Critically examining the end of life care of people with interstitial lung disease: views of patients, families and healthcare professionals.

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Abstract

Background

Interstitial lung diseases (ILDs) are a heterogeneous group of conditions causing inflammation and fibrosis of the lung parenchyma. Progressive fibrotic ILD is characterised by the development of irreversible lung fibrosis, causing progressive respiratory failure, which is associated with a poor prognosis.

People with ILD experience a high symptom burden and many experience poorly controlled symptoms at the end-of-life. There is a paucity of research considering the end of life experience of people with ILD.

Aims

This research study aimed to explore the experience of end of life for people with ILD and examine potential barriers to accessing palliative care services.

Methods

Semi-structured interviews were conducted with people with ILD (n=9), bereaved relatives (n=9) and healthcare professionals (n=12). Constructivist grounded theory methodology was employed for data collection and analysis.

Findings

Four overarching categories were constructed from the research data: i) acknowledging uncertainty, ii) accessing and organising support, iii) avoiding discussion about an uncertain future, iv) accelerating symptoms at the end of life.

Rapid deterioration of symptoms at the end of life influenced symptom control, presence of family and location of death. People with ILD prioritised a peaceful death over the location of death; central to this concept were well-controlled symptoms and that death was anticipated.

Abstract

Patient associated barriers preventing access to specialist palliative care services included prognostic uncertainty, misconceptions about the role of palliative care and avoiding advance care planning conversations. Healthcare professional related barriers included prognostic uncertainty, time limitations, remote consultations, prioritisation of malignant disease, geographical disparity and scarce community palliative care resources.

Conclusions

These findings highlight the requirement for increased explanation of prognostic uncertainty and acknowledgement that symptoms may deteriorate rapidly, meaning that end of life plans should focus on ensuring a peaceful death rather than on location of death.

Declarations

The following poster, oral presentation and publications have been produced from this thesis:

Palmer E, Bourke A, Visram S, et al 32 Critically examining the end of life care for people with interstitial lung disease: views of patients, families and healthcare professionals. *BMJ Supportive & Palliative Care* 2022; **12**: A13

Palmer E, Visram S, Bourke A, et al 0-6 Critically examining the end of life care for people with interstitial lung disease: views of patients, families and healthcare professionals. *BMJ Supportive & Palliative Care* 2022;**12**: A2-A3

Palmer E, Kavanagh E, Visram S, Bourke A-M, Forrest I, Exley C. Which factors influence the quality of end of life care in interstitial lung disease? A systematic review with narrative synthesis. *Palliative Medicine*. 2022;36(2):237-253.

Palmer E, Kavanagh E, Visram S, Bourke A-M, Forrest I, Exley C. When should palliative care be introduced for people with progressive fibrotic interstitial lung disease? A meta-ethnography of the experiences of people with end-stage interstitial lung disease and their family carers. *Palliative Medicine*. 2022;36(8):1171-1185.

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I dedicate this thesis to my family. To my wonderful husband Tom, for your unwavering love, support and technical IT abilities. To my children, Will and Layla, who disrupted my writing and thought processes on multiple occasions; you make all my days brighter.

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Chapter 1 Introduction

People with progressive fibrotic interstitial lung disease (ILD) have a poor prognosis, high symptom burden and complex palliative care needs. The majority of people with ILD die in an acute setting, however, very little is known about the quality of their end of life care. This thesis reports on a constructivist grounded theory study that explored the experience of end of life care from the perspectives of people with ILD, bereaved relatives and healthcare professionals. The principal aim of the study was to develop a comprehensive understanding of the end of life care experience for people with ILD to identify areas for reform and change in practice to improve patient care.

This chapter introduces the study by first discussing the different types of ILD and treatment approaches, followed by outlining the palliative care definitions used in this thesis. The rationale for the study is explained. This chapter ends with an outline of the structure and organisation of this thesis.

1.1 Interstitial lung disease

1.1.1 Classification of interstitial lung disease

Interstitial lung diseases are a heterogenous group including over 200 lung conditions characterised by inflammation and fibrosis of the lung parenchyma (Travis, Costabel et al. 2013, BLF 2019). See Figure 1-1 for a summary. The early stages of disease management differ between the forms of ILD and therefore establishing the correct diagnosis is paramount.

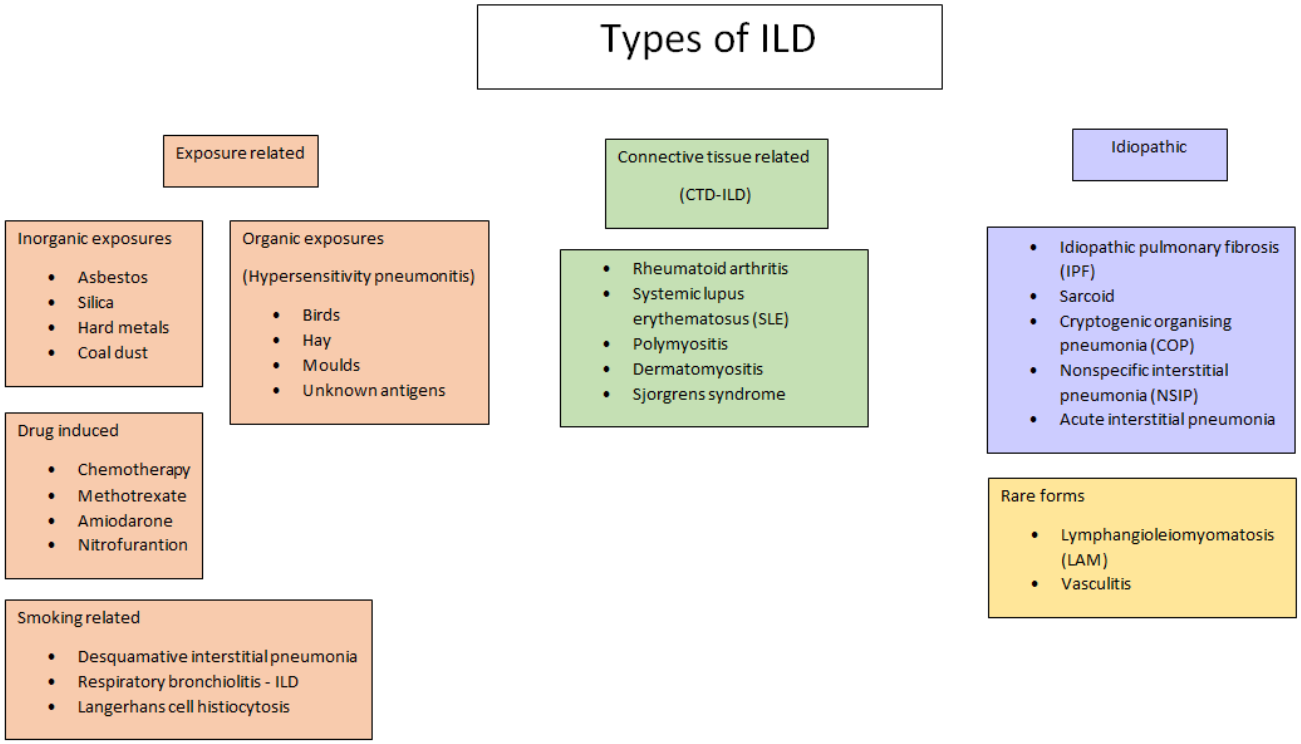

Figure 1-1 Types of interstitial lung disease

Idiopathic pulmonary fibrosis (IPF) is the most common form of ILD with 6,000 people diagnosed each year in the UK (BLF 2019). IPF causes progressive respiratory failure and has a prognosis that is worse than many cancers; the median survival in the UK is 3 years from diagnosis (NICE 2017). IPF is predominantly a fibrotic disease, whereas in other forms of ILD fibrosis is proceeded by inflammation. However, irreversible fibrosis and respiratory failure develop in the final stages of many forms of ILD. This progressive fibrosis has been reported to share similar biological and clinical features and therefore has been collectively termed progressive fibrotic ILD (Wuyts, Camerlinck et al. 2021). It is estimated that 20-30% of patients with ILD have progressive fibrotic ILD (Zamora-Legoff, Krause et al. 2017, Reiseter, Gunnarsson et al. 2018). Several criteria have been used to define progression in ILD; objective lung function measurements of decline in forced vital capacity (FVC) or gas transfer (TLCO), radiological progression seen on CT imaging, worsening symptoms or deterioration in quality of life scores (Noble, Albera et al. 2011, Solomon, Chung et al. 2016, Goh, Hoyles et al. 2017, Wells, Flaherty et al. 2020). However, there remains no universally agreed definition of progressive fibrotic ILD amongst respiratory clinicians or academics.

1.1.2 Symptoms of interstitial lung disease

The principal symptoms common to all types of progressive fibrotic ILD are breathlessness and cough which worsen with disease progression. A systematic review reported the most prevalent symptoms in IPF as breathlessness (54-98%), cough (59-100%), heartburn (25-65%) and depression (10-49%) (Carvajalino, Reigada et al. 2018). Alongside these cardinal symptoms, patients with ILD experience problems with sleep which contributes to extreme fatigue reported by approximately a third of patients (Schoenheit, Becattelli et al. 2011, Bajwah, Higginson et al. 2012, Bajwah, Higginson et al. 2013).

1.1.3 The role of the regional ILD service

The regional ILD service is based at a tertiary hospital in Newcastle upon Tyne. Patients with ILD are referred to this service from hospitals throughout the North East and Cumbria. Patients are initially discussed at the regional multi-disciplinary team meeting (MDT) which includes specialists in ILD, thoracic radiology, histology and occupational lung disease. The purpose of this MDT is to ratify diagnoses, monitor disease progression and plan treatment. Patients may then be reviewed in the regional ILD clinic or remain under follow-up with their local respiratory team. Those patients who are reviewed in the regional ILD clinic continue under shared-care with their local respiratory team. It is important to note that the regional ILD centre is the only hospital in the North East and Cumbria which is allowed to prescribe anti-fibrotic medications (Pirfenidone and Nintedanib) for patients with ILD. This means that anyone prescribed these medications must be under the care of the regional ILD service based in Newcastle.

1.1.4 Treatment strategies

Treatment for patients with ILD can be divided into three categories: disease targeted therapies, supportive therapies, and transplant surgery. Disease targeted therapies differ depending on the underlying aetiology; but are usually either anti-fibrotic medication, immunosuppression or a combination. Supportive treatment strategies are common to all types of advanced fibrotic ILD and encompass the pharmacological and non-pharmacological management of symptoms. Only a small proportion of patients with progressive fibrotic ILD are suitable for lung transplantation, which represents the only cure for progressive ILD.

The approval of anti-fibrotic medications (Pirfenidone and Nintedanib) for the treatment of IPF has dramatically changed the treatment paradigm. These medications slow the rate of decline of lung function and reduce the chance of acute exacerbations, but do not represent a 'cure' for IPF (Noble, Albera et al. 2011, Richeldi, du Bois et al. 2014). Current UK licensing for anti-fibrotic medications in IPF stipulate that patients' forced vital capacity (FVC) must fall within a predefined range (NICE 2016, NICE 2018). Recent evidence supports the use of anti-fibrotic medications for non-IPF forms of progressive fibrotic ILD and also benefit at higher FVC values (Wells, Flaherty et al. 2020). The licensing guidelines in the UK were updated in November 2021 to reflect this emerging evidence and now allow prescription of Nintedanib for people with progressive fibrotic ILD of any aetiology (NICE 2021). Immunosuppressant medications are the mainstay of treatment for connective tissue disease related ILD (CTD-ILD), hypersensitivity pneumonitis and some idiopathic forms of ILD (Adegunsoye, Oldham et al. 2017, Kelly, Nisar et al. 2020). The most commonly used immunosuppressant medications include oral or intravenous corticosteroids, steroid sparing oral agents such as mycophenolate mofetil, azathioprine or methotrexate, and intravenous cyclophosphamide or rituximab. The management of patients with ILD is complex, and for the majority disease targeted treatments merely slow disease progression rather than curing disease, therefore the early involvement of palliative care services has been recommended (Lindell, Kavalieratos et al. 2017, Pooler, Richman-Eisenstat et al. 2018). The following section explains the scope of palliative care and explains the definitions used within this thesis.

1.2 Palliative care definitions

Palliative care can be defined as "an approach that improves the quality of life of patients and their families facing the problems associated with life threatening illness, through the prevention and relief of suffering ….treatment of pain and other problems, physical, psychosocial and physical"(World Health Organisation 2020). This definition has evolved to incorporate care of patients at a stage in their disease where a cure is unlikely, but not impossible, and aims to encourage integration of palliative care earlier in the disease process (Hawley 2014). A visual model (Figure 1-2) incorporating two overlapping 'triangles' of disease management and palliative care has been developed. This model includes the

possible outcomes of surviving the illness and rehabilitation as well as end of life care. This modern description of palliative care aims to help patients and clinicians embrace the concept that palliative care can be incorporated to help with symptom management alongside active treatment.

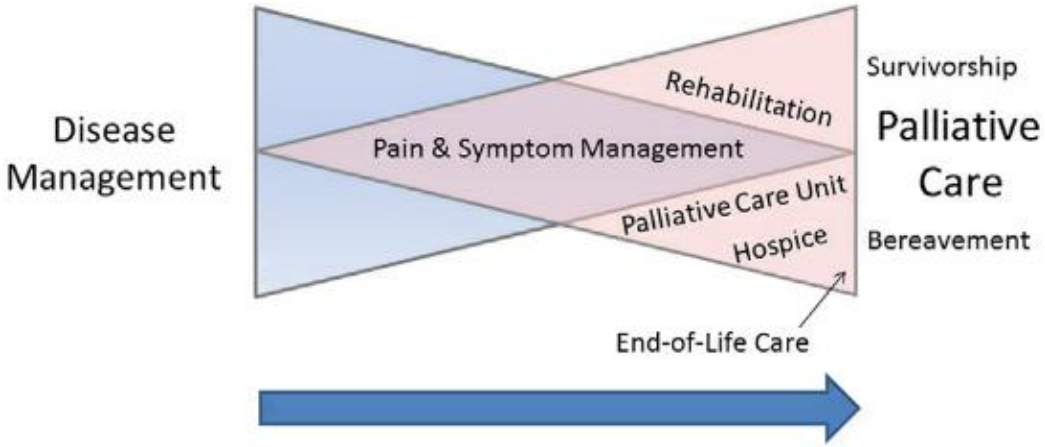

Figure 1-2 The Bow Tie model of palliative care (Hawley 2014)

The terms palliative care and end of life care are often used interchangeably resulting in confusion about what patients and healthcare professionals mean when using these phrases. Some people see a definite distinction between the phrases, with palliative care being symptom management which does not alter disease progression and end of life care being focused on patients who are expected to die imminently. NHS England refers to palliative care as a focus on making patients with uncurable illness comfortable by managing distressing symptoms and states that end of life care begins “when you need it” and may last a few days, months or sometimes over a year (NHS England 2022). The General Medical Council has issued good practice guidelines which advocate that end of life care is care received in the last 12 months of life (General Medical Council 2020). However, these definitions do not align with the perception of many patients and their relatives, who understand end of life to be a short period of time immediately prior to death. This view is reinforced by using supportive care frameworks aimed at the final weeks or days of life such as the care of the dying patient document (Northern Cancer Alliance 2020). In this research study, the term end of life care is used to refer to care in the final weeks and days of life to distinguish this from palliative care that may be received by patients from the time of

diagnosis. The focus on data from the period (days and weeks) immediately before death corresponds with patients' and bereaved carers' perspective of end of life.

Palliative care can be delivered by any healthcare practitioner, but there is a distinction between this 'general palliative care' and specialist services. 'Specialist palliative care' is provided by healthcare professionals who have undertaken specialist training in palliative medicine and specialist palliative care services encompass hospice care as well as advice, support and care provided by hospital and community palliative care teams (NICE 2011). In this study, general palliative care refers to the supportive care provided by non-specialist clinicians, such as respiratory physicians or GPs, and specialist palliative care refers to care provided by those clinicians with specialist training and qualifications in palliative care.

Advance care planning is a key component of palliative care. This refers to the process of discussing and recording patients' preferences concerning future goals of care to allow physicians to act in a way patients would have wanted if they lose capacity and can no longer communicate their wishes (Brinkman-Stoppelenburg, Rietjens et al. 2014). Low rates of advance care planning have been identified among patients with ILD and other chronic respiratory diseases (Bajwah, Higginson et al. 2013, Rajala, Lehto et al. 2016, Jabbarian, Zwakman et al. 2018).

The literature review (Chapter 2) will consider the efficacy of available symptomatic treatment approaches used in advanced ILD, the involvement of specialist palliative care and the end of life care that patients with ILD receive.

1.3 Rationale for the study

The significant symptom burden experienced by people with ILD has been well described in existing literature. People with ILD experienced a marked deterioration in symptoms during the last two years of life, with worsening symptoms of severe breathlessness and fatigue and increased reliance on informal carers (Rajala, Lehto et al. 2018). Most people with ILD die in hospital where they were often subjected to a high intensity of investigations and treatments that continued until the last few days of life (Lindell, Liang et al. 2015, Ahmadi, Wysham et al. 2016, Rajala, Lehto et al. 2016, Zou, Nouraie et al. 2019, Guo, Mann et al. 2020). Several studies have identified that referral to palliative care occurs late in the disease process, meaning that patients were unable to participate in advance care planning

discussions and consequently the quality of end of life care was affected (Lindell, Liang et al. 2015, Guo, Mann et al. 2020). People with ILD who had been involved with palliative care services and participated in advance care planning discussions were subjected to fewer investigations towards the end of life (Guo, Mann et al. 2020), demonstrating the impact that earlier involvement of palliative care services can have on the end of life care that patients receive.

Despite the poor prognosis associated with progressive fibrotic ILD and the high symptom burden experienced by patients towards the end of life, there is a paucity of research which considers end of life care. A systematic review, conducted prior to this research study, considered factors that influenced the quality of end of life care in ILD and identified a clear gap in knowledge surrounding end of life care (Palmer, Kavanagh et al. 2021). In this review five key factors were identified which influenced the quality of end of life care: i) healthcare utilisation in the last year of life, ii) involvement with palliative care services, iii) advance care planning, iv) symptom management and v) location of death. The review highlighted the inadequacy of literature relating to symptom control in the final days and weeks of life and that little is known about how patients and relatives feel about dying in an acute healthcare environment. This current research study aimed to address these gaps in the existing knowledge base.

The primary aim of this research study is to develop a comprehensive understanding of the experiences of end of life care amongst people with ILD in the North East and Cumbria region of the UK. The initial motivation for this study came from my background as trainee in respiratory medicine and my experiences of the challenges of caring for patients with ILD towards the end of life. Palliative care was identified as a priority by ILD patient groups in the region who reported that access to services was variable in different geographical locations. These motivations and the identified gaps in the literature led to the development of the research questions.

1.4 Impact of COVID-19 pandemic

This research study was conducted between January 2020 and March 2022, a two-year period in which the impact of the COVID-19 pandemic dramatically changed people's lives and the delivery of healthcare. The project was planned prior to the pandemic and required

substantial amendments prior to ethics review to incorporate remote interviewing rather than in person semi-structured interviews that were regarded as the 'gold standard' data generation method. Interviews were conducted with the majority of participants over the telephone or via video conferencing software.

The pandemic impacted on many aspects of this study. People with ILD underwent prolonged periods of shielding due to being classed as 'extremely clinically vulnerable' which had a long-lasting impact on the physical and mental health of both patient and bereaved relative participants. Healthcare services were disrupted with reduced availability of face-to-face clinical review and a reliance on remote consultations. End of life care and bereavement were particularly affected by COVID-19 restricting hospital visiting and increasing social isolation for bereaved relatives. The impact of the pandemic is described throughout this thesis, it is an important consideration when interpreting the findings, and will remain an important factor influencing service design and delivery for several years.

1.5 Structure and organisation of the thesis

This thesis explores the end of life care experience for people with ILD from the perspectives of patients, bereaved relatives and healthcare professionals. Chapter one begins with an introduction, then outlines the background and clinical context of the study.

Chapter two presents a review of relevant current literature. The research literature is summarised and critiqued, highlighting gaps in established knowledge and supporting the need for the research study. The paucity of literature relating to end of life care in ILD demonstrates the rationale for the study.

Chapter three explains the rationale for the methodological approach chosen for this study. My epistemological and ontological positions are outlined and the decision to use constructivist grounded theory approach is discussed. The purposive and theoretical sampling approaches and data generation using semi-structured interviews are explained. Data were analysed using initial coding, focused coding, development of preliminary categories, core categories and finally overarching categories using a constant comparative approach to analysis.

Chapter four presents the research findings from patient and bereaved carer participants, critically analysing the end of life care for people with ILD. These results are presented as a figurative illness journey, and the core categories that were identified from the data are discussed in detail.

Chapter five presents the findings from healthcare professionals in relation to the end of life care for people with ILD. These data are summarised into categories that examine the role of healthcare professionals in the figurative illness journey of people with ILD. Each of the core categories are discussed in detail and the relationships between categories are outlined.

Chapter six synthesises the whole data set, both patient and bereaved relative data and healthcare professional data. This chapter presents the overarching categories which emerged from these data and critiques these findings in relation to current literature. The chapter then discusses strengths and weaknesses of the study, researcher reflexivity, implications for clinical practice and opportunities for future research.

1.6 Chapter summary

The purpose of this chapter was to introduce the topic of interstitial lung disease and palliative care. This introductory chapter also outlines the rationale for conducting the study and the research aims. The main body of the thesis begins in Chapter 2 which presents an extensive review of the existing literature relating to palliative care and end of life care in interstitial lung disease.

Chapter 2 Literature Review

This chapter presents a review of literature relating to palliative and end of life care in interstitial lung disease. The disease trajectory observed in ILD is explained and its relationship to the delivery of specialist palliative care. This is followed by a review of the effectiveness of supportive treatments used in ILD, including pharmacological, non-pharmacological and psychological interventions. The next section reviews literature relating to barriers and facilitators influencing referral to specialist palliative care services, the timing of referrals and advance care planning. Literature that considered the lived experience of ILD from patient and carer perspective is presented along with healthcare professionals views. Finally, literature that specifically related to end of life care in ILD is presented, identifying significant gaps in existing knowledge and demonstrating the requirement for further research in this area.

The review was conducted through searching of EMBASE, Medline, CINAHL, Scopus and Web of Science. An example of the search strategy is demonstrated in Figure 2-1. Further sources were identified from handsearching of the reference lists of seminal articles. The results are presented narratively under the key subtheme headings.

EMBASE

(exp Lung Diseases, Interstitial/) OR (interstitial lung disease*.tw.) OR (exp lung fibrosis/) OR (pulmonary fibrosis.mp.) OR (exp allergic pneumonitis/) OR (hypersensitivity pneumonitis.mp.) OR (exp fibrosing alveolitis/) combined with (end-stage.mp.) OR (terminal* ill*.mp.) OR ((end adj6 life).mp.) OR (terminal stage*.mp.) OR (((advanced or end stage or terminal*) adj4 (disease* or ill*)).mp.) OR (exp palliative therapy/) OR (palliative care.mp.) OR (place of death.mp.) OR (end of life care.mp.) OR (terminal care.mp.) OR (dying.mp.)

Figure 2-1 Example search strategy for literature review.

In addition to the narrative literature review presented in this chapter, two systematic review articles have been published in relation to this work. The first systematic review considered which factors influenced the quality of end of life care for people with ILD (Palmer, Kavanagh et al. 2022) (Appendix E). This review concluded that there was

inadequate knowledge regarding the most appropriate location of death for people with ILD and identified a gap in literature relating to symptom management at the end of life and its association with location of death. These findings influenced the design and concept of the current research project, which was developed to explore the identified gap in existing knowledge.

The second systematic review article considered the appropriate timing for the introduction of palliative care for people with ILD (Palmer, Kavanagh et al. 2022) (Appendix F). This systematic review examined qualitative literature to understand the perspectives of people with ILD and their carers about the timing of palliative care conversations. Three interconnecting stages in the ILD patient's journey were identified: information seeking, grief and acceptance and fear of the future. The findings suggested that palliative care involvement was most appropriate in the latter two stages. People with ILD wanted referral to palliative care services to be individualised and prompted by changes in their health status rather than initiated solely based on their diagnosis or stage of disease. These findings align with current literature as discussed in section 2.3.3 of this thesis.

2.1 Disease trajectory in ILD

Fundamental issues surrounding the provision of palliative care for people with ILD are the uncertain prognosis and unpredictable disease trajectory associated with these conditions. The delivery of palliative care services has traditionally been based on the cancer disease trajectory where health declines over time and then evolves into palliative and end of life care (Murray, Kendall et al. 2005). This model suggests a linear progression of disease and deterioration in health which are relatively predictable, enabling palliative care services to be accessed at an appropriate point towards the end of a patient's life. In chronic non-malignant respiratory disease, the illness trajectory differs from the cancer model as there are often periods of stability which are punctuated by acute deteriorations and recovery (Murray, Kendall et al. 2005). Following each exacerbation patients often do not recover to the same baseline, and there is an overall gradual decline in patients' health and function.

In common with other chronic lung diseases, the illness trajectory in ILD can be unpredictable, with a background rate of lung function and symptomatic decline which is punctuated by acute exacerbations of symptoms (Figure 2-2) (Bourke and Peel 2014). The

term ILD incorporates a heterogeneous group of diseases, with significant variation in disease progression, which makes prediction of a patients' disease trajectory very difficult. Clinicians identified this uncertain disease trajectory as a major barrier to the timely referral of patients to specialist palliative care (Mc Veigh, Reid et al. 2018) and as a result late referral to palliative care services is a common theme in existing literature (Lindell, Liang et al. 2015, Guo, Mann et al. 2020) A small proportion of patients with advanced ILD are on a lung transplant waiting list and follow a further disease trajectory 'the transplant trajectory' as shown in Figure 2-2 (Bourke and Peel 2014). These patients are seriously ill with a high level of symptoms and could die from a further exacerbation, however they are hoping for a lung transplant which dramatically alters the disease trajectory (Bourke and Peel 2014). There are substantial unmet palliative care needs amongst lung transplant candidates, particularly in relation to physical symptoms and of life concerns (Pawlow, Blumenthal et al. 2020). It is crucial that these patients receive parallel planning, with involvement of specialist palliative care, for both the possible outcome of successful lung transplant, but also the possibility of further decline and death.

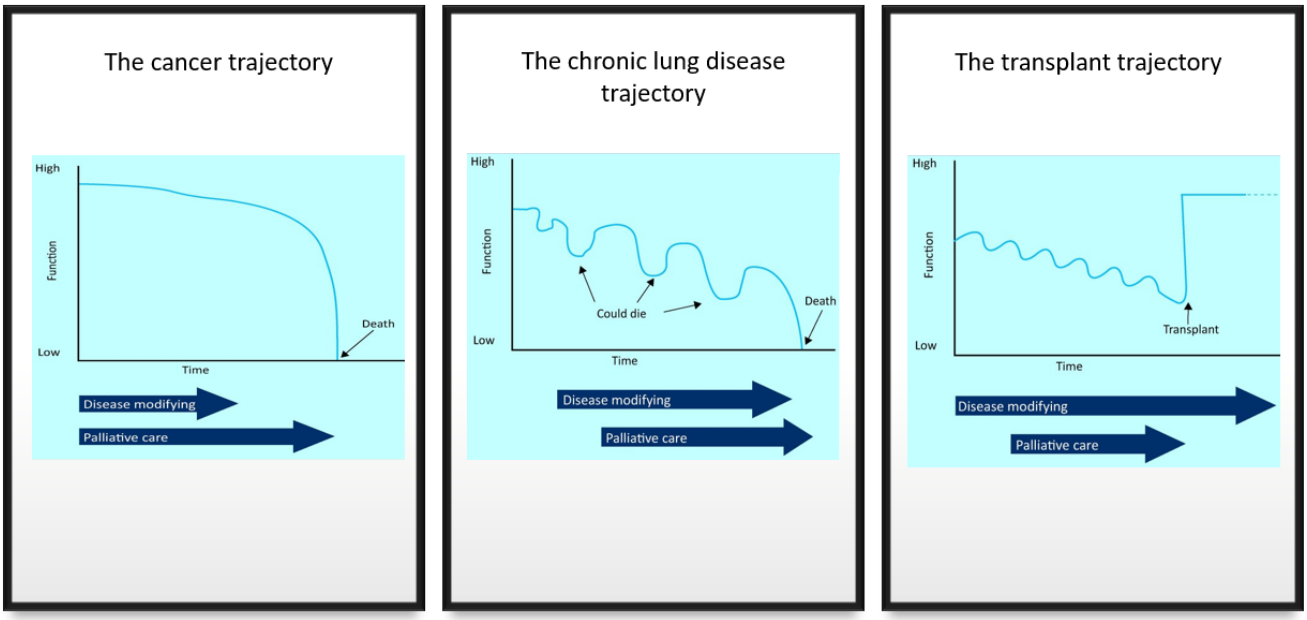

Figure 2-2 Disease trajectories in cancer and chronic progressive lung disease (Bourke and Peel 2014)

2.1.1 Predicting prognosis in non-malignant chronic disease

The timing of death in chronic severe illness remains unpredictable until late in the disease trajectory, and sometimes death is sudden and unexpected, therefore arrangements for end of life care must be triggered by severity of symptoms rather than waiting for reliable evidence that death is near. Questions remain about how to adapt specialist palliative care delivery, a service designed for rapid and predictable deterioration as in the cancer disease trajectory, to one which can accommodate the uncertainty of patients with non-malignant disease (Boland, Martin et al. 2013).

