQ-5D-5L among participant groups

Errors	Mobility	Self-care	Usual Activities	Pain	Anxiety/Depression	Total
Comprehension						
ESRD	0	0	0	0	0	**0**
COPD	0	0	0	0	0	**0**
HF	0	0	0	0	0	**0**
Retrieval						
ESRD	0	0	0	0	0	**0**
COPD	0	0	0	0	0	**0**
HF	0	0	0	0	0	**0**
Judgement						
ESRD	0	0	0	0	0	**0**
COPD	0	0	0	0	0	**0**
HF	0	0	0	1	0	**1**
Response						

ESRD	1	0	0	0	1	**2**
COPD	2	0	1	2	1	**6**
HF	3	1	4	0	1	**9**
Total	**6**	**1**	**5**	**3**	**3**	**18**

Unique across all three measures, participants completing the EQ-5D-5L had no comprehension errors indication that all attributes were clearly understood. Response errors were the most common error type across all attributes on EQ-5D-5L, particularly across questions on usual activities and mobility as can be seen in Table 13.

Most response errors were due to participants being unable to select an option from available response options. For many participants, day to day fluctuations and uncertainty about their condition appeared to be reasons for their inability to select a response option.

> "I *can walk a bit but I don't know now. I don't know, to be honest with you, if I could do it"* [PT-34H]

> *"…if my legs are bad and my breathing, it affects my walking and everything. My feet swell up and I can't get around. That's when I come under these two categories. So, you're asking me to tick any of them which is OK now but it always happens so what? I'm not ticking anyone because none of them are in my category"* [PT-59H]

> *"You can't say that though, can you? It's day-to-day, isn't it? Tomorrow, I'll probably wake up with a stupid headache"* [PT-69R]

Some response errors were due to apparent differences between verbalised experience about day to day living and the actual response option chosen.

> *"I don't have to do any of that now because my daughter does it so I don't know what I should put there. I suppose I put slight ones"* [PT-62R selected 'I have slight problems doing my usual activities']

5.8 Understanding the areas where errors were common

This section outlines the dimensions in which errors were most commonly made, and uses the qualitative data to illustrate the nature of the errors and to explore the underlying reasons for the errors.

Preparation, Emotional suffering, Mobility, Achievement, and Usual activities had the most errors across the items in the measures. Preparation had the most errors across all items with

9 of 60 participants making an error while completing the ICECAP-SCM. Emotional suffering and Mobility in the ICECAP-SCM and EQ-5D-5L respectively had the second highest number of errors with 6 of 60 participants making an error while completing the measures. Achievement and Usual activities in the ICECAP-A and EQ-5D-5L had the third highest error rates with 5 of 60 participants across the groups making an error while completing the measures. Other items in the measures had three or fewer participants with an error.

Table 14 Individual items with the highest number of errors across all participants

Errors	Preparation	Emotional suffering	Mobility	Usual activities	Achievement
Comprehension	6	2	0	0	3
Retrieval	0	0	0	0	1
Judgement	0	0	0	0	1
Response	3	4	6	5	0

Most errors for the Preparation item were comprehension errors and were due to participants conflating end of life decision making with activities that appeared to be related to day to day living.

> *"I look after myself and make up my own mind and I listen to other people and if they're better than mine then I'll take their advice, it's all according to what is being asked… Well some things I could say no to and yes if you know what I mean, the jobs that I wouldn't do I don't want to do I can do, I can do the jobs they're asking but I don't want to that's all so I'll say yes or no" [PT-42H]*

> *"I can make my own meal. I've got some frozen food in the freezer. I can do those. My daughter makes some food and keeps them in a box and I can do them"* [PT-54R]

Some response errors in preparation appeared to concern a distinct reluctance by participants to reflect on their end of life decision making and select an appropriate response option.

> *"I have my financial affairs in order…I don't know, I don't know about that question"* [PT-33R]

Most errors in Emotional suffering were response errors and some of these errors seemed to result from participants discounting the relevance of emotional suffering to their wellbeing and a reluctance to reflect on the impact of their illness on their emotional wellbeing.

> *"I don't have emotional suffering at all. I just carry on with daily life as I go. If I can't do anything I don't do it. I'm on my own all day long. I know my limits with what I can do and what I can't do. So I don't suffer with any of these"* [PT-11K]

> *"no, no, no, I don't think so … I don't know about that one"* [PT-33R]

Comprehension errors in Emotional suffering were due to participants conflating emotional suffering with symptoms that appeared related to physical distress

> "I don't know what you mean [that's fine] I mean I suffer most proper pain in my head or what, I don't know what you mean [that's fine] I don't understand that question" [PT-23R]

All errors that participants made while completing Mobility in the EQ-5D-5L were response errors and these were apparently due to uncertainty about their ability to walk about, which resulted in them selecting multiple response options or leaving the question blank due to their difficulty in quantifying their symptoms.

> *I can walk a bit but I don't know now. I don't know, to be honest with you, if I could do it*. [PT-27H-selected multiple response options]

> *"You see that's the problem if my legs are bad and my breathing it affects my breathing my walking and everything. My feet swell up and I can't get around. That's when I come under these two categories. So you're asking me to tick any of them which is OK now but it always happens so what do I? I'm not ticking anyone because none of them are in my category"* [PT-59H]

Errors made by participants while completing Usual activities were also response errors and again sometimes resulted from participants selecting multiple response options due to their terminal condition.

> *"Some days if I'm not very well I'll have to wait. I won't do it, I just can't do it. And other time when I fell alright I do it… I'm just looking at these. I could tick them all"* [PT-59H]

Some participants selected a full functioning in Usual activities despite stating that they were unable to do perform day to day activities due to their condition.

"I don't do anything. I can't do anything. I can't play with the kids. I can't take part in any activity" [PT-42H selected full functioning in Usual activities]

"I don't have to do any of that now because my daughter does it so I don't know what I should put there. I suppose I put slight ones."[PT-62R]

Most errors made by participants while completing Achievement in the ICECAP-A were comprehension errors. However participants also made retrieval and judgement errors. Comprehension errors were apparently due to an inability to understand the relevance of achievement to their wellbeing. In instances where participants appeared to understand the question, they were unsure of how to answer.

> *"I don't know what that means really. 'Progress in many aspects of my life'. What would that be? Achievements? I don't understand that"* [PT-48R]

> *"I cannot achieve and progress in any aspects of my life… No Many, many, yes my life really works on that sort of thing, without it I have got no life. Because there are some that you can never really… there are a few that you find don't seem… I don't know how to answer that one"* [PT-42H]

5.9 Are the ICECAP-SCM, ICECAP-A and EQ-5D-5L acceptable to patients with organ failure?

5.9.1 Clarity of the measures

Most participants felt the instructions and items in the measures were clear and easy to understand.

> "*It was alright, it was clear to me, I mean, what might be clear to me, might not be clear to anybody else…they were simple questions, they were straightforward to me, as I say, but everybody's different…"* [PT-38K]

> *"…it was quite straightforward I thought in my case"* [PT-43H]

> *"…they're self-explanatory. You know what you're doing"* [PT-14R]

While some admittedly struggled to complete the measures, they suggested that the items in the measures were clear and easy to understand

> "*If I was a bit younger I'd say very clear but they're a bit – I don't – my mind doesn't work so well. I've just done it as best as I think…They're all pretty straight forward"* [PT-11K]

> *"Sometimes I have really wondered whether I should answer that one or the other one and then I make up me mind and just tick… They're all clear, aren't they?"* [PT-10K]

Attributes in the measures that described the severity or frequency of more than one problem seemed to be difficult to answer. Some participants suggested that while one symptom may be present, the other can be absent thus making it difficult to reach an appropriate response.

> *"I did, I think they needed to be worded differently… like with that one there, anxiety and depression I think it should have like, am I slightly anxious, yes but do I feel a bit depressed? I know anxiety can lead to depression but a lot of people suffer with anxiety for years and years but they don't become depressed like myself, I've had you know, I've been anxious over time but I'm not depressed"* [PT-24H]

> *"…it's wrong to tick, no pain or discomfort because as you can see I'm not comfortable, even talking, I'm not comfortable, so I have discomfort but I don't have any pain. What's pain is when they're taking blood out of your arm, the bottom of your arm, like that, every five minutes [laugh]. And that's very painful"* [PT-67R]

The relevance of some items in the measures was questioned by some participants. For instance, some participants suggested that the 'achievement' attribute of the ICECAP-A was underspecified and irrelevant to their current situation. They often struggled to identity capabilities associated with 'achievement and progress' in relation to how they may achieve and progress with a terminal illness from which they may die.

> *"I can achieve and progress in all aspects of my life'. I don't know what that means really. 'Progress in many aspects of my life'. What would that be? Achievements? I don't understand that"* [PT-48K]

> *"I don't know I don't really understand the question really, achievements and progress. I don't think I can progress any, I couldn't get anything extra that I do now anyway. I don't think I could so I don't know about that question so I don't know what to put for that so do you mind if I leave that one. Progressing in what, I don't know what I'm supposed to progress with, I don't know"* [PT-28R]

Some participants struggled to identify capabilities associated with the 'achievement'. They suggested their achievements were limited due to their advanced age and terminal condition.

> *"I think it's too late for me for that now. I've run the course as you might say. I'm on the last lap"* [PT-29R]

Although some participants struggled to identify capabilities associated with achievement, many were able to identify specific issues that limited their capabilities associated with achievement. Participants identified their lack of education and health as a barrier to achievement.

> *"That's a difficult one for me because my education was not very great because I was a child during the war time and education was very bad at that particular time (I can achieve and progress) in a few aspects of my life. I think it's a lack of education."* [PT-30R]

> *I'll be happy as anything once I get out of here. Basically, that's achieving something. To have your life back"* [PT-68H]

> *"I'm hoping to achieve a lot more than I am at the moment. I will be able to restart my voluntary work. I will be able to drive after six months. I will be able to go shopping on my own and start doing my housework properly on my own because even hoovering is tiring. Ironing is tiring. I'm hoping to get all areas of my life back on track"* [PT-44H]

For one participant, there was a distinction between achievements related to their terminal condition and achievements related to general wellbeing. He viewed achievement and progress as an improvement in his terminal condition and capabilities.

> *"Achievement and progress is actually making my body move better. I'm motivated in that. At the moment that's my achievements. Achievements outside hospital are different. But at the moment my entire focus is on getting better and getting mobile"* [PT-52H]

Thus, it appears that while capabilities associated with achievement and progress were difficult to identify for some participants due to their age and terminal condition, some were able to articulate a distinction between achievements related to their illness and achievements relating to other aspects of their lives.

5.9.2 Burden of completion

Although some participants identified issues with the questionnaires, most felt the questions were not upsetting to them. Most participants also found the questions easy to answer.

> *"I didn't find them difficult. No. Quite simple. Sensible"* [PT-27H]

> *"They're all easy, all the same pretty easy really"* [PT-32H]

> *"…they were simple questions, they were straightforward to me, as I say, but everybody's different…"* [PT-38K]

> *"Very easy. A child could do that. There is nothing there is there? Nothing there"* [PT-13R]

For some participants, completing the questionnaires made them think more about functioning and capabilities associated their wellbeing.

> *"Well you do have to think. I mean if you want to do it properly you've got to think about what you do and what you might do and what you can't do and that's mainly the basis of it."* [PT-29K]

> *"…you've got to think. You can't do it straight off your head because you don't get the right answer"* [PT-35H]

Reflecting on capabilities and functioning associated with wellbeing appeared to be difficult for some participants. Some participants felt that the questions made them reflect of sensitive aspects of their wellbeing they do not normally think about.

> *"A few touched a raw nerve but I wouldn't say upsetting, I would say thought provoking more than upsetting, they don't upset me, they make you think about life…One or two questions come up, they make you think twice, they were a bit more difficult but on the whole they were well thought out and well put together questions, they were quite easy to answer"* [PT-61R]

> *"I found it difficult when you made your questions. It's making me think about things that I don't normally think about"* [PT-30K]

A few participants found the think-aloud exercise difficult. One of these participants felt the think-aloud interviewing technique made completing the questionnaires more difficult

> *"…to be able to discuss it with you and make a decision at that time is probably more difficult than having to sit and think it over"* [PT-66K]

Another participant felt that although the questions were clear, the think-aloud exercise itself was difficult due to breathlessness as a result of his terminal condition.

> "*I: How did you find answering the questions overall?*
> *IV: Very hard, [very hard, why did you find it hard?] because of my breathing, it's a constant fight. The questions are plain enough to see…"* [PT-22R]"

In some cases, participants identified attributes of the measures they found upsetting and difficult to answer. Many of these participants found questions relating to anxiety and emotional wellbeing upsetting.

> *"…the one about a burden, that's upsetting… but other than that it's alright, the questions were alright …"* [PT-23R]
> *"I found it easy. Just the bit about being anxious and things like that I felt a bit sad with them."* [PT-47K]

Although their terminal condition mainly affected their ability to perform physical activities, it appears that some participants found reflecting on the impact of their illness on their emotional wellbeing upsetting.

5.10 Summary

This chapter reports evidence on the feasibility of using the ICECAP-SCM, ICECAP-A and EQ-5D-5L in patients with end stage organ failure. Participants differed in their ability to take part in the research and some of these differences were due to reasons that appeared unrelated to their terminal condition such as logistical issues specific to the recruitment site (for example transportation arrangements for patients requiring an ambulance to get home). For some participants however, issues directly related to their terminal condition such as breathlessness were identified as reasons for non-participation. Participants with end stage heart failure were more likely to participate in the research compared to the other groups and most of these patients were recruited from the wards. Participants across the groups were generally in older age, with a median age of 80 years.

Error rates were low across all measures, suggesting that participants were able to complete the measures with few mistakes. Among the few errors made by participants, comprehension and response errors were the most common error types. Both comprehension and response errors were the most common error types in the ICECAP-SCM, and were mainly found across the two attributes of 'preparation' and emotional suffering'.

Although participants had the lowest error rates in the ICECAP-A, a few struggled with identifying capabilities associated with achievement and progress apparently due to their age and terminal illness. While participants completing the EQ-5D-5L had no comprehension errors, most of the errors made were response errors which were partly due to difficulties quantifying their physical symptoms.

Most participants felt that the instructions and questions in the measures were clear, easy to understand and were not burdensome. However, some participants found reflecting on their terminal illness difficult and upsetting, highlighting the broader effects of their illness on emotional wellbeing which warrants further investigation. Furthermore, some participants, particularly those with end stage COPD, found completing the think-aloud task difficult due

to breathlessness also illustrating the likely effects of terminal illness on physical wellbeing and capabilities associated with day to day living.

The effects of end stage organ failure on capabilities associated with both physical and emotional wellbeing as captured by the measures, underscores the need for further investigation on the impact of these end of life conditions on the wellbeing of participants across the groups. The next chapter uses data from both the think-aloud and semi-structured aspects of the interview to explore the impact of functional decline on the health of participants across the groups, and capabilities associated with both physical and emotional wellbeing.

Chapter 6

The impact of functional decline on capability wellbeing

The chapter reports responses to each attribute of the ICECAP-SCM, ICECAP-A and EQ-5D-5L and explores differences in the health and capability profiles of participants across each group. The chapter also reports the mean utility and capability scores, and the range of values across the groups. Responses for each patient were converted to QALY and capability scores using value sets derived from a survey of the general population preference-based elicitation methods (Devlin et al., 2016, Huynh et al., 2017). Summary health and capability profiles are presented in Table 15, Table 16 and Table 17.

To further explore the differences in health and capability profiles across the groups, qualitative data is used to explore the impact of functional decline and loss of capabilities on wellbeing at the end of life and the opportunity for a good death. Think-aloud and semi-structured aspects of the interview transcripts were analytically explored and themes developed to describe emerging patterns in the data. The relationship between themes were further developed as more information emerged.

End of life policies emphasise the importance of aspects of wellbeing that go beyond health, and need for measures that capture the impact of end of life care on health and wellbeing at the end of life. This chapter aims to understand the impact of end stage organ failure on capability wellbeing and capabilities associated with the opportunity for a good death. The chapter examines the impact of terminal illness and associated multi-morbidities on capabilities associated with day to day living. The reasons for loss of capabilities, and the impact of loss of capabilities on physical wellbeing, emotional wellbeing and social relationships is explored across the groups. The impact of terminal illness on the perceived rate of functional decline across the groups and its effect on terminal awareness and end of life decision making is considered.

Themes identified from the qualitative data are illustrated using quotes from participants represented by their study numbers in the same way as in the earlier chapter (for instance, PT-*n*R illustrates a participant with study number *n* with a diagnosis of end stage COPD).

6.1 Responses to the measures across the groups

This section explores responses to each attribute of the ICECAP-SCM, ICECAP-A and EQ-5D-5L across participant groups. By exploring health and capability profiles across the groups, this section aims to provide a context through which the impact of end stage organ failure on health and capability wellbeing can be understood.

6.1.1 Responses to the EQ-5D-5L questionnaire

Participants with end stage COPD generally had a worse health profile on the EQ-5D-5L compared to those with end stage heart failure and ESRD. This was particularly more severe in relation to 'mobility', 'self-care' and 'usual activities' as can be seen in Table 15. Compared to those in the end stage heart failure group, participants with end stage COPD and ESRD were more likely to report severe or complete loss of functionings in physical activities (mobility, self-care and usual activities).

The worse health profile observed among participants with end stage COPD is also reflected on their utility scores which showed a mean score of 0.37 (range: -0.09 – 0.95). Negative values are possible on the utility scale because some health states are valued as being 'worse' than dead. In contrast, participants with ESRD had an average utility score of 0.58 (range: -0.02 to 0.94) while participants with end stage heart failure had an average score of 0.60 (range: 0.19 to 1). Participants across the groups had an average utility score of 0.52 on the EQ-5D-5L. These values should be treated with caution as the sample size was not statistically powered to enable comparison between the groups. However, they provide a context through which the impact of their end of life condition on health-related quality of life can be understood.

The impact of the worse health profile observed among participants with end stage COPD on capability wellbeing at the end of life is further explored and compared with other participant groups in section 6.2.

Table 15 Response to the EQ-5D-5L questionnaire across the groups

Attributes	Freq (%)* Renal (n=18)	Freq (%)* COPD (n=21)	Freq (%)* Heart Failure (n=21)
Mobility			
• I have no problems in walking about	1 (6%)	1 (5%)	2 (10%)
• I have slight problems in walking about	4 (22%)	4 (19%)	8 (38%)
• I have moderate problems in walking about	6 (33%)	2 (10%)	3 (14%)
• I have severe problems in walking about	3 (17%)	6 (29%)	5 (24%)
• I am unable to walk about	4 (22%)	6 (29%)	1 (5%)
Self-care			
• I have no problems washing or dressing myself	9 (50%)	2 (10%)	11 (52%)
• I have slight problems washing or dressing myself	2 (11%)	6 (29%)	4 (19%)
• I have moderate problems washing or dressing myself	6 (33%)	7 (33%)	4 (19%)
• I have severe problems washing or dressing myself	0	2 (10%)	1 (5%)
• I am unable to wash or dress myself	1 (6%)	4 (19%)	0
Usual activities			
• I have no problems doing my usual activities	3 (17%)	1 (5%)	4 (19%)
• I have slight problems doing my usual activities	5 (28%)	5 (24%)	5 (24%)
• I have moderate problems doing my usual activities	3 (17%)	3 (14%)	6 (29%)
• I have severe problems doing my usual activities	2 (11%)	6 (29%)	3 (14%)
• I am unable to do my usual activities	5 (28%)	6 (29%)	2 (10%)
Pain / discomfort			
• I have no pain or discomfort	1 (6%)	4 (19%)	6 (29%)
• I have slight pain or discomfort	4 (22%)	3 (14%)	5 (24%)
• I have moderate pain or discomfort	10 (56%)	6 (29%)	5 (24%)
• I have severe pain or discomfort	3 (17%)	5 (24%)	5 (24%)

• I have extreme pain or discomfort	0	1 (5%)	0
Anxiety / Depression			
• I am not anxious or depressed	11 (61%)	5 (24%)	8 (38%)
• I am slightly anxious or depressed	3 (17%)	4 (19%)	8 (38%)
• I am moderately anxious or depressed	3 (17%)	7 (33%)	1 (5%)
• I am severely anxious or depressed	0	2 (10%)	1 (5%)
• I am extremely anxious or depressed	1 (6%)	2 (10%)	2 (10%)

*Percentage frequency values may not add up to 100% due to missing data as a result of non-response

6.1.2 Responses to the ICECAP-A questionnaire

Responses to the ICECAP-A questionnaire slightly differed across the groups. This was reflected in the ICEAP-A scores which showed participants with end stage COPD with a mean score of 0.71 (range: 0.39 – 1). Participants with end stage heart failure and ESRD had mean capability scores of 0.85 (range: 0.33 – 1) and 0.76 (range: 0.27 – 0.97) respectively. The mean capability score across the groups was 0.78. The higher capability profile and scores compared to the EQ-5D-5L scores may relate to the different evaluative space the questionnaires capture (the ICECAP-A evaluates generic capability wellbeing while the EQ-5D-5L evaluates health-related quality of life). Participants with end stage heart failure were more likely to report full capabilities across all attributes of the measures. Participants with end stage COPD were more likely to report a complete loss of capabilities, particularly across 'autonomy' and 'achievement' attributes. Again, the apparently more severe loss of capabilities reported by participants with end stage COPD group may reflect the severe impact of their symptoms on capabilities associated with day to day living.