The uncertain disease trajectory identified in ILD is not unique and has been observed in other non-malignant conditions, such as chronic obstructive pulmonary disease (COPD) and heart failure (Kheirbek, Alemi et al. 2013, Landers, Wiseman et al. 2017). People with end-stage heart failure often follow a gradually deteriorating disease trajectory, but in addition have high rates of sudden cardiac death, meaning that prognostic uncertainty is common among physicians and patients (Allen, Stevenson et al. 2012). In contrast to ILD, where disease modifying medications are the mainstay of treatment, there are multiple interventions which may change the clinical course of heart failure, e.g. high-risk cardiac surgery, percutaneous interventions, pacing device therapy as well as the potential for transplant or mechanical cardiac support (Allen, Stevenson et al. 2012). There are various suggested tools to aid prognostication that include measures of cardiac performance, exercise capacity and neurohormonal markers (Jaarsma, Beattie et al. 2009, Treece, Chemchirian et al. 2018). However, as there is no single parameter which is a perfect predicator of prognosis, numerous clinical scoring systems have been developed.

COPD is a chronic respiratory disease that follows an uncertain disease trajectory similar to that seen in ILD with gradual decline punctuated by acute exacerbations. People with COPD often experience more frequent exacerbations, but these episodes are not associated with the same high mortality as in ILD (Hoogendoorn, Hoogenveen et al. 2011, Dhamane, Moretz et al. 2015). As in heart failure, there are multiple prognostic tools used in COPD to aid clinicians with predicting an individual patient's prognosis. Again, there is overlap between scoring systems and not enough evidence to recommend one single variable or multivariable scoring system. A systematic review that evaluated COPD prognostication tools concluded that very few studies were designed to assess, or report, the prediction of mortality within

12 months and that the quality of evidence supporting existing scoring systems was low (Smith, Moore et al. 2017).

2.1.2 Predicting prognosis in ILD

The most common cause of death in ILD is the disease itself, followed by cardiac disorders and lung cancer (King, Albera et al. 2014, Kärkkäinen, Nurmi et al. 2018). Rapid deterioration at the end of life can be related to pulmonary embolism, pneumothorax, infection or heart failure (Panos, Mortenson et al. 1990) and therefore can be very difficult to predict. One study that looked at all-cause mortality established that approximately 20% of deaths of people with mild to moderate IPF were unrelated to their underlying lung disease (King, Albera et al. 2014). The significant proportion of people dying from illness unrelated to ILD and the speed with which these alternative pathologies can lead to death fuels prognostic uncertainty.

There remains a lack of standardised or validated measures to predict disease progression in ILD. A combination of clinical, physiological, function and radiological markers are used to attempt to predict disease progression. There have been attempts to integrate these elements to refine the prognostication process in IPF. However, there is little evidence to recommend a single validated tool for prognostic assessment in ILD. The common objective measurements of physiological markers (pulmonary function measurements) and radiological markers (high resolution CT-quantification on serial imaging) do not predict disease behaviour and have not been established as a reliable marker of disease progression (Robbie, Daccord et al. 2017). The potential utility of serum biomarkers as additional indicators of disease severity in IPF is likely to be a focus of future research (Molyneaux, Fahy et al. 2022).

There are a limited number of tools to help with predicting prognosis in ILD. A composite physiological index (CPI) was developed by Wells et al (2003) derived from pulmonary function measurements of FVC, FEV1 and TLCO. The CPI was shown to correlate with disease extent seen on CT imaging better than individual lung function measurements. A further widely used tool, the GAP index, was developed by Ley et al (2012). The GAP-index uses commonly recorded data (gender, age, FVC and TLCO) for people with IPF to assess risk of mortality at 1, 2 and 3 years (Ley, Ryerson et al. 2012). Its strengths are the simplicity and

that it has been shown to aid prognostication in other forms of chronic ILD (Ryerson, Vittinghoff et al. 2014, Morisset, Vittinghoff et al. 2017). However, these tools have been shown to have limited capability to accurately predict patient outcomes in a large cohort study (Lee, Park et al. 2018). More recently these prognostic tools have been developed with additional components added claiming to increase the accurate prediction of outcomes (Manzetti, Hosein et al. 2021, Zhu, Li et al. 2021).

Although the uncertain disease trajectory observed in ILD is not unique to these conditions, there are other factors which mean that this patient group present a particular challenge to providing timely palliative care. Prognostication in ILD is hampered by the lack of defined criteria for disease progression and low-quality evidence for prognostic tools. There are few disease modifying treatment options, and those which are available generally slow the rate of disease progression rather than representing a cure. The risk of death from an acute exacerbation remains high, between 33-83%, with an annual incidence of acute exacerbation reported as up to 20% (Kolb, Bondue et al. 2018). Explaining this uncertainty to patients and their families and facilitating timely involvement of palliative care remains one of the most significant challenges surrounding end of life care for people with ILD.

2.2 The role of specialist palliative care in ILD

Palliative care focuses on patients' symptomatic management rather than disease modifying treatments. It is widely accepted that specialist palliative care enhances patients' treatment and symptom control, providing a holistic approach to support people towards the end of their lives. However, there have been longstanding challenges in providing evidence of the benefit of specialist palliative care. Two systematic reviews have been conducted with the aim of reviewing the impact of specialist palliative care on patients' quality of life. Gaertner et al (2017) concluded that involvement with specialist palliative care had a small measurable effect on patients' quality of life, which was more pronounced in patients who received interventions earlier in their disease. This systematic review included randomised controlled trials of adults in hospital, hospice or community settings with any advanced illness. The size of the effect may be disappointing to advocates of specialist palliative care; however, the review included only a small number of studies (10 randomised controlled trials, reported in 12 papers) and missing data was not adequately considered by two of the studies which resulted in exclusion from the meta-analysis. The authors suggested that a

fundamental problem with the included studies was that palliative care referrals were made based on stage of disease rather than patients' symptoms and clinical need, which could have contributed to an underestimation of the effect.

These findings are supported by a recent Cochrane review which also established a small positive effect of specialist palliative care on quality of life and patient satisfaction with care for people with advanced illness (Bajwah, Oluyase et al. 2020). The review suggested that involvement of specialist palliative care increased patients' likelihood of dying in their preferred location by 22%. This is an important finding but should be interpreted with caution as although the Cochrane review was larger (including 42 randomised controlled trials) the evidence was graded very low or low quality in respect to high risk of bias, imprecision and inconsistency. Overall, existing literature showed a positive signal that specialist palliative care improved patients' quality of life and highlighted some of the inherent problems with palliative care research, namely issues with quality of evidence and low numbers of randomised controlled trials.

2.2.1 Efficacy of symptomatic and palliative treatments in PF-ILD

For people with ILD, a palliative care approach is frequently required for breathlessness, cough and fatigue as well as associated psychological symptoms (Bajwah, Higginson et al. 2013). There is a wide body of literature which advocates for early involvement of specialist palliative care for people with ILD (Lindell, Liang et al. 2015, Kalluri, Claveria et al. 2018) and support from national guidelines (NICE 2017). Palliative care interventions can be classified into three groups: pharmacological treatments, non-pharmacological treatments and psychological support.

2.2.2 Pharmacological treatments

Opioids are the most common medication prescribed for refractory breathlessness in chronic lung disease. As for many palliative measures in ILD, the evidence for use of opioids comes from studies of patients with COPD. Patients with dyspnoea related to COPD reported the use of opioids gave a sense of calm and relief from severe breathlessness, improvement in psychological symptoms and quality of life (Rocker, Young et al. 2012). An Australian randomised controlled trial found that low dose oral opiates improved symptomatic management of breathlessness for chronic lung disease patients with minimal side effects

(Abernethy, Currow et al. 2003). This study was small with only 48 participants; however, it was well conducted, and the results indicated a significant symptomatic improvement in patients' breathlessness. Five participants withdrew from the study due to side-effects from the medications with constipation reported as the most notable sustained side effect.

In contrast, a large Swedish cohort study (n= 2249) raised concerns of increased mortality associated with the prescription of higher dose opioids and benzodiazepines in patients with COPD on long-term oxygen therapy (Ekstrom, Bornefalk-Hermansson et al. 2014). A further US cohort study, which included 130,979 patients with COPD, concluded that opioid use was associated with increased risk of adverse respiratory outcomes and mortality in this patient group (Vozoris, Wang et al. 2016). A strength of both studies was the large number of participants. However, both studies were retrospective cohort design and the indication for opioid or benzodiazepine prescription, or the effectiveness of these medications, was not recorded. An alternative interpretation of the association is that increased doses of opioids and benzodiazepines were required for treatment of increasing symptom prevalence due to disease progression. This theory is supported by a Norwegian population study which found the prescription of benzodiazepine medications increased closer to death (Neutel and Johansen 2015).

A single study has reviewed the use of opioids in oxygen dependent ILD and did not find an association with either increased rates of hospitalisation or mortality (Bajwah, Davies et al. 2018). There was a reported association between the use of higher dose benzodiazepines and mortality. This study included 1603 participants who were identified from a Swedish oxygen database and therefore it was not possible to identify the efficacy of treatments for breathlessness and it was not powered to accurately assess safety of these medications. However, the observed associations support the use of opioids and lower dose benzodiazepines for patients with advanced in ILD. Although there is limited evidence surrounding the efficacy of low dose opioids in ILD, these medications appear to be safe and to improve symptom control. It is important to remember that patients who are prescribed opioids or benzodiazepines for breathlessness have severe, refractory symptoms and have limited other options to improve their quality of life. Therefore, concerns about increased mortality and hospitalisation need to be balanced with the possibility of poorly controlled symptoms at the end of life.

Chronic cough symptoms have a significant impact on the quality of life for people with ILD and cough is an independent predictor of disease progression (Ryerson, Abbritti et al. 2011). Cough can cause problems with sleeping, urinary incontinence, musculoskeletal pain and cause embarrassment leading to avoidance of public places, reduced social interaction and relationship difficulties (Swigris, Stewart et al. 2005, Morice, Fontana et al. 2007, French, Crawford et al. 2017). The cause of cough in ILD is not fully understood with many studies suggesting "multifactorial" pathogenesis influenced by architectural distortion of the lungs, increased cough reflex sensitivity and inflammation, with co-morbidities also influencing symptoms (van Manen, Birring et al. 2016, Bargagli, Di Masi et al. 2019). As a result, the evidence supporting the management of cough in ILD is limited (Birring, Kavanagh et al. 2018). Pharmacological treatments aimed at managing the effect of co-morbidities include inhaled bronchodilators, nasal corticosteroids and proton pump inhibitors (Sanchez-Ramirez, Kosowan et al. 2022). Limited data has suggested that corticosteroids and thalidomide may be beneficial for chromic cough in IPF (Hope-Gill, Hilldrup et al. 2003, Horton, Danoff et al. 2008).

Opiates are often prescribed for cough that is refractory to alternative therapies. The benefits of opiates as an anti-tussive therapy have been shown for patients with chronic cough (Morice, Menon et al. 2007). The findings of this randomised controlled trial supported opiates as a treatment of chronic cough, however, it only included 27 patients and was not specific to patients with ILD. Other authors have reservations about the use of opiates for the management of cough in ILD, primarily due to concerns about reducing the protective mechanism of cough and potential side effects (van Manen, Birring et al. 2016). At present there is little literature to support the use of opiates in refractory cough in ILD, however, they are recommended in the palliative management of cough (Raghu, Collard et al. 2011). The PAciFy trial is an ongoing UK double-blinded crossover trial that aims to answer questions about the efficacy of using morphine sulphate to manage refractory cough in people with IPF (Wu, Banya et al. 2022).

2.2.3 Non-Pharmacological treatments

Oxygen supplementation is commonly prescribed for patients with ILD, with the aim to reduce breathlessness and improve physical activity levels and is regarded as a standard of care in national and international guidelines (Raghu, Collard et al. 2011, NICE 2017).

Although patients may not experience a noticeable improvement in their symptoms, there is evidence that oxygen use increased exercise capacity and improved patients' health related quality of life (Bell, Cox et al. 2017, Visca, Mori et al. 2018). A significant problem with oxygen delivery in ILD is the often high oxygen demands, which are difficult to manage with standard modes of oxygen delivery. Issues with delivery of high-dose oxygen therapy may place restrictions on the location of patients care and could prevent patients from dying in their preferred location. Historically, it has not been possible to deliver high-dose oxygen therapy in patients' home environment or in a hospice, however, recent case reports have suggested that it is possible to overcome this barrier and allow patients more choice over location of their care (Kamp, Trudzinski et al. 2016, Kalluri and Richman-Eisenstat 2017).

Fatigue is another common symptom of ILD which is intricately linked to breathlessness. Breathlessness and fatigue limit physical activity leading to muscle deconditioning and further reduction in exercise capacity (Figure 2-3). Pulmonary rehabilitation has become the foundation of treatment for these symptoms in ILD (NICE 2017). Participation in pulmonary rehabilitation has demonstrated improvement in exercise capacity and breathlessness (Holland, Hill et al. 2008, Tonelli 2017) and reduced fatigue by increased patient awareness of coping strategies such as energy conservation (Sampson, Gill et al. 2015). Patients prefer to attend pulmonary rehabilitation sessions which included ILD-specific educational content and this may be an acceptable way to initiate discussions about advance care planning and end of life care (Holland, Fiore et al. 2015).

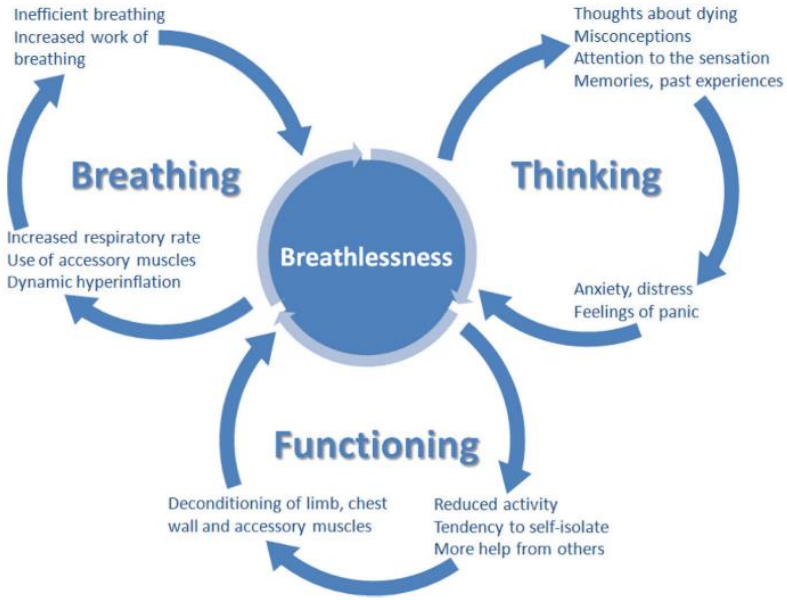

Figure 2-3 Breathing-thinking-functioning model (Spathis, Booth et al. 2017)

2.2.4 Psychological support

Palliative care is a holistic approach which addresses both the physical symptoms and emotional or spiritual needs of patients. The 'breathing-thinking-functioning' model, described by Spathis et al (2017), demonstrated the link between patients' physical symptoms and psychological symptoms (Figure 2-3). Cognitive behavioural therapy (CBT) has been successfully used to improve anxiety symptoms in patients with COPD (Heslop-Marshall, Baker et al. 2018). This randomised controlled trial was undertaken in Newcastle upon Tyne and included 279 patients randomised to either one-to-one CBT with respiratory nurses or support with leaflets. The CBT intervention reduced attendances at emergency department and hospital admissions. There are no trials which look at CBT specifically in patients with ILD, however, it is highly relevant given that anxiety has been reported to affect 58% patients with ILD (Lindell, Olshansky et al. 2010). Breathlessness support services, which provide holistic management of breathlessness symptoms and often include elements of CBT, have shown positive results in managing depression and anxiety in chronic lung disease (Bausewein, Schunk et al.).

In contrast, a US based IPF study determined that receiving palliative care did not improve patients' quality of life, anxiety, or depression compared to usual care (Janssen, Rosielle et

al. 2020). This randomised controlled trial pilot study included 22 patients and showed a trend towards increased depression in the group who received palliative care, but the findings should be interpreted with caution given the small sample size. These results compare to those described in another US study which reported increased anxiety scores for patients with IPF who participated in a 6-week support group programme (Lindell, Olshansky et al. 2010). Participants, which included patients and paired carers (n=42), were randomised for either usual care or attending a nurse led support group programme which discussed symptom control and elements of palliative care. Surprisingly, patients in the intervention group reported reduced health related quality of life and increased anxiety scores. Whereas qualitative data from interviews described patients felt "able to put my disease in perspective" and "didn't feel isolated" suggesting a positive impact of the intervention. One interpretation is that palliative care discussions exacerbated anxiety symptoms; however, this may be related to issues with timing of discussions and introducing these concepts in a group environment, rather than the content of the conversation.

2.3 Palliative care access

It is well recognised that people with ILD have a high-symptom burden and complex palliative care needs (Bajwah, Higginson et al. 2013, Carvajalino, Reigada et al. 2018). However, low levels of palliative care involvement have been reported ranging from 0% to 38% (Palmer, Kavanagh et al. 2021). This next section explores barriers and facilitating factors that influence referral to specialist palliative care.

2.3.1 Barriers that prevent patients accessing palliative care

It is important to understand the barriers which prevent engagement with palliative care in order to improve patient access to these services. A recent systematic review identified multiple barriers preventing access to specialist palliative care services (Kim, Atkins et al. 2019). This study identified that referrals to specialist palliative care were delayed as were decisions about end of life care. The findings suggested that this was related to prognostic uncertainty, patients' fear of talking about the future and confusion about the role of palliative care. However, it should be noted that this systematic review only included 10

studies; the majority based on observational data and two were not specific to patients with ILD, demonstrating the paucity of literature addressing these questions.

Confusion about the role of palliative care is widely recognised as a barrier preventing patients accessing palliative care (McIlfatrick, Hasson et al. 2014, Zimmermann, Swami et al. 2016). A Northern Ireland study, which explored public perceptions of palliative care, established expectations of palliative care were based on information from healthcare professionals or prior experience rather than an existing knowledge of this aspect of healthcare (McIlfatrick, Hasson et al. 2014). A US focus group study identified that patients with IPF had unmet educational and information needs and a poor understanding about the role of palliative care (Lindell, Kavalieratos et al. 2017). Qualitative research in the UK also highlighted that people with ILD had misconceptions about the reasons for referral to palliative care and concerns about being referred onto "that path to death" (Sampson, Gill et al. 2015). These results highlight the importance of providing patients with accurate and timely information about palliative care to address these widespread misconceptions.

Geographical disparity in access to palliative care services has been identified by a US IPF registry-based study, which found patients who lived closer to the specialist centre were more likely to be referred to specialist palliative care services (Zou, Nouraie et al. 2019). This was a retrospective cohort study with data reviewed from a 16-year period (n=828 patients with IPF), meaning that the reasons for this apparent geographical disparity could not be adequately explored. It is possible that referrals were influenced by clinician's familiarity with local palliative care services, for example a clinician with a specialist interest or the availability of local hospice services. However, patients who lived further away from specialist centres may have accessed their local palliative care services and this data would not have been captured by studies based in a central location. Geographical disparity in access to palliative care support has been identified in chronic lung disease with a noticeable difference between those living in rural and urban areas (Mc Veigh, Reid et al. 2018). This Irish study found access to palliative care support at home was particularly difficult in rural areas and some patients were not able to die in their preferred location due to inadequate support and healthcare resources. These data emphasise the importance of healthcare professionals based in centralised specialist ILD clinics being familiar with palliative care service providers throughout their region to enable appropriate referrals.

It is illogical to consider geographical disparity without acknowledging the effect of socioeconomic inequalities on patients' access to specialist palliative care services. Poorer access to palliative care resources may not be related to geographical location alone and may be part of wider healthcare inequalities. French et al (2021) conducted a systematic review exploring the socioeconomic inequities in access to palliative care and end of life care in the UK. The authors concluded that there was no clear or consistent narrative regarding socioeconomic position and the receipt of palliative and end of life care in the UK. A review of hospice care in the UK, Australia, New Zealand and Canada found that older people (aged 85 years and over), ethnic minorities, people with non-cancer illness and people living in rural areas or socioeconomic deprivation had unequal access to palliative care services (Tobin, Rogers et al. 2022). Palliative care policy makers recognise these healthcare inequalities and have issued guidance to address the disparities and ensure equality in end of life care (NHS England 2023, Office for Health Improvement and Disparities 2023).

2.3.2 Factors that facilitate access to palliative care

Factors that increased the chance of patients being referred to palliative care services have been identified from two US observational studies (Rush, Berger et al. 2018, Zou, Nouraie et al. 2019). Older age, resuscitation status and treatment at an urban teaching hospital were associated with increased access to palliative care services for patients with IPF undergoing mechanical ventilation (Rush, Berger et al. 2018). This was a large observational study including data from 3,166 patients with IPF. These data suggest that recognition of patients' poor prognosis is crucial to enable appropriate access to palliative care at the end of life. This study highlights important differences in practice between the US and UK, where very few patients with ILD are mechanically ventilated due to the recognition of poor prognosis for patients in this situation. A second US study found increased palliative care referrals were associated with older age and increased co-morbidities (Zou, Nouraie et al. 2019). Both these factors can be associated with a poorer prognosis in ILD (Ley, Collard et al. 2011) which further supports the theory that patients' access to palliative services is dependent on clinicians' recognition of their prognosis. Patient involvement with a support group and higher number of outpatient appointments also correlated with increased palliative care referrals (Zou, Nouraie et al. 2019). Support group involvement could increase patients' knowledge of their disease and empower them to initiate discussions about future care. A deterioration in patients' clinical condition would necessitate increased outpatient review

and potentially subsequent palliative care referral. However, frequent outpatient review could improve clinicians' confidence in assessment of disease trajectory and improve doctor-patient relationship allowing open discussion about advance care planning and palliative care.

2.3.3 Timing of specialist palliative care involvement

There remains debate about the most appropriate time for people with ILD to be referred to specialist palliative care. Some authors advocate that this should be considered at the point of diagnosis in IPF with the understanding that this is a non-curable, progressive condition (Lindell, Kavalieratos et al. 2017, Pooler, Richman-Eisenstat et al. 2018). However, for people with ILD of other aetiologies, it may be unknown at the time of diagnosis whether they will go on to develop progressive fibrotic disease and therefore palliative care referral is not appropriate at that time. There is also a risk of overwhelming already stretched palliative care resources with a blanket referral based on diagnosis rather than stage of disease or symptomatic need.

The CaNoPy study was a UK study of the care needs of people with IPF and their carers that aimed to establish when palliative care might be effectively introduced (Sampson, Gill et al. 2015). This study interviewed 48 participants (27 patients and 21 carers), recruited from two specialist centres, and included participants at different stages in the disease trajectory, meaning the results are generalisable and applicable to other ILD centres in the UK. The study identified 'triggers' that should prompt clinicians to consider palliative care referral; including change in physical or social function, symptom deterioration and initiation of oxygen (Sampson, Gill et al. 2015). A survey of UK respiratory healthcare professionals reported similar changes that prompted referral to specialist palliative care; poorly controlled symptoms, general deterioration, hospital admissions and end of life care (Kim, Olive et al. 2020). The concept of referral to palliative care based on 'triggers' or changes in health status is widely adopted by respiratory clinicians. However, as there remains a lack of standardised guidance to outline best practice in this area, it is largely reliant on individual clinicians' knowledge and expertise. Devising ways to target palliative care referrals, for example with Integrated Palliative care Outcome Scale (IPOS) (Murtagh, Ramsenthaler et al.

2019), or a supportive care decision tool (Sharp, Lamb et al. 2018) would ensure resources were available for the most appropriate patients.

The initiation of oxygen therapy has been established as an important 'turning point' in disease trajectory, a marker of disease progression and the point where the disease becomes highly visible to others (Schoenheit, Becattelli et al. 2011, Ahmadi, Wysham et al. 2016). Patients may view oxygen with trepidation and view requiring oxygen as a failure associated with the stigma of being ill (Lindell, Kavalieratos et al. 2017) and a significant impediment to living their life as normal (Belkin, Albright et al. 2014). Uncertainty about the purpose and proposed benefits of oxygen, physical difficulty using devices, and embarrassment about using oxygen in public have been reported by patients with chronic lung disease (Earnest 2002, Arnold, Bruton et al. 2011, Kelly and Maden 2014).The initiation of long-term oxygen was identified in a large Swedish observational study as a poor prognostic marker, with death occurring on average 8 months later; indicating that this was an important point to consider involvement of palliative care (Ahmadi, Wysham et al. 2016). Often people with ILD are initially started on ambulatory oxygen and then deteriorate further until they require long term oxygen therapy. This change in oxygen usage may be subtle but it remains an important prognostic marker and should prompt consideration of palliative care referral.

Recognition of poor prognostic features and changes in patients' disease trajectory are important to identify patients who would benefit from specialist palliative care involvement and to help to navigate the uncertainty of prognostication in ILD. Two UK specialist ILD centres adapted an existing needs assessment tool that was primarily use for patients with cancer to reflect the palliative care needs of people with ILD (Boland, Reigada et al. 2016). The NAT: PD-ILD (Needs Assessment Tool: Progressive Disease for people with ILD) was developed as a practical way to address the unidentified and unaddressed palliative care needs of patients and their carers. The tool prompted clinicians to assess the holistic needs of patients and their informal carer needs alongside clinical symptoms. The main barriers with implementation related to confidence and time constraints to fully assess psychological and spiritual needs (Reigada, Papadopoulos et al. 2017).

Bristol ILD service developed a supportive care decision aid which highlighted poor prognostic factors and concerning symptoms in IPF, the use of which was associated with

increased referrals of appropriate patients to specialist palliative care services (Sharp, Lamb et al. 2018). Interestingly, documentation of advance care planning discussions and resuscitation decisions increased for both patients for who the tool was completed and those patients for who it was not used. This is likely to be due to heightened awareness within the ILD department of the need to discuss these key issues. The study therefore demonstrates the effectiveness of increasing awareness of palliative care components and encouraging these discussions within ILD teams.

2.4 Integration of palliative care and respiratory services

Integration of respiratory and palliative care services has been associated with improved symptom management and reduced acute healthcare utilisation towards the end of life for patients with advanced respiratory disease (Higginson, Bausewein et al. 2014, Smallwood, Currow et al. 2018, Brighton, Miller et al. 2019). Several specialist ILD centres, in the UK and internationally, advocate for the integration of specialist palliative care and ILD services. There is evidence from these centres that this approach increased the frequency of advance care planning conversations, reduced acute healthcare utilisation and improved concordance with patients' preferred place of death (Bajwah, Ross et al. 2015, Barratt, Morales et al. 2018, Kalluri, Claveria et al. 2018, Archibald, Bakal et al. 2020).

The specialist ILD centre in Alberta, Canada has published a series of papers relating to its multi-disciplinary collaborative (MDC) clinic (Kalluri, Claveria et al. 2018, Pooler, Richman-Eisenstat et al. 2018, Archibald, Bakal et al. 2020, Kalluri, Lu-Song et al. 2020). Key elements of this clinic included the multidisciplinary team members (respiratory clinician, palliative care specialist, nurse, respiratory therapist, physical therapist and dietician) the early introduction of advance care planning discussions, self-management plans for management of dyspnoea and close links with community teams. The first of these papers reported that involvement with MDC clinic was associated with less acute healthcare utilisation in the last year of life and more people dying at home. The study compared data from 2012 (preceding the establishment of the clinic) with recent data from patients who were involved with the clinic. It is known from observational studies that access to palliative care services and the

proportion of people dying at home has increased over time and therefore it would be expected that in more recent years that standard care would have improved.

A qualitative study interviewed eight bereaved relatives of people who had been involved the MDC clinic (Pooler, Richman-Eisenstat et al. 2018). Participants reported a positive experience of their relatives' death (seven of whom died at home and one in a local hospice) describing feeling informed, prepared and supported when death was near. The study reports to have used purposive sampling, however the results may have been more helpful if they had interviewed bereaved relatives of patients who died in other locations to build a full picture of the impact of this service. The sample size was very small and there was no mention of data saturation which suggests that more interviews may have helped to improve the completeness of the data.

A further study was conducted in 2020 using data from the MDC clinic; the results indicated that early initiation of palliative care components such as advance care planning led to improvement in patients dying in their preferred location (Archibald, Bakal et al. 2020), and also influenced the care that patients received in acute care settings. Overall, these studies indicate that early involvement with specialist palliative care and facilitating conversations about advance care planning can influence the location of and improve the end of life care that patients receive.

A community case conference devised in London, which supported patients with ILD and carers with advance care planning, improved symptom control and quality of life for participants (Bajwah, Ross et al. 2015). The case conference model was reported to be successful and acceptable to patients and carers. However, each case conference took between one and six hours to arrange, making this a less feasible option for already stretched ILD services. Bristol specialist ILD service developed a collaborative multidisciplinary team (MDT) approach that involved ILD clinicians, palliative care specialists, nurse and pharmacist (Barratt, Morales et al. 2018). The MDT was held every six weeks, it assessed and provided physical, social and psychological support for patients with ILD and their carers. The study reported increased advance care planning discussions and documentation of resuscitation decisions, in conjunction with referrals to community teams to address other unmet palliative care needs (Barratt, Morales et al. 2018). Four studies reported the outcome of novel approaches to collaboration between respiratory, palliative

care and community teams that all resulted in increased advance care planning discussions amongst patients with interstitial lung disease (Bajwah, Ross et al. 2015, Barratt, Morales et al. 2018, Kalluri, Claveria et al. 2018, Archibald, Bakal et al. 2020). A community case conference devised in the UK, which supported patients and carers with advance care planning, improved symptom control and quality of life for participants (Bajwah, Ross et al. 2015). Another UK service developed a collaborative multidisciplinary team approach which led to increased advance care planning discussions and documentation of resuscitation decisions, in conjunction with referrals to community teams to address other unmet palliative care needs (Barratt, Morales et al. 2018). However, these studies all highlight the significant time and resources required to deliver integrated palliative care in these models. Fundamentally, there is no single way to integrate services, each centre must respond to local needs, geography and work collaboratively with existing services.

2.5 Advance care planning

Advance care planning (ACP) is a key component of palliative care and can empower people with life limiting conditions by making their wishes known. In general, current literature suggests there is inadequate opportunity provided for advance care planning for people with ILD (Rajala, Lehto et al. 2016, Liang, Hoffman et al. 2017, Guo, Mann et al. 2020, Koyauchi, Suzuki et al. 2021). This is not a unique issue in ILD and is a common theme with other chronic respiratory diseases. A systematic review of ACP for patients with chronic respiratory diseases concluded that ACP conversations are surprisingly uncommon; possibly due to uncertain disease trajectories and ambivalence amongst both patients and healthcare professionals to engage in discussions (Jabbarian, Zwakman et al. 2018). The authors suggest that these barriers could be overcome by identifying trigger points throughout disease progression to introduce ACP, and training healthcare professionals about communicating end of life issues. This review included 21 studies, but only one focused on patients with ILD. However, the conclusions remain relevant to patients with ILD, and these issues have been considered in other studies which focus on the ILD population. These findings are supported by a US study which identified a reluctance to engage with ACP conversations amongst patients with ILD (Lindell, Kavalieratos et al. 2017). Conversely, qualitative studies in both the UK and Australia have reported that people with ILD wanted further information and

embraced the opportunity for ACP discussions (Bajwah, Koffman et al. 2013, Holland, Fiore et al. 2015).

Advance care planning should be a reflexive process where clinicians discuss aspects of future care that are of importance to individual patients. There has been suggestion that ACP could be addressed in a group environment, such as pulmonary rehabilitation sessions, and that this would be acceptable to patients (Holland, Fiore et al. 2015). However, evidence from a support group intervention which included such ACP discussions was associated with increased anxiety scores in the intervention group (Lindell, Olshansky et al. 2010). This might indicate unresolved issues arising from discussing ACP in a group environment. A further argument for individual discussion rather than in a group environment is that the appropriate timing of ACP discussions will vary between patients. Clinicians may identify the need for ACP discussions from changes such as new requirement for oxygen, an exacerbation or discontinuation of anti-fibrotic medications, but it remains important to establish whether patients' wish to discuss ACP at this time. A Norwegian focus group study established that patients with chronic respiratory disease preferred to talk about prognosis and future planning at the time of diagnosis and at 'turning points' in the disease trajectory (Hjorth, Haugen et al. 2018). All participants emphasised that an individual approach was mandatory and that healthcare professionals should ask patients about their need for information about ACP. This study is limited by its small sample size as only 13 patients participated in focus groups. Participants had a range of chronic respiratory conditions including COPD, lung cancer and ILD, which makes the results generalisable to all chronic lung disease, but not specific to the views of patients with ILD. However, the conclusions are in line with ILD specific research which has established that patients were apprehensive about asking about the future, but wanted the option of gathering further information and trusted healthcare professionals to judge when it was appropriate to initiate these discussions (Bajwah, Higginson et al. 2013).

Conversations about prognosis remain critically important at the time of acute exacerbations to decide on the course of inpatient management and to ensure patients views on the priorities of treatment are established. However, planned ACP discussions are preferred in the outpatient department with someone who is well known to the patient (Hjorth, Haugen et al. 2018). Patients felt a 'warning shot' prior to discussion of ACP was important to allow mental preparation for the conversation and practically so that family members could be

involved. A further study by Hjorth et al (2020) reviewed the feasibility of introducing ACP on an inpatient thoracic medicine ward. Focusing on present and future symptoms control was felt to be an acceptable way to introduce ACP; and patients, relatives and clinicians showed a positive attitude towards these discussions. However, of the 118 people invited to participate in the study, only 51 chose to participate. The reasons that patients declined to participate were because they felt it was 'bad-timing' as their time was already occupied or they felt too ill, or felt it was 'too demanding'. These responses support the view that ACP discussions should be held primarily in the outpatient department at a time when patients feel mentally prepared for the conversation and include family members if appropriate.

The central question surrounding ACP is whether this leads to an improvement in end of life care. A systematic review concluded that there was a positive impact of ACP on the quality of end of life care (Brinkman-Stoppelenburg, Rietjens et al. 2014). Most of the included studies were observational (107 of 113 studies) and originated from the US (91 studies) where advance care planning practices may differ from the UK. This review indicated that complex ACP interventions were more effective than written documents alone. However, it is noteworthy that few of the included studies considered the congruence between end of life care received and patients' written wishes. A study which considered ACP in ILD found patients who have participated in ACP conversations or had involvement with outpatient palliative care services were subjected to less investigations during their terminal hospital admission (Guo, Mann et al. 2020). ACP was particularly influential in guiding non-specialist clinicians who may be less familiar with the disease trajectory and rely on the recognition of palliation in the community to guide inpatient decisions. Overall, evidence suggests that ACP can improve the quality of end of life care. Discussions must be guided by each individual patient's requirement for information and should be conducted by a trusted healthcare professional to avoid negative impact and patient anxiety.

2.6 Perspectives of patients, carers and healthcare professionals

The previous sections have evaluated the role of specialist palliative care in ILD and issues surrounding accessing these services. The following section reviews literature pertaining to the different perspectives of people with ILD, relatives and carers and healthcare professionals.

2.6.1 Living with PF-ILD (Patient's experiences)

Qualitative studies have provided unequivocal data describing the overwhelming burden of physical and psychological symptoms which are endured by patients with ILD (Bajwah, Higginson et al. 2013, Duck, Spencer et al. 2015, Lindell, Kavalieratos et al. 2017). A qualitative interview study, conducted in two London specialist ILD centres, considered the palliative care needs of patients with ILD and found that breathlessness was an all-consuming symptom; "I'm breathless, always breathless" (Bajwah, Higginson et al. 2013). The study interviewed patients, carers and healthcare professionals at two specialist ILD centres in London. Patients and carers recognised that breathlessness was not an isolated symptom and often co-existed alongside overwhelming anxiety and panic which led to increased distress. This study involved low numbers, only 8 interviews were conducted with patients, and it is unclear whether thematic saturation was achieved. However, the findings are supported by a larger qualitative interview study conducted in Manchester that interviewed 17 patients with IPF (Duck, Spencer et al. 2015). People with IPF struggled with symptoms of breathlessness even on minimal exertion, cough, tiredness and lethargy. They struggled with progressive symptoms as IPF affected all aspects of their lives, experiencing functional limitations and rapid disease progression. These findings are important in understanding the 'illness narratives' of patients with ILD and both studies clearly described the symptom burden experienced by people with ILD.

The progressive deterioration in symptoms and increasing function limitations described by Duck et al (2015) led to people with ILD grieving the 'loss' of their previous life. 'Loss of self' is a well described phenomenon in chronic illness where patients observe their former self-images disappearing without the simultaneous development of an equally valued new versions (Charmaz 1983). People with ILD felt cheated out of their retirement plans and mourned the activities that they enjoyed and in which they could no longer participate (Bajwah, Higginson et al. 2013, Duck, Spencer et al. 2015). Changes in roles and responsibilities around the house were particularly significant to patients, as they were viewed as symbolic of the physical constraints of their illness (Duck, Spencer et al. 2015, Overgaard, Kaldan et al. 2016). These major changes led to people with interstitial lung disease feeling isolated, lonely and lacking purpose as their lives and identities had been consumed by their illness (Bajwah, Higginson et al. 2013). However, a study which focused on the introduction of self-management strategies depicted a very different outlook, with

patients describing feeling positive about their symptom control and concentrating on meaningful activities (Kalluri, Younus et al. 2021). This study recruited patients from a specialist clinic with early integrated palliative care and an extensive patient education programme, a situation which may explain the incongruence of these results.

The high symptom burden experienced by people with ILD was also recognised by a focus group study conducted in the US that aimed to explore the palliative care needs of people with IPF (Lindell, Kavalieratos et al. 2017). This study identified the additional themes relating to frustration around the diagnostic process and the disease education received. Patients gained comfort from the increased resources available in specialist centres but were frustrated by delay in referral to this centre and limited knowledge of ILD in their communities. Confusion about the goals of palliative care was reported and the need for advance care planning was recognised but avoided as it was perceived as a loss of hope. It should be noted that the study included a relatively small number of participants (n=13), and convenience sampling was used from a single centre support group. A number of qualitative studies have highlighted the unmet information needs of people with ILD and that the desired information changed throughout the course of the disease trajectory. When symptoms started and at the time of diagnosis, patients wanted information about their lung disease, prognosis and potential treatment options (Duck, Spencer et al. 2015, Holland, Fiore et al. 2015). As time passed and the disease progressed, patients expressed a need for information about oxygen therapy, nutrition, exercise and management of cough and breathlessness symptoms (Holland, Fiore et al. 2015, Sampson, Gill et al. 2015). Several studies reported a strong desire for more specific information about disease management towards the end of life (Bajwah, Koffman et al. 2013, Holland, Fiore et al. 2015, Sampson, Gill et al. 2015).

2.6.2 The burden on informal carers

Qualitative studies have given important insight into the challenges faced by informal carers of patients with ILD. A focus group study which included 14 carers of people with ILD found that as patients adapted to the limitations imposed by their disease, carers struggled to balance performing their perceived duties as a carer and maintaining their own identities

and independence (Belkin, Albright et al. 2014). This study presented opposing views from carers, some demonstrated a 'we're in it together' attitude and others expressed a resentment towards their partner. What is clear from this study is the burden felt by informal carers who felt obligated to refocus their lives around their partner, one carer explained "Your life [as the carer] keeps getting smaller, which is really difficult" (Belkin, Albright et al. 2014)(page 3). A Canadian study that interviewed eight bereaved carers of patients with ILD also identified the significant strain felt by carers (Pooler, Richman-Eisenstat et al. 2018). Six of the participants worked full-time as well as providing care and expressed fears about leaving their relative at home alone as there were insufficient respite services available. Both studies highlight the importance of informal carers in allowing patients to continue their daily activities and the strain of this caring responsibility.

The CaNoPy study conducted in the UK has provided further appreciation of the role of informal carers in the lives of people with advanced ILD (Sampson, Gill et al. 2015). Carers reported uncertainty about their role in outpatient clinic appointments, hindering both the patients' and their understanding of the disease course and coping strategies. The study makes the important point that clinicians should change their perception of carers as passive observers to people who have an active role throughout the patient pathway. Carers encourage patients to remain active, help with symptom management and pacing of activities, therefore it is important that this active role is appreciated by clinicians in clinic appointments. Desynchrony in the information needs of patients and carers was identified throughout the patient journey but was most apparent in relation to information about end of life planning. Carers often wanted practical advice about how to manage symptoms and what to expect towards the end of life. Whereas patients did not always want to discuss things in this level of detail. These conflicting views about end of life information needs for people with interstitial lung disease demonstrate the difficulty experienced by healthcare professionals when initiating advance care planning conversations.

2.6.3 ***Healthcare professionals' views of palliative care in ILD***

Observational studies have reported patients with ILD were referred to palliative care services late in their disease trajectory (Lindell, Liang et al. 2015, Ahmadi, Wysham et al.

2016). The uncertain disease trajectory in ILD and difficulty with prognostication have been identified as reasons for late referral to specialist palliative care services (Mc Veigh, Reid et al. 2018). Healthcare professionals reluctance to refer to palliative services has been attributed to resignation that patients would invariably suffer poor symptom control, lack of knowledge about the efficacy of symptom control interventions, the discomfort associated with discussing end of life and the fear that this might diminish hope (Bajwah, Higginson et al. 2013, Colman, Curtis et al. 2013).

There is limited research that address healthcare professionals' views on providing palliative and end of life care for patients with ILD. Two surveys of respiratory physicians focused on physician-related barriers to providing palliative care for people with ILD (Akiyama, Fujisawa et al. 2020, Kim, Olive et al. 2020). A survey of UK health professionals established physician-associated barriers preventing referral to palliative care, including concern that patients might feel 'abandoned', disruption of continuity of care, perceived long waiting lists and limited beds (Kim, Olive et al. 2020). However, this was a small survey (36 respondents), limited to one area of the UK with only an approximate 15% response rate. A survey of Japanese respiratory physicians indicated that physicians experienced more difficulty providing palliative care to patients with idiopathic pulmonary fibrosis than those with lung cancer (Akiyama, Fujisawa et al. 2020). Difficulty predicting prognosis, inadequate communication, and discrepancies in understanding of goals of treatment between patients, families and physicians were all associated with difficulty in providing palliative care. This survey included a larger number of respondents (n=130) with a 96% respondent rate. A large proportion (57.7%) of respondents to the UK survey felt that specialist palliative care involvement was unlikely to add anything to their patients' management (Kim, Olive et al. 2020). The majority (71.6%) of respiratory physicians responding to the Japanese survey felt there were few established treatments to relieve patients' symptoms (Akiyama, Fujisawa et al. 2020). These findings suggest inadequate training and awareness of palliative care. This interpretation is supported by a survey of 164 Spanish respiratory physicians that identified insufficient training in palliative care, with only 46% respondents having received specific training (Barril, Alonso et al. 2018).

Palliative medicine is a growing specialty, but the demand for this expertise is often more than can be accommodated by stretched resources resulting in difficulty providing timely access to specialist palliative care. One approach, which would reduce the pressure on

palliative care services, would be following a model of 'general palliative care for all' and specialist palliative care dictated by patients' symptoms (Gaertner, Siemens et al. 2017). This method is supported by other literature that has suggested the referring all patients to specialist services could unintentionally undermine relationships with respiratory teams and cause fragmentation of care (Quill and Abernethy 2013). Respiratory physicians should feel empowered to provide general palliative care for their patients as they are likely to have seen many patients progress along similar, albeit unpredictable, disease trajectories. A survey of respiratory physicians established that incorporating a more defined role in respiratory palliative care was felt to be a desirable evolution of professional responsibility (Goodridge and Peters 2019). This survey identified some barriers to respiratory doctors providing palliative care, namely lack of training, uncertain disease prognosis and lack of time. These are common barriers reported in relation to chronic lung disease and may account for the reliance on palliative care specialists to provide palliative care (Sørensen, Marsaa et al. 2020).

2.7 End of life care in ILD

The previous section has described the patient and carer experience of living with ILD and the perspectives of healthcare professionals relating to palliative care in ILD. The next section reviews literature relating to the final weeks and days of life for people with ILD, focusing on symptom control and location of death. There is a paucity of research focused on end of life care for patients with ILD. It is known that symptoms deteriorate towards the end of life and that the majority of people with ILD die in an acute healthcare setting (Palmer, Kavanagh et al. 2021).

2.7.1 Symptom control at the end of life

Most people would agree that the ideal process of dying is when the patient is comfortable and their family are present (LACDP 2014). Symptom control is paramount to achieving good end of life care. People with ILD have a high symptom burden and their symptoms progress rapidly towards the end of life (Rajala, Lehto et al. 2018). Breathlessness was the most reported symptom in end-stage disease (66 – 93% patients) (Bajwah, Higginson et al. 2012, Ahmadi, Wysham et al. 2016, Rajala, Lehto et al. 2016, Rajala, Lehto et al. 2018). Other commonly reported symptoms included chest pain, anxiety and depression (Ahmadi,

Wysham et al. 2016, Carvajalino, Reigada et al. 2018). Good symptom control at the end of life in ILD is achievable, however, this often relies on pharmacological treatments to manage symptoms of breathlessness and anxiety. Two studies, based in Finland and Australia, have shown that pharmacological management of end of life symptoms was common with 71%-94% patients receiving opioids and 44%-73% benzodiazepines in the last week of life(Rajala, Lehto et al. 2016, Smallwood, Mann et al. 2021). A UK retrospective observational study reported that 100% of patients who received opioids or benzodiazepines for breathlessness had a documented improvement in their symptoms (Bajwah, Higginson et al. 2012). These three studies all referred to patients who died within a hospital environment where medications were easily accessible and likely to be administered without delay. There is a lack of published research relating to symptom control at the end of life for people with ILD who died in the community.

A stark contrast has been reported in the end of life care received by patients with ILD compared to those with lung cancer. Two studies, conducted in Sweden and Japan, identified that patients with ILD were more likely to experience unrelieved breathlessness at the end of life and had reduced access to specialist palliative care services despite their higher symptom burden (Ahmadi, Wysham et al. 2016, Koyauchi, Suzuki et al. 2021). An American study reviewed end of life care in ITU for people with chronic lung disease compared to metastatic cancer (Brown, Engelberg et al. 2016). This study identified that patients with chronic lung disease had access to fewer palliative care services and had a longer length of stay prior to their death. Documented review of breathlessness and chest pain did not occur as frequently at the end of life compared with patients with cancer (Ahmadi, Wysham et al. 2016, Brown, Engelberg et al. 2016) and ILD patients were more likely to have unrelieved symptoms of breathlessness, pain and anxiety (Ahmadi, Wysham et al. 2016). The deficit in documentation and higher level of unresolved symptoms may indicate other problems with the quality of end of life care for ILD patients. Overall, these studies depict a poorer quality of end of life care for people with ILD compared to those with lung cancer, with inadequate assessment of symptoms and poor symptom management.

Healthcare professionals have observed their sense of failure about being unable to adequately manage patients symptoms: “a sense of sort of hopelessness that nobody could actually do anything about it and I thought there were probably few worse ways to die than when I saw it in that instant” (Bajwah, Higginson et al. 2013)(page 873). Whereas it is

notable that poor symptom control was not a significant theme emerging from two qualitative studies which captured the experiences of bereaved caregivers (Pooler, Richman-Eisenstat et al. 2018, Egerod, Kaldan et al. 2019). Following palliative care involvement and advance care planning, carers felt able to manage predictable symptoms at home and none of them reported 'crisis' symptoms (Pooler, Richman-Eisenstat et al. 2018). However, the participants involved in this study had all been involved in multidisciplinary clinic care and may be a self-selected group of participants who had a positive experience and felt well supported by palliative and community services.

2.7.2 Location of death

Place of death was historically used as a surrogate marker for the quality of end of life care (Wright, Keating et al. 2010, Wang, Wu et al. 2016). The validity of using place of death as a quality marker has been questioned and more recently research has focused on establishing alternative methods for assessing the standard of end of life care (Hoare, Antunes et al. 2022). Established literature has suggested the majority of people wish to die in their own home (Gomes, Calanzani et al. 2013). However, this information was based on population studies reporting the hypothetical preferences of healthy individuals. Where data has been collected from patients or their caregivers, it is recognised that people change their mind about preferred place of end of life care at different points along their disease trajectories (Evans, Cutson et al. 2006). Deterioration in symptoms was the most common reason for patients and carers changing their preferences and leaving home (Rainsford, Phillips et al. 2018). It is important to remember that preferred place of care and death often changes with time and increasing burden of symptoms and requires review alongside disease progression. Routine discussion and documentation of patients' end of life preferences must occur to enable these preferences to be achieved.

The majority of people with ILD died in hospital, with studies reporting a range of between 57% and 80% of patients dying in an acute healthcare environment (Bajwah, Higginson et al. 2012, Ahmadi, Wysham et al. 2016, Rajala, Lehto et al. 2016, Higginson, Reilly et al. 2017, Zou, Nouraie et al. 2019, Guo, Mann et al. 2020). Observational studies have shown the number of patients dying with ILD in acute settings has decreased (Higginson, Reilly et al. 2017, Cross, Ely et al. 2020), which correlates with the trends in other chronic lung diseases. Independent factors which increase the likelihood of dying in hospital are an increasing

number of co-morbidities, increasing age, deprivation and living in rural areas (Higginson, Reilly et al. 2017). These findings relate to other studies which found death at home was more likely for patient who were older, married, living in rural areas and higher level of education (Zou, Nouraie et al. 2018, Cross, Ely et al. 2020). There is no evidence that patients with ILD have different end of life preferences to other diseases and when preferred place of death is ascertained, patients predominantly wish to die at home (Archibald, Bakal et al. 2020). Dying at home allowed patients to be more coherent and active and allowed extension of 'normal life' for as long as possible (Pooler, Richman-Eisenstat et al. 2018, Egerod, Kaldan et al. 2019).

People with ILD often experienced a significant deterioration in their symptoms towards the end of life leading to increased acute healthcare utilisation. A study conducted in Finland reported the final months of life for ILD patients were dominated by worsening symptoms, treatment for exacerbations and acute hospital admissions (Rajala, Lehto et al. 2016). Many patients were admitted to hospital during their final illness and received treatment for an acute exacerbation with antibiotics. A quarter of patients underwent investigations in the last 24 hours of life. These findings are supported by an Australian study reporting that many patients with ILD (36%) underwent diagnostic investigations in the final days of life. The implication from these data is that patients with ILD were not recognised as dying or deteriorated quickly and that the hope of improvement remained until the very end of life. However, criticisms of both studies include the retrospective design meaning that detailed information about patient reported symptoms was unavailable, and the inclusion of only 59 and 67 patients respectively, which is a relatively small sample size for observational studies. These studies highlight the burden of investigations and treatments experienced by people with ILD towards the end of life. However, given the unpredictable nature, acute deterioration in symptoms and high mortality from acute exacerbations it is not surprising that many people with ILD die in hospital.

There is a significant physical and psychological commitment required by informal carers to enable their relative to die at home. An observational US study which reviewed the location of death for patients with chronic lung disease found married patients were more likely to die at home (Cross, Ely et al. 2020), eluding to the carer responsibility that is taken on by relatives and especially partners when a person receives end of life care at home. Carers described a strain on relationships and the weight of responsibility when caring for relatives

towards the end of life (Pooler, Richman-Eisenstat et al. 2018). However, carers reported a sense of achievement and gratification from keeping their family member out of hospital and the end of life in accordance with their wishes. This sense of achievement and closure can only be found if patient's end of life care wishes were known, allowing planning and support to be in place for their relatives. Patients were more likely to spend the last days of life at home if their caregiver engaged in advance care planning (Archibald, Bakal et al. 2020). If patients die at home, the presence of a community nurse provided comfort for their partner and allowed them to let go of the carer responsibilities at the end of life (Egerod, Kaldan et al. 2019). This essential support can be difficult to achieve due to the unpredictable nature of ILD and the lack of community palliative care services which may lead to unplanned hospital admission at the end of life.

2.8 End of life research

End of life research poses many challenges due to the sensitive nature of the topic and ethical and practical issues surrounding working with very unwell people and their families. High rates of attrition and low levels of participation in palliative and end of life research have been widely recognised in literature (Addington-Hall 2002, Pleschberger, Seymour et al. 2011, Samuels, Schoppee et al. 2021). Major barriers affecting recruitment of participants for palliative care research included difficulty locating eligible patients, severity of illness, family and provider protectiveness, seeking patients in multiple settings and lack of resources to aid recruitment activities (Hanson, Bull et al. 2014). Practical barriers, such as travel costs, feeling too unwell to participate and lack of time due to ongoing medical appointments, have also been identified as barriers preventing people participating in research towards the end of life (Chatland, Harvey et al. 2021).

Patients involved with palliative care services are vulnerable for a variety of reasons; they are often dependent on others for care, they may be at increased risk of adverse effects from experimental therapies, and their ability to make an informed choice about involvement in research may be reduced due to cognitive impairment (Addington-Hall, Bruera et al. 2009). However, this vulnerability does not mean that patients with advanced disease should be excluded from participation in research. A systematic review that aimed to establish the perspectives of dying people about participating in research concluded that participation in end of life research was valued by dying people, who regarded their

involvement as important, particularly if it provided an opportunity to help others (Bloomer, Hutchinson et al. 2018). This review established that dying people recognised the value of research through its contribution to science and understanding and its potential personal benefits, however, the desire to helps others was the main influence on people participating in research. This review only included 10 studies, having excluded those which did not specifically state that the participants were dying, and a such may not represent the views of many participants in palliative care research. However, there remains general agreement in the literature that patients valued participation in research, as a way to engage in meaningful activity and be a person beyond their illness, and the main incentive was altruism and a desire to help others (Terry, Olson et al. 2006, Bellamy, Gott et al. 2011, Chatland, Harvey et al. 2021).

2.9 Gaps in the evidence base

Providing good palliative and end of life care in ILD is established as an important objective for ILD clinicians. The recognition of the high symptom burden experienced by patients has resulted in improved integration of respiratory and specialist palliative care services to overcome barriers and provide early access to palliative care. Holistic management of symptoms and integration of services has been shown to improve symptom control, quality of life and satisfaction with care.

However, there are unanswered questions surrounding the end of life care of patients with ILD. Most patients with ILD die in an acute care setting, despite evidence that suggests that many patients would prefer to die in their own home. This may be related to low levels of advance care planning among patients with ILD which mean that end of life care preferences were unknown. The unpredictable disease trajectory and potential rapid deterioration in clinical condition may mean that death in an acute setting is difficult to avoid if patients want active management of exacerbations. Preferred place of care is known to change as patients' symptoms deteriorate and it is possible that patients actually chose to be admitted to hospital for end of life care due to distressing symptoms of severe breathlessness, anxiety or panic or the requirement for high levels of oxygen which cannot always be provided in the community. Although many patients with ILD died in hospital, it is not known whether this was their preferred place of death or whether family members feel that they died in an appropriate place. Previous research has suggested geographical disparity in patients' access

to specialist palliative care services, leading to questions about whether the quality of end of life care is different depending on where patients live. Healthcare professionals provide valuable insight into the level of services in different areas and understanding factors which influence patients' access to specialist palliative care and good end of life care.

The most important unanswered questions surround whether the location of death was considered to be appropriate and whether families felt that good symptom control at the end of life was achieved. An appropriate place of death would enable a 'safe' death in respect to the level of medical care required and not place excessive strain on carers. There is a clear difference between patients dying in A&E or intensive care versus patients who die in a side room on a ward with input from palliative care teams in the hospital. Patients, bereaved relatives and healthcare professionals will have their own views on what level of medical involvement and support is appropriate towards the end of life. It is also important to consider the strain on relationships and psychosocial burden that can result from relatives providing end of life care at home. The more important question to benchmark the success of palliative care interventions is whether the patient died in an appropriate and safe place, rather than whether they died in their own home. This research project aims to address these highlighted gaps in knowledge through interviews with patients, bereaved carers and healthcare professionals.

2.10 Research concept (aims and objectives)

The aim of this research study was to address the gap in the literature by developing comprehensive understanding of the experiences of end of life care amongst people with ILD. The research questions were formulated from a research gap identified due to concerns highlighted by patient groups within the region and the review of contemporary literature. The initial aim of the study was to enhance understanding of factors which affected the quality of end of life care in this patient group. Over time, this aim was developed and centred around three research questions:

- What is the end of life experience of people dying with ILD?
- What factors influence patients' and relatives' satisfaction with end of life care?
- Are there any perceived barriers preventing access to palliative care services in the North East and Cumbria?

To effectively answer these questions, data was generated from three key stakeholder groups; people with ILD, bereaved relatives of people with ILD and healthcare professionals involved in caring for people with ILD. Interviews with patients aimed to capture the experience of people with end stage ILD who were managing their symptoms and planning for the future. Interviews with bereaved relatives of people with ILD gave a unique perspective on the final days and weeks of patients' lives and about their own experience of their relative's death. Healthcare professionals were interviewed to establish if there were barriers preventing patients accessing palliative care or dictating how end of life care was delivered in different areas of the region.

2.11 Chapter summary

This chapter has presented a review of the current literature and explored the role of specialist palliative care for people with ILD. The challenges faced in providing timely palliative care were outlined including factors that influenced referral to specialist palliative care services. The chapter also considered the perspectives of patients, carers and healthcare professionals in relation to living with ILD and caring for people with ILD. This critique of the literature highlighted a paucity of research concerning end of life care for people with ILD demonstrated the need for this research and the focus of this thesis. The next chapter presents the methodological approach, outlines the rationale for important decisions made in the research process and explains in detail how the study was undertaken.

Chapter 3 Methodology and Conceptual Framework

3.1 Introduction

This chapter presents the methodological approach used in this research project. It begins by outlining the research concept and the influence of this on determining the research question, guiding the methods chosen and the analytic approach. My own epistemological and ontological viewpoints are explained and the influence of my clinical background and perspectives is reviewed. The chapter explains the justification for choosing qualitative methodology and adopting a constructivist grounded theory approach. The research process is explained in three stages; i) preparing to enter the field, ii) conducting fieldwork, iii) working with the data. This chapter concludes with a discussion of trustworthiness and rigour in relation to qualitative research and explains how these requirements were met in the current study.

3.2 Theoretical approach

3.2.1 Epistemology and Ontology

Quantitative and qualitative research methodologies are often portrayed as representing very different epistemologies and ontologies. Epistemology refers to theories about knowledge and what constitutes evidence (Barbour 2014). This encompasses the principles and rules by which knowledge can be demonstrated and decisions about how social phenomena can be known (Mason 2018). Ontology refers to our views on what constitutes the social world and how we can study it (Barbour 2014). The ontological and epistemological perspectives of the researcher are essential determinants of how a research phenomenon is approached by social researchers (Denzin and Lincoln 2018). It is therefore important to outline the beliefs of the researcher and how the chosen research methodology aligns with their viewpoint, as these will inevitably influence the conclusions drawn from the research data. This section discusses the differing research paradigms and concludes in a description of my own theoretical perspective.