Table 16 Responses to the ICECAP-A questionnaire across the groups

Attributes	Freq (%)* Renal	Freq (%)* COPD	Freq (%)* Heart Failure
Stability			
• I am able to feel settled and secure in all areas of my life	9 (50%)	8 (38%)	12 (57%)
• I am able to feel settled and secure in many areas of my life	6 (33%)	6 (29%)	5 (24%)

• I am able to feel settled and secure in a few areas of my life	1 (6%)	4 (19%)	2 (10%)
• I am unable to feel settled and secure in any areas of my life	2 (11%)	3 (14%)	2 (10%)
Attachment			
• I can have a lot of love, friendship and support	11 (61%)	18 (86%)	17 (81%)
• I can have quite a lot of love, friendship and support	1 (6%)	2 (10%)	1 (5%)
• I can have a little love, friendship and support	3 (17%)	1 (5%)	0
• I cannot have any love, friendship and support	2 (11%)	0	1 (5%)
Autonomy			
• I am able to be completely independent	2 (11%)	2 (10%)	10 (48%)
• I am able to be independent in many things	12 (67%)	8 (38%)	8 (38%)
• I am able to be independent in a few things	14 (78%)	8 (38%)	3 (14%)
• I am unable to be at all independent	0	3 (14%)	0
Achievement			
• I can achieve and progress in all aspects of my life	3 (17%)	5 (24%)	4 (19%)
• I can achieve and progress in many aspects of my life	7 (39%)	7 (33%)	13 (72%)
• I can achieve and progress in a few aspects of my life	8 (44%)	4 (19%)	2 (10%)
• I cannot achieve and progress in any aspects of my life	0	4 (19%)	1 (5%)
Enjoyment			
• I can have a lot of enjoyment and pleasure	8 (44%)	6 (29%)	13 (62%)
• I can have quite a lot of enjoyment and pleasure	2 (11%)	6 (29%)	5 (24%)
• I can have a little enjoyment and pleasure	7 (39%)	7 (33%)	2 (10%)
• I cannot have any enjoyment and pleasure	1 (11%)	2 (10%)	1 (5%)

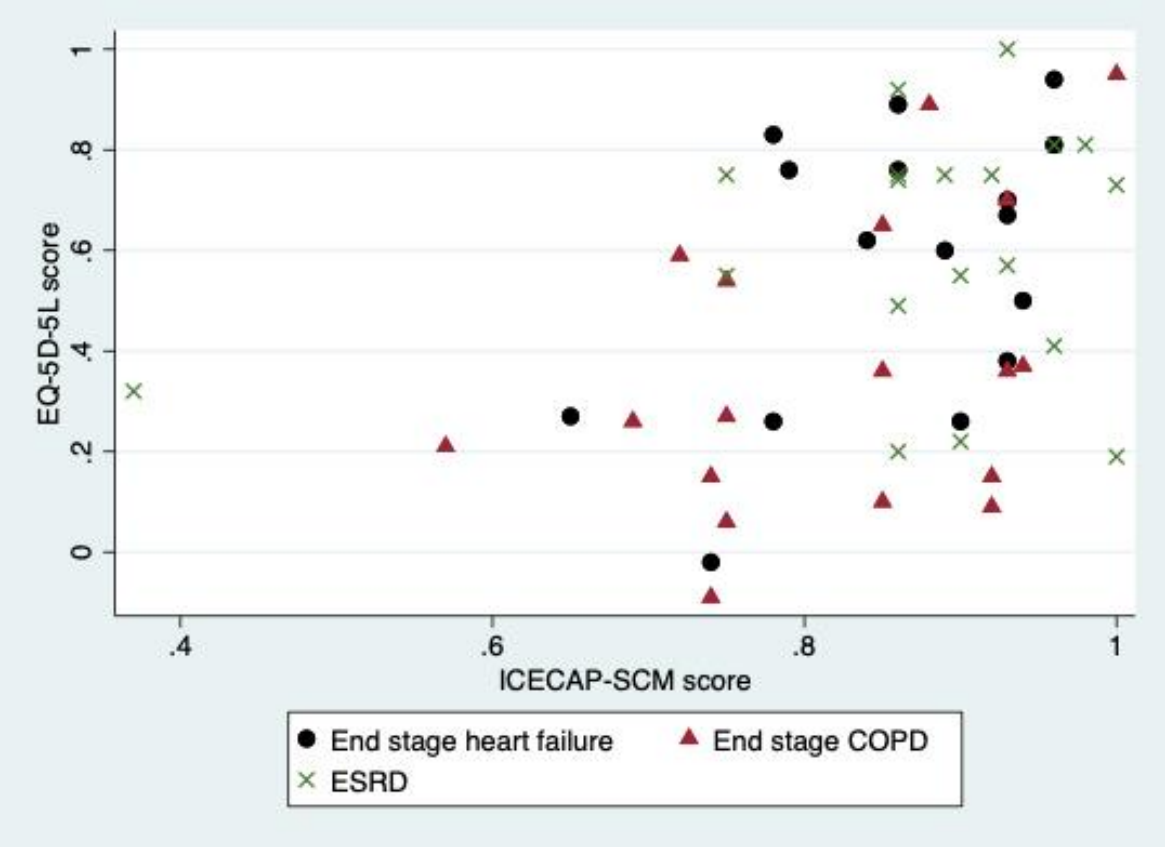
*Percentage frequency values may not add up to 100% due to missing data as a result of non-response

6.1.3 Response to the ICECAP-SCM questionnaire

Although participants with end stage COPD had a slightly worse health profile on the ICECAP-SCM compared to other groups, responses were generally similar. This was reflected in the capability scores which showed participants with end stage COPD with a mean score of 0.82 (range: 0.57 – 1) while participants with end stage heart failure and ESRD both had a mean score of 0.86 respectively. However, there were differences in the range of values. Participants with end stage heart failure had a capability score between 0.37 and 1 while participants with ESRD had a range of 0.65 – 1. Participants across the groups had a mean capability score of 0.84 on the ICECAP-SCM. Participants with end stage

COPD were more likely to have both lower capability score on the ICECAP-SCM and utility score on the EQ-5D-5L as can be seen in Figure 9 below.

Figure 9 EQ-5D-5L scores vs ICECAP-SCM scores across the groups



Participants with lower utility and capability scores were more likely to have end stage heart failure and end stage COPD compared to participants with ESRD.

Although the scores were generally similar, participants with end stage heart failure and end stage COPD were more likely to report severe loss of capabilities, particularly in relation to freedom from physical distress, emotional suffering and end of life decision making compared to the ESRD group as seen in **Table 17** below. This may reflect the impact of their distinct illness trajectory, characterised by periodic exacerbations and fluctuation of symptoms, and the acute setting of care where these symptoms are less likely to be controlled.

Using qualitative data, the next section explores the impact of their terminal diagnosis on health and capability wellbeing across the groups and how participants had coped and adapted to their terminal condition.

Table 17 Response to the ICECAP-SCM questionnaire across the groups

Attributes	Freq (%)* Renal	Freq (%)* COPD	Freq (%)* Heart Failure
Choice			
• I am able to make decisions that I need to make about my life and care most of the time	16 (89%)	15 (68%)	15 (68%)
• I am able make decisions that I need to make about my life and care some of the time	2 (11%)	2 (10%)	2 (10%)
• I am able make decisions that I need to make about my life and care only a little of the time	0	4 (19%)	0
• I am never able to make decisions that I need to make about my life and care	0	0	1 (5%)
Love and affection			
• If I want to, I am able to be with people who care about me most of the time	15 (83%)	19 (90%)	19 (90%)
• If I want to, I am able to be with people who care about me some of the time	2 (11%)	2 (10%)	1 (5%)
• If I want to, I am able to be with people who care about me only a little of the time	1 (6%)	0	1 (5%)
• If I want to, I am never able to be with people who care about me	0	0	0
Physical suffering			
• I always experience significant physical discomfort	7 (39%)	6 (29%)	5 (24%)
• I often experience significant physical discomfort	3 (17%)	4 (19%)	5 (24%)
• I sometimes experience significant physical discomfort	4 (22%)	8 (38%)	6 (29%)
• I rarely experience significant physical discomfort	2 (11%)	3 (14%)	5 (24%)
Emotional suffering			
• I always experience emotional suffering	1(6%)	4 (19%)	4 (19%)
• I often experience emotional suffering	3 (17%)	5 (24%)	3 (17%)
• I sometimes experience emotional suffering	3 (17%)	6 (29%)	6 (29%)
• I rarely experience emotional suffering	9 (50%)	5 (24%)	9 (43%)
Dignity			
• I am able to maintain my dignity and self-respect most of the time	14 (78%)	16 (76%)	18 (86%)

• I am able to maintain my dignity and self-respect some of the time	3 (17%)	4 (19%)	3 (14%)
• I am able to maintain my dignity and self-respect only a little of the time	1 (6%)	1 (5%)	0
• I am never able to maintain my dignity and self-respect	0	0	0
Being supported			
• I am able to have the help and support that I need most of the time	12 (67%)	16 (76%)	15 (71%)
• I am able to have the help and support that I need some of the time	4 (22%)	2 (10%)	2 (10%)
• I am able to have the help and support that I need only a little of the time	0	3 (14%)	4 (19%)
• I am never able to have the help and support that I need	1 (6%)	0	0
Preparation			
• I have had the opportunity to make most of the preparations I want to make	12 (67%)	10 (48%)	11 (52%)
• I have had the opportunity to make some of the preparations I want to make	4 (22%)	4 (19%)	6 (29%)
• I have had the opportunity to make a few of the preparations I want to make	1 (6%)	2 (10%)	0
• I have not had the opportunity to make any of the preparations I want to make	1 (6%)	4 (19%)	3 (14%)

*Percentage frequency values may not add up to 100% due to missing data as a result of non-response

6.2 The impact of end-stage organ failure and multi-morbidities on health and capability wellbeing

Although all participants were recruited based on a set of prognostic criteria related to the terminal phase associated with organ failure of one of three types which they managed on a day to day basis, it became apparent at an early stage of the analysis that many participants were also managing multiple other long-term health conditions which also affected their wellbeing. Many participants described multiple symptoms and impairments associated with their terminal condition which they managed on a day to day basis.

> *"I've got other things beside this kidney trouble. Like being diabetic on four injections a day, you've got to keep them until different times.... I had an accident a couple of years ago and I broke quite a lot of bones in my body and it had left one leg shorter than the other and my wrists are not very strong… If I do anything I seem to get out of breath and sometimes in my back I have pain"* [PT-47K]

> *"I suffer with rheumatoid arthritis as well as my COPD I get pain in my chest and that so I have to lie down and rest… it's something you can't get away from. As soon as you wake up in the morning you are in discomfort"* [PT-60R]

> *"I had a stroke four years ago on my left side and over the last few years, I've lost my mobility. I've got no grip in these fingers on the left side and my left leg will suddenly give way"* [PT-30K]

The extent to which these conditions were associated with their terminal condition was unclear. However, the type and number of long-term conditions managed, and the extent to which this affected wellbeing differed among the groups. Participants with ESRD were generally more likely to report managing other long-term conditions which were different to their terminal diagnosis compared to the other groups. This may be related to the fact that participants belonging to ESRD group were generally older than other groups. Most of these participants, rather than speaking about their terminal condition, often spoke about falls and accidents while engaging in day to day activities.

> *"I felt worse the past few months than I had done for a while…I was alright until I had that accident, I fell in the steps and I was in about six weeks"* [PT-47K]

Similarly, while symptoms that appear related to arthritis were reported by participants in all groups, these were more common among participants with ESRD. Most of these participants were more likely to report increased physical symptoms such as pain that appear related to arthritis which further worsened their disability.

> "*I got pain like in different parts of my body and my chest and my back…, I get it in my back and my lumbar spine and in my legs… when my back goes, I'm just stuck, I just can't move, sometimes I can't lift my arms… I can't straighten up*". [PT-38K]

Participants with ESRD also discussed their vulnerability to accidents and the effects of such accidents on their capacity to engage in simple leisure activities, limited their independence and affected their overall wellbeing.

> *"…I hit my head on the floor, I lost some memory, I was having some funny noise on my left-hand side…Sometimes when the telly on, and the news I can't understand what they are saying. Sometimes a person talking to me I can't understand what she's saying. I was hard of hearing before I fell but after that I have a bit more deafness coming in my ears"* [PT-18K]

> *"I had a fall the other week and I couldn't get up… they kept me in hospital for a while and then they gave me rehabilitation to help me with my leg, walking. I couldn't walk without assistance."* [PT-66K]

While most participants with ESRD reported managing long term conditions that appeared unrelated to their terminal diagnosis, participants in the other groups often identified symptoms related to their terminal diagnosis such as breathlessness and oedema as major issues in day to day living. These issues had apparently significant effects on their capabilities and ability to perform basic day to day activities.

6.2.1 Loss of capabilities

Participants often discussed the difficulties they faced while attempting to do basic things they were used to doing such as walking or gardening. In many cases, there appeared to be an apparent decline in function and a progressive loss of independence as they lived with their terminal condition. This may have been due to the increasing burden of their terminal illness and other long-term conditions being managed. Participants in all groups reported on their loss of capabilities and how it affected different aspects of their lives.

> *"I can no longer work in the garden… I can't walk in the park. I'm very lucky I happen to live in [place removed] with the park backing onto [place removed], so, we used to do a lot*

of walking and climbing, although I can't do these things any more. Can't fly anymore. I'm not allowed to fly" [PT-36H]

"I'd like to be a lot more independent but I can't be, I'd like to be a lot more in control of myself but I can't be, I have to go through other people, which I don't like, I mean even if I want money out of the bank I have to ask. I like to do my own thing but I can't any more so it's just I can't do what I want to do" [PT-23R]

I'm not as quick as I used to be…there are things that I can't do now that I could do before. I miss my walking, when I could walk, I walked about quite a lot, you know, which I don't do now" [PT-38K]

These three participants identified how their loss of mobility and independence had a limiting effect on their everyday lives.

Reasons for loss of capabilities

Symptoms related to their terminal diagnosis and other long-term conditions were the main reasons for loss of capabilities among participants. Although participants with ESRD were less likely to report severe loss of capabilities in day to day living compared to the other groups (**Table 16**), accidents and falls were often identified as the main reasons for loss of capabilities.

"I had a fall the other week and I couldn't get up…they kept me in hospital for a while and then they gave me rehabilitation to help me with my leg, walking" [PT-66K]

On the other hand, participants with end stage HF and end stage COPD were more likely to report severe loss of capabilities in day to day living. Symptoms related to their terminal illness such as breathlessness and oedema were often identified as major reasons for loss of capabilities. Participants with end stage COPD regularly identified breathlessness as the main reason for their loss of capabilities.

"I have trouble breathing and I can't walk where I want to walk because my breathing won't allow me" [PT-69R]

"I'm out of breath just and it's getting worse and worse, any small thing I do, it knocks me out of breath and it's getting worse, I can't do anything about it, I'm just sitting like a cabbage" [PT-31H]

While breathlessness was also reported by a few participants with end stage HF, many identified oedema as a major reason for their loss of capabilities. Participants spoke of how oedema affected their breathing and mobility.

> *"…my legs just start swelling again. All the water gets to the end. That's when you get the problems, you can't breathe, your legs start filling with water. It's all down to the water. My feet swell up and I can't get around..."* [PT-59H]

> *"Because of the water on my ankles and that, I think that makes my feet go like this, the waters come out in me body uneven. If you look at me ankles, they've come out uneven. I walk down and it feels like somebody's just kicked my legs from underneath me [laugh]. It happened to me the other day…*." [PT-37H]

Although the specific causes of the loss in capabilities varied across the groups, there appeared to be similar and considerable impact on physical wellbeing, emotional wellbeing and social relationships. The impact of loss of capabilities on these aspects of wellbeing is considered below.

The impact of loss of capabilities on physical wellbeing

Many participants reported how their terminal condition limited their ability to work and perform physical activities such as shopping, gardening and to pursue other interests and leisure activities. Some of these participants expressed regret and sadness over the things they had wanted to accomplish but were unable to due to their illness

> *"What's happened to me, at my age, I think is that I'd got my life planned. I wanted to do this, that and the other. To think, after 79 years, you finish up and you're going to be restricted on what you can do for the rest of your life... it's such a downer." [PT68H]*

> *"You must have some hobbies and interests and whilst you can do them and pursue them life is alright but when things change so that you can't… I played golf for years and years and years and I absolutely loved it but I couldn't play it anymore and that was very sad and I used to go on country rambles because I enjoy the countryside but that had to stop. I can't do the things I want to do …" [PT-45R]*

> *"I had to pack up my job completely because I couldn't do it no more, I can't drive and I can't do none of it no more, I haven't got the energy and people don't realise that, you're on the scrap heap before you notice it and people don't you know, I've had to go to college now at sixty odd years of age and learn something different, it's hard, when you walk into a class you're the oldest one by 30 years, you're older than the teacher… " [PT-61R]*

The impact of loss of capabilities on physical wellbeing appeared to be more severe among participants with end stage COPD who were also more likely to report severe or complete

loss of functionings while completing the EQ-5D-5L due to breathlessness. Many spoke of how breathlessness limited their ability to perform simple tasks and engage in their usual activities.

> *"I won't be able to lift the hoover up the stairs or, what else? Simple things, if I drop a pen, it will take me a while to pick it up due to my breathing, yeah... I couldn't go and run for the bus or I couldn't go to the gym and I used to love going to the gym, before I was sick. I have severe problems with doing simple things"* [PT-33R]

The severe and unpredictable nature of breathlessness appeared to limit their achievements and ability to make long-term plans for their future and career.

> *"it's only a certain amount of things I can do, there's been so much in my life that I would have liked to have achieved like my daughter, my youngest one, when she finished school I thought that's it, I can go back to work and everything now and then I ended up with my COPD so that sort of restricted me from being able to go to work because you have your good days and you have your bad days and the bad days tend to tower over the good days" [PT-60R]*

Participants also discussed their difficulties with personal care due to the limitations of their terminal condition. Participants with end stage HF and end stage COPD were however, more likely to report loss of capabilities associated with personal care than those with ESRD. Participants with end stage heart failure and end stage COPD identified breathlessness and oedema as the main factors that led to loss of capabilities associated with personal care. Participants reflected on their difficulties with walking to the bathroom and engaging in intimate personal care due to breathlessness and oedema.

> *"I need help with undressing and dressing because I can't bend down to get my socks on… getting out of the chair, getting out of bed, going to the toilet you'd be amazed how difficult it is, when you're lying down you won't be able to go to the toilet and these things you take for granted… it might sound ever so petty but when you've done it all your life and you have to get somebody else to do it and you never needed help with it..." [PT-45R]*

> *"I have to have help washing and dressing…Again it's all to do with breathing and I get very short of breath bending and pretty well bending is the worst part. And I'm really short of breath now"* [PT-28R]

> *" I can't bend to put my socks on or my shoes or my trousers. Walking to a bathroom. My bathroom is upstairs. It takes me ages and by the time I get up the stairs, I'll either wet myself or I'll be too knackered to go to the bathroom and I'll have a rest. That's what been like the last 14 months. Sometimes I've been just knackered getting into bed. And if you wanna go to toilet all you gotta do is take a bucket. Because I can't make the toilet. You know when I can't make toilet I've gotta piss on the bed"* [PT-59H]

> *"…if I go to the toilet I sit down for five minutes to get my breath back and then I have to come back another five minutes to get my breath back so going to the toilet is over half an hour. That's why I have these bottles here because it's less work and less work mean I can do it easier but I'm out of breath"* [PT-31H]

For most participants, their difficulties with physical activities such as walking, gardening, and their difficulties with certain aspects of their personal care such as walking to the bathroom, washing and dressing often made them frustrated and angry at their loss of independence. The next section examines the impact of loss of capabilities on emotional wellbeing and social relationships.

The impact of loss of capabilities on emotional wellbeing and social relationships

Symptoms related to their terminal illness and other long-term conditions affected the emotional wellbeing of participants, their relationships with family members, friends and the wider community. While struggling with loss of capabilities due to breathlessness, some participants with COPD suggested that people often failed to empathise with the complexities of their condition and the impact of breathlessness on their ability to perform basic activities.

> *"I can hardly [climb]down the stairs can only get to the top of there and I'll stop and people just look at you and think what's the matter with you because they haven't got a see-able injury and nobody thinks about it, nobody thinks about things like that. With COPD people can't see it, it's a disability that people can't see."* [PT-61R]

> *"Everybody thinks I'm lazy and I'm not, I'm not putting it on. A lot of people think I'm putting it on but I'm not, I want to do things and go and do it and get up and you know, walk around and do things but I'm out of breath all the while or I fall over so well"* [PT-23R]

Many participants reported that their worsening health and reliance on family members to help them cope with loss of capabilities made them feel like a burden to them. This was particularly common among participants with end stage heart failure and end stage COPD who were more likely to report severe loss of capabilities associated with their emotional wellbeing (**Table 17**). Some of these participants felt that their terminal condition limited the freedom and capabilities of their partners and children.

> *I do feel like a burden sometimes. I feel I am a burden to them all. Do you know what I mean? I decided that's why I wouldn't go on, if anything happened to me I'd just go, I wouldn't go any further because they've done enough for me, I don't want them to keep*

doing it all their lives, not them, you know my sons and my husband they've got a life, I am a burden... I'm a nuisance!!" [PT-23R]

"I'm a burden to people, I feel like I'm a burden, I feel like I make, I should be fit and like I used to be but I can't because they're doing things for me… I'm unable to do anything, my daughter's even got to talk on the phone for me, I can't talk so she talks for me …they're all working and they've got their own jobs and they've got their own things to do" [PT-31H]

Others reported feeling like a burden because family members were already managing significant health challenges of their own and should not be further burdened with their disability.

"I'm a burden to everybody at the moment, my mom and everyone, it's not fair on them. My mom's just got over cancer when she's having to come up to the hospital and look after me, really it should be the other way around, it's not fair" [PT-37H]

"I'm a burden on my husband, because he…he can't handle himself, with his heart bypass, so…I just feel a burden, and he'd goes barmy if I say that" [PT-41R]

Indeed, in some cases, a sense of being a burden on family members appeared to make some participants unwilling to seek help from family members and close persons which apparently worsened their condition.

"You're reliant on getting to hospital. You have to rely on different things, the ball is (not) in your court. With the COPD you can't do what you wanted to do, you can't always be there, you keep wanting to go out somewhere and you can't rush round now and get ready, you can't do things. It's horrible sometimes no matter what you do …they've got their own lives to lead, you don't want to drag them down, you don't want to have them to have to come to you all the time… that's why I'm in here because I wouldn't tell anybody how bad I was getting and I have ended up in here because I don't want them to worry" [PT-61R]

"I feel like a burden. I do feel like that if I ask anybody for help. That's why I don't asking any of them for help. When you start asking them you are a burden to them. You feel guilty... you feel... oh its fine you go and answer it its dad, he might want something. That's how you feel" [PT-59H]

In addition to the guilt of being a burden to family members, some participants also reported being angry and frustrated at their loss of capabilities and ability to do things they were used to doing. A sense of defeat at the immutability of their condition and reliance on others to perform basic tasks often led them to express feelings of anger and frustration especially when they were unable to do the things they had become accustomed to doing.