3.2.2 Research Paradigms

The term research paradigm encompasses the set of common beliefs and agreements shared between scientists about how problems should be understood and addressed (Kuhn 1962). The research paradigm can be characterised by the response to three essential questions: the ontological, the epistemological and the methodological questions see Figure 3-1.

1. Ontological: What is the nature of the "knowable"? Or what is the nature of "reality"?
2. Epistemological: What is the nature of the relationship between the knower (the inquirer) and the known (or knowable)?
3. Methodological: How should the inquirer go about finding out knowledge?

Figure 3-1 Generating Inquiry Paradigms (Guba 1990)

It consists of a framework of concepts, theories, standards and methods which outline what constitutes a legitimate contribution to the distinct scientific field (Pope and Mays 2020). Two research paradigms which are frequently presented as distinct and opposing perspectives are positivism and constructivism/interpretivism. Barbour (2014) argues that in practice many researchers borrow from more than one tradition or approach and that the distinction between these approaches is not as clean or neat as it is frequently portrayed.

Positivism is the dominant research concept underlying clinical and basic sciences research. The basic belief system of positivism is a 'realist ontology', i.e. the belief that a single reality exists which is driven by indisputable natural laws (Guba 1990) and empiricist epistemology that we can know and understand phenomena by observation (Pope and Mays 2020). "Positivism attempted to replicate the natural sciences in seeking to control the natural world, through establishing universal laws, which could be further tested through a set of scientific rules of method" (Barbour 2014). This paradigm aligns with quantitative research methodology where the researcher is objective, and variables are recognised and controlled for in the research design. A Positivist approach was not appropriate for this study due to the complexity of the research question and the numerous factors which are important in influencing the quality of end of life care. Positivism can be problematic for studying the

views and opinions of research participants who are capable of self-reflection and can change their behaviour and opinions (Pope and Mays 2020).

In contrast, interpretivism/constructivism highlights the contingent nature of knowledge and reality and argue that there is no distinct objective reality (Barbour 2014). These paradigms favour relativist ontological beliefs that "realties exist in the form of multiple mental constructions, socially and experimentally based, local and specific and dependent for their form and content on the persons who hold them" (Guba 1990)(page 27). The subjectivist epistemology assumes that the researcher is inherently part of the object being investigated and that it is important to understand the phenomena from the viewpoint of those being studied (Pope and Mays 2020). The influence of the researcher is acknowledged with recognition that knowledge generated during an interview is the co-creation of the researcher and participant. An interpretivism/constructivism research paradigm was adopted in this study, encouraging the researcher to reflect upon their subjective understanding and potential biases.

The following section will outline my theoretical perspectives and how these support the chosen research methodology.

3.2.3 Researcher's theoretical perspective: Critical realism

The concept of critical realism emerged in the 1970s – 1980s associated with the work of Bhasker (1975) (1989) and was further advanced by other scholars in this tradition (Archer 1998). Critical realism combines realist ontology (the belief that a real world exists independently from our beliefs and constructions) with constructivist epistemology (our understanding of the world is influenced by our own perspectives and specific viewpoint) (Maxwell 2012). This approach allows for acceptance that there is an objective external reality, whilst acknowledging that much of the human reality is a social construction and therefore dependent on a person's specific standpoint. "Critical realism rejects the theory of 'multiple realities', in the sense of independent and incommensurable worlds that are socially constructed by different individuals, but is compatible with the view that there are different valid perspectives on reality" (Maxwell 2012). Realism supports the concept that an individual's social and physical circumstances influence their beliefs and perspectives (Maxwell 2012).

I hold firm realist ontological beliefs about the world owing to my background in clinical medicine. However, medicine teaches us that although two patients may have the same pathological disease, their experiences of their illness are shaped by their own beliefs, perspectives, and social situation. Therefore, I also maintain a constructivist epistemological position which appreciates an individual's experience of the world from their perspective and unique situation. The chosen methodology (described below) aligned with this ontological and epistemological standpoint and allowed exploration and understanding of patients', carers' and healthcare professionals' unique experiences.

The study described here focused on perceptions of end of life care for people with ILD; it was concerned with experiences of accessing or delivering end of life care and perceived problems with services. Reality in this research is therefore the perspective of the participants and gives priority to their individual experiences and the meanings that they attach to things.

3.3 Research methodology

3.3.1 Rationale for constructivist grounded theory

Qualitative research methods are generally best suited to answer questions which seek to understand human behaviour, opinions and experiences; information which is difficult to obtain from quantitative methods of data collection (Guest 2013). Good end of life care is a complicated construct involving many different components which would be difficult to identify and measure using quantitative methodology. Therefore, this study used interpretative qualitative methodology to answer the research questions.

The literature review described in chapter two revealed few publications which considered the quality of end of life care for people with ILD, demonstrating a paucity of knowledge in this area and the need for explanatory theory. The exploratory and inductive style of constructivist grounded theory is appropriate when little is known about the topic with the aim to generate new knowledge in the form of theory (Mills 2015). Constructivist grounded theory (Charmaz 2014) was chosen as an appropriate methodology for this study with the aim of building substantive theory about end of life care for patients with ILD.

3.3.2 Grounded theory approaches

Grounded theory was first described by Glaser and Straus (1967) in their seminal text *The Discovery of Grounded theory*. At this time, the dominant research paradigm was positivism using quantitative research methods to verify theory after logical deductions were made based on testing hypotheses (Denzin and Lincoln 2018). Glaser and Strauss presented revolutionary systematic methodological strategies which advocated developing theories emerging from qualitative data rather than deducing hypotheses from existing theories (Charmaz 2014) . Their method offered a robust and structured approach to qualitative research which aligned with the positivist paradigm of the time.

Since this first description of grounded theory, Glaser and Strauss have taken grounded theory methodology in divergent directions. *Classic* grounded theory refers to Glaser's view of the researcher as an objective, neutral observer who gathers data to discover theory as an external reality (realist ontology, objectivist epistemology), whereas Strauss adapted this methodology to allow for the acceptance of multiple realities (relativist ontology, interpretivist epistemology)(Corbin 2008). Grounded theory has been developed by many scholars to move away from the positivist stances used in these earlier methods. Charmaz (2014) developed a constructivist grounded theory approach which adopts the inductive, comparative, emergent and open-ended style of the original description by Glaser and Strauss. However, the constructivist approach recognises research as a construction, acknowledges that it occurs under specific conditions and that the researcher's position, perspective and interactions are an inherent part of the research reality (Charmaz 2014). The view that the research is constructed rather than discovered encourages the researcher to reflect on their actions and decisions when generating and analysing the data.

The prerequisite to *classic* grounded theory, that the researcher should not have prior theory based on existing knowledge, is impossible to reconstruct in healthcare research undertaken by researchers who are specialists in the field being studied. Charmaz (2014) agrees that a researcher approaching a project with a 'blank slate' is no longer a realistic proposition. Frequently, funding applications and ethical approval processes require a literature review at the outset of a research project. Engagement and understanding of core

literature can be useful, rather than a hindrance, providing the researcher ensures they maintain an open mind when analysing their own data (Dunne 2011). Strubing (2007) advises that prior knowledge and experience gained from clinical work should be used effectively in the application of essential grounded theory methods. Acknowledging existing assumptions, experience and knowledge of the research area is an effective method of establishing the researcher's own perspective in relation to the research study (Mills 2015) and helps to avoid the researcher imposing their own preconceptions on the developing theory. Constructivist grounded theory methodology was chosen for this study as this approach both acknowledges the theories which motivated the choice of research question and allow for theories to be developed by review of relevant literature. These different versions of grounded theory agree on the core principles and actions which underpin grounded theory methodology. Charmaz (2014) views the major versions of grounded theory as constituting a constellation of methods, rather than an array of different methods. Birks and Mills (2015) suggest the following to constitute a set of essential grounded theory methods:

i. Initial coding and categorization of data
ii. Concurrent data generation and analysis in an iterative process
iii. Writing memos
iv. Theoretical sampling
v. Constant comparative analysis
vi. Theoretical sensitivity
vii. Intermediate coding
viii. Identifying a core category
ix. Advancing coding and theoretical integration

Figure 3-2 depicts the grounded theory process from the point of development of the research question to completion and dissemination.

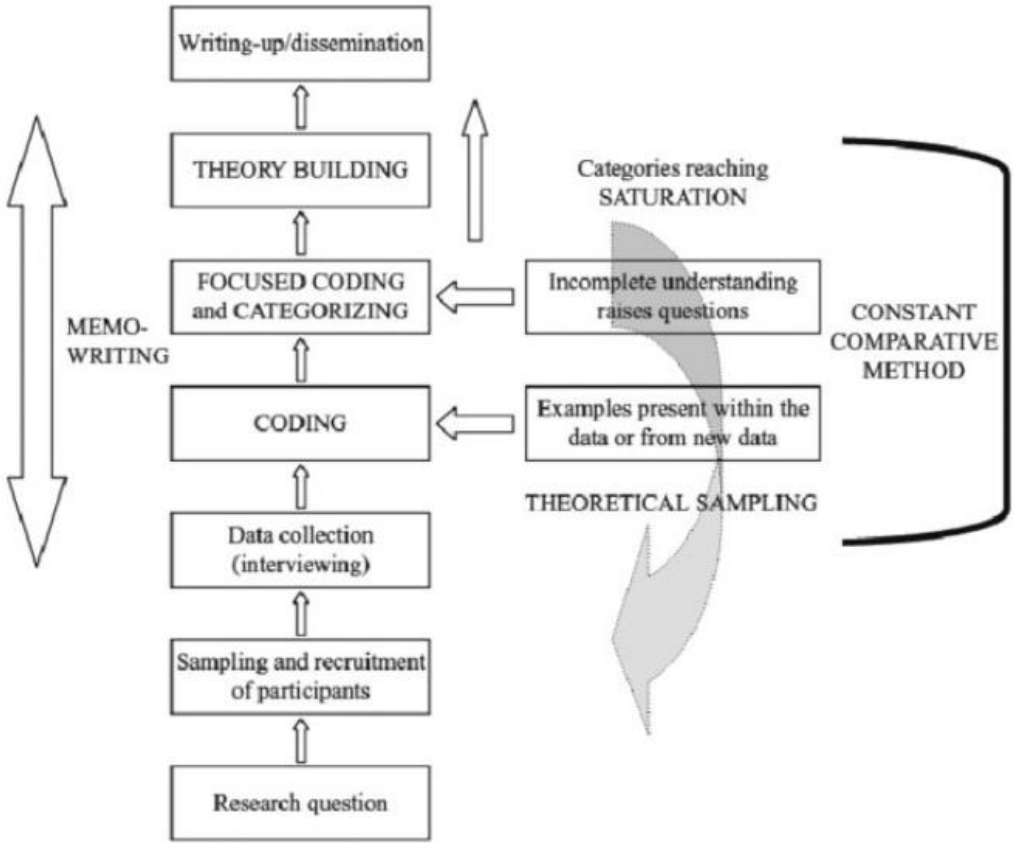

Figure 3-2 A visual representation of Grounded theory (Charmaz 2014)

Fundamental to grounded theory research is the process of concurrent data generation and analysis (Mills 2015). To achieve this the researcher collects data from an initial purposive sample and data from these early encounters is coded and analysed before more data is generated and the process is repeated. This allows analysis to inform the collection of subsequent data through theoretical sampling. It is this principle of concurrent data generation and analysis which differentiates grounded theory from other research methodologies.

In the sections below I set out the research process undertaken in this study, organised into three key stages i) preparing to enter the field, ii) conducting fieldwork, iii) working with the data.

3.4 Stage one: preparing to enter the field

3.4.1 Ethical approval

Ethical approval was sought from the Health Research Authority (HRA) and the study application was reviewed at the West Midlands Solihull Research Ethics Committee meeting on 14th October 2020 (REC reference 20/WM/0267). The integrated research application system form is included in the appendices. A favourable opinion was issued by the Research Ethics Committee subject to specific changes made to the protocol and patient facing documents. HRA approval was granted on 16th November 2020 (Appendix A). Following HRA approval, Newcastle Hospitals NHS Foundation Trust agreed to sponsor the study and Confirmation of Capacity and Capability was finalised on 14th December 2020. Recruitment to the study began on 1st January 2021.

3.4.2 Ethical considerations

There were several ethical considerations in relation to this research study; these were considered as anticipated risks to either the participants or to me as the researcher. The main anticipated risk to participants was causing undue upset and stress when talking about sensitive subjects during interviews. Research which considers issues around death and dying is judged a sensitive topic. Topics that intrude into a participants 'private sphere' or delve into deeply personal experiences are considered to be sensitive research topics (Lee 1993). Research regarding death and dying may not be regarded as private, but may be emotionally taxing for participants (Lee 1993, Dickson-Swift 2008). Brannen (1988) argued that when researching sensitive topics, it is important to allow participants to define the problem in their own terms, rather than prejudging and labelling or defining the issues too precisely. The qualitative research methodology applied in this study allowed the exploration of sensitive topics with the participant controlling what was discussed and allowed them to guide the interview towards the areas they felt were most important.

The main commitment from the participants was time. The interviews were scheduled to last 30-45 minutes for patient and bereaved relative participants. Participants were advised prior to the interviews that they could pause or end the interview at any time and restart or reschedule as necessary. Participants were advised that they did not have to answer questions that made them feel uncomfortable, and if there were topics they would rather

not discuss this could be agreed prior to starting the interview. I adopted a policy of reaffirming consent throughout the interview and the interview would have been ended if the participant was no longer willing or able to give consent. In the event of participants experiencing anxiety or distress due to the interview topics, follow up and support was made available with appropriately trained staff from their usual clinical team or bereavement support services.

Another risk to participants related to protection of their identifiable information to preserve their anonymity and confidentiality. The participant information sheet advised participants that the information provided was kept strictly confidential and was stored securely. Participant identifiable information (e.g., names and address) were removed from the interview transcript and the original digital recordings were destroyed once transcription was completed. The transcribed interview data were referred to by a unique reference code or the purpose of analysis. All research data, documents and reports (including digital recordings) were manged and stored in compliance with Caldicott guidelines (Caldicott 2013), the Data Protection Act (Mullock 2000) and local policies and procedures.

I worked as a respiratory doctor within the regional ILD service during the time I was undertaking this research project. However, I did not have significant direct contact with the patient or bereaved relative participants through my clinical work. This approach was adopted to avoid influencing data generation in the interviews or coercion of participants to be involved in the study. Several of the healthcare professional participants were my colleagues and well known to me through my clinical work.

3.4.3 Inclusion and exclusion criteria

The inclusion and exclusion criteria were used to determine participants' eligibility to participate in the study. The inclusion and exclusion criteria are detailed in Table 1.

Inclusion Criteria	**Exclusion Criteria**
People with advanced interstitial lung disease with diagnosis confirmed by regional multi-disciplinary team.	Under 18 years of age Unable to give informed consent.

Bereaved relative of a person who had died from interstitial lung disease in the past 12 months. Healthcare professional who had direct involvement in the care of patients with interstitial lung disease Adult aged over 18 years	

Table 1 Inclusion and exclusion criteria

3.4.4 Sampling strategy

The research sample comprised of people with ILD, bereaved relatives of people who had died with ILD and healthcare professionals who worked within the discipline of ILD. Qualitative methodology does not require a pre-determined assessment of sampling size, instead data generation is completed when a judgement is made related to the concept of data saturation (see section on theoretical saturation 3.4.5). Suggestions of appropriate sample sizes vary in the literature from 20-30 interviews (Creswell 2017) to 30-50 interviews (Morse 1994).

The initial sampling technique used in the study was purposive sampling. Purposive sampling involves identifying and selecting individuals who are knowledgeable about or interested in the phenomenon of interest (Creswell 2017). The focus of this study was exploring the end of life care for people with ILD and identifying potential barriers to accessing palliative care services. The emphasis was on identifying participants who provided 'maximum variation' with the intent that important shared patterns would be identified which cut across cases and derived significance from having emerged out of heterogenicity (Palinkas, Horwitz et al. 2015). For patients and bereaved relatives this meant that participants were invited to participate who were likely to have different experiences or perspectives, e.g., died at home or in hospital, specialist palliative care involvement, expected or unexpected death. The timing of decedents' death in relation to the coronavirus pandemic and social distancing restrictions was identified as a further factor which needed to be addressed through purposive sampling. Healthcare professionals who were involved in the care of patients with ILD were invited to participate. Purposive sampling was employed to sample a mixture of respiratory clinicians, specialist nurses and palliative care clinicians who worked in different geographical locations in the region.

Theoretical sampling was undertaken following the collection and analysis of preliminary data obtained through the purposive sampling. Theoretical sampling is the hallmark of constructivist grounded theory methodology and is the essential method responsible for making the process emergent (Mills 2015). In contrast to conventional methods of sampling as it is driven by emerging categories and gaps in the analysis and is responsive to the data, not determined in advance based on a predefined theoretical framework. Charmaz (2014) argues that theoretical sampling is only possible once preliminary categories have been identified and is used as a technique to develop and refine these categories. Theoretical sampling can be undertaken by selecting participants and by modifying the questions asked in data generation to explore gaps in knowledge, clarify uncertainties, test interpretations of the data and develop emerging theory (Sbaraini, Carter et al. 2011). In this current study, data from the initial interviews were analysed and based on the findings further participants were approached for participation in the study who either contributed to data emerging on potential patterning or attempt to cover gaps in the knowledge.

Coding following the initial interviews with people with ILD and bereaved relatives, identified a number of related initial codes which focused around wanting a peaceful death (Table 2). Theoretical sampling was utilised to explore the concept of a peaceful death in more depth in subsequent interviews through revision of the interview questions. Recruitment was targeted at bereaved relatives of people who had died in non-acute medical environment to allow further exploration of the potential association between location of death and the notion of a peaceful death. This theory directed sampling maximised the opportunity for the concept of peaceful death to be examined from various angles and from the viewpoint of multiple participants.

Participant	**Initial coding**	**Focused coding**	**Theoretical sampling**
BR_01	Struggling to breathe and frightened	Wishing for a peaceful end	Recruitment of bereaved relatives of people who had died in different healthcare environment i.e., hospice, to explore impact of location on achieving a peaceful death.
BR_02	Died 'the way he wanted'		
PT_03	Wanting to be		

	comfortable		Revised interview schedule for patients to ask about priorities at end of life and specific question for bereaved relatives about how/where their relative wanted to die.
PT_02	Not wanting to be alone		

Table 2 Example of theoretical sampling

Table 3 provides a further example of theoretical sampling in relation to data generated from interviews with healthcare professionals. Preliminary interviews with healthcare professionals identified several related initial codes centred around the comparison of the care of people with ILD to those with other diseases. In general, these comparisons were made in relation to access to community services for patients with COPD and malignancy. Theoretical sampling was employed to explore the concept of disparity of care compared to other diseases by selective recruitment of healthcare professionals who had experience of interaction with community palliative care services in different regions. The interview schedule was revised to incorporate questions about the view of palliative care for people with ILD compared to other diseases.

Participant	**Initial coding**	**Focused coding**	**Theoretical sampling**
HCP_01	Limited access to palliative care services	Comparing with care in other diseases	Recruitment of HCPs who had experience of interaction with community palliative care teams throughout the region, i.e., respiratory specialist nurse from rural hospital and palliative care doctor from south of region. Revised interview schedule to include questions about view of
HCP_02	Lack of knowledge of ILD in community		
HCP_04b	Palliative care origins in cancer care		
HCP_05	Difficulty accessing services for non-malignant disease		

			palliative care access for patients with ILD vs other conditions.

Table 3 Example of theoretical sampling

Theoretical sampling guided the questions used to generate data in successive interviews to interrogate the key concepts emerging from the data. This was reflected in the evolving questions included in the interview schedules (Appendix C). Theoretical sampling allowed progression of the analysis, providing the opportunity to define categories, delineate the properties of categories and clarify the relationship between categories (Charmaz 2014). This sampling approach allowed the evolving main categories to develop, and relationships were identified which led to the emergence of the core categories.

3.4.5 Theoretical saturation

Theoretical saturation is defined as the point "when gathering fresh data no longer sparks new theoretical insights, nor reveals new properties of your core theoretical categories" (Charmaz 2014)(page 113). This definition was intended for use specifically in relation to building a theoretical model and the broader term 'data saturation' has been adopted to reflect the wider application of the term and concept in qualitative research (Guest, Namey et al. 2020). Data saturation is used to describe the point in data collection and analysis where new data produces little or no new information to address the research question (Guest, Namey et al. 2020), and is used as a marker that data collection is adequate. Data saturation is often referred to as a 'point' suggesting that it is a precise event which can be recognised by the researcher (Saunders, Sim et al. 2018). However, it can be argued that new data, especially that which is theoretically sampled, will always contribute additional knowledge, but that there is a point of diminishing returns from continuing data collection (Mason 2010).

The use of theoretical saturation as an indication of completed data generation in constructivist grounded theory remains contentious. Dey (2007) critiques the use of the term 'saturation' as being incongruent with a method where not all data is coded and instead relies on the researcher's assumption that the properties of categories are saturated. Data saturation is frequently adopted as marker of sampling adequacy, however,

accepting saturation as a generic quality marker is inappropriate (O'Reilly and Parker 2012). An alternative suggestion is that sample size should be decided from 'information power' where the amount of information a sample provides on the topic of interest dictates how many participants are required (Malterud, Siersma et al. 2016). This approach is congruent with Charmaz's thoughts that theoretical saturation is achieved at different points depending on the complexity and originality of the research question (Charmaz 2014). Despite the criticisms of theoretical saturation, it remains the conventional approach used to define when data collection is adequate in constructivist grounded theory research and therefore has been used in this study.

3.5 Stage two: conducting fieldwork

3.5.1 Sample recruitment

The identification and recruitment of patients and bereaved relatives took place at the North East regional interstitial lung disease service. This site was chosen for recruitment as it is a regional referral centre, covering a large geographical area, including both densely populated urban areas and rural/isolated areas. There was scope within this patient population to obtain data from participants with different aetiologies of interstitial lung disease from varied geographical areas and socio-economic backgrounds. An initial purposive sample (see section 3.4.4) were sent an invitation letter, and participant information pack. Theoretical sampling was undertaken subsequently following initial data generation (see section 3.4.4).

Initial contact with participants was indirect and made via the regional ILD service. An invitation letter and participant information pack were sent to potential participants who were then required to return their completed consent form to be involved in the study. An opt-in approach is generally favoured in end of life research (Hanratty, Lowson et al. 2012), but this can lead to a reduction in sample size (Ewing, Rogers et al. 2004, Hunt, Shlomo et al. 2013). End of life research has observed higher rates of recruitment when the participants received a recruitment phone call (Stiel, Heckel et al. 2015). However, Stiel and colleagues primarily collected quantitative data via a questionnaire and therefore did not require participants to share the same level of personal detail as in an interview. I considered

contacting potential participants via phone following receipt of the information pack, however, I felt this was not appropriate given the sensitive topic and that a phone call might be perceived as intrusive and introduce coercion.

Bereaved relatives were contacted after three months and within a year of their relatives' death. There is debate about the most appropriate time to approach grieving relatives about participation in research. Existing literature suggests that most relatives felt able to participate in research interviews in the first five months following bereavement (Bentley and O'Connor 2015). The opt-in approach implemented for recruitment in this current study allowed bereaved relatives to decide if and when they felt they wanted to be involved in the study.

Patients with end-stage disease were difficult to identify from clinical databases therefore, instead I decided to prospectively identify patients who had advanced disease from their review in the regional clinic. Potential participants were identified by ILD consultants who were not involved in data collection to prevent any coercion or confusion arising from me acting in dual professional roles for a particular patient as both their doctor and as a researcher.

3.5.2 Consent

Written informed consent is fundamental to conducting ethical research and was granted by all participants prior to commencing the research interview. The participant information leaflet (Appendix B) provided participants with an explanation of the purpose of the research and that it was being conducted as part of a doctoral thesis at Newcastle University. It detailed the time commitment required by the participant and emphasised that participation was voluntary and that a participant had the right to withdraw from the study at any time before the interview or until 14 days after the interview had been conducted. After 14 days interview data may have been included in the final analysis and it would not be possible to remove their data from inclusion in the study.

Participants were required to opt-in to involvement in the study by returning a completed consent form (Appendix B). The consent form confirmed that the participant had understood the information included in the participant information leaflet, agreement to the interview being digitally recorded and anonymously transcribed and to storage of the anonymised

data. I reviewed the consent form with the participant immediately prior to their interview to allow them the opportunity to ask any further questions.

3.5.3 Data generation

The previous sections considered the sampling approaches, recruitment and consent of participants, this next section follows on to discuss data generation via semi-structured interviews.

3.5.4 Semi-structured interviews

Qualitative research employs a range of methods of data generation including interviews, focus groups, ethnography and other approaches including document analysis, visual methods and online research (Barbour 2014). The most commonly utilised method is qualitative interviewing, which generates a body of knowledge that is conceptual and theoretical and is based on the meaning that life experiences hold for the participant (DiCicco-Bloom and Crabtree 2006).

Individual semi-structured interviews were the preferred method of data generation for this study as this aligned with the methodological approach and was considered appropriate for addressing the research questions. Charmaz (2014) suggests that semi-structured interviews focus attention on the topic whilst providing interactive space to enable the research participants' views and insights to emerge. Individual interviews are widely used in qualitative healthcare research to co-create meaning with interviewees by reconstructing perceptions of events and experiences related to healthcare and healthcare delivery (DiCicco-Bloom and Crabtree 2006). Individual interviews allowed in-depth exploration of the sensitive and personal experiences surrounding the topic of end of life care.

A semi-structured interview approach was taken to grant the participant the flexibility and control to guide the enquiry process, allowing participants to express their experiences completely and to deviate from the questions to explain their priorities and their story. It is not possible to gather data from qualitative interviews in a completely unstructured way; as the decisions, judgements and phrasing of questions by the researcher shape the data generation process (Mason 2018). An interview schedule was developed for this study that aligned with the research questions and helped to focus on the significant and emerging

themes from interviews. I allowed the content of interviews to be led by the participant with the interview schedule utilised as a prompt to guide follow-up questions and to generate comparable qualitative data. The interview schedule was revised throughout the course of the study in relation to the incoming data from interviews and to allow exploration of aspects of the developing categories.

I conducted all the semi-structured interviews myself. As an experienced respiratory physician, I was familiar with speaking to patients and relatives in a clinical setting and had transferrable skills from my clinical role but limited experience of conducting research interviews. Therefore, prior to commencing the research study interviews I attended qualitative interview training and undertook a 'mock' interview with a member of the supervision team to practise these interview skills. This training helped me to recognise my influence on the participants' narratives and that my preconceived ideas from working in healthcare influenced follow-up prompts and questioning. Mason (2018) argues that it does not improve our understanding to view social interaction in an interview as 'bias' which can potentially be eradicated or regard the interviewer's role as a stimulus which can be standardised. Instead, it is better to appreciate the complexities of the interaction and develop a sense of how context and situation may influence interview interactions. I was conscious that my reaction to participants stories would influence the direction of the interview and tried to offer responses and prompts without detracting from the participants narrative.

Dyadic interviews were conducted with patients and bereaved carers who wanted support from a relative during the interview. Dyadic interviews were conducted with one patient who requested their carer was present, one bereaved relative interview which involved two participants (spouse and daughter of the deceased), and one healthcare professional interview with two individuals who shared a job role. However, dyadic interviews reduce the amount of data generated from each respondent and may reduce the participants' self-disclosure of sensitive topics as they involve the disclosure to both the researcher and additionally the other participant (Morgan 2016).

3.5.5 Interviewing bereaved relatives

Interviewing bereaved carers is an established method used in end of life research. In this study, bereaved carers were interviewed to provide insight into the final weeks, days and hours of life for people with ILD. A concern about interviewing bereaved relatives was the risk of causing distress by asking them to recount an upsetting and sometimes traumatic experience. However, previous research has suggested that the risk of distress from interviews with bereaved carers was lower than anticipated by researchers and that this risk was outweighed by the benefits to relatives who were given the opportunity to discuss their experience (Seamark, Gilbert et al. 2000, Emanuel, Fairclough et al. 2004, Takesaka, Crowley et al. 2004). The process of undertaking the interview may be cathartic; providing the participant with a chance to reflect and assess their experiences and giving a sense of purpose, empowerment, and healing (Hutchinson, Wilson et al. 1994, Dickson-Swift 2008).

Alongside the ethical considerations, concerns have been raised about the validity of interviewing bereaved relatives as surrogates for patients' (Addington-Hall and McPherson 2001). It has been suggested that there was little congruence between relatives' perception of symptoms and patient reported symptoms towards the end of life (Higginson, Priest et al. 1994). However, more recent studies indicate that bereaved relatives are an adequate proxy for patient experiences of care and are useful to provide insights and perceptions of care at the end of life (Tang and McCorkle 2002, Seow, Bainbridge et al. 2016). It is important to acknowledge that relatives will have their own views on this experience and that heightened emotions around the time of death may influence their memories. As there is frequently some uncertainty about the disease trajectory in ILD and often patients die unexpectedly, interviews with bereaved relatives were felt to be an important way to capture data about the last weeks and days of patients' lives. Good end of life care should be attentive to the experience of patients and their family members (GMC 2010, LACDP 2014), therefore how a relative perceived a patient's death is crucial information when evaluating end of life care.