"I've worked all my life. I have been able to do things myself and for everyone. The way I am, I can't do bloody nothing and when you can't do it and you're frustrated. I'm stuck in bed all day with my legs up. Who wants to be stuck in bed all day? I don't! But I don't want

it being increased back again. The way you have it... it sort of really pisses you off because you just cannot do anything. That's when you get frustrated." [PT-59H]

"I panic when I can't breathe and I get really depressed in myself because I get angry with myself because I can't do what I want to do in life and that really gets to me, it really makes me angry because I think well rather than keep relying on people all the time, I should be doing it but I just can't do it with the COPD" [PT-60R]

"I can't do anything, I can't change anything, I'm very depressed I'm hoping I go quick with the wife you see but they keep telling me hang on.. there is a new baby coming in.. so I'm hanging on, hanging on, hanging on and they keep coming to see me.. and half the time I can't concentrate on my mind because it's not the same as it was before. I used to make things, repair things, be active. I can't concentrate on things…" [PT-31H]

"I've always been kind of independent and you know when I've had to rely on people that sort of upsets me and anything to do with that, which I'm sure you must have seen that you know [PT-50H Participant cried while reflecting on the impact of his illness on his family members]

The impact of loss of capabilities on emotional wellbeing went beyond overt emotional reaction to loss of capabilities and a sense of self-perceived burden on family and friends. It also affected the ability of some participants to establish and maintain valued social relationships. This was particularly common among patients with ESRD who were likely to report loss of capabilities around attachment as can be seen in **Table *16***. In some cases, loss of capabilities around attachment made some participants feel isolated and lonely.

I mean sometimes I cry for no reason which is very rare which I think sometimes it's loneliness… I think it's mainly being alone all the time and not seeing anyone… I've always been used to going out and doing loads of things... I haven't been on a bus for three years now, I think if I was to see more people it would probably be better" [PT-47K]

"I do get anxious and depressed at times, especially when I'm on my own. There's a lot of time when I'm on my own I think about things… I know all I've got to do is get on the phone and I'll have somebody. But up to now I've caught it you know and carried on." [PT-15K]

Symptoms related to their terminal illness and other long-term conditions managed daily had considerable impact on the physical and emotional wellbeing of most participants. However, many participants also expressed considerable concern about the rate at which their condition had worsened and the extent to which this affected their capabilities in day to day living.

6.2.2 The rate of functional decline and loss of capabilities

Participants across all groups discussed their concerns and uncertainties regarding their symptoms, the long-term conditions they managed, and the prognosis of their terminal

illness. This was particularly common among participants in the late end of life phase (participants who died within four months of being interviewed) and may be related to an increased illness burden as they approached the end of their lives. Most of these participants discussed their concerns over their deteriorating health and other conditions they managed.

> *"I am slightly anxious because it seems to be a long time clearing up this infection. And I'm still positive that it will clear but it just seems a long time, it is making me a little bit down*" [PT-67R, Female, 71, died 5 days after interview]

> *"I had a cardiac arrest back in February… and I literally went down at a steady [rate]. I was going down every single day of my life and to a stage where my wife said, you look as though you've been poisoned. We've had an absolutely dreadful year" [PT-50H, Male, 70, died three weeks after interview]*

> *"I had to go in to hospital because I had bleeding from my lungs and [had] a bronchoscopy and I've got to go again on the 13th. Because for about 19 years, I was taking warfarin, and the warfarin stopped. So, I'm going through things with that, which does give me a concern. But then again, I've got to wait until the 13th of November to see what they say about it."* [PT-66K, Male, 87, died 3 months after interview]

Some participants however, expressed concern over a lack of clarity over the prognosis of their condition.

> *"I don't have anxieties, no. Fears maybe? Of how long, how long could my lungs stay like this, the way it is, if it makes sense?*" [PT-33R]

> *"I'm not getting the answers I want to know and that's what's making me depressed. They keep giving x-rays but nothing comes about it"* [PT-59H]

Participants with end stage HF and end stage COPD were more likely to express concern over their rate of deterioration in health and loss of capabilities. For some of these participants, their sudden deterioration in health and the rate at which capabilities were lost was unexpected. They discussed their shock at their rapid decline and impact of being suddenly unable to do things they were used to doing.

> *"I love gardening, so I was always in the garden putting plants in, digging plants up and throwing plants away. I didn't feel bad or nothing like that and then I collapsed. Basically, it was such a shock to me. This is why I can't really say that I felt ill. It just sort of came straight on me like that. As far as that, I was active. I didn't know this was coming on to me. It was the most terrible shock when you finish up in hospital. Basically, you don't know what's hit you."* [PT-68H]

> *"I have been ill and its sort of took me off my feet and it's knocked me out… everything has just gone from what it used to be"* [PT-39R]

The apparent difference in the rate of deterioration among participant groups may be related to their respective illness trajectory and pattern of functional decline. While reflecting on their health state over time, there were clear differences regarding the 'stability' of the condition across the groups. Most participants with ESRD reported that their condition had remained stable over time.

> *"Still the same, really. I don't think it's deteriorated much, really"* [PT-11K].

> *"Well, for me, I feel okay. I still feel the same as when I first went"* [PT-48K]

> *"Not much change at all, is there? I'm pretty well the same"* [PT-15K].

In contrast to reports among participants with ESRD, most participants with end stage HF and end stage COPD felt that their condition had rapidly deteriorated over time. Indeed, participants with end stage HF and end stage COPD who were in the late end of life phase felt that their condition had significantly deteriorated and expressed concern over their prognosis and the rate of deterioration of their terminal condition.

> *"I just had one prognosis after another where it's gone very bad indeed. I can't do anything about the lungs or heart"* [PT-45R, Male, 76, died 4 months after interview]

> *"I've had this chest infection. I just got a bit tired and I've been trying to bring myself up but it seems to be only down and down" [PT-67R, Female, 71, died 5 days after interview]*

> *"Terrible, absolutely terrible, I never thought I'd be like this, I never thought people could, it could happen to people like it does. I keep just looking for breath you see, no it's getting worse and worse and worse*" [PT-32H, Male, 76, died 3 weeks after interview]

For many participants with end stage HF and end stage COPD, breathlessness, chest infections and oedema were the main reasons for their deteriorating health. There was a sense that their symptoms and terminal condition had deteriorated over time.

> *"It's getting worse and worse and worse and things like that. For four weeks now I don't go to church, my foot swell up full of water" [PT-21H]*

"My breathing has become very, very difficult. I have real difficulties breathing, that's right, that's why I'm in here. My heart's been getting weaker. I have a leaky valve; I've got a defibrillator" [PT-36H]

"It started off with asthma, then it went to bronchitis, then it went to COPD, and obviously now I've got the worst of the worst" [PT-40R]

Indeed, despite apparent attempts to self-manage their condition, and other interventions by health care professionals, they felt that their condition still deteriorated.

"I have my steroids and everything but they don't work. Two weeks ago I came in. A week before that I came in. You could say at least once a month for six years and I sometimes stop in up to six weeks. I try ever so hard not to come." [PT-23R]

"I came in here and I went right downhill. I don't know what they're doing to me. I said to him this morning, 'Before I came in here you wouldn't have thought anything was wrong with me. Now look at me. What have you done to me? I'm worse now than what I was at home. I feel worse now than when I came in the very first time" [PT-13R]

For many of these participants, there appeared to be a recognition that an apparent lack of improvement in their health may ultimately lead to death. Although some participants struggled with the possibility of death, they also reflected on their preparations for death and other treatment preferences at the end of life.

6.2.3 Terminal awareness and end of life decision making

Many participants demonstrated an awareness that their advanced age, and increasing illness burden could ultimately lead to death. This was often initiated by bereavement or reflection on their own terminal condition.

"…I'm here one day and gone the next. I know I haven't got much. I've just lost my sister. She was 93. I'm coming up to 93. We'll have to see. I haven't got much longer. I'm lucky I suppose to be this old". [PT-11K]

"When you're going to die eventually, and some of us are going to die sooner than eventually, you do reflect on a lot of things. But that's it, isn't it? That's life" [PT-14R]

For some of these participants, the recognition of death as a likely outcome of their terminal condition was often initiated by discussions with health professionals in charge of their care and their worsening condition. This led to reflections on wellbeing, treatment options and preferences for dying such as the desire for a pain free death.

> *"They have come and discussed the situation of resuscitation and have advised against it. It's things like jumping on my chest because I've got a bit frail through not eating properly, they could break my ribs and cause me more problems"* [PT-45R]

> *"They can't do anything with me because it's untreatable so they just keep me alive really, I've got worse than it was before… I nearly died three times I've come back again and I think the next time I won't come back because the last time I came back twice and then still held on and everything is black and blurred, my feet was numb, ah God.. it was a nice way to go. There was no pain or nothing. I'm not worried about dying as long as it goes pain-free."* [PT-32H]

However, some participants, particularly those who very near the end of life, struggled to come to terms with their imminent death and cried while reflecting on the things they wanted to accomplish and the lives they would shortly leave behind.

> *"I understand how these professional footballers feel when their glittering career comes to an end and the crowd are completely gone. I don't think I'm any different to anybody else on that, I think everyone is like that because nobody on this planet likes losing, nobody likes to go earlier than they should do… it's gone ever so quick* [cries]*"* [PT-45R Male, 76, died 4 months after interview]

> *"I left my little girl with my older sister and my little boy with my mom. There's only a few of us but, we do love each other. I love her, I love to say good bye. I know that I'm going to die, and I don't want to die, I don't want to leave here, sorry* [*cries*]" [PT-37H, Female, 35, died 4 months after interview]

Despite apparent difficulties in coming to terms with their imminent death on the part of some participants, many discussed their preparations for dying, legal and funeral arrangements. Indeed, PT-37H who cried while reflecting on her coming death also spoke of her desire to be buried and her intention to pick a song for her funeral.

> *"My funeral is paid for and it's all settled…12, 15 years I've paid"* [PT-11K]

> *"We've made a will, we've done our power of attorney, we're in the process of paying for our funerals so, you know, little bits at a time. The biggest problem is sorting the house out, 50 years' worth of rubbish"* [PT-67R]

> *"I spoke to the Priest, the only thing I have do is pick my song for my funeral, and I've made sure that everybody knows that I do not want to be cremated because now, even though you're a Catholic you can now be cremated whereas if you were Catholic we never used to. I still don't believe that you should be, but all these new age Catholics think they should be and I don't, I'm very old fashioned like that."* [PT-37H]

For some participants, end of life decision making appeared to be partly driven by a desire to reduce the burden of funeral arrangements on their family members and prevent problems in the administration of their estates after death. As noted earlier, many participants reported a sense of self-perceived burden while reflecting on their loss of capabilities in day to day

activities. It appears, that participants were also concerned about being a burden after death, underpinning the pervasiveness of self-perceived burden as an issue for many participants.

> *"…I've made all the arrangements. My son's got nothing to worry about. All he's got to do is phone them up and they come down and they will do it. It's been paid for"* [PT-11K]

> *"I made a will because when my wife died we didn't have a will and it caused a massive problem, massive problem and it was very expensive and so I made a will because my first marriage ended after two and a half years and my second marriage lasted for 40 years until she died"* [PT-45R]

> *"I have already paid for my funeral…, I have already paid for the cars, the flowers, the coffin, that's all sorted, all I have got to do is turn up basically everything is up and paid for that's a burden you don't want to leave your children with"* [PT-61R]

> *"The will and everything like that's all in place, we're financially sound. The only thing that we said we'd do would be to make our funeral arrangements so it wasn't a burden on the family"* [PT-50H]

Participants with end stage COPD were more likely to report loss of capabilities associated with end of life decision making as reported in **Table *17***. The apparently severe and unpredictable nature of their symptoms appeared to limit their ability to make important end of life decisions

> *"I'm trying to sort it out but I haven't done it yet, to make a will. Every time I try to do something, I end up back in here. I end up back in hospital so I'm not doing anything alright, about 25 time I've been in here. 25 times in the last 12 months"* [PT-40R]

> *"I haven't sort of got everything right in my head yet. There's things I want to do but I can't do it because I'm here all the while but I mean there's lots of places I've got to go, the bank and that"* [PT-23R]

Despite the various issues related to their symptoms and other long-term conditions managed on a day to day basis, most participants spoke of various ways they had coped with the effects of their condition and in some cases, were able to achieve some degree of independence.

6.3 Coping with terminal illness and multi-morbidities

6.3.1 Support from family members, friends and carers

Participants discussed the various ways they managed the effects of their terminal illness and multi-morbidities on their capabilities and wellbeing. Family members, carers and health care professionals were important in managing the effects of their terminal illness

and long-term conditions. In most cases, friends and family members were important in identifying needs, providing emotional support and maintaining important social relationships.

> *"We've got two lads and a girl. They've been absolutely marvellous. One comes every day…. Like I say, when you're in this hospital, you appreciate what they're doing. They've brought me from nothing to what I am now"* [PT-68H]

> *"I'm able to have the support that I need... my great friend she is… she comes up more than anybody. She's only a friend. She's golden, she is. I wouldn't know where I'd be without her.* [PT-13R]

> *I've got friends I've had for 60 years. I've known people for 60 years. My own brother-in-law, for instance, I get on, he comes down and sees me when he can, occasionally. He's very good. I've got one friend who lives in* (PLACE REMOVED). *I've known him 60 years. I was best man at his wedding and he was best man at mine. We'll still get in touch now, occasionally, by phone. I've got another friend in* (PLACE REMOVED). *He phones me about once a fortnight to see how things are going… my relationships with people are as well as they could be, I've always had good relationships since very long with regards that side of things.* PT-30K]

> *"…I've got my son and I rely on him. If my son sees anything in my mobility or anything I think he'll help me to move and then he'll get it… if I can't get hold of him I phone my brother and he will come down. So between the two of them I rely on those two… We're all close and we all keep in contact with one another."* [PT-11K]

Family members and friends were also important in assisting with aspects of day to day living where capabilities have been lost. Participants described how family members helped them cope with episodes of deteriorating health, and assisting with various household activities.

> *"sometimes I can wake up and I don't feel at all well and I'm not, I can't sort of do anything really. That's when my wife has to help me… people who care about me more than anyone is family, mostly my wife, my daughter, grandchildren"* [PT-28R]

> *"When she came in to see me she said to me I'm not going to leave you alone she said I've got to get you in hospital, I wouldn't go, she made me come, I'm glad she did because I was really ill. I didn't realise how ill I was"* [PT-54R]

> *"There's things I can't do; Bending down, opening cupboards and things like this, I need help there. And of course, any of the maintenance of the house, I can't do any more, nor housework, so on and so forth. So I have to rely on my wife, she's very good at it, she helps me"* [PT-66K]

> *"…My family come and help out with the cleaning. I do rely on my daughter-in-law to come and do my cleaning of my toilets and places like that. I don't do that. I think she prefers to do it to make sure that I'm clean…if I want to have help, I'd ask for it and I'd get it from my son. If I need curtains put up, I ask my son or the daughter-in-law to come and do it and they do it"* [PT-48K]

An atmosphere that allowed participants to clearly communicate their needs was an important aspect of support which appeared to improve the emotional wellbeing of participants.

> *"…the people around me doesn't make me feel different, you know what I mean, I'm able to communicate with certainty what my problem, my atmosphere, my family, my family gives me support like I'm on my feet, yeah they don't sit up and say oh you're this or you're that… I get help and support, really and honestly I'm spoiled... they do a good job with me, they do a good job making me feel like a young man*" [PT-53K]

Family members and other close persons were also important in assisting with intimate personal care and maintaining personal hygiene

> *"…as regards dressing myself, trying to make beds, I can't do so I get help from my family, I get help dressing, cleaning… they'll do for me, if I'm really poorly they'll come and they'll bath me, wash my hair and do whatever…"* [PT-60R]

> *"I have to have nurses come and wipe me and clean me up and things like that, I can't do it myself."* [PT-51R]

However, some participants did have some reservations regarding the role of family members and other close persons in providing care due to their loss of capabilities. Some participants spoke of their unease with the type of support received from health professionals to manage their condition and their embarrassment at having family members and other close persons help with intimate aspects of their personal care

> *"if I've got a problem I can phone my GP or I can phone up (consultant). If I've got a problem about anything to do with my illness, I can call the secretary and they will get back to him and it's quite good… I find that because of my illness I'm under a lot of doctors. Because I'm here, I'm at (hospital name), then I'm at (hospital name) and I'm at (hospital name) as well, at each of them, so, I feel I'm kind of the guinea pig, and then everyone else is doing their, well, which they're doing a good job but it's like I'm meant to have some appointment, but then it gets cancelled and stuff like that"* [PT-33R]

> *All my children are grown up now and when I'm poorly they come round, they bath me and that and it's embarrassing for me as it is for them, you know, to get their mother in the bath and bath her and shower her and dress her and that and I don't think it's very fair on them to do that, whether I'm their mom or not, they shouldn't be doing that"* [PT-60R]

> *"I don't like nurses coming to the toilet with me but it's just one of those situations"* [PT-45R]

Some participants however, wanted their family members involved in assisting with intimate aspects of their personal care.

> *"I've got the support that I need, I've got certain staff that I can go to, I know they would go over and beyond for me. But I would still like to see my mom to come up and help me go to the shower and things like that"* [PT-37H]

Hence, while some participants felt embarrassed about the role of family members and health professionals in helping cope with loss of capabilities, particularly those associated with intimate personal care, some felt they needed more help from family members in helping them with intimate personal care.

6.3.2 Learning to cope with loss of capabilities

Although family members and other close persons were important sources of help in coping with loss of capabilities, many participants discussed how they had learned to manage their symptoms and cope with the effects of their terminal condition on their wellbeing. For example, they discussed their use of breathing exercises to manage breathlessness, and subtle body movement to manage symptoms related to arthritis.

> *"I've got exercises to help me with my breathing, positions and resting and things like that, to help slow my breathing down so, it's better"* [PT-67R]

> *"…at the top of the spine where my vertebrae's have gone, if it gets locked, I can turn it one way, but it won't turn back the other way, I can't move my head... but they told me, when I've got it in the top of my spine to rotate my head and do it, which I do, do it from side to side and front and back and do it about four, five times a day. I try to do the exercises so hopefully, keep my fingers crossed, it will stop it from locking"* [PT-38K]

Despite struggling with symptoms related to general frailty and loss of capabilities in day to day living, some participants explained how they had learned to avoid falls and achieve independence in certain aspects of their lives. They discussed their use of walking aids in

learning to walk and their efforts to achieve independence in managing aspects of their personal care.

> *"I can move and hold things I want to and sometimes if I get up and try and pick things up I might fall down but I don't fall down now. I've taught myself not to fall down*" [PT-63K]

> *"Last week, I couldn't even walk and then they give me a Zimmer frame and now I'm on a walking stick. I am trying to help myself. It's going to be a bit of a job"* [PT-68H]

> *"I have to be washed and dressed, I'm trying to learn to do it, I could sit down and wash but I can't stand up and take a shower with soap and this and that but when it comes to dressing I am now getting my socks on, I couldn't do that for a while*" [PT-53K]

Learning to cope with loss of capabilities also appeared to involve managing the effects of terminal illness on emotional wellbeing. Managing symptoms and the effects of long-term conditions on wellbeing was a source of anxiety to many participants. For one participant, psychotherapy was a way of managing his anxiety due to his terminal condition.

> *"I do get anxious and one of the problems that I have is when I want to go to the toilet you know for a wee I get tired so that's an anxiety that I think that's built into part of my problem with my heart you know. So that issue hopefully is going to be addressed because I've joined the Healthy Minds from [Place removed]"* [PT-50H]

For some participants, however, learning to cope with loss of capabilities appeared to involve a desire to understand the limits of their capabilities and adjust both their day to day activities and the activities of family members

> *"Your body obviously determines what you can do and what you can't do and I've been carrying quite a lot of fluid round so obviously that affects how the heart works. My heart is really in a bad condition so I'm only going to have a limited sort of, hopefully not a limited life but a, limited things that I can do within my life. And once I know what I can do then I can, we can build our life round it and that's not a problem"* [PT-50H]

Through an understanding of the limits of their capabilities, some participants discussed the various changes they had made to their usual activities to cope with the effects of their terminal condition such as a deliberate attempt to avoid physical activities.

> *"At the moment, because of my heart condition, I'm not as able to be as independent as I want to be because I get tired very easily. My driving is restricted because I have an ICD which could go off if I get below a certain heartbeat. I restrict where I go and what I do. Unfortunately, I've had to give up quite a lot of my voluntary work because it's physical and I can't do physical work. I do more paperwork"* [PT-44H]

> *I don't really go out with my family much now because of mainly the walking, I get out of breath and you know, when you get there you are exhausted and you don't want to sort of participate in it all, whereas it's better if they came to my house where we can enjoy ourselves and have a family meal and whatever rather than me having the effort to get dressed because by the time I've done all that I'm too tired to do anything so I prefer them to be around me and around things like that way.* [PT-60R]

Hence, it appears that some participants explored the limits of their capabilities and when such limits were understood, adapted day to day activities in order to cope such as reducing physical activity

6.3.3 Adaptation to loss of capabilities

Adapting to end stage organ failure and other long-term conditions that limit capabilities in day to day living appeared to be an important aspect of coping with loss of capabilities; and participants in all groups discussed the ways they had adapted to their condition. However, participants with ESRD were more likely to report adapting to the effects of their terminal condition compared to the other groups; this may be related to the different rate of decline compared to the other groups and the apparently less debilitating nature of the illness. Despite reporting severe loss of functionings in relation to other groups (**Table 15**), many spoke of adaptive mechanisms that improved their capabilities in day to day living.

Participants spoke of using assistive aids that ranged from temporary structures set up in the bathroom, to relatively simple items such as the use of a chair while bathing to avoid standing. Participants however, discussed how they had effectively used changes in their bathroom to adapt to prior difficulties with intimate personal care.

> *"at first I used to have some problems in the bath. It was very difficult to come out I used to fall down on the floor, then the girl she was helping me to get out. Then social worker they come and put a board in, I can sit on a board and put one leg in, now it is easy to have a bath as well"* [PT-18K]

> *"Well I've got a walk-in shower, at the minute… if I can't stand up, I use, you know, I'll use the plastic chair, so that's the way I do it. I sit on a plastic chair and do it that way"* [PT-38K]

For some participants, despite acknowledging reduced functioning, adaptive changes in intimate personal care was reflected in their self-reported functioning.

> *"I can't stand and wash myself but I have a shower every day because I've got a long walk-in shower and I've got a seat in there which I sit down and I've got an overhead shower or a spray"* [PT-11K] (Ticked I have no problems washing or dressing myself on EQ-5D-5L)

To adapt to loss of capabilities associated with mobility, participants across the groups used mobility aids such as walking sticks, Zimmer frames and mobility scooters. Many reported improvements in their capabilities and general wellbeing as a result of the use of such aids.

> *"Since my heart attack after that I'm little weak at some areas mostly walking problem... I was walking and fell over so many times in the garden and I went to the people who give the stick for walking and I got walking stick. I use it a lot for walking"* [PT-18K]

> *"I can walk about as much as I can. I've got my frame there. I use that quite a lot now, especially at home because I fell over and I broke my hip. That's how it all started. I was stupid when I was drawing the curtains at night and I tripped over the television wire… I can walk about very slowly, very carefully. I've got my mobility scooter and all that and I get out in it"* [PT-58H]

> *"When I was very ill the first time I was dreadful you know I got nothing in the legs, nothing in the tank, I couldn't do anything...now I have a stick and I get up on my own. I don't fall over like I did when I started"* [PT-55H]

Although physical aids and assistive technology were important in adapting to loss of capabilities in day to day living, participants were often aware of the limits of these aids. For instance, one participant reflected on the difficulties of using physical aids to adapt to symptoms such as breathlessness.

> *"If you're not be able to walk or anything like that, like if you lose a leg, you can get a plastic one or with a blade, you can learn to run again. With COPD you can't. I can't walk more than a few yards without being completely out of breath. It just drains you.* [PT-61R]

Recognising the limitations of physical aids, many participants discussed practices and learned routines that enabled them to adapt to their loss of capabilities. For one participant, this involved a deliberate attempt to avoid sitting for long periods of time and morning routines to avoid falls and other risks associated with her condition.

> *"I can't sit for a long, long time, because if I sit for a long, long time, when I come to get up, I can't move because I've sat in that one position too long… I can't just get up out of bed in the morning because I can't stand until I've been up a bit and I've righted myself and I've tried to move my feet and my legs, so that I can try to get my balance. If I just get right up out of the bed, I will fall over"* [PT-38K]

For another participant, adapting to loss of capabilities appeared to involve discounting previous capabilities as relevant to current capabilities sets. Instead, he talks about being able to 'manage' them.

> *"I don't think I have any problems (with usual activities). I just try to walk a bit and do what I can, there's things I can't do like the garden and things like that, too strenuous, I can't do it, I'm doing them in a fashion but things get done, I manage them"* [PT-47K] (Ticked I have no problems doing my usual activities)

Adapting to loss of capabilities went beyond the use of physical aids and lifestyle changes, participants appeared to re-evaluate their capabilities in light of their terminal condition, which led to discounting the relevance of previous capabilities. Many participants discussed other ways they had adapted to their loss of capabilities through a change in the way they viewed and engaged with their terminal condition and loss of capabilities. One such way was to accept the limitations of their condition and its effects on their daily lives.

> *"I have adjusted to my condition, my condition hasn't adjusted to me, I know what I can't do and I accept that. I have got to be realistic about it. I'm not fighting it… I'm just sat back in bed…it's easier to rest. I accept my limitations"* [PT-45R]

> *"I just carry on with daily life as I go. If I can't do anything I don't do it. I know my limits with what I can do and what I can't do"* [PT-11K]

Acceptance of the effects of their terminal condition and loss of capabilities also appeared to involve viewing such effects as part of a normal ageing process rather than a disability. Identifying their terminal condition as a product of ageing enabled them accept pain, aches and other limits on their physical activities.

> *"I'm 60, you can go to bed and then get up with an ache and pain that wasn't there when you got in to bed, you see what I'm saying? You can get headaches, aches and pains. My age now, I'm 60 now not 16, you see what I'm saying? So, your body wears out, as you get older*" [PT-49H]

> *I think everybody does experience physical discomfort to a certain degree, you know, even you. Even the doctor… I mean, they do, don't they? Unless there's somebody like, well, like God or Jesus or whoever.... I think everybody gets a bit of pain …that's how it goes.* [PT-29K]

> *"don't forget that I'm nearly 81 you know, it isn't like when you're 40 or 42, you know, like that, in your thirties and forties, but as you get older, everybody gets it* (pain)*… I just do what I can and what I can't do, I just have to leave it and that's it"* [PT-38K]

While some participants acknowledged and accepted their loss of capabilities, they appeared to express a resolve to engage with the limits of their capabilities and strive for independence in their daily lives.

> *"… I'll have a go, if I can't do it, I'll still have a go… I try to be independent in all things if I can. I mean I'll have a go…it's never beaten me up to yet, but I'm not going to say I'm always going to be able to do it"* [PT-38K]

> *"I don't let it get to me as much as, you know, it tries to because I think you've got to have that little bit of strongness in you to say, it's not going to beat me I'll move forward in whatever way whether it's slow or fast or whatever but I'll move forward and move that way"* [PT-60R]

> *"… If I want to do it, I'll do it one way or another. If I want to do plants, I'll go up to the greenhouse. I'll make sure that I do. I intend to get something out of life. I'm not one of those who is going to be sitting there watching the telly all day"* [PT-68H]

Such resolve can involve adapting to and discounting the effects of pain and discomfort on day to day living.

> *"Well I've always got pains, so, but you get used to it, if you stop doing things, because you've got pain, you'll never do anything would you,"* [PT-38K]

> *"I'm continually in pain but it's pain I would expect so I don't really consider it"* [PT-52H]

> *"I've got this tube in that is a bit of discomfort and all I do is I have to pull my trousers down a little bit, otherwise, it cuts into me. But not enough to complain about. I wouldn't class that as a discomfort. Basically, I don't have any discomfort"* [PT-68H]

For some participants an unease with relying on others for support and a fear of being let down led to a determination to engage with the limits of their capabilities.

> *"I can't rely on people so I have to do these things. If you start relying on people and they let you down you're stuck. So you've automatically got to do it yourself and you do the best you can*" [PT-11K]

> *"you can't sort of just not do anything and then expect other people, because other people, they've got their own lives to live as well as you've got your life to live, so as long as I can keep going, that's it… I've either got to try and do it myself or go without, one or the other"* [PT-38K]

Adapting to loss of capabilities appeared to involve reciprocal trading of capabilities that relied on complementary strengths and skills of close persons. For instance, one participant with a relatively higher level of physical functioning compared to his disabled partner, discussed the trade-off in capabilities that exists between them Ih enabled him to achieve independence.

> *"I look after the wife. She's got arthritis and disabled so I look after her and of course I have to look after myself. because there are things that I do get some help with, for instance, the wife, although she's disabled she does the clerical work like answering the phone and figuring out what the house insurance is and paying the bills and things like that because she went to a good school with a good education. So I do the manual things like what's got to be done and she does on the phone and sorts the bills out and stuff like that. So I should say I'm able to be independent"* [PT-29K]

While some participants adapted to their loss of capabilities through various physical, psychological and behavioural mechanisms, it appears that adaptation can involve a mutually beneficial relationship that enables a degree of independence that would otherwise be difficult to achieve.

6.4 Summary

This chapter has explored the impact of end stage organ failure, alone or in combination with other multi-morbidities, on health and wellbeing. The chapter examines the responses to the individual attributes of the measures across the groups. Participants with end stage COPD were more likely to report severe loss of functionings and capabilities profiles compared to other groups. This was also reflected in the lower health QALY and capability scores observed among participants with end stage COPD.

Participants with end stage COPD, and to a lesser extent end stage heart failure, spoke of the pervasive nature of breathlessness which severely limited their functionings and capabilities in daily living. Qualitative analysis was used to explore in greater depth, the impact of their terminal condition on capability wellbeing at the end of life and the differences between the groups.

Participants spoke of their loss of capabilities in day to day living and its impact on their physical wellbeing, emotional wellbeing and end of life decision making. Participants reported their loss of capabilities associated with mobility, personal care and other activities of daily living. The reason for loss of capabilities differed across the groups; participants with end stage heart failure and end stage COPD identified symptoms that appeared related to their terminal condition while participants with ESRD reported other long-term conditions.

While the impact of their terminal condition on capability wellbeing differed across the groups, it was apparently more severe in the end stage COPD group. Participants with end stage COPD struggled to cope with the impact of their symptoms on their wellbeing. For many, this led to anger and frustration at their loss of capabilities and inability to perform simple tasks. In many cases, the severe and apparently less controlled symptoms affected capabilities associated with end of life decision making, particularly in the end stage heart failure and end stage COPD group. The sharp contrast between participants with ESRD

compared to the other groups, in relation to the impact of their terminal condition on their wellbeing, may relate to their distinct illness trajectory. Participants with ESRD are not particularly associated with the classic organ failure trajectory characterised by periodic exacerbation of illness but rather functional status remains relatively stable (Murtagh et al., 2011a). Indeed, most participants felt that their condition remained stable, even among those who died shortly after being interviewed.

Due to the unpredictable nature of symptoms among participants with end stage heart failure and end stage COPD; and the more enduring limitations on capabilities seen in those with ESRD, participants spoke of the various ways they had coped with their symptoms and adapted to the effects of their terminal illness and other multi-morbidities. Family members, friends and carers were important sources of support in coping with loss of capabilities. For many participants, coping with effects of terminal illness involved adapting to their physical limitations and loss of capabilities. While participants with ESRD reported severe loss of functionings in day to day living, they were more likely to report adaptive changes and mechanisms that enabled them cope with capabilities associated with their wellbeing. Adaptation involved the use of physical aids to cope with loss of capabilities associated with mobility and personal care. Adapting to loss of capabilities also involved acceptance of limitations and changing physical activities to cope with disability. There also appeared to be reciprocal trading of capabilities that enabled participants to achieve some degree of independence.

Chapter 7

Discussion

This thesis explored the feasibility of using the ICECAP-SCM, ICECAP-A and EQ-5D-5L with patients with end stage heart failure, end stage COPD and ESRD and used the analysed data to examine the relative impact of functional decline on health and capability wellbeing. Measurement of health outcomes is often used to inform resource allocation decisions (Edlin et al., 2015) and the role of measures such as the EQ-5D-5L in evaluating health and wellbeing at the end of life is the focus of considerable debate in the literature (Coast et al., 2018a, Normand, 2009a, Hughes, 2005). The EQ-5D-5L with its focus on health ignores other aspects of wellbeing that may be important in the evaluation of end of life care interventions. End of life interventions may not necessarily extend life but may be directed towards symptom control and improving broader aspects of wellbeing. Consequently, health maximisation as a normative rule in judging the desirability of social states may be inappropriate in evaluating end of life care interventions.

The ICECAP-SCM was developed as a self-complete tool to evaluate capability wellbeing at the end of life. However, there has, as yet, been little research to evaluate the feasibility of completing the ICECAP-SCM among patients near the end of life. Research to date has explored the feasibility of using the ICECAP-SCM among patients receiving care in a hospice, most of whom had a cancer diagnosis (Bailey et al., 2016). No previous research has investigated the use of the ICECAP-SCM in patients with end stage organ failure which is the most common cause of death, both globally (Naghavi et al., 2017) and in the UK (Public Health England, 2018). This study aimed to determine the feasibility of completing the ICECAP-SCM, alongside the EQ-5D-5L and ICECAP-A, for patients with end stage organ failure. Participants were asked to think-aloud while completing the measures and

their responses were examined for errors in comprehension, retrieval, judgement and response. Following the think-aloud interview, a short semi-structured interview was conducted during which responses were probed for clarity and issues around acceptability of the measures and burden of completion were further explored. Thematic analysis of both the think-aloud and semi-structured interviews identified reasons for errors made during completion of the measures and provided an in depth understanding of the impact of terminal illness on health and wellbeing.

This chapter discusses the key findings in the context of the wider literature. The chapter reflects on the methodological approach, including the strengths and weaknesses of the research. The broader implications of the research for policy and practice are discussed. Lastly, the key contributions of the research and areas for further research are outlined.

7.1 Summary of results

7.1.1 Error rates across the measures

Think-aloud sections of the interviews were examined for errors in comprehension, retrieval, judgement and response. Comprehension and response errors were the most common error type. The EQ-5D-5L had the most response errors while the ICECAP-SCM had the most comprehension errors.

The total error rates (across all four categories) in the ICECAP-SCM and EQ-5D-5L were similar at 5.7% and 6.3% respectively. The ICECAP-A had the lowest error rate at 3%. Participants with end stage heart failure had the highest error rate across all three measures at 5.9%. Participants with ESRD and end stage COPD had the lowest error rate across all three measures at 4.6% and 4.8% respectively. Thus, it is clear that error rates across all the measures and participant groups were generally low and similar. While participants with ESRD had lower error rates, the differences in error rates compared to the other groups were quite small.

Looking further into the data, there were slightly larger group differences in error rates across the three measures. Participants with end stage heart failure and end stage COPD had higher error rates when completing the EQ-5D-5L (6.7% and 9.5% respectively) compared to the other two measures completed. Most of these errors were response errors which were partly as a result of difficulties in quantifying their symptoms due to day to day fluctuations in their condition. Participants with end stage heart failure and end stage COPD had relatively lower error rates in the ICECAP-SCM at 4.1% and 4.8% respectively. In contrast, participants with ESRD had higher error rates in the ICECAP-SCM (8.7%) while having the lowest error rate in the EQ-5D-5L. The higher error rates in the ICECAP-SCM were mainly due to apparent difficulties in understanding and responding to questions related to advanced care planning and end of life decision making.

Most measures were acceptable, clear and easy to complete. Despite there being no apparent comprehension issues with the EQ-5D-5L, some had difficulties with the response options due to their fluctuating symptoms. While the ICECAP-A had the least error rates across all measures, many suggested that the question on achievement was unclear and struggled to understand the importance of achievement to their wellbeing and identify capabilities associated with achievement given their age and terminal condition. Although more participants preferred the content of the ICECAP-SCM questionnaire over the ICECAP-A and EQ-5D-5L, many had difficulties with questions on 'preparation' and 'emotional suffering'. These participants struggled with reflecting on their end of life arrangements and the impact of terminal illness on their emotional wellbeing and the wellbeing of their family members.

Most participants found completing the think-aloud task beneficial and helpful. However, some felt that talking through their answers made them reflect more on their wellbeing. In many cases, this led to sadness at their loss of capabilities and the impact of their illness on their wellbeing and the wellbeing of their family members. The practical task of thinking

aloud the questions was also difficult for some participants with respiratory failure who experienced breathlessness, highlighting the considerable impact of terminal illness and functional decline on health and broader aspects of wellbeing.

7.1.2 The ICECAP-SCM is feasible to use with patients with end stage organ failure.

The ICECAP-SCM was developed as an economic tool to evaluate capability wellbeing at the end of life, thus questions about its feasibility are relevant to this thesis. Participants had few errors while completing the ICECAP-SCM. While the error rate in the ICECAP-SCM was slightly higher than the E-5D-5L and ICECAP-A, the absolute difference in error rates across these measures were small. Participants across all groups felt the questions in the ICECAP-SCM were appropriate, clear and easy to understand. Among those who expressed a preference between the ICECAP-SCM, ICECAP-A, and the EQ-5D-5L on the most appropriate measures for evaluating their wellbeing, a slightly higher number preferred the ICECAP-SCM (four participants) over the ICECAP-A (two participants) and the EQ-5D-5L (three participants). While the sample sizes within the groups are too small to draw firm within groups conclusions, this thesis has shown that the ICECAP-SCM can be completed by patients on the organ failure trajectory with minimal error.

7.1.3 Responses to the measures

Participants with end stage COPD had lower utility and capability scores across all measures compared to those with end stage heart failure and ESRD. The lower scores observed among participants with end stage COPD was apparently due to the severe and restrictive nature of breathlessness in day to day living. Participants with end stage COPD were more likely to report severe or complete loss of functioning in relation to basic day to day activities while completing the EQ-5D-5L, compared to the other groups. This was also reflected in the ICECAP-A where participants with end stage COPD were more likely to report severe or

complete loss of capabilities particularly in relation to autonomy and achievement due to debilitating symptoms of breathlessness.

Participants across the groups had similar capability scores in the ICECAP-SCM and slightly different scores in the ICECAP-A. Participants with end stage heart failure, however, had higher scores in the ICECAP-A compared to the other groups. Participants with end stage COPD and end stage heart failure were more likely to report worse profiles in relation to capabilities associated with emotional wellbeing and end of life decision making. Most of these participants felt that their apparently poorly controlled symptoms and loss of capabilities in day to day living limited their opportunity to make important end of life decisions and made them feel like a burden to their family members. Participants with ESRD, on the other hand, were more likely to report full capabilities in relation to end of life decision making but were more likely to report loss of capabilities around attachment which made them feel isolated and lonely. Their loss of capabilities around attachment was due to a severe loss of capabilities associated with autonomy which impacted their ability to maintain valued social relationships. Thus, while the capability scores were similar across the groups, there were clear differences in the reported capability profiles across the groups which largely reflects the different ways symptoms associated with their terminal condition affected capabilities associated with day to day living.

7.1.4 The impact of functional decline on health and wellbeing

Most participants spoke of managing their terminal illness and other long-term conditions on a day to day basis amidst progressive functional decline and loss of capabilities. Although the reasons for loss of capabilities varied within the groups, these were often related to common effects of their end stage organ failure and other long-term conditions that participants were managing. Breathlessness was commonly reported by participants with end stage COPD and end stage heart failure as the reason for their loss of capabilities. Participants with end stage heart failure identified oedema as another major reason for their

loss of capabilities in day to day living. However, among participants with ESRD, accidents and other long-term conditions that did not appear to be directly related to their terminal condition such as stroke and arthritis, were often identified as reasons for their loss of capabilities.

Loss of capabilities impacted both the physical and emotional wellbeing of participants, and their ability to maintain desired social relationships. Participants spoke of the various ways their symptoms limited their mobility, ability to perform their personal care, and do tasks they were previously capable of doing. In many cases, capabilities were lost at a rapid pace which left participants feeling shocked at their rapid decline, uncertain about their current capabilities and the level of future care that they might need. In many cases, an inability to perform basic tasks left participants angry and upset about their lack of capability and reliance on others to perform such tasks. Indeed, many participants reported feeling like a burden to family members, friends and carers, and in a few cases, participants talked about under-reporting their symptoms to these close persons to due to guilt associated with self-perceived burden. Furthermore, many participants reported that their worsening symptoms and regular need for care affected their ability to make important end of life decisions.

It is clear that symptoms related to terminal illness and other long-term conditions had a profound effect on the wellbeing of participants. However, many participants spoke of various ways they had managed their symptoms and coped with their loss of capabilities. Most participants relied on family members and close persons to help them cope with the effects of their terminal condition and loss of capabilities in day to day living. On the other hand, some also discussed how they independently managed the effects of their terminal condition.

Many participants appeared to adapt to the effects of their terminal illness on their wellbeing. Due to the more subtle and predictable nature of symptoms among participants with ESRD, they were more likely to report strategies developed for controlling symptoms

and adapting to the effects of their terminal illness and other long-term conditions. On the other hand, participants with respiratory and heart failure were less likely to report adapting to their terminal condition due to the unpredictable nature of their symptoms which ultimately resulted in regular hospital admissions. Adapting to their disability involved the use of physical aids to cope with loss of capabilities associated with mobility and personal care. However, in some cases, physical aids were apparently insufficient and participants spoke of changes in behavioural practices and usual activities to cope with their terminal condition.

For many, accepting their limitations and interpreting their terminal condition as an inevitable consequence of ageing rather than their terminal illness helped them adapt to their condition. Adapting to loss of capabilities also appeared to involve reciprocal trading of capabilities between partners that enabled a degree of independence than would otherwise have been difficult to independently achieve.

The relationship of these findings to existing literature is discussed in the next section.

7.2 Relationship to wider literature

This section discusses the completion of the measures across participant groups and the impact of functional decline on health and wellbeing. Findings are discussed in the context of existing literature.

7.2.1 Error rates across the measures

This is the first study to explore error rates in the completion of the ICECAP-SCM, ICECAP-A and EQ-5D-5L in patients with end stage organ failure. There are limited studies on the use of economic outcome measures in patients near the end of life, thus the extent to which findings can be directly compared to similar literature is severely limited. Only one study has explored completion of all three measures in patients near the end of life who were receiving care in a hospice setting (Bailey et al., 2016). This section directly compares the

error rates across the three measures reported in this study to error rates reported in the study by Bailey et al. (2016).

In this study, the higher error rate in the EQ-5D-5L compared to other measures, were similar to the results of their study which showed the EQ-5D-5L with the highest error rate. However, the ICECAP-SCM had the second highest error rate at 5.7%, differing from the results of their study which showed the ICECAP-SCM with the lowest error rate the three measures. Across items of the ICECAP-SCM, while 'preparation' and 'dignity' had the highest and second highest error rate in their study, this study reported 'preparation' and 'emotional suffering' as items with the highest error rates. Indeed, emotional suffering was error free in their study.

One reason for the contrasting findings, particularly in relation to the higer error rates in the ICECAP-SCM may be that patients involved in the study by Bailey et al. (2016) were explicitly aware of their impending death due to the fact they were receiving care in a hospice whereas patients in this study may not have recognised or accepted their end of life prognosis because they were being treated in a large acute hospital and may have felt they were not yet approaching the end of life.. The ICECAP-SCM, focuses on capability wellbeing at the end of life, explicitly addresses end of life decision making compared to the other measures; hence patients may have been more reluctant to reflect on or discuss their end of life preferences. Furthermore, both studies differed with regards their sample size, patient population and care setting.