3.5.6 Remote interviewing

It is usual for in-depth interviews to be conducted in person, especially those which concern sensitive topics; however, due to restrictions in place during the COVID-19 pandemic this

was not possible. Therefore, the study protocol was adapted prior to ethics review to allow interviews to be conducted remotely, via telephone or video conferencing, to comply with COVID-19 restrictions. Semi-structured interviews are often the foundation of qualitative research and conducting interviews in person has been considered the "gold standard" (Opdenakker 2006, Novick 2008). Interviews in a participant's home can lead to a more 'giving' environment and strategies such as preamble chat about the family and home environment are effective in building rapport and support a good interview (Sivell, Prout et al. 2019). I found that conducting interviews remotely made it difficult to build rapport and establish a relationship with participants, as the more informal discussions prior to and following the interview were shortened by the remote platform of the interviews.

Arguments against remote or telephone interviewing are the loss of visual and non-verbal cues, loss of contextual data and difficulty building rapport which contribute to data loss or distortion (Novick 2008). However, telephone interviews give more privacy and may allow respondents to feel more relaxed and able to talk about sensitive topics (Carr and Worth 2001, Drabble, Trocki et al. 2016). Interviews were undertaken with patient and bereaved carer participants via telephone and video conferencing according to the participants' preference. I recognised the advantages of video conferencing, particularly the ability to appreciate visual cues, clearer audio recording and potential to build greater rapport with the participant. However, some of the participants appeared to be more uncomfortable talking about sensitive topics in front of a video camera than over the phone. Only a few interviews were conducted in person, these were with healthcare professionals only. These interviews felt comfortable for both the participant and me, however, they were all conducted with people I knew professionally, meaning there was less focus on building rapport and the nature of the conversations was not an emotive or sensitive topic.

3.5.7 Field notes

Field notes were documented immediately after the interview concluded. These were short memos that captured my immediate observations and thoughts about the interview including the setting and location, interaction with the participant, non-verbal communication and any emerging ideas from the interview. Potential new categories or theoretical concepts that were inspired by the interview were also documented. These memos were reviewed as part of the constant comparison data analysis to identify ideas or

concepts which could contribute to the emerging categories. An example of part of the field notes are included below Figure 3-3.

Field Notes: Patient_04
This man had very advanced ILD and extensive palliative care support with regular involvement of district nursing team, Macmillan nurses, telephone support from specialist palliative care and a recent 10-day inpatient admission at a local hospice. Despite the considerable involvement of these teams, he was disappointed with the communication and infrequent appointments with the tertiary ILD service because of the covid-19 pandemic. His last appointment had been a telephone review a few days previously with someone he didn't know who "had to read his notes" and he perceived this person as being dismissive of his deteriorating symptoms. The doctor had planned a further telephone review which had upset him as he felt he should be reviewed in person at the next opportunity. The interview highlighted to me the importance that people with ILD place on their appointments with the specialist service and the reassurance they derive from these interactions. Although this man had fantastic continuity of care from the palliative care teams, it was important to him that he was not forgotten or abandoned by the specialist team. He did not view remote/telephone consultations a substitute for face-to-face review.

Figure 3-3 Example of field notes

3.6 Stage three: working with the data

3.6.1 Data Analysis

This section explains the constructivist grounded theory approach to data analysis employed in this research study. Constructivist grounded theory employs iterative strategies of going back and forth between the data and analysis, uses constant comparison methods which keep the researcher interacting with the data and emerging analysis (Charmaz 2014). It is recommended that initial analysis should occur alongside data generation and analysis should occur concurrently and guide subsequent sampling and data collection (Mills 2015).

The following diagram (Figure 3-4) illustrates the stages of the data generation and analysis in this research study. The diagram depicts these stages as occurring in a linear fashion, however, in reality they took place concurrently and alongside theoretical sampling.

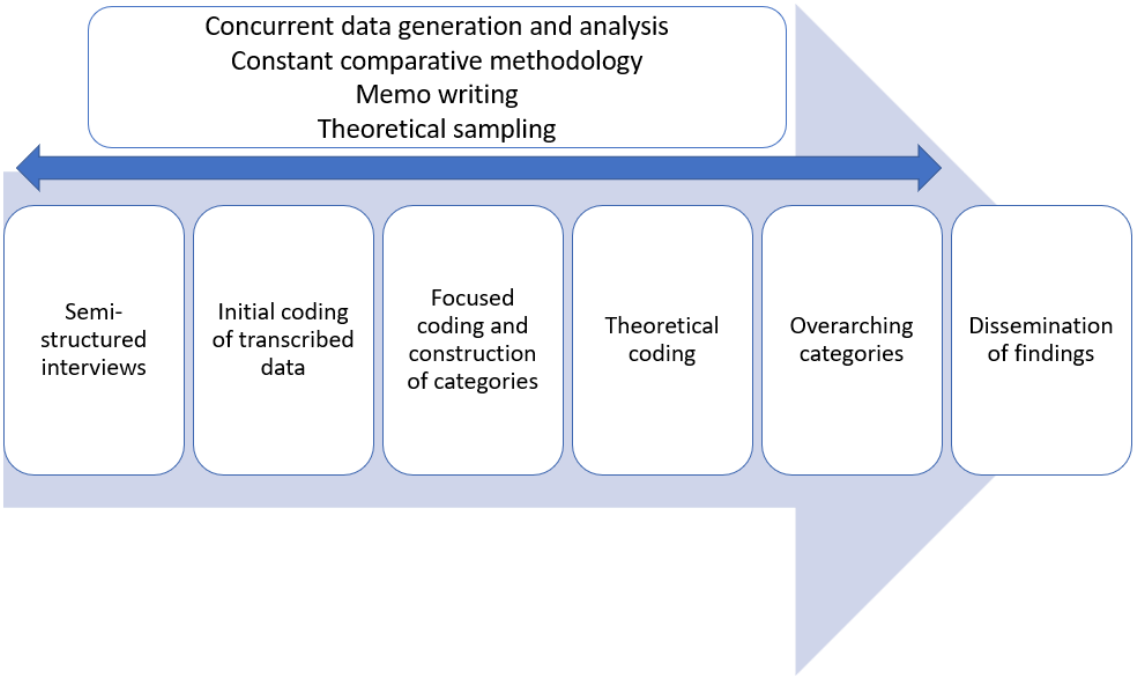

Figure 3-4 Stages of data generation and analysis

Analysis of the initial interview transcripts occurred concurrently with data generation using continuous constant comparative methods. This method of analysis generates successively more abstract concepts and theories through the inductive processes of comparing data with data, data with code, code with code, code with category, category with category, and category with concept (Charmaz 2014). Data were analysed using a systematic approach proceeding through three coding phases: initial coding, focused coding and categorising and theoretical coding (Charmaz 2014). The initial codes were refined, expanded and collapsed to develop focused codes, which in turn led to the construction of categories through connecting and grouping related codes. Analysis of the initial interview transcripts and the developing categories informed the recruitment of participants for subsequent interviews and the questions that were asked in these interviews. Data generation from subsequent

interviews was used to examine and refine the emerging analytic theories (see section 3.4.4).

3.6.2 Computer software packages

nVivo software was used to manage the transcribed data and organise the coded data. This computer software package was utilised for data management; however, I found that it limited the conceptualisation of the data. Instead, I favoured a more 'traditional' approach to explore the relationships between codes, categories and emergent theoretical codes and overarching categories using a research diary, mind map diagrams, sticky notes and highlighting. This approach using low-tech pen and paper methods to aid creativity in the analysis process and supporting with nVivo for data organisation has been successful for qualitative researchers in other disciplines (Maher, Hadfield et al. 2018). Buckley and Waring (2013) gave examples of how to use diagrams to support different aspects of the research process. Diagrams proved valuable to assist the process of data analysis and to present the final outcomes of the study.

3.6.3 Coding

Coding is the cornerstone of constructivist grounded theory methodology and is the pivotal link between generating data and developing theory to explain these data (Charmaz 2014). Data were analysed using a systematic approach proceeding through three stages of coding: initial, focused and theoretical coding. Data analysis began with detailed reading of interview transcripts and field notes that were documented during and immediately after each interview. This allowed immersion in the data and appreciation of the broader picture of each interview prior to undertaking initial coding. Initial coding relied on remaining open to exploring all possible theoretical directions that were indicated by this preliminary reading of the data. Data were reviewed line by line with initial codes attributed to sections of the data. The process of coding involved breaking down the data into component parts, considering these parts individually and comparing and contrasting with other segments of data.

I employed a 'lumping' coding approach as described by Saldana (2015), rather than the line-by-line coding favoured by Charmaz (2014) which Saldana refers to as 'splitting'. Lumping

coding involved coding sentence-by-sentence and in some situations coding several sentences 'lumped' together. This allowed for comparison of data between interview transcripts at an earlier stage in the coding process. Initial coding methods used were descriptive, process and in-vivo coding. I endeavoured to use gerunds to describe these initial codes as it is suggested that coding using gerunds helps to detect processes and stay close to the data (Glaser 1978). The following examples of initial coding are taken from the semi-structured interviews (Table 4, Table 5).

Researcher	**Participant (BR_01)**	**Initial coding**
	And he said, you know what's happening here, don't you? And I said yeah, I think I do. And he said Well we can only keep him comfortable and it's only going to be a matter of hours. So, I said okay, he said do you want to ask me anything? I said I just don't want him to be struggling and being frightened and he said no it'll be it'll be fine it'll you know it'll calm down.	Avoiding direct discussion of dying Managing symptoms Answering questions Wishing for a peaceful end Reassuring that symptoms will improve
	So, I just sat there, and it got to about quarter to five and his breathing seemed to change. It stopped moving as much, and it was a bit shallower. There was a buzzer thing, so I pressed the buzzer in the two nurses came in the male nurse and young lady nurse. And….Um. The male nurse said he's taking his last breaths. So, and then eventually he said that's it he's passed away, he says, you can stay with him, if you want to you know. And so, I rang my son's and told them, and [name of son] wanted to come back and I	Noticing changes Looking for support Recognising dying Recognising death Offering support Telling family Barriers of PPE/Covid ward preventing support from

	said it's pointless, you're going to have to get all gowned up and everything. Because he'd gone home and changed all his clothes, put them in the washing machine had a shower because he was on the Covid ward. Which was frightening. I don't know why he was on the covid ward, no idea…. but…...that last day just haunts me.	family Frightening being on Covid ward Feeling haunted
Interviewer: Yes, I can tell……What is it that haunts you about it?		
	Just…. the fact that I wasn't there…. That was a terrible day because I was pacing the floor most of the time waiting for someone to get back to me…. I didn't know what was happening to him…. I knew he would be frightened.	Feeling guilt Chasing information Waiting for information Worrying he would be frightened
	The fact that I've been I've been with him 24/7 for so long…and I couldn't be with him, you know. (crying) I didn't know what was happening in the hospital and he would have been scared. Because we both said we didn't want him to go to a hospital, you know. I wanted to keep him at home.	Excluded by Covid restrictions Not knowing what was happening Feeling guilt about place of death

Table 4 Examples of initial coding

Table 5 demonstrates how sections of text from different interview transcripts were given a code based on their conceptual meaning. The initial codes used were derived closely from the participants own words to preserve the context of their meaning.

Quotations	**Description**	**Analytical Meaning**	**Initial code**
BR_01: That last day just haunts me….	Replaying the events	Grief that her husband	Feeling haunted

BR_01: It was awful. Was cruel to watch, you know. I think if they'd calmed him down quicker…it just haunts me to think of him struggling.	of the last day of her husband's life. Traumatised that he suffered overwhelming symptoms at the end of life.	had not had a peaceful death.	
BR_03: Yes, that really played heavily on my mind at the beginning. It still does, you can see it still does. It's just my only big qualm is not knowing what happened when I wasn't there.	Grief that she was unable to be present at the end of her husband's life and that he died alone.	Fixated on the unknown–- whether her husband died peacefully.	
BR_05: Okay, from my point of view, it was very traumatic. It was a shock. I still keep occasionally having a little flashback.	Suffering flashbacks to performing CPR on her husband at home. Traumatised by the experience.	Replaying the traumatic events surrounding her partner's death.	

Table 5 Examples of initial coding

Focused coding was the second phase in coding, a process which advanced the theoretical direction of the research findings. Focused coding involved further refinement of the initial codes; the original codes were collapsed and merged, as further data analysis and incoming data revealed that larger sections of text were better suited to one key code rather than several smaller ones (Saldaña 2015). This phase required decisions about which initial codes made the most analytic sense to categorise these data incisively and completely (Charmaz 2014). Focused coding highlighted the codes which were found to be most important in the emerging analysis and to answer the research questions.

Focused coding involved studying and assessing the initial codes and concentrating on the relationships between codes and their representation of the meaning of the data. Engaging in focused coding brought me further into the constant comparative process, going backwards and forwards through the codes and emerging categories to make sense of the data and ensure that the focused codes and categories considered plausible relationships

between the data. An example of the move from initial coding through focused coding to the construction of the category 'wishing for a peaceful end' is included in Table 6.

Constructing the category 'wishing for a peaceful end'			
Participant quote	**Initial code**	**Focused code**	**Category**
PT_07: I'm not scared of the moment of death, I'm scared of everything that goes up to it.	Afraid of suffering	Fear of suffering	Wishing for a peaceful end
PT_08: And hopefully I'll have a big, massive heart attack and that will be it.	Wanting to die fast		
PT_07: I've done the advanced planning stuff and all of that. I don't want to be in denial, and I don't want to be afraid.	Don't want to be afraid		
BR_04a: All that day the family went in and out. Most of us stayed with him.	Always family present	Family presence	
PT_02: With being on my own, I just didn't want to be left on my own. So, towards the end, I would like hospice care.	Not wanting to die alone		
BR_06: He played his favourite rock and roll music on his phone, next to his dad's ear.	Playing his favourite song		
BR_03: He had been very poorly, as poorly as he had ever been over the last couple of months.	Increasing symptom burden	Symptom control	

BR_01: She put a cannula in, and she put something in it. And then, he seemed to calm down a little bit, not much.	Medication efficacy		
BR_01: And his eyes were nearly coming out of his head, he was struggling so much to breathe.	Struggling to breathe		
BR_08: We waited a little bit and then his breathing got worse, and I had to ring for the nurse.	Responding quickly to changes		

Table 6 Constructing the category 'wishing for a peaceful end'.

As the focused coding phase progressed, preliminary categories were generated that summarised several focused codes under the same heading. These categories were deemed to be provisional to enable them to be changed and developed allowing new ideas to be incorporated as further data was available from subsequent interviews. Theoretical sampling (see section 3.4.4) was employed to develop and test the strength of these preliminary categories, maximising the opportunity to approve or disprove the contribution of these categories to the developing theory.

3.6.4 Core categories

Initially two distinct analyses were conducted for the patient and bereaved relative data and separately for the healthcare professional data. This approach was adopted as the data generated through interviews with these participants was diverse, with patients and relatives describing the lived experience of dying with ILD whereas healthcare professionals commenting on this experience from a well-informed outsider perspective. It was apparent from healthcare professional data that they had an appreciation of the broader picture and issues that affected many patients throughout the region.

The core category for each analysis were constructed towards the end of the focused coding process. The focused coding process organised the initial codes, forming sub-categories and then clustering these sub-categories into main categories which supported the construction of the core category for each analysis. These core categories were realised by utilising

theoretical sampling, constant comparative analysis and theoretical memoing to refine and integrate categories. Figure 3-5 illustrates this process in relation to patient and bereaved relative data and Figure 3-6 demonstrates this in relation to healthcare professional data.

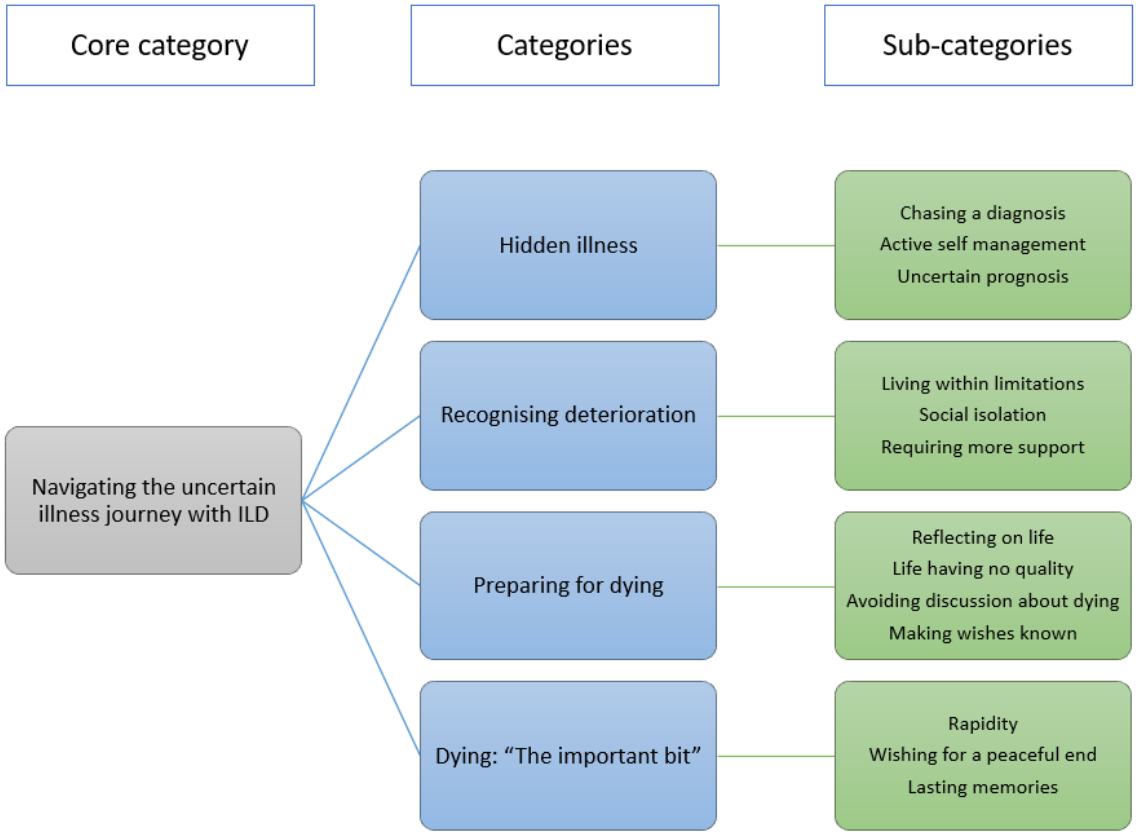

Figure 3-5 The relationship between the core category, categories and sub-categories for patient and bereaved relative data

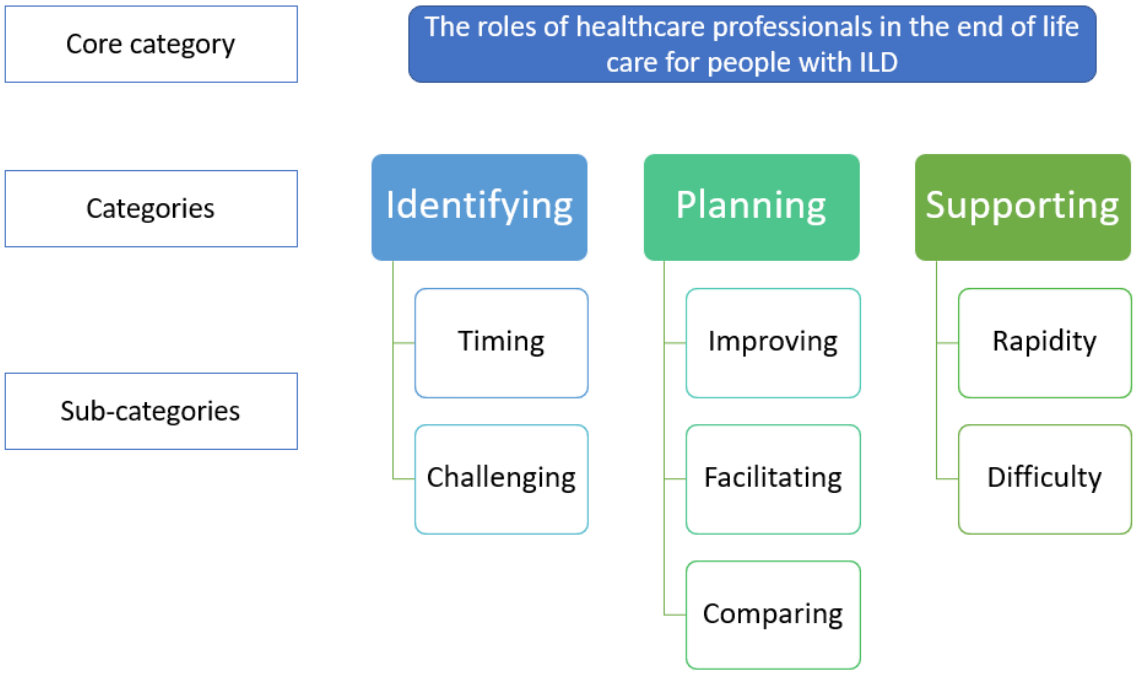

Figure 3-6 The relationship between the core category, categories and sub-categories for healthcare professional data

3.6.5 Theoretical coding

The final coding process used in constructivist grounded theory is theoretical coding. Glaser (1978) states that theoretical codes give an integrative scope, the broader picture and a new perspective. There is debate about whether theoretical coding is an emergent process, or an application of the researchers' pre-existing knowledge and theoretical sensitivity (Charmaz 2014). In this study, theoretical codes were constructed from the synthesis of the patient and bereaved relative and healthcare professional analyses. Constant comparative methodology was again used to identify the codes and categories that were important in both analyses. This led to the development of four overarching categories that answered the research questions and explained the experience of end of life care with ILD: i) acknowledging uncertainty, ii) accessing and organising support, iii) avoiding discussion about an uncertain future, iv) accelerating symptoms at the end of life.

3.6.6 Memos

The writing of theoretical memos is a core activity throughout the constructivist grounded theory process and has been described as 'The theorising write-up of ideas about codes and

their relationships as they strike the analyst whilst coding.' (Glaser 1978)(page 83). The use of memos prompts the analysis of data and codes early in the research process. Charmaz (2014) agreed that memos play a central role in constructing theoretical categories and the use of memos expedites analytic work and accelerates productivity.

Memos are records of thoughts, feelings, insights and ideas relating to the research project written throughout the data collection and analytical process (Mills 2015). In this research study, analytic memos were used to document my thoughts during the process of coding and generation of categories. These memos were particularly useful in making links between codes and categories and relating these to the research questions. Coding and memo writing are regarded as concurrent analytical activities as there is a reciprocal relationship between developing a coding system and starting to understand a research phenomenon (Weston, Gandell et al. 2001). Saldana (2015) agreed that a code should be thought of as a prompt or trigger for written reflection on the deeper and more complex meanings it evokes.

In this research, memos were used in three ways: i) to record thoughts about the codes attributed to data and reasons those codes were later refined and collapsed or expanded; ii) to explore how I personally related to the participants and how listening to and reading their experiences made me feel; and iii) to reflect on emergent patterns, categories, concepts and theory and future directions for the study. Theoretical concepts and questions were identified through memoing which informed theoretical sampling, sorting and development of theoretical codes. Lempert (2007) reasons that memos should be regarded as data and this was the attitude adopted in the current study. The re-reading of memos from earlier in the research process and writing further memos based on these data was instrumental in developing theoretical concepts.

Memoing was used throughout the research process to document procedural and analytical decision making and to explain the scope of codes and categories and how these related to the developing theory. At the start of the research process, I utilised memos to explore my own philosophical position, feelings and assumptions about the research topic. Most memos were impromptu and were based on reflections during the analytical process. However, memos were also employed as a form of debriefing following each of the research interviews and after coding each transcript to document thoughts about the participants' individual experience and the relationship to other data. These memos also reflected on the

questions asked during interviews and the response of participants and how this could be improved or altered for subsequent interviews. Memos are an effective mechanism for raising data to a conceptual level and are recognised as a technique for initiating and maintaining momentum throughout the analysis process (Charmaz 2014, Mills 2015). Memoing was crucial in initiating ideas and maintaining focus on emerging ideas and concepts particularly when work on this study was interrupted by clinical commitments. The following memos (Figure 3-7, Figure 3-8) demonstrate early theoretical memos and how this research tool was utilised in the current study.

Memo: 'Living with ILD' rather than 'dying with ILD'

I initially started this project expecting to talk to people who were dying of end stage ILD and although I expected some people to be reluctant to talk to me about the future, I thought that the majority (having consented to be involved in an EOLC study) would talk to me openly about their plans/fears about the future. However, the majority of the patient transcripts are talking about the challenges and burden of living with the symptoms of ILD and actually very little of the conversation was about dying. In fact, I was talking to patients who viewed themselves as *living with progressive ILD* rather than *dying with progressive ILD*.

I have a lot of interview data about living with ILD, but very little about preparing for dying and even less about the experience of EOLC and the final days/hours. However, only the bereaved carer participants could talk to me about the final days of life and therefore there will be less interview data about this specific area. The challenges I found were to move past the descriptions of death to allow understanding of what influenced decisions and feelings in those final days. Sadly, many relatives had not been present at the time of death or death had been unexpected and sudden – this meant that many of the participants had unresolved questions.

Initially I felt that asking about bereavement and how relatives were coping after the death of a loved one was straying off the topic. However, one patient participant explained to me that his priority was knowing that his wife would be OK after he died – leading me to evaluate and realise that a relative's grief reaction if a reflection on the quality of EOLC.

Figure 3-7 Example of theoretical memo: 'living with ILD' rather than 'dying with ILD'.

Memo: Developing candidate categories

I have spent the weeks prior to my second progress review attempting to 'summarise' the data in a way that I can articulate and explain to other people. This has led to the development of some candidate categories:
Living with ILD
Preparing for dying
Dying, "the important bit"
Lasting memories

Living with ILD: This category is about how people with ILD manage their symptoms, try to maintain normality in their life and continue living their lives despite their disease. As their disease progresses this theme encompasses unrelenting symptoms, increasing reliance on others and tolerating oxygen for survival.

Preparing for dying: This category relates to whether people with ILD are truly prepared for dying. Whether there is understanding and acceptance of prognosis and whether people have engaged with advance care planning. The support available from palliative care services is also incorporated into this theme.

Dying "the important bit":
The title of this category comes from a carer who remarked that she had missed the important bit, the end of her husband's life. This theme relates to the emphasis and importance placed on the final days/hours of life, what people with ILD wish for the end of life and how getting it right is so important for them and their families.

Lasting memories:
This category relates to the grief and bereavement process and how the timing and process of dying can leave lasting memories for bereaved relatives.

These four categories have a linear relationship and do not encompass all of the sub-categories - for example I need to review accessing healthcare resources, covid-19 restrictions, information needs to see if codes within these sub-categories map to other sub-categories which are included in the candidate categories.

Living with ILD is a very broad theme and may be better splitting into multiple categories e.g.
Living with ILD / Hidden illness - relating to the time of initial onset of symptoms and diagnosis focus on self-management of symptoms, keeping active, wellbeing.
Living with ILD / Recognising deterioration - relating to disease progression, worsening symptoms, increasing reliance on others, increasing carer responsibilities, tolerating oxygen for survival, finding activities exhausting. (i.e., illness has progressed and is no longer hidden)

Figure 3-8 Example of theoretical memo: Developing candidate overarching categories.

3.6.7 Theoretical sensitivity

Theoretical sensitivity is 'the ability to recognise and extract from the data elements that have relevance for the emerging theory'(Mills 2015)(page 181). Charmaz (2014) suggests that theoretical sensitivity is attained through a practice of seeing possibilities, establishing connections and asking questions of the data. This process allows the researcher to reach down into the fundamentals of data, probe participant experiences and generate abstract concepts (Charmaz 2014).

A literature review and systematic review were conducted at the beginning of this research project, which established a gap in existing knowledge, and informed and shaped the research aim and questions to develop the research proposal. Charmaz (2014) advises that the literature review material should be set aside until after the development of analytical categories and relationships between the data are considered. I undertook the literature review with the understanding that I would aim to prevent the knowledge influencing the research process. This was achieved by keeping engaged with the data and allowing the emergent theory to determine the relevance of the literature and then locating my work within the existing literature.

The second aspect of theoretical sensitivity is nurturing the ability to develop theoretical insight into the area of research combined with the ability to construct something from these insights (Glaser and Holton 2004). This includes the ability to conceptualise, organise and make abstract connections. Constructivist grounded theory methods give the researcher theoretical openings that avoid importing and imposing established ideas from extant theories (Charmaz 2014). Engaging with initial and focused coding with an open mind allowed the identification of numerous theoretical possibilities during data analysis. Subsequent theoretical coding identified overarching categories which were constructed from further creative analysis of the data. These theoretical codes were not preconceived and were grounded in the data.

3.6.8 Reaching theoretical saturation

The end point of data analysis in constructivist grounded theory is theoretical saturation. Theoretical saturation refers to the concept that categories are 'saturated' when gathering fresh data no longer sparks new theoretical insights, nor reveals new properties of these

core theoretical categories (Charmaz 2014). However, it can be difficult to ascertain when saturation has been achieved.

Following 30 participant interviews being conducted, compared and analysed, a judgment was made that the categories were well-defined and there was explanation of the relationship between categories and the range of variation within and between categories. At this point no new relevant concepts were being identified that were important to the developing categories. It was considered that the study had reached saturation based on the repetition of the main concepts and decreasing heterogenicity. It should be noted that the planned recruitment period for this study was extended by two months to allow time for additional theoretical sampling and data generation as theoretical saturation was not yet attained. At this stage 24 participant interviews had been conducted. Saturation is a judgment, but also takes into account the situation of the research, including running out of time or money (Wiener 2007). Although the timeframes of the research project dictated that study recruitment could not extend further, theoretical saturation was deemed to have been satisfied after 30 participant interviews.