The higher error rate in the EQ-5D-5L, compared to other measures, were due to apparent difficulties in the ability of patients to quantify their health states due to their fluctuating symptoms; similar to findings by Bailey et al. (2016) which reported that patients struggled with the descriptive level of the EQ-5D-5L due to fluctuating symptoms. Among participants completing the ICECAP-A, their study reported that 'achievement' had the highest error rate similar to findings reported here. In their study, errors in the EQ-5D-5L

were spread across all five items of the measure while most errors in this study occurred while participants completed ‘mobility’ and ‘usual activities’.

7.2.2 Completion of the measures

Despite the limited literature on the use of all three measures in patients near the end of life, a number of studies have used cognitive interviewing techniques to evaluate the feasibility of using disease and end-of-life specific quality of life measures in patients near the end of life (Wilkinson et al., 2014, Watanabe et al., 2008, Bergh et al., 2011).

One study using the ICECAP-A and EQ-5D-5L in a sample drawn from the general adult population showed the ICECAP-A with a lower error rate than the EQ-5D-5L (Al-Janabi et al., 2013), similar to the results obtained here. Thus, findings of the thesis further inform existing evidence that economic measures expressed in terms of capability can perform competitively with other widely used measures, while capturing broader aspects of wellbeing that may be useful to decision makers. It should be noted however, that differences in error rate across the measures were quite small and thus differences in these error rates may not provide the most important basis for making choices regarding the use the measures in a particular clinical or end of life setting.

Utility and capability scores reported in this study were similar to the findings reported in a study by Bailey et al. (2016) which used similar think-aloud interviewing methods among patients completing the ICECAP-SCM, ICECAP-A and EQ-5D-5L in a hospice setting. Their study reported an average EQ-5D-5L utility score of 0.55, similar to the total average score of utility score of 0.52 reported by this study. However, this value was higher than the average utility score of the end stage COPD group which had an average utility score of 0.37, reflecting the severe health profile of participants in the end stage COPD group. The average capability score of 0.78 in the ICECAP-A reported here, is identical to the capability score reported of 0.78 by Bailey et al. (2016). Participants in the end stage heart failure group however, had a slightly higher average capability score in the ICECAP-A (0.85). The

average capability score among participants completing the ICECAP-SCM (0.84) was identical to the average score obtained by Bailey et al. (2016) which reported an average score of 0.84 suggesting that the different setting of care had no impact on capability wellbeing at the end of life. This is surprising as research suggest that patients in a hospice setting achieve better end of life outcomes compared to patients in acute settings (Connor et al., 2007).

Despite the similarities in the utility and capability scores in both studies, there were some differences in the response to individual attributes of the measures. Participants in this study were more likely to report severe loss of functionings in relation to day to day activities, and a lack of support to cope with their loss of capabilities compared to the study by Bailey et al. (2016) reflecting the hospice setting of care where physical needs are more likely to be met. Participants in this study also had worse health profiles in relation to physical and emotional distress; and were more likely to express anxiety regarding their terminal condition and the impact of their illness on the wellbeing of their family members. These differences are again, likely due to the hospice setting of care where symptoms are better controlled and the psychological needs are more likely to be met (Steele et al., 2005).

Participants in the study by Bailey et al. (2016) were however more likely to report loss of capabilities in relation to end of life decision making compared to the findings reported here. This is surprising given that the hospice model of care in the UK explicitly encourages end of life decision making (Faull et al., 2012). One reason for the differences may be the relatively older participants in this study. About 50% of participants in this study were over 80 years old compared to about 30% of participants in their study; hence participants may have had a longer time to reflect on their end of life preferences. Furthermore, research suggests that older people are more likely to engage in end of life decision making compared to younger people (Cook et al., 2017). Another reason for the differences in both studies may be due to the relatively younger nature of participants in the hospice setting who may

have grown up in a society with less deference to professionals; and their expectations in relation to choice in end of life decision making may be much higher given their hospice setting of care. Indeed, research suggests a generational shift in attitudes towards end of life care as more people experience friends and relatives ageing and dying (Clarke et al., 2017).

Participants in the ESRD were mainly recruited from out-patient palliative care services compared to participants in the other groups who were mainly recruited from in-patient wards. It is unclear however, the extent to which their actual terminal condition rather than care setting may have been responsible for differences in capability and utility scores and profiles reported across the groups. Participants in the ESRD group were more likely to report poor management of symptoms such as pain and loss of capabilities in relation to physical distress. This was despite the fact that participants in the end stage COPD group were more likely to report severe loss of capabilities in day to day living due to the physical effects of their terminal condition. The apparently better control of physical symptoms in the end stage COPD and end stage heart failure group compared to the ESRD group may reflect the acute setting of care where pain are better managed (Office for National Statistics, 2015). Participants in the ESRD group were more likely to report full capabilities in relation to decision making about their treatment preferences compared to the other groups which may reflect their active choice in declining potential life prolonging interventions such as dialysis and renal transplant. Participants in the study by Bailey et al. (2016) were more likely to report full capabilities in relation to choice over their treatment preferences compared to participants in the COPD and heart failure group and these similar to those reported in the ESRD group. Capabilities associated with dignity at the end of life appeared to be similar across the groups irrespective of setting of care and were slightly higher in the end stage heart failure group. Indeed, values in the end stage heart failure group were similar to those reported in the study by Bailey et al. (2016) suggesting that care setting may not particularly impact capabilities associated with dignity at the end of life. The comparisons within the groups reported here, and those reported by Bailey et al. (2016), should however

be treated with caution as the sample sizes in both studies (60 patients roughly split between three groups in this study compared to 33 patients in their study) are insufficient to make statistically significant inferences. However, they raise important questions regarding the extent to which the setting of care and end of life practices at these setting improves capabilities associated with outcomes important to patients, and warrants further investigation.

Health outcome measures, particularly measures useful for economic decision making are generally required to have sufficient psychometric properties before their wide spread use in any given population (Brazier and Deverill, 1999). However, it is important that such measures are acceptable, clear and easy to understand. Most health outcome measures, including measures developed for the evaluation of end of life interventions, mainly focus on psychometric characteristics, ignoring the issues around interpretability of the items, burden and acceptability (Albers et al., 2010). Researchers working in the field of outcome measurement have strongly argued for a greater use of qualitative methods, including cognitive debriefing to assess administrative burden, interpretability and practical issues that can affect the use of quality of life measures (Turner et al., 2007, Cano and Hobart, 2011).

Cognitive interviewing techniques were useful in providing greater insight into the acceptability of the measures among participants, their ease of use and the burden of completion. These metrics are instrumental to evaluating the feasibility of using quality of life measures particularly in patients near the end of life where worsening symptoms may seriously affect participants' abilities to complete quality of life measures (Emanuel et al., 2004). Although the questionnaires were generally viewed as acceptable, clear and easy to complete, questions relating to anxiety and depression in the EQ-5D-5L and emotional wellbeing in the ICECAP-SCM appeared to be sensitive for many participants. The sensitivity of these questions may relate to a sense of self-perceived burden common among many participants who struggled with uncertainties associated with their illnesses and the

impact of their terminal condition on the wellbeing of their family members. Conversations around end of life care are generally challenging (Pontin and Jordan, 2013) and reflecting on health and wellbeing at the end of life can be upsetting (Pessin et al., 2008). Indeed, previous studies have reported that patients often had difficulties responding to questions on advance care planning and end of life decision making while completing quality of life measures (Bailey et al., 2016, Harding et al., 2010, Wilkinson et al., 2014). Questions that explore sensitive issues may require careful attention as research show that questions deemed sensitive by respondents are more likely to be left uncompleted (Korevaar et al., 2002, Addington-Hall et al., 2014). Previous research using quality of life measures in people near the end of life has reported that questions relating to anxiety were difficult and upsetting to answer due to concerns over the impact of their illness on their family members (Watanabe et al., 2008).

Error rates across the groups differed at both the item level and the person level. Item level errors represent the number of errors across the attributes of a measure while person level errors indicate the number of persons who make errors in a measure irrespective of the number of times the errors occurred. A slightly lower percentage of participants with ESRD (27.7%) had errors in the ICECAP-SCM compared to the end stage COPD (28.6%). However, the item level error rate in the ESRD group (8.7%) was significantly higher than the end stage COPD group (4.7%). The higher person-level error in the end-stage COPD group represents a higher number of participants with single item errors while the greater item-level error in the ESRD group reflects a higher number of participants with errors in multiple items. The disparity between person and item level error rates raises important questions such as the relative significance of both level of errors to the use of the measures. Error rates are rarely reported in think-aloud studies and most studies are largely focused on improving questionnaire content (Patrick et al., 2011, Murtagh et al., 2007, Addington-Hall et al., 2014) hence the implications of differences in error rates on an item and person level basis is largely unaddressed in the literature. However, a useful guide to understanding the

relevance of both person and item level error may relate to their informational content. While person-level errors provide information on the number of people able to complete a questionnaire without errors, it does not account for differences in the number of questions contained in a measure. For instance, the ICECAP-SCM contained 7 items while the ICECAP-A and EQ-5D-5L each contained 5 items; thus, there were more 'opportunities' for errors in the ICECAP-SCM. Item-level error rates on the other hand, do account for differences in the number of items in a questionnaire and the nature of errors made by participants across individual items, thus providing a more informed basis in evaluating the completion of these measures.

In conclusion, the low error rates and ease of completion of the measures show that all three measures can be used in patients with end stage organ failure. Although some questions were upsetting to some participants, the sensitivity of the questions may reflect the end of life setting in which the research was conducted and the impact of participants' illnesses on broader aspects of their wellbeing. Most participants were recruited from an acute setting where there may be less end of life support for emotional and psychological aspects of wellbeing compared to other end of life settings such as hospices (Virdun et al., 2015).

7.2.3 Comprehensibility and interpretation of the items

For measures to be meaningful to participants, its items have to be clearly understood and defined in relation to relevant aspects of wellbeing. Most errors and non-response in questionnaires arise from problems in cognitive tasks required to generate an appropriate response (Tourangeau, 2018). While errors were generally low across all measures, some items had a higher error rate than others mostly due to comprehension and response issues.

In the ICECAP-SCM, some participants had problems understanding and providing a response to questions on 'preparation' and 'emotional suffering'. Comprehension errors in preparation were due to an apparent inability to distinguish between preparation related to end of life decision making and preparation in the context of day to day living, interpreting

the item in relation to specific household activities unrelated to end of life decision making. The description of 'preparation' in the ICECAP-SCM (**Appendix *5***) provides cues that explicitly link preparation to end of life decision making suggesting that some participants might have been reluctant to fully engaged with the question and reflect on the possibility of death. End of life conversations are notably difficult (Pontin and Jordan, 2013) and previous studies have reported that patients often had difficulties responding to questions on advance care planning and end of life decision making while completing quality of life measures (Bailey et al., 2016, Harding et al., 2010, Wilkinson et al., 2014). Similarly, the higher number of errors in emotional suffering may be related to the strong emotional impact experienced by patients due to their terminal condition (Soto-Rubio et al., 2018)

Some participants questioned the relevance of 'achievement' in the ICECAP-A to their wellbeing suggesting that their terminal condition and advanced age made the question irrelevant to their circumstance. This is similar to findings from a previous study using the ICECAP-A in an older population reported that some participants struggled with achievement and questioned its relevance to their wellbeing (Keeley, 2014). In some cases, however, participants identified different barriers to achievement such as educational accomplishments, improvement in health and general wellbeing. The different contexts in which achievement was framed in the ICECAP-A was also reported in a previous research using the ICECAP-A (Keeley et al., 2013). Differences in the way patients interpret items in a questionnaire have also been reported by other studies (Iris et al., 2016, Watanabe et al., 2008). Research show that differences in interpretation of words or phrases in a question may be influenced by respondent characteristics such as demographic and cultural factors (Iris et al., 2016). While what constitutes reasonable achievement may be similar among people who share similar characteristics such as age, they are likely to differ among people with different age and social condition.

The differences in the normative relevance of aspects of wellbeing such as 'achievement', where participants often questioned its importance to their wellbeing, may underscore the need for an approach that explicitly accounts for stages of the life-course and patients on different end of life phases (Coast, 2019).

7.2.4 The impact of end stage organ failure on health and broader aspects of wellbeing

The benefits of end of life interventions often go beyond their impact on health status and affect other aspects of wellbeing that are relevant to end of life decision making (Kehl, 2006, Clark, 2017). Breathlessness, oedema and other symptoms, despite being managed on a day to day basis, led to loss of capabilities across the groups, in the areas of physical wellbeing, emotional wellbeing, social relationships, and end of life decision making.

Breathlessness is a common symptom among people living with end stage organ failure (Currow et al., 2010). Breathlessness was reported by most participants, particularly those with end stage heart failure and end stage COPD. Breathlessness led to loss of capabilities associated with mobility, personal care and basic day to day activities. The regular need for medical attention to manage breathlessness and other symptoms among participants in the end stage heart failure and end stage COPD group appeared to further limit the ability of participants to make financial and legal end of life decisions. Despite attempts at self-management there was a sense of continued and often rapid decline. Worsening symptoms and a progressive decline in capabilities was a source of anxiety for many participants. The effect of breathlessness on wellbeing seen in this study has also been reported by previous studies. For example in a study of patients with COPD it was found that despite active interventions to reduce breathlessness, deterioration in social, emotional and physical functioning were common among people with end stage COPD (Elkington et al., 2005). Similarly, for people with end stage heart failure, breathlessness was often associated with fear, fatigue and a sense hopelessness (Horne and Payne, 2004). Severe breathlessness has

also been linked to depression, anxiety, social isolation (Jones et al., 2004) and inability to perform basic day to day activities (Hasson et al., 2008, Fraser et al., 2006).

Most participants with ESRD reported a relatively stable condition including those who were very near the end of life – a contrast to reports among participants with end stage heart failure and end stage COPD. Previous research on the functional status of patients with ESRD reported a relatively stable functional decline which subsequently becomes rapid as the patient approaches death (Murtagh et al., 2011a). While participants with end stage heart failure and end stage COPD mostly managed symptoms related to their terminal diagnosis, participants with ESRD often spoke of other long-term conditions that appeared unrelated to their terminal illness such as arthritis, stroke and falls. The apparently greater impact of co-morbidities on wellbeing observed among ESRD participants has also been reported by Wachterman et al. (2017) who found that patients with ESRD were more likely to have a greater burden of co-morbidities compared to those with cancer. Despite a relatively stable physical condition, most participants were often anxious over the impact of long-term conditions on their wellbeing. Anxiety over the effects of long-term conditions on day to day living, sleeplessness and fatigue were often reported by participants in the ESRD group. Research suggest that patients with ESRD are likely to suffer from emotional and psychological distress due to the impact of unanticipated co-morbidities on their wellbeing (Damery et al., 2019), and uncertainty in attributing symptoms to their renal disease (Selman et al., 2019). Previous research identified anxiety, sleeplessness and fatigue as symptoms commonly reported by patients with ESRD (Traue and Ross, 2005, Yong et al., 2009)

The impact of self-perceived burden on wellbeing is of growing concern particularly at the end of life. Participants across the groups regularly expressed concern over the impact of their terminal condition on the wellbeing of their family members. Most of these participants, particularly in the end stage heart failure and end stage COPD group, felt that their worsening symptoms made them a burden on their family members. A sense of self-

perceived burden was evident in financial, legal, and other aspects of end of life decision making. This is similar to findings from previous studies which showed that patients near the end of life report a sense of self-perceived burden while reflecting on their wellbeing, often becoming distressed (Wilkinson et al., 2014, Iris et al., 2016). Self-perceived burden has been closely linked to negative physical and mental health outcomes (Wilson et al., 2007), depression and poor quality of life (Chochinov et al., 2007). Indeed, research show suggests that self-perceived burden can influence end of life decision making with some patients reluctant to receive home care due to considerations of the impact of their illness on family members (Rakic et al., 2018).

7.2.5 Coping with loss of capabilities at the end of life

Patients near the end of their lives often use a combination of physical, emotional, cognitive and behavioural strategies to manage their disability and the effects of their terminal illness on their wellbeing (Benkel et al., 2010, Sand et al., 2009). Family members and close persons provided immediate support to most participants that enabled them cope with their loss of capabilities. Family members also provided a supportive environment that enabled participants to cope with the impact of their terminal illness on their emotional wellbeing. The role of family members, friends and the wider community in providing a supportive network to cope with loss of capabilities is increasingly being recognised as an important aspect of end of life care provision (Rumbold, 2010). Previous studies have shown that family members and close persons regularly undertake important informal caring roles in providing support and care to older relatives particularly those with severe disability near the end of life (Minkler, 2014, Burns et al., 2013). This research provides further evidence on the specific roles of family members and close persons in assisting patients with end stage organ failure cope with the effects of their terminal condition.

Most patients used a combination of physical, cognitive and behavioural adaptive changes that enabled them to cope with their loss of capabilities. Walking sticks and various assistive

devices were used by the participants to help them adapt to loss of capability in mobility and personal care. Breathing exercises and rest stops during walks were used by many participants to cope with breathlessness; and in some cases, usual activities and daily routines were changed. These findings are similar to the results of other studies which reported that patients used breathing exercises (Bausewein and Simon, 2013), took breaks during long walks and changed daily routines to cope with breathlessness (Barnett, 2005). In some cases, adapting to the effects of their terminal illness involved accepting symptoms and disability as a normal aspect of existence rather than a disability. Acceptance is a common coping mechanism exhibited by people with progressive chronic illnesses near the end of life (Telford et al., 2006). Research shows that individuals with degenerative long-term conditions tend to view their loss of capabilities as a normal aspect of their wellbeing in order to maintain their sense of identity (Lloyd et al., 2016). Research also shows that patients with end stage COPD adapt to their terminal condition by viewing their disability as normal rather than a limitation (Habraken et al., 2008).

7.2.5 Measuring end of life care outcomes in organ failure

Measuring end of life care outcomes involves considerable conceptual, practical and ethical challenges given the increasingly complex realm of palliative care delivery and broad spectrum of needs at the end of life. The greater cognitive impairment and functional decline among people near the end of life, compared to non-end of life patients, pose serious challenges to the use of outcome measures at the end of life (Head and Ritchie, 2004). Measuring end of life care outcomes is particularly important in patients on the organ failure trajectory where care is often delivered in non-specialist settings where the quality of palliative care may be poor. A systematic review of the literature in chapter three showed insufficient evidence on the feasibility of using economic measures in patients receiving palliative care. Indeed, no study has investigated the feasibility of using economic measures exclusively in patients on the organ failure trajectory. Hence, this research provides much

needed evidence on the feasibility of measuring end of life care outcomes in patients on the organ failure trajectory. The majority of patients approached agreed to participate in the research, particularly among patients with end stage heart failure where 21 of 24 patients approached consented to complete the participate in the research. Similar studies in patients near the end of life have reported much lower consent rates (Bailey et al., 2016, Flythe et al., 2019). Even among patients very near the end of life, there was no apparent difference in the ability of patients to complete the measures compared to patients in the earlier stages of their terminal illness. Although there were health-related issues such as breathlessness that influenced the ability of the COPD group to complete the measures, these challenges were due to the think-aloud method which required verbalisation of responses.

The organ failure trajectory presents a unique challenging environment where rapidly fluctuating health state may affect patient-measure completion (Evans et al., 2013). Measuring health outcomes when health states fluctuate has been identified as an issue in conditions with episodic exacerbations such as multiple sclerosis (Lorenz et al., 2007). Outcome assessment particularly at times when health states fluctuate has been shown to lead to inaccurate estimation of quality of life in patients with episodic symptoms (Sanghera and Coast, 2018). Most errors in the EQ-5D-5L were response errors and a considerable number of these errors were due to participants selecting multiple response options, particularly among participants in the heart failure and COPD group. Some of these participants suggested they belonged to more than one response option due to their fluctuating symptoms. These findings are similar to the results of a previous study which reported that patients receiving care in an acute setting had difficulties quantifying their health state due to their fluctuating symptoms (Wilkinson et al., 2014). A similar study using all three measures in a hospice setting also reported that some participants struggled with the descriptive level of the EQ-5D-5L due to daily fluctuations in their terminal conditions (Bailey et al., 2016). Another study reported that some patients near the end of life selected a range of response options due to their fluctuating health states (Watanabe et al., 2008).

Indeed, some studies report that some participants have suggested increasing the recall period of the quality of life measures (Iris et al., 2016) and administering measures repeatedly over time to capture their fluctuating symptoms (Flythe et al., 2019). The fluctuating nature of symptoms particularly among patients on the organ failure trajectory suggests that the descriptive level of the ICECAP-SCM, expressed in terms of frequency may provide a more appropriate basis, compared to the EQ-5D-5L expressed in terms of severity, for evaluating end of life care outcomes.

The value judgements that inform economic decision making, and outcomes considered relevant at the end of life underlies the conceptual challenges involved in measuring end of life care outcomes (Coast et al., 2018a). Researchers have questioned whether a measure that goes beyond health to include a broader aspect of wellbeing, may provide a more appropriate basis for evaluating end of life care outcomes. For most participants, the effects of their terminal illness and other long-term condition managed on a day to day basis went beyond health-related aspects of their wellbeing. Most participants spoke of the various ways their terminal illness impacted on their social relationships and ability to make relevant end of life decisions. These issues, in addition to the impact of self-perceived burden discussed in the previous section, may underscore the need for broader aspects of wellbeing such as social and emotional and psychological wellbeing in evaluating end of life care outcomes.

While adapting to the effects of terminal illness is an important aspect of living with end of life conditions, the prospect of adaptation raises important methodological questions around measuring end of life care outcomes due to the possibility of response shifts (Li and Rapkin, 2009). Research show that individuals with long-term disability tend to value their quality of life higher than proxy of hypothetical valuations (Menzel et al., 2002). Indeed, some participants suggested their disability was a normal consequence of ageing, choosing to select a response option that indicated full functioning in relation questions about their usual

activities despite admitting an inability to perform such activities. Such clear examples of adaptive responses may compromise the ability to identify health needs and deficiencies, a core aim of measuring health outcomes. Furthermore, it can also undermine the use of economic measures in resource allocation decisions when values do not truly reflect a health state (Joore et al., 2002). However, questions remain on the extent to which such choices constitute 'real adaptation' or reflects the changing priorities of patients (Mitchell, 2018). The capability framework, underpinned by distributive concerns and the impact of response shifts on resource allocation (Hick, 2012), is theoretically immune to response shifts due to adaptation. Hence, the ICECAP measures based on the capability framework may be more appropriate in end of life and long-term care settings where adaptation and response shifts are likely. Indeed, research by Coast found that while there is evidence of adaptation among people near the end of life, this was not reflected in self-assessments of capabilities among some people with end of life conditions (Coast et al., 2018b).

7.3 Reflections on the research

This section reflects on the qualitative method used and data collection process. It reflects on the data collection set-up and procedure and the feasibility of using a think think-aloud interviewing technique in patients with end stage organ failure. It also reflects on role of the researcher and the sensitivity of conducting research in people near the end of life. Lastly, it reflects on the strengths and limitations of the research.

7.3.1 Reflection on data collection and think-aloud interview procedure

It was expected that participants would differ in their ability to understand the think-aloud exercise hence a preliminary think-aloud task was performed before the main exercise to enable participants to become familiar with the technique. Despite this, participants differed in their ability to understand and perform the task. However, some difficulties in the ability to perform the think-aloud task were directly linked to the effects of their terminal illness. For instance, breathlessness was a major issue for participants with end stage COPD who

struggled with talking through their answers. The effect of breathlessness on the ability to think-aloud can be seen in the higher number of refusals to participate in the research due to breathlessness (5 of 17 patients approached declined to participate due to breathlessness); it also makes these interviews particularly challenging to conduct. Participants with ESRD and end stage heart failure had no apparent problems with the think-aloud task that was related to their terminal condition. The inability of some participants with end stage COPD to perform the think-aloud tasks raises questions about the suitability of the think-aloud method for people with this condition. It also raises the possibility that patients with severe breathlessness may have been excluded. To reduce the possibility of patients being excluded due to breathlessness, patients were encouraged to remain on oxygen therapy as they completed the questionnaires. Although the noise from the nebuliser reduced the clarity of the recordings from these think-aloud interviews, this decision enabled patients who struggled with severe breathlessness to participate in the research. This approach proved to be effective as a third of all patients in the end stage COPD group were in the late end of life phase (that is, died within four months of being interviewed) including two participants who died within a few days of being interviewed.

There were some participants who required assistance in reading the questionnaires for reasons such as fatigue, difficulties in reading, blindness, and loss of fingers. However, these participants were willing to participate in the study and asked for the questionnaires to be read aloud by the interviewer. There may be differences between the responses to self-administered and interviewer-administered questionnaires among respondents (Levine et al., 2005). However, any influence on the interview is likely to have been minimal, as care was taken to ensure that the questions were read verbatim and efforts made to ensure that no word or phrase was emphasised. Furthermore, interviewees were encouraged to think-aloud their responses as they would have done if they had been completing the questionnaires themselves.

The use of a think-aloud method may have prompted participants to engage with the questions more than they would in a usual self-complete situation. Indeed, some participants reported that talking through their answers made them reflect more on their wellbeing than they would have otherwise. While a deliberative reflection on wellbeing provides useful qualitative data that underscores the importance of the think-aloud process in evaluating the feasibility of completing a questionnaire, it raises questions about its impact on the cognitive process and broader applicability. Questions on whether the act of thinking aloud changes the response to a question is an ongoing debate in the use of think-aloud methodology (Güss, 2018). Previous studies evaluating the validity of a think-aloud methodology have, however, found no evidence of change in the cognitive process due to a think-aloud procedure (Leow and Morgan-Short, 2004).

7.3.2 The influence of the researcher

The identity of the researcher in relation to qualitative research is the subject of considerable debate in the literature (Galdas, 2017, Harvey, 2013). Questions about the influence of the researcher's opinions and values on the research questions, methodology, data analysis and results are particularly important in qualitative research. To reduce the potential for bias, continuous self-reflexivity, and critical reflection on one's opinions and values, is recommended throughout the research process (Stannard, 2012). There are certain areas of my background, training and opinions and values that may have influenced the research process. These include my training as a health economist; my background as a black African conducting qualitative research in a different culture with predominantly white British participants; and my lack of prior experience in qualitative research and sensitive interviewing. Theorised subjectivity, a reflexive approach that critically examines the influence of the intellectual and personal biographical characteristics of both the researcher and respondent, is encouraged throughout the research process (Letherby et al., 2012). However, any influence related to my identity is likely to be minimal as a rigorous process

was established in developing the research questions, data collection and analysis. The research questions were developed in collaboration with well-established researchers in both the School of Nursing and Health Economics Unit of the University of Birmingham with a broad range of research experience. Furthermore, to ensure rigorous data collection procedures, a protocol and topic guide were developed in collaboration with consultants and nurses in the recruitment site. The topic guide was adhered to throughout the interview process. Themes developed during the analysis were regularly reviewed and critiqued by members of the research team.

My lack of experience in think-aloud interviewing and qualitative research may have impacted on the quality of the think-aloud interview process. To familiarise myself with the think-aloud interview procedure, mock think-aloud interview sessions were done with colleagues and critical feedback was received. When data collection commenced, my confidence and skill in interviewing improved as I conducted interviews with participants. Interviews were actively listened to, in order to identify areas for improvement. Guidance was received from more experienced members of the supervisory team on the use of prompts to improve the quality of the data collected from the think-aloud interview. My ability to communicate the think-aloud procedure effectively and manage the interview process also improved as I conducted more interviews with participants. Furthermore, critical feedback was received from researchers experienced in the use of the technique on ways to improve the quality of the data collected during the study.

7.3.3 Sensitivity of conducting the interviews

The sensitivity of conducting interviews was a particular concern in this research given the nature of the study. All participants were near the end of life and some questionnaires administered to them, in particular the ICECAP-SCM, covered sensitive issues. Hence, considerable skill was required in managing emotional reactions to sensitive topics that were discussed during the interview. Training in sensitive interviewing was undertaken before

data collection commenced and a protocol was developed for dealing with emotional reactions that might occur during the interview. Informal arrangement to debrief was offered by a member of the supervisory team. The opportunity to debrief with my supervisor was particularly helpful as it provided a situation where my thoughts about the patients I had spoken to could be expressed, particularly in situations where patients died a few days after being interviewed. A journal was kept where thoughts about patients interviewed were written. The journal provided an opportunity to explore the emotional toll of the interview and served as a debriefing tool particularly following interviews where participants verbalised their helplessness in the face of severe disability and spoke of their fear of dying.

Two participants cried while reflecting on some questions contained in the measures during the think-aloud interview. Opportunities were offered to both participants to stop the interview and seek further help which they both declined. In both situations, the participants completed the think-aloud interview and upon reflecting on the think-aloud process expressed views that while the questions were sensitive, they were helpful and relevant to their wellbeing.

7.3.4 Reflections on data analysis

The think-aloud segments of the interviews were analysed for completion errors followed by a thematic analysis of both the think-aloud and semi-structured segments of the interview. This section reflects on theoretical and practical issues associated with data analysis. The section reflects on the role of the independent raters in error analysis and emergent theoretical issues associated with the use of a think-aloud method.

Five independent raters assessed the think-aloud aspects of the interviews for errors in completion. Of the five raters, one was involved in developing the attributes of the ICECAP measures. Their involvement is unlikely to lead to bias in error classification across the measures as clear rules on the assignment of errors were determined using established guidelines (Beatty and Willis, 2007). Furthermore, errors were assigned following a

deliberative process which involved all five raters. The number of raters in the study was either similar to or higher than the number of raters in previous studies using similar methods (Al-Janabi et al., 2013, Bailey et al., 2016, Horwood et al., 2014). There are no specific guidelines on ideal number of independent raters in a think-aloud study. However, expertise and knowledge of the underlying content of assessment is recommended (Li and He, 2015). Indeed, one rater (JC) was involved in the development of two of the three measures (ICECAP-SCM and ICECAP-A), and two other raters (CB, PK) have used both measures in research.

Error rates were quantitatively presented and descriptively used to explore differences in error rates both within and between participant groups. Given the large number of sub-samples (i.e. 60 patients each completed 3 questionnaires), questions remain about whether the degree to which error rates differ across the measures can be explored through statistical methods.

This analysis was focused on descriptive statistics of the error rates to support the primary study objective of determining the feasibility of completing the measures. The study sample size was large enough to achieve this primary objective. However, I was aware from the outset of both the logistical difficulties of recruiting patients near the end of life and the additional burden on health care staffs and department that would be required to recruit a sample size large enough to conduct a formal comparison of error rates between the measures. Using a paired t-test to identify a difference of 5% in the error rates between any two questionnaires, assuming the standard deviation of the error rate of approximately 15% and a power of 0.9, for a 5% significance test; between 88 and 119 responses to each questionnaire would be needed. As each questionnaire has a maximum of 60 responses, any analysis would likely be underpowered (i.e. give a false negative). Furthermore, increasing the number of statistical tests conducted also increases the likelihood of a false positive (Type I) error.

7.3.5 Strengths and Weaknesses of the research

This thesis explored the feasibility of using the ICECAP-SCM with patients in end stage organ failure. The strengths and weaknesses of the research are discussed in relation to aspects of the research methodology such as the sample size and data collection and analytical techniques.

7.3.5.1 Sample size and characteristics

One of the major strengths of the research is the number of participants recruited to the study. Sixty participants with end stage organ failure participated in the research making it the largest patient-completed think-aloud study using any of the ICECAP measures to date. It is also the largest patient-completed think-aloud study using economic measures of health and wellbeing. The sample size is also large for a think-aloud study (Phillips, 2014). The high number of participants is a clear strength of the work given the known significant challenges in recruiting participants for end of life research (Kirchhoff and Kehl, 2008, Campbell et al., 2016), and has resulted in a significant amount of data that strengthens the generalisability of findings.

Another strength of the research was the success in recruiting people who were very near the end of life. End of life care was operationally defined, in line with current guidelines, as the care received by people in the last twelve months of life. Hence, prognostic criteria were developed in collaboration with health care professionals to facilitate the recruitment of participants who were likely to be in their last twelve months of life. The accuracy of the application of the prognostic criteria in selecting participants for the study is confirmed by the fact that 23 of 60 participants had died within six months of being interviewed. The study was closed six months after data collection ended following the recommendation of the recruitment site research and development unit so it was not possible to determine the number of participants who died within 12 months of being interviewed. Some of these participants died within a few days of being interviewed, ensuring the inclusion of people

at different phases of the organ failure trajectory. Recruiting patients very near the end of life has proved difficult in the past due to greater illness burden and rapidly changing prognosis as patients approach the end of life (Mackin et al., 2009).

A third strength of the research was the recruitment of participants with different conditions causing end-stage organ failure, from different and non-hospice settings. For example, participants with ESRD mainly received care from specialist nurses working in the community while participants with end stage heart failure and end stage COPD were recruited mainly from the wards in the hospital. Some participants with end stage heart failure were recruited in the community and received care from palliative care nurses working in the community. The different care settings from which participants were recruited was important in ensuring generalisability of findings given contemporary evolution in palliative care delivery models.

There are however some limitations of the study. One limitation of the study is the recruitment of participants from a single geographical location, thus raising some questions about the generalisability of the results. The research may have benefitted from recruiting participants from multiple locations across different regions of the UK. Words and phrases can be interpreted differently across geographical areas. Previous research has identified comprehension errors due to differences in questionnaires due to differences in the way words are used between cultures (Weis et al., 2013). Hence, this research may have benefitted from recruiting patients from diverse geographical locations. However, due to financial and time constraints associated with this doctoral work, this was not feasible but may be considered for further research.

A second limitation of the research is the limited ethnic and religious diversity of the research participants. Black and other ethnic minorities made up about 10% of the sample size. However, this is not too dissimilar to official figures which show that people from ethnic minorities make up about 13.9% of the population (Office for National Statistics,

2018a). The majority of research participants identified as Christian, except for one participant who identified as Sikh. Given the growing religious diversity in the UK (Office for National Statistics, 2013), and the importance of spiritual and religious domains to end of life considerations (Koenig et al., 2012), greater religious diversity among research participants may have increased the generalisability of the findings. Future research may consider recruiting patients from settings with large proportion of religious minorities.

7.3.5.2 Data collection and analysis

The use of think-aloud methodology alongside retrospective verbal probing minimised interference during the interview and allowed for further clarification of unclear responses. The semi-structured interview provided an opportunity to further explore aspects of completion that may not have been captured by the think-aloud process such as issues around acceptability and burden of completion.

The use of a think-aloud technique has some limitations, many of which relate to theoretical issues with the technique. Questions remain regarding the extent to which the use of a think-aloud technique reflects working memory (Charters, 2003) and the effects of the greater cognitive demand of think-aloud interviewing (Phillips, 2014). Furthermore, the apparent lack of a standardised framework for evaluating qualitative data from think-aloud studies may lead to questions about the objectivity of the findings in relation to the assessment of errors. Despite the limitations of the technique, think-aloud methodology has proved popular among researchers and has been widely used to evaluate the use of health outcome measures in various settings.

The research may have benefitted from data analysis alongside data collection. Simultaneous data collection alongside analysis informs the data collection and can improve the quality of the research (Ruona, 2005). Due to time constraints and significant delays in obtaining ethical approval and commencing data collection, an accelerated recruitment protocol was implemented which did not allow sufficient time for data collection alongside

analysis. However, whilst simultaneous data collection alongside the analysis may have influenced the thematic analysis and allowed for the possibility of adjusting the questions posed to participants in an iterative way, it is unlikely to have influenced the error analysis of the think-aloud data.

7.4 Implications of the research for policy and practice

The research presented in this thesis primarily investigated the feasibility of using the ICECAP-SCM in patients on the organ failure trajectory. The results of the thesis are relevant to end of life care research particularly in relation to evaluation of end of life care interventions. The broader evaluative scope of the ICECAP-SCM has important implications for health care decision and policy making.

This thesis provides much needed research on the feasibility of using economic measures in evaluating health and wellbeing at the end of life. The research has clearly demonstrated that the ICECAP-SCM can be used to evaluate end of life care outcomes in patients on the with end stage organ failure receiving care in different settings. The ICECAP-SCM was acceptable, clear and easy to understand. There is also clear evidence that the ICECAP-SCM does indeed go beyond health to include aspects of wellbeing relevant to the end of life such as psychological and social wellbeing and end of life decision making which are neither captured nor explicitly addressed in widely used measures such as the EQ-5D-5L. The results therefore, offer an alternative for researchers working in end of life research interested in the use of a more comprehensive and relevant measure for evaluating end of life interventions and services. The ICECAP-SCM provides researchers and policy makers with a tool to evaluate both health and social care outcomes of interventions directed at people near the end of life.

This thesis has shown that the ICECAP-SCM (and indeed the other two measures) can be used with patients very near the end of life. Evaluating end of life care outcomes in patients in the last days and weeks of life has proved difficult in the past due to a range of health-

related and ethical issues(Chen et al., 2014). The ICECAP-SCM provides an opportunity for decision makers and health care professionals interested in the impact of interventions and services provided to patients in the last few days and weeks of life. Routine evaluation of end of life care interventions and practices in the last few weeks of life can improve quality of palliative care services provided and inform economic decision making.

The ICECAP-SCM has not been tested in other common end of life care populations and dying trajectories such as patients on the frailty trajectory who may be found in different care settings such as nursing homes. Hence, researchers interested in using the ICECAP-SCM should be mindful of the relatively small amount of research which has used the measure and that further research is needed, particularly in relation to its construct validity and sensitivity to change (Streiner et al., 2015).

This thesis has underscored the importance of broader aspects of wellbeing rather than just health, in the evaluation of end of life care interventions.

7.5 Recommendations for Practice

The results of the thesis have relevant policy and decision-making implications. The National Institute for Health and Care Excellence (NICE) is currently exploring new ways to capture the wider benefits of health care interventions, particularly through measures that are relevant across both health and social care (NICE, 2017). There is increased recognition of the importance of social care at the end of life among policy makers, due to increased vulnerability and loss of functionings associated with people near the end of life (UK Parliament, 2015). Hence, the evaluative scope of the ICECAP-SCM, which measures capabilities associated with broader wellbeing may be relevant to the strategic objectives of NICE and the Department of Health and Social Care.

Currently, health-based instruments such as the EQ-5D-5L are being used in decision making of health interventions including end of life of life care interventions. This research provides useful information that informs the growing debate on the role of QALYs and

capabilities in evaluating outcomes of health interventions (Mitchell et al., 2017, Cabrera, 2018). The use of the ICECAP-SCM with its broader evaluative scope offers the prospect of an alternative measure that can inform the decision-making process at different levels of policy making and provide a more appropriate estimation of the benefits of end of life care interventions.

7.6 Directions for future research

This is the first research to compare the EQ-5D-5L, ICECAP-SCM and ICECAP-A in patients with end stage organ failure. A number of areas of further research are identified below.

7.6.1 Research in different care setting and illness trajectories

The ICECAP-SCM has been investigated in both the terminal illness trajectory and the organ failure trajectory. However, there is a need to investigate the feasibility of using the ICECAP-SCM in patients on other illness trajectories and in other care settings such as the frailty trajectory (e.g. dementia). Patients on the frailty trajectory experience a different pattern of decline and receive care in settings (nursing homes) that are different from patients on the organ failure and terminal illness trajectories. Research in these areas would help determine the suitability of ICECAP-SCM and its applicability to broad aspects of end of life care.

7.6.2 The use of the measures within the groups

Although the total error rates across all three measures were similar across the groups, there were significant differences in the completion of the measures across the groups. Future research may seek to explore the reasons for such differences particularly the higher error rate in the ICECAP-SCM observed among participants with ESRD. The reason for the higher error rate, mainly across emotional suffering and preparation, are not entirely clear,

underscoring the need for research to explore in greater depth issues around completion of the ICECAP-SCM in patients with ESRD.

Another area for further research may be the role of the EQ-5D-5L in evaluating conditions with fluctuating symptoms. Most participants with end stage heart failure and end stage COPD reported difficulty in completing the EQ-5D-5L due to their fluctuating symptoms that made it hard to select an appropriate response option. Hence, further research is necessary to better understand the impact of fluctuating health states in quality of life measurement.

7.6.3 The impact of care setting in improving end of life care outcomes

Patients in the study compared to the study by reported similar scores in relation to capability wellbeing at the end of life. As suggested earlier, patients receiving care in a hospice setting were expected to achieve better end of life care outcomes given the exclusive focus on end of life wellbeing in a hospice setting. Although physical and psychological distress appeared to be better controlled in the hospice, capabilities associated with dignity were similar in both studies. Indeed, capabilities associated with end of life decision making were worse in the hospice study compared to this study irrespective of the setting from which patients were recruited from. However, the very small sample size and the single location from which patients were recruited from in both studies, severely limits the inferences that can be made from these findings. Investigating the extent to which care setting can improve outcomes important to patients may be an interesting area for future research, and can inform the transferability of optimal practices across settings to improve end of life care outcomes.

7.7 Conclusion

This thesis reports the first assessment of the ICECAP-SCM, the EQ-5D-5L and ICECAP-A in patients on the organ failure trajectory towards death. This thesis provides much needed evidence on the feasibility of using the ICECAP-SCM in patients at the end of life, adding to the only existing study of patients, primarily with cancer receiving care in a hospice (Bailey et al., 2016).

This research has shown that the ICECAP-SCM, the EQ-5D-5L and ICECAP-A are feasible to use in patients with end stage organ failure. The measures were found to be clearly understood and were not difficult to complete. While error rates differed across the measure, the differences were quite small. Furthermore, although a slightly higher number of participants preferred the ICECAP-SCM above other measures, most participants had no preference regarding the three measures.

This thesis has also identified and explored in greater depth, the impact of end stage organ failure and other long-term conditions on capability wellbeing and capabilities associated with the opportunity for a good death. Findings from the thesis underscores the importance of broader aspects of wellbeing to end of life decision making and the practical and policy significance of adaptation in relation to wellbeing (Dobríková et al., 2016) and outcome measurement (Schwartz et al., 2018).

This study contributes to existing gaps in the literature on the feasibility of measuring end of life care outcomes using economic and health outcome measures. A systematic review of the literature showed that research to date has not considered the feasibility of using economic measures in evaluating health and wellbeing among patients on the organ failure trajectory. Hence, this research provides evidence on the feasibility of using economic measures in evaluating health and wellbeing at the end of life, and provides greater insight into the practical and methodological concerns that may influence the feasibility of using such measures.

The goals of palliative care interventions and end of life policy currently emphasise a comprehensive approach aimed at improving outcomes that go beyond health. Hence, economic evaluation of palliative care services should include measures that are appropriate for capturing the broader benefits of end of life interventions. However, decision makers may also be interested in the impact of interventions on improving health outcomes for people near the end of life, and may choose to use measures that offer comparability across the health system such as the EQ-5D-5L. the ICECAP-SCM was developed specifically to evaluate capability wellbeing at the end of life, and there is still little research on its feasibility and other psychometric properties. Hence, the most appropriate use of the ICECAP-SCM currently, is alongside other available measures to support economic decision making among people near the end of life and to obtain further evidence on its psychometric properties.

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Appendix

Appendix 1 Data extraction form

Review title or ID
Henry Nwankwo

Study ID:

General Information

Study number	
Date form completed	
Name/ID of person extracting data	
Study title (title of paper/ abstract/ report that data are extracted from)	
Publication type	
Possible conflicts of interest (for study authors)	
Name of author	
Year of publication	

Eligibility

	Review Inclusion Criteria	Yes/ No / Unclear	Location in text
Type of study	Randomised trial/Non-RCT	...	
	Qualitative, Qualitative or Mixed Methods	...	
	Other design (specify):	...	
Participants	End Stage organ failure (e.g. end stage COPD, end stage HF, ESRD) Self-completed questionnaires	... ...	
Types of questionnaire:	Quality of life measures	...	
Addresses the feasibility of using measuring instruments:		...	
Decision:			

	Review Inclusion Criteria	Yes/ No / Unclear	Location in text
Reason for exclusion			

DO NOT PROCEED IF STUDY EXCLUDED FROM REVIEW

Population and setting

	Description	Location in text
Population description (from which study participants are drawn)		
Setting (including location and social context)		
Method/s of recruitment of participants		
Terminal diagnosis		

Methods

	Descriptions as stated in report/paper	Location in text
Aim(s)/Objective(s) of study		
Design		
Summary study objective		

Participants

Provide overall data and, if available, comparative data for each intervention or comparison group.