3.6.9 Enhancing rigour

The requirements of reliability, replication and validity which are typically associated with determining rigour in quantitative studies are less applicable to qualitative studies (Maher, Hadfield et al. 2018). There is no consensus regarding the validity of qualitative studies with academics outlining different criteria which must be satisfied to ensure rigour (Guba and Lincoln 1989, Beck 1993, Charmaz 2014). These authors agree on the main principles; the research conclusions should be credible and trustworthy, the methods should be described in a way that allows others to understand the decisions made at each stage of the analysis, the results should be transferable to another setting and have meaning outside of the specific world in which the research was conducted. Table 7 reviews how the standards of rigour were considered in this study.

Standards of rigour **(Guba and Lincoln 1989, Beck 1993)**	**Definition**	**Evidence: how it was considered in the study**
Credibility	Credibility refers to the trustworthiness of the findings and correlates with 'how vivid and faithful the description of the phenomenon is' (Beck 1993).	• Allowing participants to guide the enquiry process • Using participants' actual words in the theory • Checking theoretical constructs against participants' meanings of the phenomenon • Clearly articulating my personal views and insights about the phenomenon through analytical memos • Triangulation–- Member of the supervision team independently analysed a sample of transcripts
Auditability and confirmability	Auditability reflect the consistency of the research and refers to the ability of another researcher to follow the methods and understand decisions made at every stage of the analysis (Beck 1993). Confirmability of findings means that the data has been accurately represented and the interpretations of those data are not invented by the researcher (Polit and Beck 2012).	• Specifying how and why participants were recruited (purposive and theoretical sampling) • Outlining the criteria used in my thinking (e.g., questions asked about the data and the way in which coding relationships were identified)
Fittingness and transferability	Fittingness and transferability refers to the possibility that the findings would have meaning to	• Delineating the scope of the research in terms of sample, setting and levels of theory generated

	another group or could be applied in another context (Speziale and Carpenter 2011).	• Providing thick and rich descriptions to allow readers to ascertain how findings might relate to other situations • Describing how the literature relates to categories which emerge in the theory
Dependability	Dependability means that the findings are repeatable if the research occurred with the same cohort of participants, coders and context (Forero, Nahidi et al. 2018).	• Rich description of the study methods • Establishing and audit trail of data collection process • Stepwise replication of data – measuring coding accuracy

Table 7 Standards of rigour

Charmaz (2014) describes four standards of rigour which differ from those in the table above; credibility, originality, resonance and usefulness. *Credibility* begins with having sufficient data to ask perceptive questions and develop theory and requires robust reflexivity throughout the research process (Charmaz and Thornberg 2020). Research can demonstrate *originality* by providing new insights or conceptualisation of a problem and importantly establishing the significance of the results. *Resonance* considers the requirement for theory to represent the participants' experiences, but also provide insight for others. The *usefulness* of the results is demonstrated in the theory's relation to knowledge development and practical application. Charmaz argues that *credibility* and *originality* generate *resonance* and *usefulness* demonstrating the importance of a rigorous research process in ensuring quality in the development of constructivist grounded theory. Memoing (see section 3.6.6) is fundamental to the development of grounded theory and has been argued to be the most significant factor in ensuring quality in constructivist grounded theory (Mills 2015). Memos were used to map activities and decisions during the research process and form an audit trail demonstrating methodological congruence and procedural precision.

3.6.10 Maintaining reflexivity

One crucial aspect of qualitative researching is acknowledging the influence of the researcher's own views and prior knowledge of the topic on data generation and analysis. Reflexivity is recognising who we are and how we are part of and interact with the worlds that we study (Allan, Arber et al. 2018). Reflexivity is more than a reflection on the research

process, it is an active critical engagement which requires evaluation of the researcher's own biases as they inform both the data collection and the environment in which the research is taking place (Addington-Hall, Bruera et al. 2009).

Throughout the research process I engaged with 'self-reflexivity', reflecting on how my own embedded influence may shape the research outcomes, and 'collaborative reflexivity' which involved sharing information, feedback and reflecting on my practice with both my supervision team and qualitative researchers outside of my study (Alvesson and Sköldberg 2017). Charmaz (2014) recommends keeping a methodological journal to engage in reflexivity and avoid preconceiving the data. My research diary and fieldnotes written at the time of interviews were crucial in guiding my reflexivity, prompting ideas for memos and documenting my dilemmas, directions and decisions. These notes and explanations reminded me of my emotional interpretation of what had been discussed in participant interviews and gave me an insight into my thought processes immediately after each interview. The research diary also outlined how my research idea had changed and developed during the process of data generation and analysis.

Researchers in healthcare settings may have multiple identities, such as clinician and researcher, within the social world which they are studying (Allan, Arber et al. 2018). It is widely acknowledged that researchers with dual roles often find difficulty in maintain the boundary between their two roles (Dickson-Swift 2008). In particular, knowledge that the researcher is a healthcare practitioner may raise expectations that help will be forthcoming (Richards and Schwartz 2002) and participants may expect their questions about access to services and symptom management to be answered. My background as a respiratory doctor and my continued work within the Newcastle ILD team and Marie Curie hospice during this project created some difficulty maintaining boundaries during interviews with patients and bereaved carers. Patients with whom I had significant involvement within a clinical setting were ineligible for recruitment into the study to prevent confusion between my role as a researcher and as an NHS clinician. At times, I felt uncomfortable being unable to offer practical advice on symptom management and grief counselling. However, it has been argued that maintaining a neutral stance when interviewing patients with advanced stage disease may be counterproductive and creating boundaries within the research interview need not necessarily exclude a clinical encounter (Sivell, Prout et al. 2019). As my confidence with interviewing increased, I found that I was able to avoid straying into a therapeutic

discussion during the interview and instead answered any clinical questions from participants after the research interview was completed.

Engaging in reflexivity about preconceptions is especially significant when undertaking focused coding because it is these codes which shape the analysis (Charmaz 2014). As a respiratory medicine trainee, I have experience of both working within the regional ILD service and in other respiratory departments throughout the region and it is likely that I would look more favourably upon places where I had previously enjoyed working. My prior experience of patients dying with ILD was largely limited to patients who died in hospital and were very symptomatic and therefore this may have led to cognitive bias when interpreting the data. The data analysis was undertaken with both these points in mind, and I discussed my interpretation of data with other members of the supervision team to try to minimise my preconceptions influencing the coding process.

3.7 Chapter summary

This chapter outlines the research aim and questions, the philosophical approach of critical reasoning and explains the linkages made between this and the resulting choice of constructivist grounded theory as a methodology. The choice of semi-structured interviews has been discussed and justified and the sampling strategy, data generation and analysis detailed. Finally, this chapter describes the measures taken to ensure rigour and maintain reflexivity throughout the research process.

Chapter 4 Patients' and bereaved relatives' views and experiences of end of life care in ILD

This chapter presents the findings from the patient and bereaved relative data. The healthcare professional data analysis is presented in the following chapter (Chapter 5) and the findings of these chapters are synthesised and summarised in the discussion chapter (Chapter 6). Figure 4-1 shows an overview of the categories and sub-categories generated, which formed the core category of 'navigating the uncertain illness journey with ILD'. The categories outline the figurative illness journey experienced by people with ILD.

Throughout the chapter, illustrative words or phrases from the participants' own accounts are integrated within the text and key quotations from the participants are used at various points to ground meaning to the findings.

4.1 Sample characteristics

In total, thirty participants were interviewed for this study. This included nine patients (six men, three women) and nine bereaved relatives (one man, eight women). To achieve this recruitment a total of 28 patients were contacted with 10 responses. One patient was excluded from the study as he was unable to communicate effectively over the phone due to previous cerebrovascular accident affecting his speech. A total of 58 bereaved relatives were contacted with 10 responses. It was not possible to contact one of the people who returned their consent form to arrange an interview.

The sample characteristics of the patient and bereaved relative participants are outlined in the tables below (Table 8, Table 9). The index of multiple deprivation decile is included to provide information about participant'' socioeconomic status (Office of National Statistics 2019). Deciles range from the most deprived 10% (decile 1) of small areas nationally, to the least deprived 10% (decile 10). The geographical location of participants in relation to the tertiary ILD centre is shown in the maps included in Appendix D.

Sample characteristics: patient participants					
Participant code + pseudonym	**Age**	**Diagnosis**	**Ethnicity**	**Index of multiple deprivation decile**	**Palliative care involvement?**
PT_01 Susan	81	IPF	White-British	9	Yes
PT_02 Lorraine	69	CPFE-IPF	White-British	1	Yes
PT_03 Peter	83	CPFE-IPF	White-British	5	No
PT_04 Alan	72	IPF	White-British	5	Yes
PT_05 William	70	IPF	White-British	10	No
PT_06 Robert	78	IPF	White-British	7	Yes
PT_07 Pamela	69	CHP	White–- Any other White background	8	Yes
PT_08 Christopher	61	IPF	White-British	2	Yes
PT_09 Henry	73	IPF	White-British	4	No

Table 8 Sample characteristics: patient participants

Sample characteristics: bereaved relative participants									
Participant information				**Deceased relative's information**					
Participant code + pseudonym	**Age**	**Ethnicity**	**Relationship to deceased**	**Diagnosis**	**Ethnicity**	**Index of multiple deprivation decile**	**Location of death**	**Death anticipated?**	**Palliative care involvement?**
BR_01 Dorothy	78	White-British	Partner	IPF	White-British	1	Hospital	Yes	Yes
BR_02 Jane	59	White-British	Partner	IPF	White-British	4	Home	Yes	Yes
BR_03 Ruth	60	White-British	Partner	IPF	White-British	6	Hospital	No	Yes
BR_04a Rita	83	White-British	Partner	IPF	Asian–- Indian	8	Hospital	Yes	No
BR_04b Deepta	47	Mixed – White and Asian	Daughter						
BR_05 Maureen	73	White-British	Partner	IPF, PPFE	White-British	10	Home/Hospital (cardiac arrest)	No	Yes
BR_06 Eileen	77	White-British	Partner	Asbestosis	White-British	6	Hospital	Yes	Yes
BR_07 Mark	51	White-British	Son	Asbestosis	White-British	8	Hospital	No	No
BR_08 Julia	60	White-British	Partner	IPF (familial)	White-British	10	Hospice	Yes	Yes

Table 9 Sample characteristics: bereaved relative participants

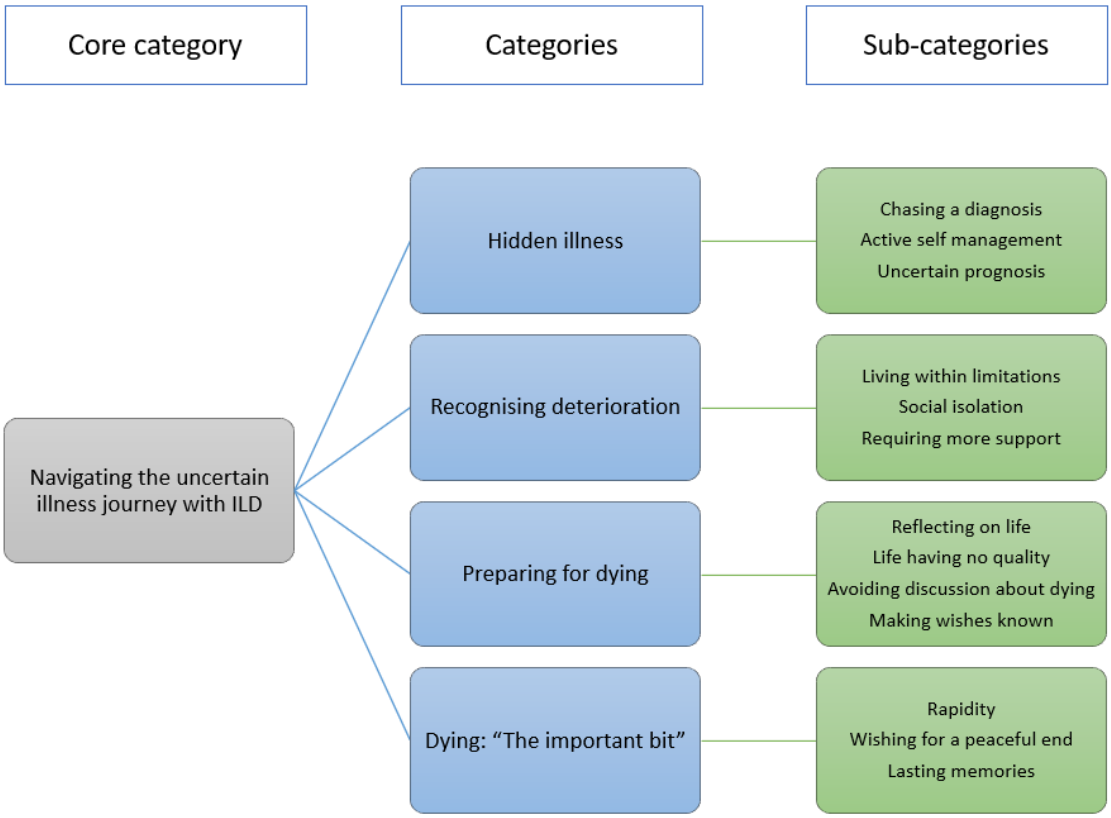

Figure 4-1 Navigating the uncertain illness journey with ILD.

4.2 Hidden illness

The category hidden illness refers to aspects of living with ILD which mean that the impact of the disease is hidden from family, friends and wider society. Many participants had not heard of interstitial lung disease prior to their diagnosis and felt that these conditions were a 'hidden illness' within society. The diagnostic process was often lengthy with some people being treated for alternative conditions before the final diagnosis was confirmed. The sub-category 'chasing a diagnosis' refers to the wait and uncertainty that contributed to the feeling of ILD being a 'hidden illness' that was poorly understood outside of specialist clinics.

Hidden illness also refers to the visibility of the disease; the initial symptoms of ILD were frequently mild and not immediately visible to others. The sub-category 'active self-management' explores the ways in which people with ILD strived to maintain their health and normal life despite their illness. The sub-category 'uncertain prognosis' explains how people with ILD understand their diagnosis and measure their progress against an uncertain timeline of their terminal diagnosis. Anxiety about the future and disease progression were often hidden symptoms of ILD which people did not reveal to their relatives or healthcare professionals.

Figure 4-2 depicts the sub-categories and focused codes that contributed to the main category hidden illness.

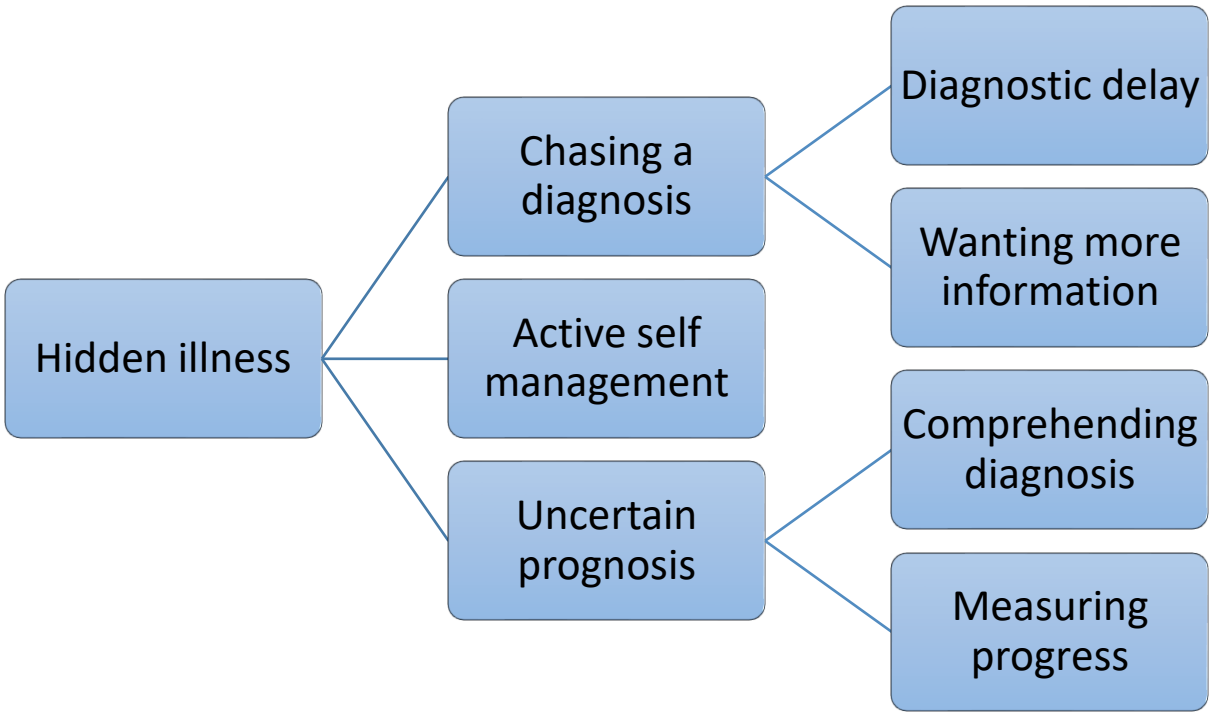

Figure 4-2 Constructing the main category 'hidden illness'.

4.2.1 Chasing a diagnosis

I asked people with ILD about their lung disease and how it affected their lives. For most people this led to discussion of the initial development of symptoms and the diagnostic process. They recalled the difficulties they experienced as they 'wanted more information' and were chasing appointments which were often delayed or cancelled. Initial symptoms were often very minor, such as *'a little dry cough' (Alan, person with IPF),* irritating rather than worrying, which initiated a protracted diagnostic process as they were referred from their GP to local respiratory teams and then onto the specialist clinic.

> *For quite a while, he kept getting a different diagnosis of what his lung problem was, basically, because they obviously didn't know. (…) They kept reassuring him it wasn't lung cancer. (Maureen, Bereaved relative)*

> *The CT scan obviously was worrying, because nobody could explain what the results meant, at that particular time. Hence, we were chasing up the consultants, to take things further. (Henry, person with IPF)*

Initial treatment for alternative lung conditions was common and not unexpected as ILD is a relatively rare condition. The uncertainty surrounding diagnosis meant that many people were chasing clinicians in an attempt to drive forward the diagnostic process. Anxiety was heightened for people who were given an idea of the short prognosis associated with their lung disease without the opportunity to discuss this at length with a professional. This led to people placing significant weight on their review by a specialist to discuss medications and speak to someone who really understood their illness.

> *I felt really nothing was done until I saw the people in [specialist centre] […] and I came out of there literally feeling, "Well, at least somebody knows what they're talking about. Somebody has given me a little bit of hope and they've talked to me as though I'm a, sort of, person, rather than a statistic," which I had felt beforehand. (Susan, person with IPF)*

> *And then I had a telephone conversation with [name of healthcare professional] at the [specialist centre], which was more informative and more reassuring, if it can be reassuring, that the situation was terminal. And I felt happier, after speaking to him, to be quite honest, that was quite a nice, relaxed telephone conversation really. And he took time to explain everything to me, or as much as he could at that time. (Henry, person with IPF)*

> *Then we were prescribed this all singing, all dancing new drug. We researched it ourselves as well and it really boost his confidence. (Ruth, bereaved relative)*

Such comments provide examples of the reassurance people gained from seeing a specialist who was able to answer their questions about their illness and prescribe anti-fibrotic medications. Conversely, one person described significant anxiety associated with his clinic appointments as this forced him to focus on his ill-health, an aspect of his life which he preferred to avoid thinking about.

> *Again, it's like, if you don't hear things for a while, and then I spoke to [name of consultant] and, after that, I was down in the dumps. […] And when he said, "You're in a very vulnerable position," and you think, "Well, it sounds like I'm going to die any time now." You know, that's… But that's probably me, just picking out the worst bits. And these are probably just general information, you know, generic stuff. Once I've got over it, after about four or five days, I think- Because I'm getting used to it, if you like, the shock. But it still affects me. I didn't want it to. (William, person with IPF)*

People with ILD observed a lack of knowledge about their disease amongst healthcare professionals outside of specialist centres. One woman described how the district nursing team had mistaken her diagnosis for cancer which caused frustration and reduced her trust

in this service. People with ILD compared their disease to cancer and COPD which they felt were much better understood by their friends, relatives and healthcare professionals.

> *As you probably know yourself, there's very little research into it compared to cancers and other diseases. It's not very well funded, I believe. (William, person with IPF)*

> *I mean, people come in the house, and they look at me, and they say, "There's nothing wrong with you," but if I went to the door to open it to let them in, I'd be out of breath before I got there, even with the oxygen on. So, it's not obvious to anybody else that I'm ill at all. (Peter, person with CPFE-IPF)*

> *The times I've said to people, "I do not have COPD," even going to the exercise class and the physios who are brilliant, but all the handouts that you got all referred to COPD. I was there crossing it out and putting, "I have IPF." (Susan, person with IPF)*

In the final quote above Susan demonstrated her frustration with the lack of knowledge of her disease by amending the resources given to her at pulmonary rehabilitation classes. She was advocating for individualised assessment of patients during these classes and highlighting her requirement for more information about managing her disease.

4.2.2 Active self-management

In the initial stages of disease, people with ILD and their relatives looked for ways to improve their health, such as keeping active, maintaining a healthy weight and using home remedies for relaxation and treatment of cough.

> *Obviously walking any distance was a problem, although he did keep active because he was always told walking's the best exercise he could actually do. (Jane, bereaved relative)*

> *I went through a lot of things with him, saying, "When you get into bed, do the breathing exercises, relax." I sprayed the pillow with lavender oil, all the, if you like, old wives' remedies, but they do work, things like lavender. (Maureen, bereaved relative)*

It was important for people with ILD and their relatives to be actively involved in the management of their disease and these self-management strategies were a way of trying to regain control over their lives. Some were unable to manage activities such as pulmonary rehabilitation or physiotherapy exercises and felt that they were not following the advice from healthcare professionals and not helping themselves. One person referred to herself as a 'bad patient' when she was unable to follow medical advice.

> *You try, and think, you know they must think I'm lazy, or I've given up, I'm a bad patient, whatever, but even talking like I am to you now, it does make you very tired. (Susan, person with IPF)*

Maintaining a healthy weight was a priority for both patients and their families. Most people with ILD experienced problems with weight loss and were advised to increase their calorie intake, advice which was sometimes contrary to what they perceived as a healthy diet.

> *And he advised to increase his calories. You know, plenty of eggs and bananas, everything contrary to what we had done all the years with his heart problem, when it had to be low fat and… (Eileen, bereaved relative)*

However, poor appetite limited their calorie intake and was often a source of worry for their family, especially spouses, who viewed maintaining weight as one thing they could help with in the face of deteriorating disease. One patient was overweight, and this had been a barrier to his referral for consideration for lung transplantation.

> *Basically, I cannot do exercise, because I cannot brush my teeth without having to have my oxygen on up my nose. […] Fair enough, you need to be burning calories, but I can't burn calories, because [of] my lungs. (Christopher, person with IPF)*

Christopher felt let down by healthcare teams who *'never pushed me to lose weight'.* He perceived a missed opportunity to pursue a lung transplant, and towards the end of his life this remained a significant regret and he directed the blame towards the specialist ILD team.

4.2.3 Uncertain prognosis

Many people with ILD described their shock when average prognosis was discussed at the time of diagnosis. *"Well, you've got three to five years." Which sort of hit me like bang. (William, person with IPF).* Until this point, as symptoms were often initially mild, many people had little comprehension of the serious nature of their underlying lung conditions.

> *So, when you're sitting there, in your opinion, perfectly alright and somebody tells you you've got a terminal illness, it's very disbelieving […] So, the shock, it is a shock, but it's a peculiar sort of… a shock that doesn't… it didn't set in until about a day after. […] It came over as though I only had two or three weeks to live and so I rushed out and I did all my will and I even went to the funeral director, got my funeral organised and headstones, everything. (Alan, person with IPF)*

This excerpt highlights the anxiety that was associated with a new diagnosis of idiopathic pulmonary fibrosis and how confusion about prognosis led to significant anxiety and panic. This man was not prepared for a conversation about prognosis and was clearly shocked by the information that he had a terminal illness. In one sentence he describes feeling 'perfectly

alright' and disbelieving the prognosis, and yet he was so disturbed by the information he perceived that he was imminently dying and hastily arranged his funeral.

People with ILD reacted to their uncertainty and 'wanting more information' by researching their condition on the internet, however, this often led them to websites which contained information they would have preferred not to read.

> *The first thing I did, and I'm sure a lot of other people did, was look it up on the internet to see what it entailed, and I frightened the life out of myself because there were some awful things. It was more or less, "Well, you've got this. That's it," sort of thing. You know, people talked about the awful way that they'd seen relatives die and I thought, "Oh God, that is concerning, not just for me, but for relatives as well." (Susan, person with IPF)*

> *You had people going on who have had mothers or fathers, whatever, and they've died of it, and they've made a diary, written everything in. You start reading it and you're thinking, "Oh," it is quite frightening. So, maybe there are some things maybe you don't want to dwell on. (Lorraine, person with CPFE-IPF)*

> *But I've done the worst thing, haven't I? I've read everything about it, absolutely everything, all the research, all the things. So, I know what's coming, it's just I don't know how it's going to come and when it's going to come. (William, person with IPF)*

Researching on the internet was viewed as a way to manage anxiety and address the unknown and uncertainty of the disease. In fact, it had the opposite effect and exposed people to knowledge about disease progression that they were not prepared to contemplate.

People with ILD sought ways of 'measuring progress' and disease progression. The unpredictable nature of their symptoms made it difficult to monitor their progress.

> *That can be a bit scary because you don't know, because you do understand that this illness, you can go down very quickly or you can just go down very slowly, but it can change. You don't know whether it's, sort of… you get up in the morning and think, "My goodness, things have moved on." You don't know whether, if they move on, that they're going to stay moving and very quickly or whether they're going to plateau again. (Susan, person with IPF)*

As a result, considerable value was placed on their clinical review at the specialist ILD clinic as this provided objective information from lung function tests, radiological imaging and the opinion of specialists regarding disease progression.

> *Now, it's a bit difficult to assess, from my point of view, whether it's doing me any good or not, I can only go by what the hospital and the doctors say. And they seem to think I'm reasonably stable at the moment. (Peter, person with CPFE-IPF)*

> *Anyway, I don't know if I'm that bad. I think, maybe, I might be, I don't know. I know that when my CT… I don't understand this, my CT scans show a lot of stability, but my lung function is just steadily going down and I don't know why. (Pamela, person with CHP)*

People with ILD found it difficult to comprehend the short average life expectancy associated with their illness, especially those with IPF who were told an average prognosis of three to five years. However, it was often the patients with other causes of ILD who followed a more uncertain illness trajectory. These people were less aware of the serious nature of their condition at the time of diagnosis as for many this was not portrayed as a terminal illness. As their illness progressed the advanced nature of their disease still came as a shock.

> *As the letter that came it said, it's advanced. I think that was quite a shock to me because I don't really think of it that way but, at the same time, it is. (Pamela, person with CHP)*

In the above excerpt, Pamela is referring to the information leaflet sent out for recruitment into the current study. She had been identified as having advanced disease and therefore a potential participant by healthcare professionals at the specialist ILD clinic and had also been regularly involved with palliative care services. Her understanding of her condition and how she chose to view her disease highlights the emphasis people place on living with progressive ILD rather than dying with ILD.

4.3 Recognising deterioration

The category recognising deterioration encompasses the events and changes in symptoms that resulted in ILD becoming a more visible illness, both to the person with ILD and to family, friends and wider society. Recognising deterioration often occurred in stages, when people looked back on what they could do previously and compared their current health to the past. It also occurred acutely, with a sudden deterioration in symptoms or the initiation of oxygen therapy. The sub-category 'living within limitations' refers to the worsening symptoms and deteriorating health status faced by people with ILD that led to increasing reliance on relatives as informal carers.

Alongside these deteriorating symptoms, people with ILD experienced a loss of self and the sub-category 'social isolation' explores the psychological effects of living with ILD. The sub-category 'requiring more support' refers to the increasing need for support from both family members and healthcare professionals. Recognising deterioration was frightening for people

with ILD and introduced thoughts and fears about the future, concepts that were frequently not something that people were ready to process or talk about. Patients' and relatives' views on support from palliative care services are explored in this section.

Figure 4-3 depicts the sub-categories and focused codes which contributed to the emergence of this category.

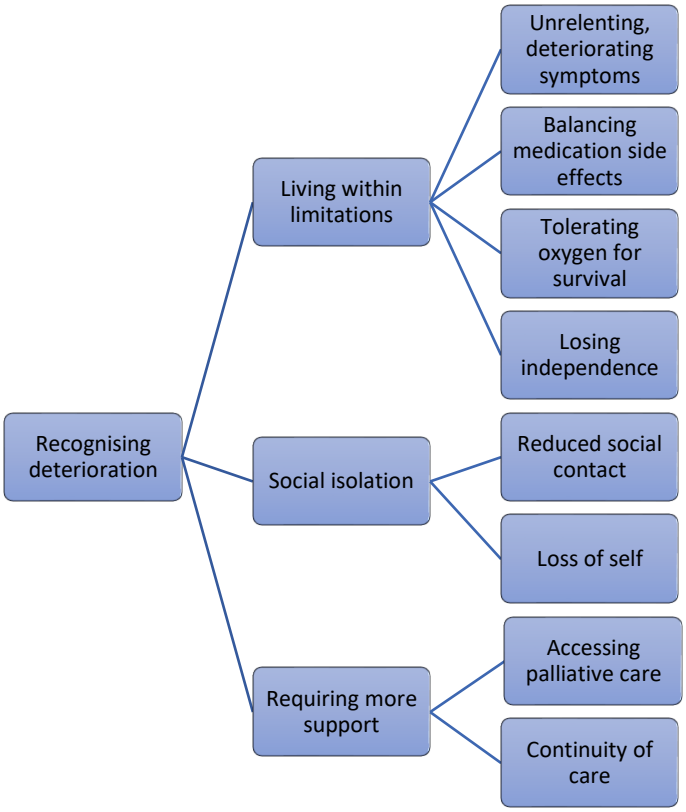

Figure 4-3 Constructing the category 'recognising deterioration'.

4.3.1 Living within limitations

People with progressive ILD experienced unrelenting symptoms that affected all aspects of their lives and led to people re-shaping their lives around the limitations placed on them by their disease. The overwhelming symptom burden experienced by people with ILD was evident; symptoms reported were breathlessness, cough, fatigue and weight loss.

> *He used to pant terribly. Towards the end, he panted. It was, "Hah, hah, hah, hah, hah." That was panting. I used to make him sit, and breathe in, and hold, and breathe out, and breathe in and hold, but I used to… If I wasn't with him to make him do that, he would go back to the panting. (Maureen, bereaved relative)*

> *Tiredness, sheer exhaustion. A lot of the time, you don't feel 100%. You feel this weariness and you feel just not quite right, and it's a very hard thing to come to terms with because you can have a good night's sleep and you get out of bed, and half an hour later, all you want to do is go back to bed. It really hits you out of the blue sometimes. […] You know, people say, "Oh, I get tired," or, "I get out of breath," and you think, "Well, yes, but not like I do, surely." It's a really, really difficult thing to put into words. (Susan, person with IPF)*

Breathlessness had a significant impact on mobility and reduced the level of activity for many people with ILD.

> *when I do decide I'm going to do something, I either can't do it because I get out breath even just sometimes only thinking about the things I'm going to do before I even do them, if that makes sense. You know, I think, "Oh, I'm going to do so-and-so. Oh, just a minute, that means I've got to stand up, and if I stand up, I'll have to walk over there, and if I walk over there, I'm going to be out of breath. So, well, I won't bother." (Laughter) I'll sit down and watch the television. Which is very easy with the remote. (Peter, person with CPFE-IPF)*

> *His mobility suffered terribly, just doing everyday things like dressing, bathing, things like that, became really challenging. He was out of breath, and it was awful to watch because you start trying to breathe for them, almost. […] It was awful, you know, it was a vicious circle, really. I did try to take him out, just for a drive in the car, but getting to the car, even, like, pulling the car into the driveway, walking a matter of 10m, it was an awful battle. (Mark, bereaved relative)*

As demonstrated by these quotes, the impact of worsening breathlessness was wide ranging. It prevented people with ILD from managing their own activities of daily living without assistance, contributed to overwhelming fatigue and reduced the possibility for leaving the house and socialising. Breathlessness was the most common and most disabling symptom experienced by people with ILD.

The importance placed on maintaining a healthy weight has been previously discussed in relation to 'active-self management'. Continued and unmanageable weight loss was very

distressing for patients and relatives as this was a sign of illness progressing causing a deterioration in physical appearance making the illness more visible.

> *I was losing weight at such a rate. I mean, every time I looked in the mirror, I thought, "My God, I'm getting thinner and thinner here," and it was worrying. (Peter, person with CPFE-IPF)*

> *And I think the weight loss was, by the end of his life, that was the biggest issue, you know. When he went in, he was starving. I mean there were bones out everywhere. For a man who was always slim, I mean, I think he must've weighed like 4 stone, or something. (Deepta, bereaved relative)*

> *no matter what food he got, he'd not put on weight. He wouldn't put on weight, because his body was, as I say, consuming itself to try and fight a disease it couldn't really fight. (Maureen, bereaved relative)*

The emotive language used in these quotes to describe weight loss, highlights the significance of this symptom to people with ILD and their relatives. The relatives interviewed were mostly women who saw cooking as their role in the house; it represented an important way of caring for their relative and feeling less helpless. Weight loss was seen as a failure of their caring role and a sign that the disease was 'consuming' their relative. Relatives raised these concerns at clinic appointments and were disappointed when they were given advice that was difficult to incorporate into their lives, as discussed previously in the sub-section 'active self-management'.

Recognising deterioration in health status forced many people to re-evaluate their priorities in life. One participant recalled her husband saying, *"I'm here to live, I don't want to exist." (Maureen, bereaved relative)*. This change in priorities was highlighted by their approach to balancing medication side effects.

> *Really, I felt as if I was permanently […] permanently seasick. I couldn't eat at all. I wasn't eating and I had chronic diarrhoea. So, the side effects were horrendous. So, I only took it for a week, and I just phoned and said, "Look, I can't cope with the side effects." I know they said they might give you a few extra months of life, but if that is the price you've got to pay, it is not- wasn't worth it for me. […] I thought, "Well, it is not helping us do anything." It is not helping us get out the house or go for a walk, or whatever, it is just- I couldn't see any advantage to it myself. (Lorraine, person with CPFE-IPF)*

Here the intolerable side effects of anti-fibrotic medications are described, which led to some people deciding to discontinue this treatment. People acknowledged that they would probably not live as long without the medication, but that the quality of life was their priority.

The initiation of oxygen represented a significant decline in health and another point where their illness became visible to others. Many people were reluctant to start on oxygen, concerned that oxygen meant they were closer to dying *"Once I go down that road, there's no turning back" (Susan, person with IPF)* and they wanted to avoid this prescription for as long as possible. However, others accepted and tolerated oxygen as a means of increasing their survival. *"I realise I've got to take it because, otherwise, I wouldn't survive" (Peter, person with CPFE-IPF).* Using oxygen was terrifying, a constant reminder of illness which forced people with ILD to confront their ongoing deterioration.

> *I mean, when somebody suddenly says to you, "You're on 24-hour oxygen for the rest of your life," it is quite a kick in the stomach. It is quite frightening. It is quite a frightening thought. (Lorraine, person with CPFE-IPF)*

The prescription of oxygen was *"such a huge life change" (Lorraine person with CPFE-IPF)* that came with the need to meticulously plan excursions and resulted in reduced social activity and further isolation, an issue which is discussed further in the next sub-section. None of the participants found that oxygen greatly improved their symptoms and often they were unclear on the intended benefits of its use.

> *I just started on oxygen about six weeks ago, and I'm finding it a little bit helpful, but it's certainly not the panacea for everything. […] I get leaflets and you've got pictures of men striding down a country road with the oxygen strapped to their back. Well, I'm afraid that is never going to be me. (Susan, person with IPF)*

> *I thought the oxygen would help us. When I was still gasping for breath, I was panicking and phoning the hospital. I was saying, "Is this the way it is supposed to be?" (Lorraine, person with CPFE-IPF)*

Several people reported their disappointment with oxygen, as highlighted in these excerpts, as it did not improve their breathlessness symptoms or allow them to increase their physical activity. Overall, oxygen provided some relief from symptoms but was viewed as something required to survive rather than improving quality of life.

As people with ILD suffered deteriorating symptoms, they started to lose their independence and relatives took on increasing carer responsibilities. Initially, this was a subtle change in roles around the house with an often-unspoken recognition of deterioration.

> *He always used to wash up, that was one of his jobs. He had stopped doing the washing up. So, anything that involved standing had become … he never said anything though, he just stopped doing it (Deepta, bereaved relative)*

> *Fortunately, my wife's taken a keen interest in the garden, and she's got it looking lovely, but I feel badly about not being able to help her in the garden. We have someone to come and cut the grass and to do the heavy work. (Peter, person with CPFE-IPF)*

As the disease progressed, symptoms worsened and the burden on relatives increased, compelling many to take on the role of an informal carer. Towards the end of life, many people with ILD were unable to perform their activities of daily living without help from informal carers.

> *In the last month or so – month, six weeks – he couldn't do anything for himself. I was having to dress him, shave him, do his hair, bathe him, put him to bed, get him up, whatever, so for the last, maybe, six weeks, four weeks, before he died. (Maureen, bereaved relative)*

> *So, every other day I used to have to bed bath him and help him to shave yeah and dress him and managed to get him downstairs, but he always got dressed until the very day before he died. And just trying to keep everything normal the days in between, we just have a wash and take the perching stool into the bedroom. He had the commode in there, even though the toilet was only steps away he couldn't manage it, you know so um it was just all my mornings were taken up just getting him prepared. (Dorothy, bereaved relative)*

These examples highlight the significant carer strain experienced by relatives in the study. Many had declined help from social services due to the cost or concerns about the risk of having strangers in their house during the coronavirus pandemic. There was a common feeling of *"We'll just get on with it and cope with it" (Maureen, bereaved relative).*

4.3.2 Social isolation

People with ILD were frustrated that their 'hidden illness' was not recognised or appreciated by their friends. They gradually withdrew from social activities as attending these events became more difficult due to worsening breathlessness and fatigue.

> *I've got friends who come around and say, "Hey, there's nothing the matter with you. Are you coming out for a pint tonight?" "No, I'm sorry, I can't come out. I couldn't get there. If I did, I couldn't get up the steps. If I did, I couldn't get to the bar to pay my round, you'd have to buy my beer all night." And they say, "Huh, well, that's no good, is it? You'll have to stay at home then." (Peter, person with CPFE-IPF)*

These initially small changes to their lifestyle demonstrated the requirement to live within limitations imposed by their disease. Those who worked at the time of diagnosis and later were forced to retire due to deteriorating health, greatly missed this aspect of their life.

> *And then obviously, after that, it just seemed to get worse. I think it was my mood, you know, frightened, concerned, sort of all that. And I loved my job, so I didn't want to give up work. (William, person with IPF)*

As the disease progressed, people with ILD faced the stark reality of increasing limitations on both their lives and their relatives' and social isolation became an escalating issue.

> *so gradually you're aware that his world was getting smaller and smaller, and what he was able to do was getting less and less. (Deepta, bereaved relative)*

Many missed previous hobbies and interests, others missed social interaction from outings outside their house as many were now no longer leaving the house. This could be due to anxiety about COVID, practical inability to leave or feeling that activities were 'too much effort'.

> *Maybe if I were iller, I probably wouldn't care, but you just sit as if you want to…. I used to dream about- I used to go on a lot of holidays, going on cruises and all sorts. Now, I'm dreaming about going to Asda. (Laughter) Just actually going into Asda and picking a few of your own groceries, you know. […] It just seems the height of luxury to be able to go into a shop. (Lorraine, person with CPFE-IPF)*

> *I think what I realised was he managed to, so he did less and less, and I think the lockdown meant that the few little things that he had left to do, like, he used to do a bit of shopping and he used to, all that stuff had stopped, so I think then it was the four walls. (Deepta, bereaved relative)*

Those who lived alone were particularly affected by social isolation and the effects of shielding during the COVID-19 pandemic lockdown.

> *So, that has been the worst thing I've got to say about this illness, but I know it is because of COVID as well. If maybe COVID wasn't here, they might have been able to do more things, but just the pure isolation, it just drives me around the bend. You get up, well, that is half the time, trying to get ready. Where am I going? You watch the telly and go to bed. You try to potter and do little things in the house, but I would think that out of everything has been the worst. It has been the worst thing. (Lorraine, person with CPFE-IPF)*

Living in an ever-decreasing world with reduced social interaction led to a feeling that people with ILD had lost the aspects of life that they enjoyed. People with ILD described a 'loss of self' either directly related to their symptoms because of inability to do certain activities and role reversal concerning jobs within the house, or indirectly as increasing fatigue meant they were unable to continue with previous hobbies, interests and important aspects of their life.

> *The other strange thing… You know, I used to do yoga and meditate every day. I had this fantasy that I was going to end my life in a meditative bliss because that's how I was going to handle the end of life. I've found I don't have the energy for it, I don't have the energy for anything. To even think about all these… The spiritual*

> *aspect of my life is just gone, I don't know, it's just gone, I don't even think about it. (Pamela, person with CHP)*

> *He got really upset because he couldn't sing. We always sang in the car together. He struggled because he couldn't get his breath. Just silly things like that, I suppose. It, kind of, took everything away, it took everything away from him. (Julia, bereaved relative)*

These extracts describe the sense of 'loss of self' and that the illness had taken away the joy of life. They highlight how the interplay between symptoms and the effect on physical function destroys a person's sense of self.

4.3.3 Requiring more support

As their disease progressed, people with ILD recognised a deterioration in their overall health status and their increasing requirement for more support from family members and healthcare professionals. Increased support was sought for both declining physical symptoms and the additional psychological burden caused by deteriorating health and anxiety about the future. People with ILD were unsure who to contact for further support and relied on guidance from the specialist ILD clinic, palliative care services or their GP. Some people with ILD had misconceptions about palliative care, as illustrated by the quotes below. However, they were reassured about being referred to these services after discussion with the specialist ILD team who they trusted.

> *He didn't fully understand what palliative care was. He kept thinking that it was end of life and I was reassuring him all the time, "No, no, no, this is not, this is just keeping you comfortable with this illness, that's basically what it is." It's not because you're going to peg out on me or anything. (Ruth, bereaved relative)*

> *when you say 'palliative care', I think an awful lot of people think end of life care and end of life means literally end of life next month, say, or next week even. I think that perhaps should be advertised a bit more widely that palliative care does kick in quicker. (Susan, person with IPF)*

> *I think, maybe, people don't want to take the help, again, because they think, when it's involved with a hospice, it's a death thing. You get this disease, unless a miracle happens, it is going to end in death. Inevitably, it'll be death. For however long you have, just to have that help is a big thing. (Julia, bereaved relative)*

Conversely, palliative care involvement was viewed positively by some people with ILD and their carers *"it's to get everything right … it's all part of the package, you're really lucky." (Ruth, bereaved relative).* Relatives appeared to be more prepared for the discussion of palliative care, as they had been silently worrying about ongoing deterioration, and often were involved in reassuring their relative about the benefits.

> *Palliative care, when that was mentioned, that sent shockwaves through mam and dad. I said, "Look, it doesn't mean that this is it, it's imminent, it just means that we're trying to improve the quality of your daily living, you know?" (Mark, bereaved relative)*

The support provided by palliative care specialists ranged from symptom management to practical help with accessing community services and psychological support.

> *This was, obviously, before he was really ill, so it was breathing exercises, relaxing exercises, things to do to help: what to do with your coughing and what to do if you're panicky, whatever. (Maureen, bereaved relative)*

> *You know, she was really good, I'm going to talk to her again because I've been having a lot of problems mentally lately. (Pamela, person with CHP)*

In general, the involvement of palliative care and community teams was appreciated by people with ILD and their relatives. They valued the regular phone calls or visits and particularly the 'continuity of care' that this regular contact provided.

> *Well, God bless them, [they] ring me every week on a Wednesday, one of the nurses rings me to see how I am, just have a chat, you see. (Alan, person with IPF)*

> *so perhaps two to three months prior we did receive a phone call from the district nurses to say this is who we are, this is our telephone number, we're 24/7, don't hesitate to call us. All of that was all put into operation long before we actually required it […] it was really useful because who else would I have phoned? (Jane, bereaved relative)*

> *The day will come when I will need them, but as [name of healthcare professional] said, "Well, at least you can get to know our faces. When you do need us, we'll be familiar to you," which is fair enough, I suppose. (Susan, person with IPF)*

> *They say they're coming long before you're really, really ill, so they get to know you. (Lorraine, person with CPFE-IPF)*

These quotes reveal that despite having advanced disease and a significant symptom burden, many of the participants felt that it was too early for them to need input from palliative care services. However, they accepted the possibility that this might be required in the future and the introduction to palliative care services was well received if phrased as being introduced early and before people were 'really, really ill'.

Palliative care involvement was not ubiquitous amongst all patients or indeed prior to death for the relatives of bereaved carers. Some people with ILD remarked that palliative care had not been directly discussed with them at any point in their illness and therefore they had not accessed any palliative care services. Others had chosen not to engage in discussions about

palliative care. For some, events at the end of life happened too quickly to allow timely palliative care involvement.

> *It's been mentioned, but there's been no follow-up, and I've never followed it up at all. (Peter, person with CPFE-IPF)*

> *If it hadn't happened as quickly, I think if the [palliative care] nurses had got involved, or the palliative care team at the [name of local hospital] had got more involved, then there might have been more information forthcoming, but it happened very quickly – relatively quickly – as I understand it. (Maureen, bereaved relative)*

Maureen explained that her partner had started what she assumed was planned to be a series of conversations with specialist palliative care prior to his death. However, he died unexpectedly at home, without resuscitation decision in place. This highlights that people with ILD can die unexpectedly and there is not always the luxury of multiple conversations to address palliative care issues.

The impact of the COVID-19 pandemic was felt deeply by people with ILD as they were a group of highly vulnerable people who were required to shield from society. In addition to the strain and anxiety of shielding, support services were closed, and respiratory staff redeployed which meant a significant reduction in contact with healthcare professionals. As reported in the hidden illness section, people with ILD valued their close relationship with healthcare professionals and this sudden change in frequency of face-to-face contact was difficult, especially for those whose health continued to deteriorate during the lockdown period. These people felt forgotten by their medical teams and were deteriorating quietly at home.

> *they cancelled all my appointments. I had no appointments whatsoever with the hospital because they had cancelled them all. They didn't even give us a phone appointment. So, I was phoning my GP and my GP was saying, "Phone the hospital. Ask to see for an appointment for the hospital." The hospital was telling us to phone my GP. (Lorraine, person with CPFE-IPF)*

> *Obviously, he never saw his respiratory consultant. He never saw the specialist nurse practitioner. He talked to the nurse practitioner on the phone, but you can't really judge how people are progressing, really, over the phone. (Maureen, bereaved relative)*

> *I'm saying, "COVID could have made it better." I'm not sure it would, but at least… Because he did used to keep saying, "I've never seen a consultant. I've never seen a consultant." He hadn't seen the consultant for about a year and a half. (Maureen, bereaved relative)*

> *To be honest, it was somebody who I didn't know who was looking at my files for the first time and I didn't… in fact, I was a bit disappointed. They needn't have rang me, needn't have rang me at all … he was going to put the phone down. It was going to be another telephone consultation ___ or something. I said, "No," I said, "Hang on." I think I said, "I really do think that you should have a face-to-face twice a year." I'm not expecting too much, twice a year. (Alan, person with IPF)*

These quotes illustrate the damaging effect of the COVID-19 pandemic on the clinician-patient relationship within the specialist ILD service. People with ILD perceived that healthcare professionals had given up on them as there was nothing further that could be offered in terms of active or disease modifying treatment options. Alan mourned the previous good relationship he had with healthcare professionals which he perceived to have been tainted by pressures on the system and the inability to provide what he felt were adequate follow-up arrangements.

Many recognised that there was often little change in their treatment following review by the specialist ILD team, but there was comfort from talking to someone about their illness journey and discussing any progression of symptoms and worries with a healthcare professional. Even a remote consultation held the 'power' to help people feel less alone and provide ongoing support and continuity of care.

> *okay, they can't do anything to you, they can't do anything for you, but they don't realise the power that a little chat every two months or three months, and I know they've got loads of patients, but just a chat over the phone from somebody you know just to say, "are you okay?" just a friendly thing like that. It means so much to people. (Alan, person with IPF)*

Interestingly, Alan was having regular review by the specialist palliative care team who were ringing him every week and he had also been an inpatient at a local hospice for a short period for symptom management. Despite this regular contact with healthcare professionals who were closely affiliated with the ILD service, he still felt *"cast adrift"* and let down by healthcare services.

4.4 Preparing for dying

This category explores the stage in their illness journey where people with ILD have recognised ongoing deterioration and they start to regard dying from ILD as a more tangible possibility. For some people this realisation occurred relatively early in the disease process, however, there were others who never accepted their terminal diagnosis.

This category encompasses the symptoms and psychological status of people with ILD during the last months of life. The sub-category 'reflecting on life' explores how some people with ILD felt at peace towards the end of their life, enjoying time spent with family and finding enjoyment in a smaller world. The second sub-category 'life having no quality' considers another group of patients who unfortunately suffered a significant symptom burden which left them feeling that life had no quality and was no longer worth living. The dichotomy between these viewpoints is explored and potential influencing factors are suggested. A significant aspect of preparing for dying is making wishes known for the end of life through advance care planning conversations. However, the majority of people with ILD 'avoided discussion about dying' and did not engage with advance care planning conversations.

Figure 4-4 demonstrates the relationship between the sub-categories and focused codes that constructed the main category preparing for dying.

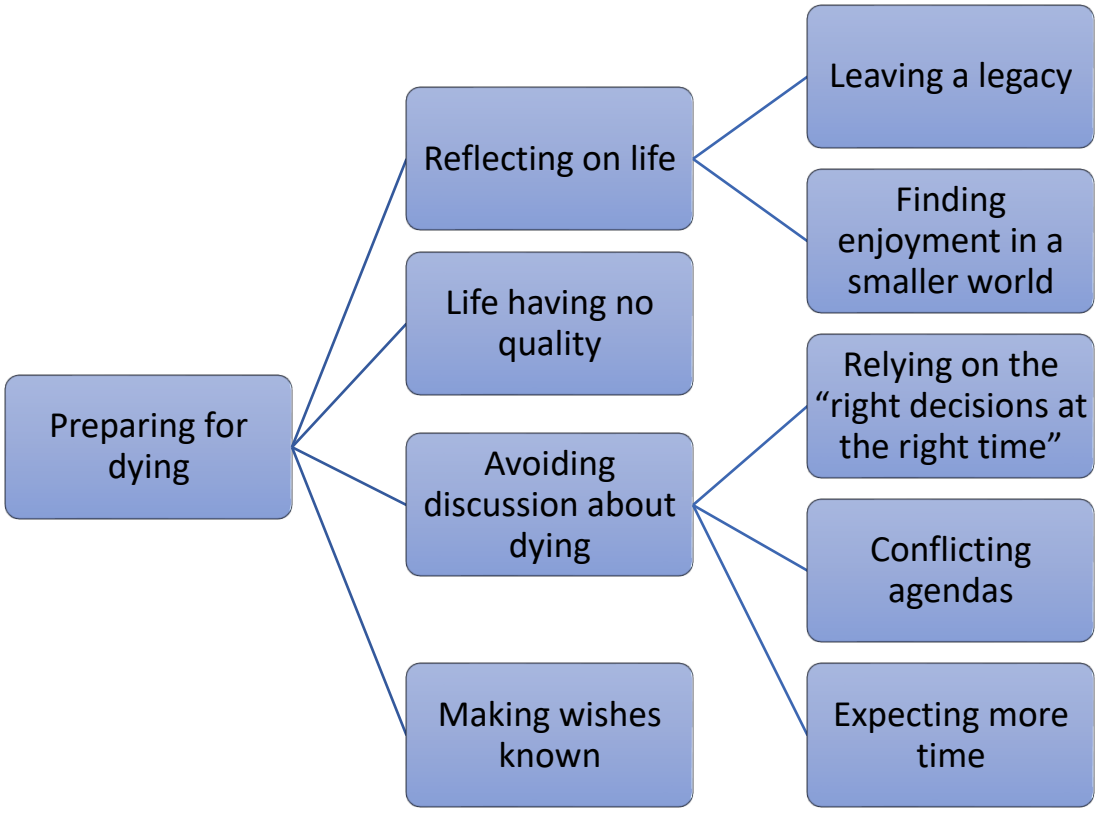

Figure 4-4 Constructing the main category 'preparing for dying'.

4.4.1 Reflecting on life

Towards the end of life people with ILD reflected on their lives, the things they enjoyed doing and the important people in their social circle. Many found 'enjoyment in a smaller

world' allowing them to develop hobbies, spend time with family members and concentrate on their personal version of 'leaving a legacy'.

> *there aren't that many people in the world who do the work that I do. […] My colleagues can't do the analysis […] it's really up to me. I have to make a decision about whether this is what I want to leave as my legacy. It's what I wanted to leave as my legacy when I started working on it. (Pamela, person with CHP)*

> *And I think in some ways I felt like the last couple of years of his life were in some ways the most content, you know? … I just think he could see that they [his grandchildren] were both in a good state, and I felt like he felt, "Okay, I can checkout now," you know, like, "Whatever happens, everybody's good, everybody's set up, everybody's happy." So, I think that was the key thing that changed for him. (Deepta, bereaved relative)*

> *She likes gardening, and she's got it looking lovely at the moment. So, I go and sit out there, and we can chat, and I can look at the flowers, and talk to her, read my book, and order cups of tea and coffee whenever I want one, you know. (Peter, person with CPFE-IPF)*

> *she had a very particular relationship with her grandfather; it was a recent relationship of sharing cooking and tips, and I know that his last 2 years of his life, he had felt more and more close to his grandchildren. (Rita, bereaved relative)*

This facet of contentment and acceptance of their illness was not limited to people who were happy to engage in advance care planning discussions, however it did seem to be associated with those who had fewer symptoms or appeared to be more accepting of their prognosis.

4.4.2 Life having no quality

For some people with ILD the escalating symptoms towards the end of life became overwhelming and distressing for them and their relatives. This had a significant impact on their quality of life and resulted in feelings that 'this isn't living'.

> *So, um I don't think he wanted to carry on, you know he mentioned a couple of times that this isn't living, you know. (Dorothy, bereaved relative)*

> *And, and then we'd get him downstairs, and he would sit in the conservatory he had a problem with his eyes, so he couldn't read or see very well, so all he could do really in the last few months was just sitting the conservatory or in the living room watching television. And that was about it, you know, life wasn't very good at the end you know. (Dorothy, bereaved relative)*

> *but I think he had no quality of life at all, really, or hardly any quality of life at all. He'd gone from somebody, as I say, who enjoyed his food and really enjoyed food, to not wanting food. He just sat. […] It was getting to the stage where it was a mercy, in a way, the way he went. (Maureen, bereaved relative)*

> *If it wasn't for my wife (coughing) I wouldn't be here. […] There is times when I've thought to myself, I wish I could get hold of Dignitas you know in Sweden [sic]and give them a ring and end my life because it came to that stage like. But obviously that's something I can't do. (Robert, person with IPF)*

The unspoken aspect underlying these quotes is that people with ILD were often living their last weeks and months of life through periods of national lockdown with very little social contact or support. This isolation, the uncertain disease trajectory and worries about the future all contributed to a tremendous psychological burden of symptoms.

4.4.3 Avoiding discussion about dying

There was a stark contrast between whether people with ILD experienced relative contentment or a high psychological burden and distress in the final stage of their life. There was general understanding and acceptance amongst people with ILD that time was short, but most had not participated in advance care planning conversations and did not want to talk about what would happen at the end of life.

> *And eventually the doctor said, "You've got to-" he didn't use that term, but I knew what he meant, "you've got to address end of life care," and [name of husband] very correctly said, "Yes, yes, yes, I'll do that." Didn't look at me. I nodded vigorously, "Yes, of course we have." […] We never had that conversation. (Rita, bereaved relative)*

> *He wouldn't talk much about things. Again, he would bottle things up, which is why, when the GP came, I don't think he accepted what the GP was saying, and he wouldn't talk about it a lot. (Maureen, bereaved relative)*

> *Yes. Maybe I choose to ignore it, not really face it. But I know I have to. But I'm facing it every day. (William, person with IPF)*

For some this was a *"what will be will be" (Peter, person with CPFE-IPF)* attitude and they trusted healthcare professionals to make the right decisions and the right time. Others did not accept their prognosis and expected to have more time'.

> *I'm not sure that [name of partner] ever came to terms with the fact that what he had was terminal. I did, but I think [he], maybe, at the back of his mind, knew but thought it would be many months down the line, or many years down the line or whatever. (Maureen, bereaved relative)*

> *I knew that [name of husband] was on borrowed time, but we grasped every positive that was given by the professionals, and we thought he would have much more time. But obviously we were deluding ourselves. (Eileen, bereaved relative)*

Relatives often recognised the unrelenting deterioration in their spouse's health, and many tried to encourage participation in advance care planning discussions with healthcare

professionals. At times they had conflicting agendas leading to frustration on the part of the relative as they were unable to ask questions for fear of causing distress.

> *I wanted to discuss his defibrillator when the Macmillan nurse came one time, about when it was time to deactivate it, but it was a sensitive subject and never discussed. […] All the professionals appeared to skirt around the subject. The Macmillan nurse said, "Patience. A patient has to ask the question," and he asked [name of husband] if he wanted to ask, and [he] stated categorically no. So, the subject was closed. (Eileen, bereaved relative)*

This excerpt suggests that some healthcare professionals appeared to avoid engaging in advance care planning discussions, even surrounding important aspects like defibrillator deactivation, expecting patients to begin the conversation. This created tensions as many people with ILD trusted healthcare professionals to initiate appropriate discussions at what they judged to be 'the right time' and did not actively pursue advance care planning. One person died without writing his will and left his partner with significant financial issues to resolve after his death. This again echoes the idea that people with ILD struggle with the uncertain prognosis, and many can only manage this uncertainty by avoiding discussion of the future. Relatives however often recognised dying and were not surprised when their spouses deteriorated or died quickly.

Although people with ILD had often not discussed end of life care with healthcare professionals, and in some cases did not have a 'Do not resuscitate' form in place in the community, they had expressed to their family that they did not want their life prolonged.

> *Well, I'll have treatment for chest infections. If there's a chance I'm going to survive in some sort of meaningful condition, I'll have that. I'm not going to say I'm not going to have any treatment, but I don't want to be just kept alive for the sake of it. (Peter, person with CPFE-IPF)*

> *if ever I'm having a heart attack, just leave me, just put me in the recovery position, but don't start pumping on my chest, just let me die. I'd sooner die like that. (Christopher, person with IPF)*

Many had accepted the inevitable decline in their symptoms but had not found the right time to speak to their family about the future. Some were frustrated that family members did not allow them to talk about their worries and fears about how their disease would progress.