	Description as stated in report/paper	Location in text
Total number		
Age		
Sex		
Race/Ethnicity		

MEASURING INSTRUMENT

	Description as stated in a relevant research paper	Location in text
Instrument Name		
Target population		

Results

	Description as stated in report/paper	Location in text
Issues during completion		
Completion time		
Response rate		
Length and burden of questionnaire completion		
No of participants missing/unable to complete study with reasons		

Applicability

Have important populations been excluded from the study?	... Yes/No/Unclear	
Does the study directly address the review question? (any issues of partial or indirect applicability)	... Yes/No/Unclear	
Notes:		

Other information

	Description as stated in report/paper	Location in text (pg & ¶/fig/table)
Key conclusions of study authors		
References to other relevant studies		
Correspondence required for further study information (what and from whom)		
Further study information requested (from whom, what and when)		
Correspondence received (from whom, what and when)		

Appendix 2 Quality assessment form

Study Number			
Title			
Author			
Year			
	Yes	No	Can't tell
Was there a clear statement of the aims of the research?			
Was the methodology appropriate?			
Was the research design appropriate to address the aims of the research?			
Was the recruitment strategy appropriate to the aims of the research?			
Was data collected in a way that addressed the research issue?			
Has the relationship between researcher and participants been adequately considered?			
Have ethical issues been taken into consideration?			
Was data analysis sufficiently rigorous			
Is there a clear statement of findings?			
How valuable is the research?			

Appendix 3 Quality assessment summary of included studies

Title	Author/ Year	Was there a clear statement of the aims of the research?	Is a qualitative methodology appropriate?	Was the research design appropriate to address the aims of the research?	Was the recruitment strategy appropriate to the aims of the research?	Was the data collected in a way that addressed the research issue?	Has the relationship between researcher and participants been adequately considered?	Have ethical issues been taken into consideration?	Was the data analysis sufficiently rigorous?	Is there a clear statement of findings?
Development and initial validation of a new outcome measure for hospice and palliative care: the St Christopher's Index of Patient Priorities (SKIPP)	Addington-Hall 2014	Y	Y	Y	CT	Y	CT	Y	Y	Y
Hospice patients' participation in choice experiments to value supportive care outcomes	Bailey et al 2019	Y	Y	Y	Y	Y	CT	Y	Y	Y
"The ICECAP-SCM tells you more about what I'm going	Bailey et al 2016	Y	Y	Y	Y	Y	CT	Y	Y	Y

through': A think-aloud study measuring quality of life among patients receiving supportive and palliative care										
Validation and Clinical Application of the German Version of the Palliative Care Outcome Scale	Bausewein et al 2005	Y	Y	Y	Y	Y	CT	Y	Y	Y
The McGill Quality of Life Questionnaire: A measure of quality of life appropriate for people with advanced disease. A preliminary study of validity and acceptability	Cohen et al 1995	Y	Y	Y	Y	Y	CT	CT	CT	N
Development and content validity of a hemodi	Flythe et al 2019	Y	Y	Y	Y	Y	CT	Y	CT	Y

alysis sympto m patient-reported outcom e measure										
Validati on of a core outcom e measure for palliativ e care in Africa: the APCA African Palliativ e Outcom e Scale	Harding et al 2010	Y	Y	Y	Y	Y	CT	Y	N	Y
Develop ment and validati on of a core outcom e measure for palliativ e care: the palliativ e care outcom e scale. Palliativ e Care Core Audit Project Advisor y Group	Hearn et al 1999	Y	Y	Y	Y	Y	N	Y	N	Y
Discove ring the hidden benefits of cognitiv e intervie wing in two languag es: The first phase of	Iris et al 2016	Y	Y	Y	Y	Y	CT	Y	Y	Y

a validation study of the Integrated Palliative care Outcome Scale										
What does the answer mean? A qualitative study of how palliative cancer patients interpret and respond to the Edmonton Symptom Assessment System	Bergh et al 2011	Y	Y	Y	Y	Y	CT	Y	Y	Y
The feasibility, reliability and validity of the McGill Quality of Life Questionnaire-Cardiff Short Form (MQOL-CSF) in palliative care population	Lua et al 2005	Y	Y	Y	Y	Y	N	Y	CT	Y
The value of cognitive interviewing techniques in	Murtagh et al 2007	Y	Y	Y	Y	Y	CT	Y	Y	Y

palliative care research										
The applicability of quality-of-life assessment in palliative care: comparing two quality-of-life measures	Pratheepawanit, et al 1999	Y	Y	Y	Y	CT	CT	Y	Y	N
Assessing psychological and supportive care needs in glioma patients - feasibility study on the use of the Supportive Care Needs Survey Short Form (SCNS-SF34-G) and the Supportive Care Needs Survey Screening Tool (SCNS-ST9) in clinical practice	renovanz et al 2016	Y	Y	Y	Y	Y	CT	Y	Y	Y
Health-related quality of life and well-being in people over 75	shah et al 2019	Y	Y	Y	Y	N	N	Y	N	N

years of age with end-stage kidney disease managed with dialysis or comprehensive conservative care: a cross-sectional study in the UK and Australia										
Validation and application of a module of the M. D. Anderson Symptom Inventory for measuring multiple symptoms in patients with gastrointestinal cancer (the MDASI-GI)	Wang et al 2010	Y	Y	Y	Y	Y	N	Y	N	N
The Edmonton Symptom Assessment System (ESAS): what do patients think?	Wantanabe et al 2008	Y	Y	Y	Y	Y	Y	Y	Y	Y
Development of	Weis et al 2013	Y	Y	Y	Y	CT	CT	Y	Y	Y

an EORTC quality of life phase III module measuring cancer-related fatigue (EORTC QLQ-FA13)										
Exploring the Quality of Life at the End of Life (QUAL-E) Instrument with Australian Palliative Care Hospital Patients: Hurdles and directions	Wilkinson et al 2014	Y	Y	Y	Y	Y	CT	Y	Y	Y

Y-Yes
N-No
CT- Can't tell (Unclear)

Appendix 4 Characteristics of measures included in systematic review

Author/Year	Instrument name	Target population
Addington-Hall 2014	SKIPP and POS	Palliative patients
Bailey et al 2019	ICECAP-SCM	Palliative patients
Bailey et al 2016	ICECAP-SCM, EQ-5D-5L, ICECAP-O	Palliative patients
Bausewein et al 2005	POS	Palliative patients
Cohen et al 1995	MQOL	Palliative patients
Flythe et al 2019	SMaRRT-HD	ESRD patients
Harding et al 2010	APCA African POS	Palliative patients
Hearn et al 1999	POS	Palliative patients
Iris et al 2016	IPOS	Palliative patients
Bergh et al 2011	ESAS	Palliative patients
Lua et al 2005	MQOL-CSF	Palliative patients
Murtagh et al 2007	GDS POS MSAS-SF	Older and palliative patients
Pratheepawanit, et al 1999	MQOL, PEPS	palliative patients
renovanz et al 2016	SCNS-SF34-G and SCNS-ST9	palliative patients
shah et al 2019	KDQOL SF-6D and ICECAP-O	older adults, general adult population and disease specific measure for patients with kidney disease
Wang et al 2010	MDASI-GI	Cancer patients
Wantanabe et al 2008	ESAS	Cancer patients
Weis et al 2013	EORTC QLQ-C30	Cancer patients
Wilkinson et al 2014	QUAL-E	Palliative patients

Appendix 5 ICECAP-Supportive care measure

ID role [ ] number [ ] [ ]

ABOUT YOUR WELL-BEING

Please place a tick (✓) in ONE box in EACH group below, to indicate which statement best describes your situation at the moment.

1) Having a say – Your ability to influence where you would like to live or be cared for, the kind of treatment you receive, the people who care for you

I am able to make decisions that I need to make about my life and care **most of the time**	4
I am able make decisions that I need to make about my life and care **some of the time**	3
I am able to make decisions that I need to make about my life and care **only a little of the time**	2
I am **never** able to make decisions that I need to make about my life and care	1

2) Being with people who care about you – Being with family, friends or caring professionals

If I want to, I am able to be with people who care about me **most of the time**	4
If I want to, I am able to be with people who care about me **some of the time**	3
If I want to, I am able to be with people who care about me only a little of the time	2
If I want to, I am **never** able to be with people who care about me	1

3) Physical suffering – Experiencing pain or physical discomfort which interferes with your daily activities

I **always** experience significant physical discomfort	1
I **often** experience significant physical discomfort	2
I **sometimes** experience significant physical discomfort	3
I **rarely** experience significant physical discomfort	4

4) Emotional suffering – Experiencing worry or distress, feeling like a burden

I **always** experience emotional suffering	1

I **often** experience emotional suffering	2
I **sometimes** experience emotional suffering	3
I **rarely** experience emotional suffering	4

5) Dignity – Being treated with respect, being spoken to with respect, having your religious or spiritual beliefs respected, being able to be yourself, being clean, having privacy,

I am able to maintain my dignity and self-respect **most of the time**	4
I am able to maintain my dignity and self-respect **some of the time**	3
I am able to maintain my dignity and self-respect **only a little of the time**	2
I am **never** able to maintain my dignity and self-respect	1

6) Being supported – Having help and support

I am able to have the help and support that I need **most of the time**	4
I am able to have the help and support that I need **some of the time**	3
I am able to have the help and support that I need **only a little of the time**	2
I am **never** able to have the help and support that I need	1

7) Being prepared – Having financial affairs in order, resolving things that are important to you, having treatment preferences in writing or making a living will

I have had the opportunity to make **most** of the preparations I want to make	4
I have had the opportunity to make **some** of the preparations I want to make	3
I have had the opportunity to make a **few** of the preparations I want to make	2
I have **not** had the opportunity to make **any** of the preparations I want to make	1

Appendix 6 ICECAP-A

ABOUT YOUR OVERALL QUALITY OF LIFE

Please indicate which statements best describe your overall quality of life at the moment by placing a tick (✓) in **ONE** box for each of the five groups below.

1. Feeling settled and secure

I am able to feel settled and secure in **all** areas of my life	4
I am able to feel settled and secure in **many** areas of my life	3
I am able to feel settled and secure in **a few** areas of my life	2
I am **unable** to feel settled and secure in **any** areas of my life	1

2. Love, friendship and support

I can have **a lot** of love, friendship and support	4
I can have **quite a lot** of love, friendship and support	3
I have **a little** love, friendship and support	2
I **cannot** have **any** love, friendship and support	1

3. Being independent

I am able to be **completely** independent	4
I am able to be independent in **many** things	3
I am able to be independent in **a few** things	2
I am **unable** to be at all independent	

4. Achievement and progress

I can achieve and progress in **all** aspects of my life	4
I can achieve and progress in **many** aspects of my life	3
I can achieve and progress in **a few** aspects of my life	2
I **cannot** achieve and progress in **any** aspects of my life	1

5. Enjoyment and pleasure

I can have **a lot** of enjoyment and pleasure	4
I can have **quite a lot** of enjoyment and pleasure	3
I have **a little** enjoyment and pleasure	2
I **cannot** have **any** enjoyment and pleasure	1

Appendix 7 EQ-5D-5L

Under each heading, please tick the ONE box that best describes your health TODAY.

MOBILITY

I have no problems in walking about ❑

I have slight problems in walking about ❑

I have moderate problems in walking about ❑

I have severe problems in walking about ❑

I am unable to walk about ❑

SELF-CARE

I have no problems washing or dressing myself ❑

I have slight problems washing or dressing myself ❑

I have moderate problems washing or dressing myself ❑

I have severe problems washing or dressing myself ❑

I am unable to wash or dress myself ❑

USUAL ACTIVITIES *(e.g. work, study, housework, family or leisure activities)*

I have no problems doing my usual activities ❑

I have slight problems doing my usual activities ❑

I have moderate problems doing my usual activities ❑

I have severe problems doing my usual activities ❑

I am unable to do my usual activities ❑

PAIN / DISCOMFORT

I have no pain or discomfort ❑

I have slight pain or discomfort ❑

I have moderate pain or discomfort ❑

I have severe pain or discomfort ❑

I have extreme pain or discomfort ❑

ANXIETY / DEPRESSION

I am not anxious or depressed ❑

I am slightly anxious or depressed ❑

I am moderately anxious or depressed ❑

I am severely anxious or depressed ❑

I am extremely anxious or depressed ❑

Appendix 8 Information sheet

UNIVERSITY OF BIRMINGHAM

Evaluating care for patients with Organ Failure

Patient information sheet



Information about the research

You are invited to take part in a research study which aims to improve the way that quality of life is measured. This research is supported by the University of Birmingham. To help you decide if you wish to take part, this leaflet explains the purpose of the research and how you would be involved. A researcher will be available to go through this information sheet with you and answer any questions you may have. This is likely to take about five minutes.

What is the purpose of the study?



The aim of this study is to find out what you think about a number of questions designed to assess quality of life. This research aims to improve these questions.

Why have I been invited?

You have been invited to help with the research because you attend the heart failure clinic. We hope that about sixty (60) patients will take part in the study.



Do I have to take part?

No; it is entirely up to you, but your help would be much appreciated. A researcher will discuss the study with you and go through this information

sheet before you make a decision. You may wish to discuss this with your family or friends first. If you agree to take part, you will be asked to sign a consent form and the interview can be arranged at a time that is convenient for you.



You are free to withdraw from the study at any time and you do not need to explain why. Please be assured that if you withdraw it will in no way affect the standard of care you receive.

What will I have to do?

First you will be asked a few introductory questions. You will then be asked to complete three short questionnaires with a total of 17 questions, talking through your answers. This interview will be recorded to make sure your views are accurately reported. The interview is expected to last about 30 minutes.



What are the possible disadvantages of taking part?

It is possible that some people may find some of the questions difficult to answer and might find talking about their health upsetting. If this happens, you are free to stop the interview at any time and will be given any additional support you may need.

What are the possible benefits of taking part



Your views as a patient will provide a very valuable contribution to our research. Your particaption will help in designing more appropriate questionnaires for furtehr research studies. The study will also try to find out if the questions are useful in assessing your wellbeing.

What will happen if I don't want to carry on with the study?

You are free to withdraw from the interview at any time and your recording can be deleted, if you wish.



What if there is a problem?

This study has been reviewed by the NHS Ethics Committee. If you have any concern about any aspect of this study, please feel free to speak to the researcher Henry Nwankwo (Tel.) or Dr Phil Kinghorn (Tel.). If you remain unhappy and wish to complain formally, you can send a written complaint to and address it to Cardiology Unit,

At any point, if you have further questions or complaints regarding any aspect of the research, the Patient Advice and Liason Service (PALS) offers confidential advice and information which you may find useful. They can be found at: . Tel: .

Will my taking part in this study be kept confidential?

If you agree to take part and have the interview recorded, I will use a non-personal code to identify the recording so that you cannot be recognised. Any names or places you mention during the interview will be anonymised. The questionnaire you complete will be identified by a study number.

However, in the event of a disclosure of malpractice or abuse during the interview, this will be brought to the attention of health professionals involved in the research (who are separate from those professionals providing care to you at the clinic), and their advice followed.

Involvement of the GP

We will send a letter to your GP to inform them of your participation in this research.

What will happen to my personal data and audio recordings?

An encrypted password protected digital recorder will be used for the interview and interviews will be downloaded to secure University servers at the first opportunity and deleted from the portable digital recorder. Your personal data will only be accessible to members of the research team. At the end of the study, personal data collected for the study will be destroyed in line with the Data Protection Act. The anonymised research data collected

for the study will be stored for ten years in line with the University of Birmingham policy"

What will happen to the results of the research study?

The results will be useful in helping to develop better questionnaires. A summary of the results will be provided to the cardiology unit. As a participant in the research you and your family would be entitled to get the results of the study.

In developing a measure of quality of life, it is important to ask people themselves what they think and not just rely on the views of professionals or the government.

Who is organising and funding the research?

This study is being carried out by the University of Birmingham and is funded through the College of Medical and Dental Sciences.

Who has reviewed the study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given a favourable opinion by North Wales Research Ethics Committee.

Further information and contact details

General information about this research can be found on: http://www.icecap.bham.ac.uk/

Specific information about this research project can be provided by Henry Nwankwo (Researcher: [redacted] or tel. [redacted]) or Dr Phil Kinghorn [redacted]

If you are unhappy with the study you should approach either Henry or Phil (see above for details) or for a formal complaint contact the cardiology unit at [redacted] (see address above).

Following the interview you may find that there are issues that have been discussed about which you would like further support. If this is the case, you can talk to staff in the clinic where further services can be offerred. Or you may find the following national resources helpful:



CRUSE Bereavement Care (Daytime helpline 084 4477 9400 or email: helpline@cruse.org.uk).

Age UK: www.AgeUK.org.uk or tel. 0121 437 0033

If you decide to participate you will have a copy of this information sheet and a signed consent form to keep.

Appendix 9 Topic guide

Evaluating care for patients with Organ Failure

Aim of think-aloud interviews

To explore through think-aloud interviewing, the nature of any completion problems people have with ICECAP-SCM and to compare these with the ICECAP-A and EQ-5D-5L

1. INTRODUCTION

Take Consent

- Explain the aims of study

Thanks for agreeing to take part in this interview. The interview today is part of a larger study that I am involved in at the University of Birmingham, about measuring quality of life in patients with renal failure, chronic obstructive pulmonary disease and heart failure. The aim of the work is to develop questionnaires that could be used to give us information we need when choosing which forms of care are best for patients.

This particular interview will find out how valid the questions are in evaluating patient's quality of life in Chronic Obstructive Pulmonary Disease/ renal failure/ heart failure (Depending on the site the patient is recruited from).

- Explain what is going to be asked about in this interview

In this interview I am going to ask you to "think aloud" as you complete three very short questionnaires with a total of 17 questions. By thinking aloud I ask you to talk through your thought process as you answer the survey question.

I want you to tell me everything you are thinking from the time you read out each question until you have given your final answer. I don't want you to plan what you are going to say, just act as though you are alone in the room speaking to yourself. There are no right or wrong answers. I will start with a couple of example questions.

To start, though, I am going to ask a few basic questions, about *yourself and your family* / *yourself and [patient's name]*.

- Check ok to have interview recorded

Can I just check that you are happy to have the interview recorded? This will help me to record your views accurately.

- Reminder about anonymity and withdrawing from study

Just before I start I would like to remind you that although quotes may be used from your interview when I write up the findings I will not include your name or any details that

could identify you. Finally, you are free to stop the interview at any time and withdraw. I will destroy the recording, if that is what you wish.

2. DEMOGRAPHICS & BACKGROUND INFORMATION

- **Patient**

OK, as I mentioned, I want to start by asking a few basic questions about you and about your family...

- **What is your age?**
- **What is your religion?**
- **How would you describe your ethnicity?**
- **Are you married or do you currently have a partner?**

Thank you. Now we will move on to the main task.

3. WARM-UP

- Counting exercise:

The process of thinking aloud can be unfamiliar, so I would like to start you off with a warm-up practice task.

<u>For patients:</u>

For the first task I will ask you to think aloud while you are counting how many times you have had contact with health professionals over the past week (or how many windows are in your home)

This sort of question has been used before to get people familiar with the process of thinking aloud. Only tell me what comes into your mind. I won't interrupt you unless you are silent for more than ten seconds. Ok, now please count up how many times you have

had contact with a health professional today (how many windows are in your home) and think aloud as you go.

Feedback/ encourage/ example.

***If unable to think-aloud, give them an example of think aloud.**

Good! Now onto the main task.

SECTIONS 4 AND 5 ALLOCATED TO EQ-5D AND ICECAP-SCM IN RANDOM ALLOCATION

4. QUESTIONNAIRE 1

In a moment I will give you a copy of the first questionnaire. As explained at the beginning I would like you to complete this questionnaire, thinking aloud while you complete it. If you wish, I will sit outside your line of sight while you do this so as not to distract you. If not, I will sit quietly with you while answer the questions. Just to recap, what I mean by 'Think Aloud' is that I want you to tell me everything you are thinking from the time you read out each question until you have given your final answer. I don't want you to plan what you are going to say, just act as though you are alone in the room speaking to yourself. I would like you to think aloud constantly. If you are silent for any long period of time I will prompt you to keep talking. Please try and speak as clearly as possible, as I shall be recording what you say. Don't worry about hurting my feelings if you want to criticize any of the questions. My job is to find out if there are any problems with the questions. There are no right or wrong answers. I will now give you the questionnaire and would like you to start completing it, in your own time, thinking aloud as you go.

Thank you.

5. QUESTIONNAIRE 2

For the second task I would like you to do the do the same with this second questionnaire I am about to give you.

~REMIND IF NECESSARY ABOUT~

- ***Thinking aloud from beginning to end.***
- ***Speaking as clearly as possible.***
- ***There are no right or wrong answers.***
- ***No need to plan, rush, or go any slower than normal.***

6. QUESTIONNAIRE 3

For the third task I would like you to do the do the same with this third questionnaire I am about to give you.

~REMIND IF NECESSARY ABOUT~

- ***Thinking aloud from beginning to end.***
- ***Speaking as clearly as possible.***
- ***There are no right or wrong answers.***
- ***No need to plan, rush, or go any slower than normal.***

7. CLARIFYING DISCUSSION

That is the end of the think-aloud exercise. For the final section of the interview I would, just briefly, like to discuss with you how you found completing the questionnaires.

- **You seemed a bit unclear about the instructions. Was this confusing?**

- **You seemed a bit unsure about which box to tick on [question] Can you say why that was?**

- **You seemed a bit unsure by what was meant by the term [term]**
  - **What do you understand by that word?**
  - **How did you interpret the word in your answer?**
  - **How did that affect how you answered the question?**

8. SEMI-STRUCTURED INTERVIEW

Thank you. Just before the end I would like to ask you some questions. First, I would ask you questions about your recent health state. Second, I would like to ask you some questions on your opinion of the questionnaire.

Questions about health state

- **How would you describe your current health state in the last six months?**
- **When was the last time you required admission as a result of your condition?**

Questions on the questionnaires

- **Did you find any questions off-putting?**
- **Did you find any questions confusing?**
- **How clear did you find the instructions?**
- **How easy or difficult did you find answering the questionnaires overall?**
- **Did you find that answering the [1st measure] first affected your answers to the 2nd measure?**
- **Do you have any further comments about either questionnaire in terms of their**

ability to record your health and general quality of life?

9. INTERVIEW END

Thank you. That is the end of the interview. I hope you found it ok.

I will be writing a short summary of the findings for participants: would you be interested in receiving a copy?
Give them GP letter.
Greeting and leave.

UNIVERSITY OF BIRMINGHAM

Evaluating care for patients with Organ Failure

Patient consent form

Clinic: .....

Patient Identification Number for this Study: .....

Title of Project: Evaluating care for patients with Organ Failure

Name of Researcher: Henry Nwankwo

Please initial the box

1. I confirm that I have read and understood the information sheet dated 05 March 2017 (version 1.3) 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 my participation is voluntary, I am free to withdraw at any time without giving any reason and without my care or legal rights being affected. ☐
3. I understand that data collected during the study, may be looked at by the study Research Team at the University of Birmingham. ☐
4. I agree to having the interview recorded. ☐
5. I agree for the use of anonymised quotations in publications. ☐
6. I agree to take part in the above study. ☐
7. I agree to my General Practitioner being informed of my participation in the study. ☐
8. I understand that information on malpractice or abuse that may arise during the study will be disclosed to relevant professionals. ☐

Name of Patient	Date	Signature

Name of person taking consent	Date	Signature

When completed: 1 for participant, 1 for researcher site file, 1 to be kept in medical notes.

Appendix 11 Sponsorship and ethical approval letters

UNIVERSITY OF BIRMINGHAM

FINANCE OFFICE

To Whom It May Concern,

5th December 2016

Dear Sir / Madam

Project Title: **Evaluating care for patients with organ failure**

Sponsor Reference: **RG_16-173**

The University currently arranges Clinical Research Insurance through membership of UMAL, this cover is renewable annually on 1st August and the University ensures that there are no gaps in cover.

We confirm that the University will arrange insurance for the above Research Trial for the duration of the Research Project involving human participants including legal liability cover. This will be subject to the terms, conditions and exceptions of the relevant liability insurance policies as detailed in the current trial documentation.

The University will provide notification if there are any changes to the provider in a particular year or if the indemnity limits change. Copies of the trial specific annual confirmatory letters will be available upon request.



Dr Sean Jennings
Research Ethics and Governance Manager
Research Support Group

University of Birmingham Edgbaston Birmingham B15 2TT United Kingdom
w: www.finance.bham.ac.uk

NHS
Health Research Authority

Mr Henry C Nwankwo
University of Birmingham
Institute of Clinical Sciences College of Medical and Dental Sciences
The Medical School
B15 2TT

Email: hra.approval@nhs.net

08 March 2017

Dear Mr Nwankwo

Letter of HRA Approval

Study title:	**A think-aloud study of the feasibility of economic measures of end of life care in the organ failure trajectory and associated care settings, and the impact on the opportunity for a good death.**
IRAS project ID:	**212259**
Protocol number:	**RG_16-173**
REC reference:	**17/WA/0022**
Sponsor	**University of Birmingham**

I am pleased to confirm that **HRA Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications noted in this letter.

Participation of NHS Organisations in England

The sponsor should now provide a copy of this letter to all participating NHS organisations in England.

Appendix B provides important information for sponsors and participating NHS organisations in England for arranging and confirming capacity and capability. **Please read *Appendix B* carefully**, in particular the following sections:

- *Participating NHS organisations in England* – this clarifies the types of participating organisations in the study and whether or not all organisations will be undertaking the same activities
- *Confirmation of capacity and capability* - this confirms whether or not each type of participating NHS organisation in England is expected to give formal confirmation of capacity and capability. Where formal confirmation is not expected, the section also provides details on the time limit given to participating organisations to opt out of the study, or request additional time, before their participation is assumed.
- *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* - this provides detail on the form of agreement to be used in the study to confirm capacity and capability, where applicable.

Further information on funding, HR processes, and compliance with HRA criteria and standards is also provided.

It is critical that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details and further information about working with the research management function for each organisation can be accessed from www.hra.nhs.uk/hra-approval.

Appendices

The HRA Approval letter contains the following appendices:

- A – List of documents reviewed during HRA assessment
- B – Summary of HRA assessment

After HRA Approval

The 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.

In addition to the guidance in the above, please note the following:

- HRA Approval applies for the duration of your REC favourable opinion, unless otherwise notified in writing by the HRA.
- Substantial amendments should be submitted directly to the Research Ethics Committee, as detailed in the *After Ethical Review* document. Non-substantial amendments should be submitted for review by the HRA using the form provided on the HRA website, and emailed to hra.amendments@nhs.net.
- The HRA will categorise amendments (substantial and non-substantial) and issue confirmation of continued HRA Approval. Further details can be found on the HRA website.

Scope

HRA Approval provides an approval for research involving patients or staff in NHS organisations in England.

If your study involves NHS organisations in other countries in the UK, please contact the relevant national coordinating functions for support and advice. Further information can be found at http://www.hra.nhs.uk/resources/applying-for-reviews/nhs-hsc-rd-review/.

If there are participating non-NHS organisations, local agreement should be obtained in accordance with the procedures of the local participating non-NHS organisation.

IRAS project ID	212259

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please email the HRA at hra.approval@nhs.net. Additionally, one of our staff would be happy to call and discuss your experience of HRA Approval.

HRA Training

We are pleased to welcome researchers and research management staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

Your IRAS project ID is **212259**. Please quote this on all correspondence.

Yours sincerely

Miss Helen Penistone
Assessor

Email: hra.approval@nhs.net

Copy to: *Dr Sean Jennings, University of Birmingham*
Ms Helen Langston, University Hospitals Birmingham

Appendix A - List of Documents

The final document set assessed and approved by HRA Approval is listed below.

Document	*Version*	*Date*
Contract/Study Agreement [Sponsorship agreement]		05 December 2016
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Sponsor indemnity]		18 July 2016
GP/consultant information sheets or letters [GP Letter]	1.0	10 October 2016
Interview schedules or topic guides for participants [Topic guide]	1.1	18 November 2016
IRAS Application Form [IRAS_Form_20122016]		20 December 2016
Letter from sponsor [Confirmation of Insurance]		05 December 2016
Non-validated questionnaire [ICECAP-SCM]	1.2	03 February 2012
Non-validated questionnaire [ICECAP-A]	2	01 January 2010
Non-validated questionnaire [ICECAP-SCM Adjusted version]	1.2	03 February 2012
Participant consent form	1.3	05 March 2017
Participant information sheet (PIS) [Cardiac]	1.3	05 March 2017
Participant information sheet (PIS) [Renal]	1.3	05 March 2017
Participant information sheet (PIS) [Respiratory]	1.3	05 March 2017
Research protocol or project proposal	1.3	15 February 2017
Response to Request for Further Information		
Summary CV for Chief Investigator (CI) [Curriculum Vitae]		
Summary CV for supervisor (student research) [Fiona Irvine CV]		
Summary CV for supervisor (student research) [Joanna Coast CV]		
Summary CV for supervisor (student research) [Cara Bailey CV]		
Summary CV for supervisor (student research) [Philip Kinghorn CV]		
Validated questionnaire [EQ-5D-5L Questionnaire]		

IRAS project ID	212259

Appendix B - Summary of HRA Assessment

This appendix provides assurance to you, the sponsor and the NHS in England that the study, as reviewed for HRA Approval, is compliant with relevant standards. It also provides information and clarification, where appropriate, to participating NHS organisations in England to assist in assessing and arranging capacity and capability.

For information on how the sponsor should be working with participating NHS organisations in England, please refer to the, *participating NHS organisations, capacity and capability* and *Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria)* sections in this appendix.

The following person is the sponsor contact for the purpose of addressing participating organisation questions relating to the study:

Name: Mr Henry Nwankwo

Tel: 01214142286

Email: hcn486@bham.ac.uk

HRA assessment criteria

Section	HRA Assessment Criteria	Compliant with Standards	Comments
1.1	IRAS application completed correctly	Yes	No comments
2.1	Participant information/consent documents and consent process	Yes	Non-substantial amendments were made to the Participant Information Sheets and Consent Form following REC favourable opinion to clarify the use of data.
3.1	Protocol assessment	Yes	Non-substantial amendments were made to the study protocol following REC favourable opinion.
4.1	Allocation of responsibilities and rights are agreed and documented	Yes	This is a single site study taking place in the NHS where the single site and study sponsor have an existing joint research arrangement. Therefore, no agreement is expected.

Section	HRA Assessment Criteria	Compliant with Standards	Comments
4.2	Insurance/indemnity arrangements assessed	Yes	Where applicable, independent contractors (e.g. General Practitioners) should ensure that the professional indemnity provided by their medical defence organisation covers the activities expected of them for this research study
4.3	Financial arrangements assessed	Yes	No external funding will be available to support this study at site.
5.1	Compliance with the Data Protection Act and data security issues assessed	Yes	No comments
5.2	CTIMPS – Arrangements for compliance with the Clinical Trials Regulations assessed	Not Applicable	No comments
5.3	Compliance with any applicable laws or regulations	Yes	No comments
6.1	NHS Research Ethics Committee favourable opinion received for applicable studies	Yes	No comments
6.2	CTIMPS – Clinical Trials Authorisation (CTA) letter received	Not Applicable	No comments
6.3	Devices – MHRA notice of no objection received	Not Applicable	No comments
6.4	Other regulatory approvals and authorisations received	Not Applicable	No comments

Participating NHS Organisations in England

This provides detail on the types of participating NHS organisations in the study and a statement as to whether the activities at all organisations are the same or different.
This is a single site study. Participants will be recruited at site but may complete the research activities either at site or in their own home depending on which is most convenient. If this study is subsequently extended to other NHS organisation(s) in England, an amendment

should be submitted to the HRA, with a Statement of Activities and Schedule of Events for the newly participating NHS organisation(s) in England.

The Chief Investigator or sponsor should share relevant study documents with participating NHS organisations in England in order to put arrangements in place to deliver the study. The documents should be sent to both the local study team, where applicable, and the office providing the research management function at the participating organisation. For NIHR CRN Portfolio studies, the Local LCRN contact should also be copied into this correspondence. For further guidance on working with participating NHS organisations please see the HRA website.

If chief investigators, sponsors or principal investigators are asked to complete site level forms for participating NHS organisations in England which are not provided in IRAS or on the HRA website, the chief investigator, sponsor or principal investigator should notify the HRA immediately at hra.approval@nhs.net. The HRA will work with these organisations to achieve a consistent approach to information provision.

Confirmation of Capacity and Capability

This describes whether formal confirmation of capacity and capability is expected from participating NHS organisations in England.
This is a single site study where the sponsor and site have an existing joint research arrangement. The R&D office will confirm to the CI when the study can start.

Principal Investigator Suitability

This confirms whether the sponsor position on whether a PI, LC or neither should be in place is correct for each type of participating NHS organisation in England and the minimum expectations for education, training and experience that PIs should meet (where applicable).
It is expected that there will be a local collaborator at site to facilitate the access of externally employed researchers. The sponsor should clarify any training expectations they have of members of the research team. GCP training is not a generic training expectation, in line with the HRA statement on training expectations.

HR Good Practice Resource Pack Expectations

This confirms the HR Good Practice Resource Pack expectations for the study and the pre-engagement checks that should and should not be undertaken
Where arrangements are not already in place, externally employed researchers requiring access to site will be expected to obtain a Letter of Access based on standard DBS and occupational health checks.

IRAS project ID	212259

Other Information to Aid Study Set-up

This details any other information that may be helpful to sponsors and participating NHS organisations in England to aid study set-up.
The applicant has indicated that they intend to apply for inclusion on the NIHR CRN Portfolio.

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

Gwasanaeth Moeseg Ymchwil
Research Ethics Service

Ariennir gan **Lywodraeth Cymru**
Funded by **Welsh Government**

Pwyllgor Moeseg Ymchwil Cymru 5
Wales Research Ethics Committee 5
Bangor

Clinical Academic Office
Ysbyty Gwynedd Hospital
Betsi Cadwaladr University Health Board
Bangor, Gwynedd
LL57 2PW

Website : www.nres.nhs.uk

Please note:
This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

06 February 2017

Mr Henry C Nwankwo
University of Birmingham
Institute of Clinical Sciences College of Medical and Dental Sciences
The Medical School
B15 2TT

Dear Mr Nwankwo

Study title:	**A think-aloud study of the feasibility of economic measures of end of life care in the organ failure trajectory and associated care settings, and the impact on the opportunity for a good death.**
REC reference:	**17/WA/0022**
Protocol number:	**RG_16-173**
IRAS project ID:	**212259**

Thank you for your letter of 31 January 2017, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact hra.studyregistration@nhs.net outlining the reasons for your request.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).
Guidance on applying for NHS permission for research is available in the Integrated Research Application System, www.hra.nhs.uk or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publically accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion").

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	*Version*	*Date*
IRAS Application Form [IRAS_Form_20122016]	-	20 December 2016
Research protocol or project proposal	1.2	23 January 2017
Participant consent form [Consent form]	1.2	23 January 2017
Participant information sheet (PIS) [Cardiac Version]	1.2	23 January 2017
Participant information sheet (PIS) [Renal Version]	1.2	23 January 2017
Participant information sheet (PIS) [Respiratory Version]	1.2	23 January 2017
Interview schedules or topic guides for participants [Topic guide]	1.1	18 November 2016
GP/consultant information sheets or letters [GP Letter]	1.0	10 October 2016
Non-validated questionnaire [ICECAP-SCM]	1.2	03 February 2012
Non-validated questionnaire [ICECAP-A]	2	01 January 2010
Non-validated questionnaire [ICECAP-SCM Adjusted version]	1.2	03 February 2012
Validated questionnaire [EQ-5D-5L Questionnaire]	-	-
Non-validated questionnaire [ICECAP-SCM]	1.2	03 February 2012
Non-validated questionnaire [ICECAP-A]	2	01 January 2010
Non-validated questionnaire [ICECAP-SCM Adjusted version]	1.2	03 February 2012
Summary CV for Chief Investigator (CI) [Curriculum Vitae]	-	-
Summary CV for supervisor (student research) [Fiona Irvine CV]	-	-
Summary CV for supervisor (student research) [Joanna Coast CV]	-	-
Summary CV for supervisor (student research) [Cara Bailey CV]	-	-
Summary CV for supervisor (student research) [Philip Kinghorn CV]	-	-
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Sponsor indemnity]	-	18 July 2016
Letter from sponsor [Confirmation of Insurance]	-	05 December 2016
Response to Request for Further Information	-	-

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at http://www.hra.nhs.uk/hra-training/

17/WA/0022	**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project.

Yours sincerely

[redacted]

Dr Philip Wayman White, MBChB, MRSM
Chair, Wales REC 5

E-mail: rossela.roberts@wales.nhs.uk

Enclosures: *"After ethical review – guidance for researchers"*

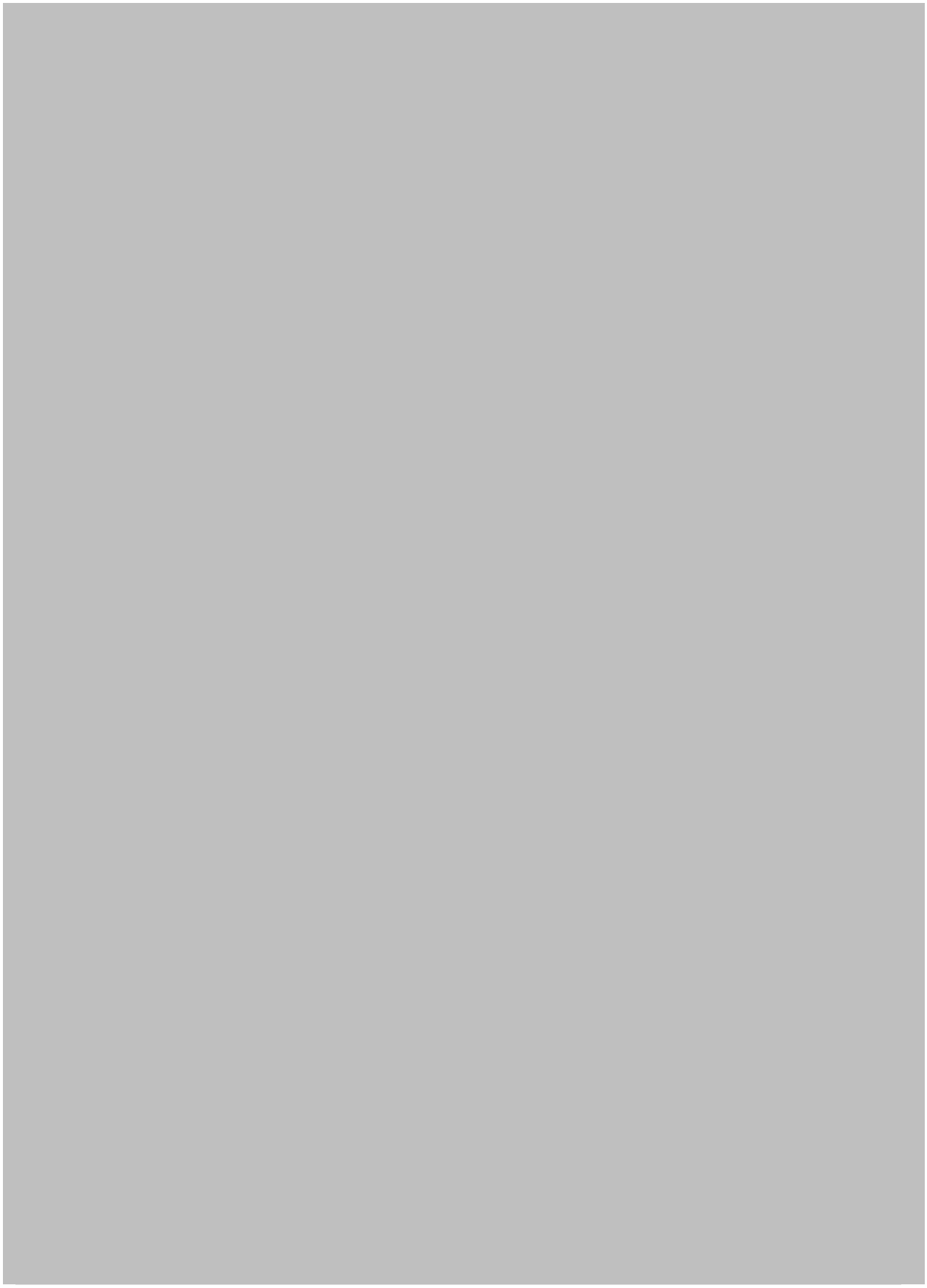


SL-AR2 After ethical review - research oth

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R&D Office: Ms Helen Langston
University Hospitals Birmingham
[redacted]

Academic Supervisor: Professor FE Irvine
University of Birmingham
[redacted]



Appendix 12 Standardised classification scheme for error rating

ID	X	X	X

Measure?	4	3	3	3	4

NB:
4=best
1=worst

Rater	HN1	JC2	PK3	AH4	CB5

No	Item	(a) Comp	(b) Retrieval	(c) Judge	(d) Response	(e) Struggle	(h) Comments
B1	Attachment (love and friendship)						
B2	Security (thinking about the future without concern)						
B3	Role (doing things that make you feel valued)						
B4	Enjoyment (enjoyment and pleasure)						
B5	Control (independence)						

Comprehension
any misunderstanding of a word, phrase, or response option
any words or phrase that the participant does not understand.
Retrieval
a recall error or a miscalculation of the time frame stated in the question,
Judgement
the participants response does not match that of the investigators intent for the question item,

any recalled relevant experiences that the participant questions as irrelevant or inadequate,
Response
participants desired response is missing from the written survey response choices,
any response which is felt to be socially desirable answer
participants response is inconsistent with the personal experience expressed
Participant's answer is inconsistent with previous answers
Struggle
the participant has had difficulty answering the question, even when their final response is correct.

	Error	Not error
Comprehension	Love? I'm not sure what that means. [And no box ticked]	Love? I'm not sure what that means. I will assume it's about having my family around. [Box ticked]
Retrieval	Love? Well, I had a lot of love a few years ago. It's all changed now, but I think I will go with "a lot of love".	Love? Well, I had a lot of love a few years ago, it's hard to say now, but I think I will go for that box.
Judgement	Love? Well it's asking about how much love I can have, which isn't much… from people anyway… but I love lots of things, I love going to the cinema, so I will say "a lot of love".	Love? Well I love lots of things and can get lots of love. I love going to the cinema… I will say "a lot of love".
Response	Well I would say I can have some love and friendship, only that option is missing [No box ticked, tick in between level 2 and level 3] Well I would say I can have a lot of love and friendship [where responses earlier indicate that this is clearly untrue, or being said because informant perceives this to be socially desirable]	Well I would say I can have some love and friendship…I guess that's closest to "quite a lot", so I will tick that box [Box ticked] Well I would normally say I can have a lot of love and friendship, because you wouldn't normally want to own up to not being able to [response to measure is NOT 'a lot of love and friendship']]
Struggle	Love? It's just such a difficult concept…can I have a lot? What does "can" mean – I don't know! I'm just going to go with this answer, but I find	Love? It's a difficult concept…can I have a lot? What does "can" mean? I can have support in my life that's fine. [Answer ticked]

	it a confusing question. [Answer ticked]	

Error key:
C = Comprehension

T = Retrieval

J = Judgement
R = Response

S = Struggle

X? - possible error
Decisions:
Majority principle requiring >2 rating it as an error to count. Soft rates (i.e. with ? count as half). Errors confirmed if for example, 3 firm rates, 2 firm rates & one soft rate, 1 firm rate and 3 soft rates

When 2 rates - group decision
When <2 rates for specific error, but >2 for an error of some sort - group decision