> *I mean, without telling my family, I have decided on what sort of a service I want and what sort of music I want. I have decided that, but we haven't talked about it yet. I don't think there's been an ideal time. Maybe there never will be. I think that's*

> *probably what I've been waiting for, but it may never occur. (Susan, person with IPF)*
>
> *It's difficult not to be able to talk to anybody. I realised nobody wants to know, nobody wants to hear about it, nobody wants… Every now and then I get very frustrated, and I shout, "I'm dying, you know." Then the response is, "You know, everybody dies eventually." "No, I'm dying." (Pamela, person with CHP)*

It seems that people with ILD had an understanding that time was short, but that some did not feel empowered to discuss this further with healthcare professionals. They may not have been given adequate opportunity, although in many cases it did seem that there was an active decision to avoid this discussion.

4.5 Dying: "The important bit"

The title of this category comes from a relative who remarked that she had missed "*the really important bit" (Ruth, bereaved relative)*, the end of her husband's life. This category relates to the emphasis and importance placed on the final days and hours of life, what people with ILD wish for the end of life and how getting it right is so important for them and their families. There remains a focus on this short time at the end of a person's life because it can never be repeated, and mistakes cannot be corrected.

People with ILD frequently deteriorated quickly towards the end of life which has a significant impact on the quality of end of life care provided. The first sub-category 'rapidity' refers to the challenges of providing good end of life care in this situation and reviews the associated impact on location of death.

The second sub-category 'wishing for a peaceful death' incorporates both patients' wishes for their end of life care and also the reality of the bereaved relatives experiences. The sub-category 'lasting memories' considers the impact that the process of death has on family members. Positive lasting memories of this experience may provide some comfort for grieving relatives but in contrast others may be haunted by unanswered questions.

Figure 4-5 demonstrates the relationship between the focused codes and sub-categories that contributed to the emergence of this category.

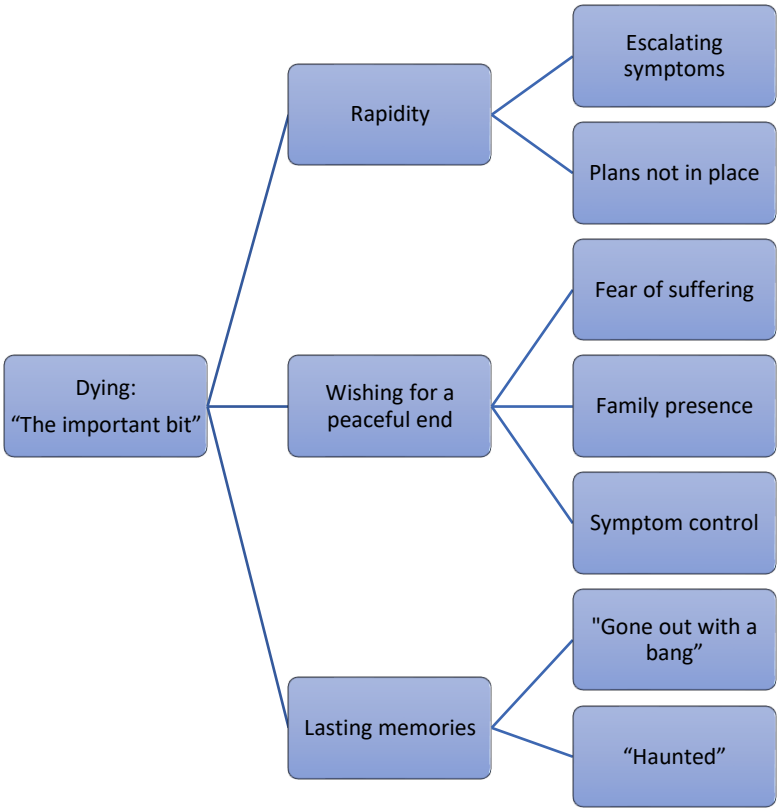

Figure 4-5 Constructing the category 'dying: the important bit'.

4.5.1 Rapidity

All patients and bereaved carers interviewed were aware of the short prognosis associated with their lung disease, however, death was often unanticipated and quicker than expected.

> *I think it might've been just literally the day before he died, and he was in quite, well, you wouldn't have known anything was wrong, you know. He was talking fluently, asking intelligent questions. […] And then literally, you know, he didn't get up the next day, he wasn't in a good state at all the next day, and he went straight into hospital. (Deepta, bereaved relative)*

> *I think it was quicker than I expected it to be. I expected more of a gradual downhill thing, process, where it wasn't, it was just bang, we're here. (Jane, bereaved relative)*

The rapid deterioration and escalating symptoms experienced by most people with ILD towards the end of life frequently resulted in hospital admission. Many resorted to hospital admission due to difficulty managing symptoms, fear of worsening symptoms, or hope of improvement with treatment. Plans were often not in place in the community to allow people with ILD and their relatives to feel confident remaining at home, even if that was previously what they had wished. One man did not have a 'Do not attempt resuscitation' form in place when he collapsed unexpectedly at home, his wife performed

cardiorespiratory resuscitation and he died shortly afterwards in hospital. Another man only had a resuscitation decision made when he was admitted to hospital the day before he died. The absence of advance care planning discussions was evident from interviews with people with ILD and their relatives who viewed dying as a topic to be avoided and as a result few plans were put in place for end of life care.

> *The ambulance people came in and said, "Has he got a Do Not Resuscitate? Has he got this, has he got that? Has he got an end of life?" None of those things, he didn't have any of that stuff, and so really the only option – and there was no oxygen in the house; the oxygen helped him be more comfortable, but you know, there was nothing in place, so they had to take him in at that point. (Deepta, bereaved relative)*

This quote reveals that without clear plans in place for the end of life, people with ILD and their families did not know who to turn to when symptoms rapidly deteriorated. This led to there being no other option than to call an ambulance and for hospital admission. Despite the recognition that their relative was dying, as demonstrated by questions from the ambulance crew about end of life medications at home, nothing was in place to support him at home and hospital admission was inevitable.

Rapid decline in the last few days of life often led to changes in preferences for end of life care as people with ILD and their families were frightened by escalating symptoms. When asked about where they would prefer to die, many people with ILD wished to die at home, with the recognition that their plans would be influenced by the circumstances and speed of their symptom deterioration.

> *what has happened in the past to people I know, is that when your time comes, it's obviously very quick. […] I don't know how you know, but when it happens, you ring up the ambulance and they take you [hospital]. That's where people normally go and you're there for about three or four days before the good lord comes and takes you by the hand. So, that's, in reality... I mean, I would like to think I could be at home, but, in reality, I think I'll be in hospital, again, with nobody that I know. (Alan, person with IPF)*

Alan refers to dying in hospital without his friends or family present, a powerful quote that represents the change in healthcare during the COVID-19 pandemic, and patients' fear of dying alone.

Location of death was unimportant for most of the bereaved relative participants, with some acknowledging that they would not have been able to manage end of life care at home. Some bereaved relatives viewed dying in hospital as the only way to ensure that their

relative received the care needed to adequately manage their symptoms; the continuous presence of healthcare staff allowed rapid response to symptoms and carers to relinquish their caring responsibilities.

> *It was probably the best way that it could've been because we were together. If he had been home, it probably wouldn't have been, if you can say, as nice an experience. I mean it wasn't a nice experience, but I couldn't have wished for any more. (Julia, bereaved relative)*

However, hospital admission for some meant being alone and isolated from family at the end of life. The additional psychological trauma caused by COVID-19 restrictions on visiting was evident from bereaved relatives who described their anguish at not knowing what was happening, as illustrated in the first quote below. Sadly, one person died in hospital unexpectedly during the COVID-19 pandemic, meaning his wife was unable to visit him in hospital due to restrictions. She discussed her plans to sleep in the carpark of the hospital just to remain as close to him as possible.

> *That was a terrible day because I was pacing the floor most of the time waiting for someone to get back to me…. I didn't know what was happening to him…. I knew he would be frightened. […] The fact that I've been I've been with him 24/7 for so long…and I couldn't be with him, you know. (crying) (Dorothy, bereaved relative)*

> *I said that I would go… Oh God. I said I would go to the carpark and stay there, but they said that I didn't need to, that I was to stay here. I just wanted to be close. (Ruth, bereaved relative)*

4.5.2 Wishing for a peaceful end

Dying without suffering was the most important aspect of end of life care for people with ILD and their relatives. Unsurprisingly, people with ILD wanted to be comfortable, with well controlled symptoms and surrounded by their family. They were afraid of escalating symptoms at the end of life and that they would suffer overwhelming and frightening breathlessness symptoms before they died.

> *And hopefully I'll just have a big, massive heart attack and that will be it. Because I certainly don't want to suffer with regard to gasping for breath, as if you're drowning, you know. (Christopher, person with IPF)*

> I'm scared, I'm scared. I'm not scared of the moment of death, I'm scared of everything that goes up to it. I think I would be lying to myself if I said, "Oh yes, I'm fine with it, I'm going to die. Okay." No, I'm very afraid because I don't think… It's more like, "How long am I going to be lying there in bed, is it going to be months?"

> How long does it take? I don't know. I don't want to lie there for months, it sounds horrible. Come on, get it over with. If I can't enjoy anything about being alive, if I'm just lying there drugged because I'm so breathless because my lungs are so bad… *(Pamela, person with CHP)*

The location of death was not an important factor for those patients who were interviewed. People with ILD had differing views about whether they would wish to die at home or in a healthcare environment. Those who had prior experience of local hospices expressed a wish to die in a hospice, however, they were also realistic that this may not be possible if deterioration was quick and unexpected.

> *Yes, we have talked about it. I did say I did want to go into the hospice. With being on my own, I didn't just want to be left on my own. So, towards the end, I would like hospice care. (Lorraine, person with CPFE-IPF)*

> *He had said that he didn't want to die at home because he didn't want to die in bed where I would be sleeping afterwards. He didn't want that. (Ruth, bereaved relative)*

> *A number of our friends […] have died here in hospital alone. I don't want to be in a ward surrounded by strangers and nobody who cares for me there, it just seems like a terrible way to die. (Pamela, person with CHP)*

> *I think she knows that all being well, I'd like to be at home, rather than in hospital, but sometimes that's taken out of your hands, isn't it? (Susan, person with IPF)*

> *Yes, I want to be comfortable, but… Yes, alright, I don't want to be on a bed of nails, or anything, if that's what you're thinking. But, yes, I mean, I've already told you, I don't really want to be cared for in a home, or anything. I'd rather spend as much time at home as I can, provided my wife and daughter can manage me, and I'm not a raving lunatic and throwing things about. (Peter, person with CPFE-IPF)*

These extracts describe that the presence of family and not dying alone were important factors that influenced where people with ILD wanted to die. Bereaved relatives took comfort in being present at the end of their relatives' life and for those whose symptoms were well controlled this was a positive experience. Relinquishing the caring responsibilities was also important to some carers to allow them to spend more meaningful time with their relative at the end of life.

> *Now I realise there were so many people that never got that opportunity with their relatives, and it's so important, I think, to have the last few months, the last few…hours when you're just around them. (Deepta, bereaved relative)*

> *So, I went, I sat with him, and held his hand. He was out, but they do say hearing is the last sense that goes, so I sat and talked to him until he flat-lined. That was it, really. […] You never know when your time is up. At least [name of partner] and I were together, and I was with him at the end. (Maureen, bereaved relative)*

Symptom control at the end of life was crucial to whether relatives felt their loved ones died peacefully. Symptom burden, management of symptoms and availability of support were significant factors influencing the location in which people with ILD died. Only one person died peacefully at home, despite this being the preferred location of death for many of the participants in the study. For this man, his symptoms declined rapidly over a few days, but he had good support from his GP and community teams and never experienced severe breathlessness symptoms.

> *it was very pain free, very peaceful […] It was just so quick, I don't think anybody could have done anything any differently or whatever. As I say I think it is, like you say, a lot of it being down to the GP being so honest, being so open and being so helpful. Putting into place what he actually put into place. (Jane, bereaved relative)*

The majority of people with ILD died in hospital, however, the experience of hospital care was very variable. Factors which influenced satisfaction with end of life care included whether death was expected, communication between healthcare professionals and relatives, symptom control and location within the hospital (i.e., accident and emergency versus a private side room). The experience of healthcare staff was important in developing trust and providing reassurance to patients and relatives.

> *Um, I feel like the young nurse wasn't experienced enough, because when [name of husband] was dying, I said to her, "I haven't seen anyone die before". She said, "Neither have I". Then I thought oh crikey. How experienced was she? You know that you're dealing with the end the life of my husband. […] I think it would have been helpful if it was someone who knew his history, she obviously didn't know how ill he was. … You know, she just didn't have the experience, I think. (Dorothy, bereaved relative)*

As discussed above, people with ILD were afraid of suffering extreme breathlessness at the end of life, a fear which often prompted hospital admission. The experience of Dorothy highlighted the difficulty with managing end of life symptoms in ILD. Her husband was admitted to hospital and received medications for management of breathlessness, but he continued to struggle with his symptoms and did not die peacefully as he would have wished.

> *they can't get their breath, you know, this disease. Obviously, they're going to panic. I didn't want that to happen to him I wanted something to happen just to make him go to sleep and not struggle. I used to say to friends and family, I really don't want him to be struggling at the end and be frightened and panicking. You know, you just want a peaceful end. (Dorothy, bereaved relative)*

4.5.3 Lasting memories

The importance of the final days and hours of a person's life are highlighted by relatives' lasting memories. Relatives were comforted by the knowledge that their relative died peacefully, the way they wanted, and they were not alone.

> *I said if he'd written a list of how he wanted to die and everything, I could have ticked every box that he wanted, which, don't get me wrong, I've been upset, and you do get upset, but I think that's helped me. (Crying) (Jane, bereaved relative)*

Again, the place of death was not as important to relatives' memories as being present and spending meaningful time with their relative. One family described their relatives' death as "*gone out with a bang" (Rita, bereaved relative)* as he had died on New Year's Eve and there had been fireworks in the sky as they approached the hospital to say goodbye. This family described a positive experience where multiple family members were able to gather at the bedside in a side room on a medical ward and they were laughing, reminiscing and spending meaningful time together.

> *And sometimes we made a lot of noise, talking and laughing, which seems terrible now, but we felt that [he] with his mask on could hear us. I mean, gosh. (Crying) It was a very moving 12 hours or more, and the children were there. (Rita, bereaved relative)*

The death of a relative is a deeply personal time and amongst the anguish and heartache of the situation, relatives recalled small pieces of the experience which reminded them of their loved one.

> *He [his son] played his favourite rock and roll music on his phone, next to his dad's ear, and [name of husband] responded, by playing air piano to Jerry Lee Lewis. (Eileen, bereaved relative)*

When relatives were unable to be present at the end of life, they were left with unanswered questions which frequently led to a more complex and prolonged grieving process. Sadly, there were two relatives who were unable to be with their relative at the end of life as they died 'unexpectedly' in hospital. One relative remarked that she *"hadn't been there for the really important bit" (Ruth, bereaved relative)* a fact which left her with unanswered questions.

> *I keep thinking, "I'm over that, I don't need to know it now," but I'm not over that bit because as soon as I get upset and start thinking about it, I've got a list of questions in my head. (Ruth, bereaved relative)*

Relatives relied on healthcare professionals to recognise that their relative was dying. This recognition allowed plans to be put in place at home or hospital visiting restrictions to be eased for a patient approaching end of life. Communicating even the uncertainty of the situation would allow relatives to prepare themselves.

> *I never saw anything that was too similar to the end of life protocol, so I thought, "No, it's not going to happen yet, the doctors would have said." (Ruth, bereaved relative)*

> *The frustration around the visiting was we didn't get a chance to speak to doctors as frequently as you would have wanted, to find out, you know, "Look, what are we looking at here? Is this man going to come home or is he…" (Mark, bereaved relative)*

These excerpts indicate the difficulty experienced by relatives visiting hospital during the COVID-19 pandemic, with restrictions placed on visiting times that resulted in reduced chance for communication with healthcare professionals. In both cases, failure of healthcare professionals to recognise the uncertainty about prognosis and communicate this with family resulted in relatives being unprepared and not present at the time of death. In contrast, a woman whose husband died in a local hospice recalled the excellent communication from healthcare professionals that enabled her and her family to fully appreciate the uncertainty of the situation and the realisation that her husband was sick enough to die.

> *We had to have a conversation with the doctor, who was amazing, and she said that she didn't think [name of husband] would make it home because he'd had quite a few episodes where he'd had to have more help. […] We had a lot of conversations with the doctor, …. She spoke to all the boys and explained about what was going on with their dad and what we wanted to do. (Julia, bereaved relative)*

There were others who were 'haunted' by their relative's death as the process of death had been a traumatic experience. Maureen performed CPR on her husband who collapsed unexpectedly at home and had not previously discussed resuscitation with healthcare professionals. She recalled the trauma of his death but took comfort from the fact that she had been present at the end of his life.

> *It will be some time before I forget doing that C'R … It's different doing it on a person. He had so little flesh on him I could actually hear his ribs when I was doing it, and it will be a time. […] It was very traumatic. It was a shock. I still keep occasionally having a little flashback. (Maureen, bereaved relative)*

Dorothy remembered the overwhelming symptoms experienced by her husband at the end of life and how distressing this had been to witness. Although he died in hospital, his symptoms were difficult to manage and there was a delay in administering medications for symptomatic relief.

> *It was awful. Was cruel to watch, you know. I think if they'd calmed him down quicker…it just haunts me to think of him struggling. […] he was in a terrible state just struggling so much to breathe, you know. […] the whole time [he] was sitting up moving his head back and forward and staring, his eyes were really staring you know. And he tried, he tried to speak to me and took the mask off. The young nurse was there, and she said something awful, she said […] don't do that, she says, don't take that off or you'll die. […] Anyway, he wanted to tell us something and I'll never know what it was. (Dorothy, bereaved relative)*

4.6 Chapter summary

This chapter has presented the analysis of data from people with ILD and bereaved relatives. It has explained the four main categories that were identified from the analysis: i) hidden illness, ii) recognising deterioration, ii) preparing for dying and iv) dying "the important bit". These categories represent the figurative illness journey experienced by people with ILD and their relatives and formed the core category 'navigating the uncertain illness journey with ILD'.

Patients and bereaved relatives recalled a frustrating and protracted period of time prior to diagnosis. The uncertainty surrounding disease trajectory and prognosis weighed heavily on many people with ILD who found it difficult to measure where they were on their illness journey. People with ILD experienced a high symptom burden and the progressive deterioration of their symptoms led to 'loss of self' and increasing reliance on informal carers.

People with ILD avoided talking about dying or end of life care, relying on family and healthcare professionals to make the right decisions at the right time. Towards the end of life, some people reflected on their life experiences and created positive memories, whereas others were overwhelmed by their symptoms and reported a dismal quality of life. Rapid deterioration in symptoms in the final weeks and days of life frequently dictated location of death and patients and family were often unprepared for death.

The next chapter (Chapter 5) presents the results of data analysis from healthcare professionals. The collective findings from all interview data are synthesised in the discussion section (Chapter 6).

Chapter 5 Healthcare professionals' views and experiences of end of life care in ILD

This chapter outlines the constructivist grounded theory analysis of healthcare professionals experience of end of life care in interstitial lung disease. Three main categories were derived from the interview data relating to the roles of healthcare professionals in the end of life care for people with ILD: i) identifying, ii) planning, iii) supporting. Figure 5-1 shows the sub-categories which constructed each category. Each of these categories and sub-categories will be presented in the remainder of this chapter and discussion of the relationship between the categories.

5.1 Sample characteristics

Twelve healthcare professionals were interviewed. These professionals worked in multiple hospitals and hospices around the region and were from a mixture of disciplines, including respiratory consultants, palliative care clinicians and specialist nurses. Fourteen healthcare professionals were contacted and twelve participated in an interview. The sample characteristics of these participants are outlined in the following table (Table 10) and their geographical distribution in relation to the tertiary referral centre is shown on the map in Appendix D.

Participant	**Job role**
HCP_01	Palliative care specialist nurse
HCP_02	Palliative care doctor
HCP_03	ILD / respiratory consultant
HCP_04a	ILD specialist nurse
HCP_04b	ILD specialist nurse
HCP_05	Respiratory specialist nurse
HCP_06	ILD / respiratory consultant
HCP_07	ILD / respiratory consultant

HCP_08	Palliative care consultant
HCP_09	ILD / respiratory consultant
HCP_10	ILD / respiratory consultant
HCP_11	Respiratory specialist nurse

Table 10 Sample characteristics: healthcare professional participants

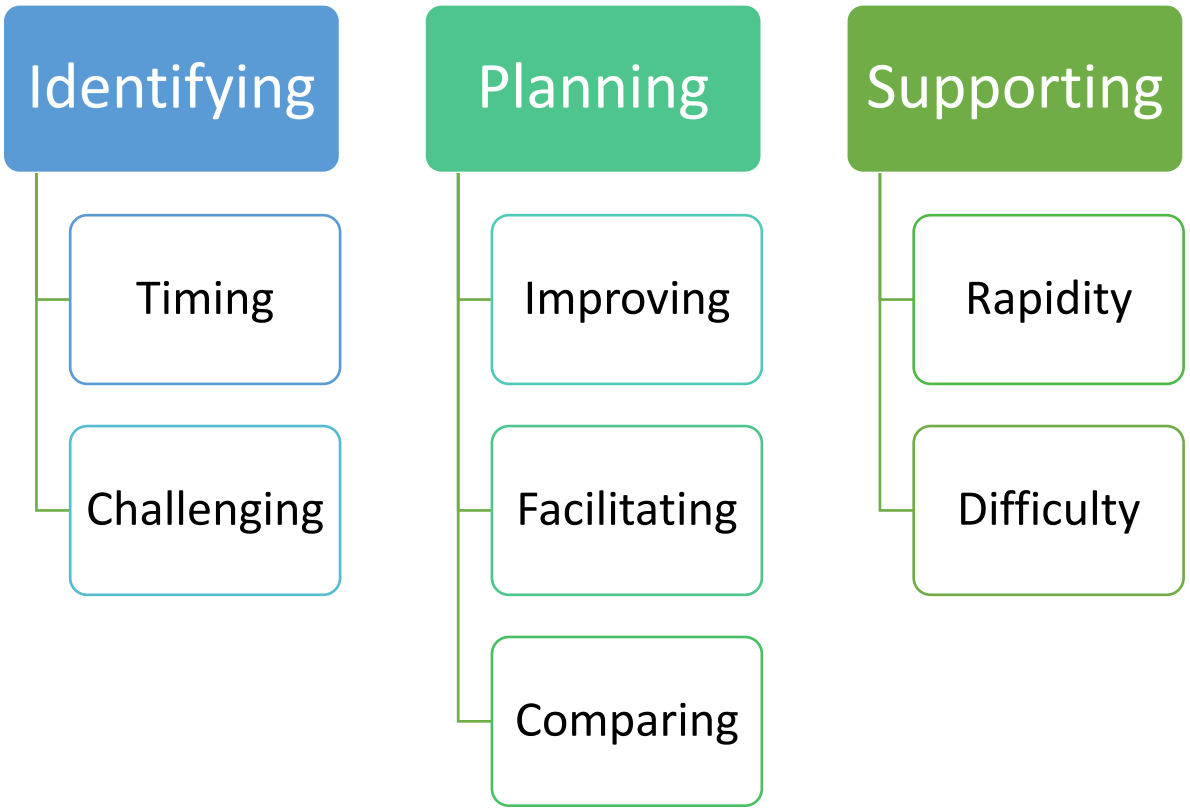

Figure 5-1 Roles of healthcare professionals in the end of life care for people with ILD

5.2 Identifying who needs palliative care

The first category generated from healthcare professional interviews was named 'identifying' (Figure 5-2). This category incorporated the timing of initial discussions with patients about palliative care and the change in focus from active treatment to a more symptom management and palliative approach. This category also encompassed challenges

and barriers perceived by healthcare professionals when involving patients in discussions about palliative care.

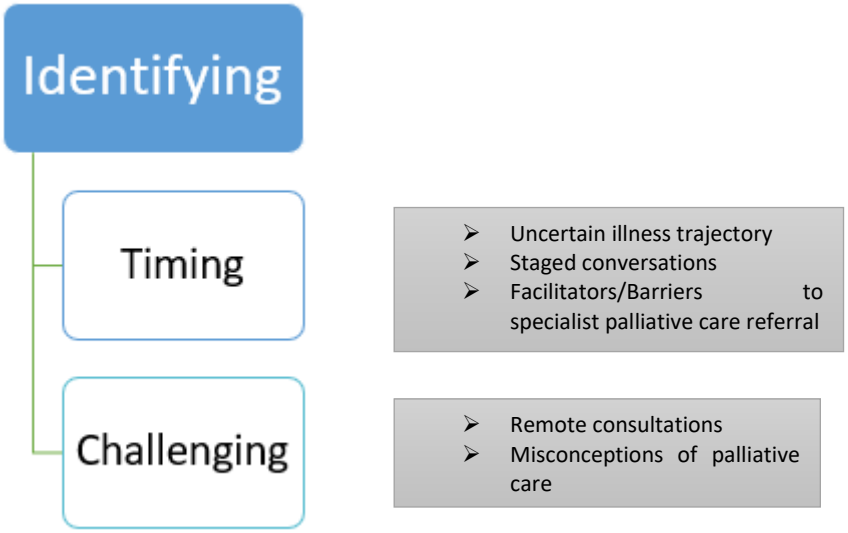

Figure 5-2 Constructing the category 'identifying'.

5.2.1 Timing

All the healthcare professionals were confident in their ability to recognise which patients would benefit from palliative care input. However, the underlying type of ILD influenced clinicians' certainty in predicting disease trajectory and therefore their confidence in discussing palliative care with patients in a timely manner.

> *I think it's always in the back of your mind because we know they've got a life-limiting condition, but some of them die a lot quicker than others. Some of them, you can see they just steadily decline over years. I've been here for three years and there are people that are 'slowly, slowly, slowly', but there are other people who have just gone along on a nice, even keel and then they've just – well, kind of – dropped off their perch. (HCP_04, ILD specialist nurse)*

The steady decline over years allows gradually introduction of palliative care at a time appropriate for each individual patient. However, the description of patients who 'dropped of their perch' references an unexpected decline from both healthcare professionals' and patients' perspective with little time to involve palliative care.

Some clinicians also remarked that palliative care was not appropriate for all patients with ILD. Specifically, they felt that it may not be appropriate to involve palliative care teams in the management of newly diagnosed patients with rapidly progressive disease due to the

expediated nature of their deterioration and the belief that their condition could be stabilised or improved.

> *our main focus in dealing with such an unwell patient was actually to get him the treatment that might actually reverse the condition. […] But actually, getting palliative care involved might not be the right thing for that particular patient. And even though you know they're very unwell, like this chap, we didn't actually get palliative care involved. (HCP_03, Respiratory consultant)*

There was recognition that palliative care involvement was appropriate for many patients soon after they were diagnosed and potentially at the point of diagnosis.

> *I am quite frank at the very beginning, and say, "Look, you are aware, you have been told, that this is not a curative condition. I will be talking to you about things that you think are not pertinent today, but I need to know what's important for you at this moment in time." It's a part of my questioning when I do see them – "What are the important things this week, for you and as a family?" (HCP_11, Respiratory specialist nurse)*

> *But definitely, we try and come in as early as we can to support them through the whole thing, look at quality of life, and support the ILD team as well to support them. (HCP_01, Palliative care specialist nurse)*

Healthcare professionals were convinced of the benefits of palliative care involvement for their patients. However, they preferred to wait until they knew a patient better and had built a trusted relationship before discussing this topic. The decision to refer to palliative care was viewed as a 'shared decision' between the patient and clinician.

> *And I think it's a balance between what you think is best for the patient, and it has to be at the right time. So, I think it's a very individual decision to make, a shared decision between you and the patient. (HCP_03, Respiratory consultant)*

This extract recognises that referral to palliative care services needed to be an individual decision for each patient and the role of healthcare professionals was to initiate the discussion at the right time in the illness journey. However, as previously mentioned the uncertain disease trajectory affected clinicians' ability to identify the 'right time' for some patients. Some healthcare professionals mentioned specific changes in a patient's condition, such as the initiation of oxygen, as a prompt for considering palliative care referral.

> *So, it's mostly if the patient has significantly deteriorated, if I start putting them on oxygen, so if I realise, actually, no, they're now needing oxygen and that's the time when I address it, and if patients decline, but if they're happy, then I said, "Listen, this isn't because you're dying. This is to make introductions, so that, at some point down the line, you're not meeting a new time when you're really suffering quite badly," and then I refer them also to our community respiratory nurse. (HCP_10, Respiratory consultant)*

There was also recognition that some patients with ILD do not want to be referred to palliative care services, however, these patients often receive palliative interventions under the care of their usual respiratory teams.

> *Some patients will say, "I'm not having that," "Is that the Macmillan nurses?" or "That's what you get when you die" or "You go in one of them and you don't come out." You do still get that, or sometimes you're still met with: "Oh I don't want to think about death and dying." (HCP_04, ILD specialist nurse)*

> *So, I do think that sometimes, for some patients it is a really hard thing to accept. I mean, I think for some there is a difference knowing they have got a life limiting illness, but then suddenly palliative care is involved. (HCP_01, Palliative care specialist nurse)*

For some patients who were reluctant to accept referral, healthcare professionals were able to provide reassurance and introduce palliative care gradually over a series of appointments. The gradual and slow disease progression which was observed for many people with ILD allowed *"the luxury of maybe having a bit more time, just to slowly introduce the idea where you transition from a solely treatment- based approach to palliation" (HCP_03, Respiratory consultant)*. The phrasing of the initial discussion influenced whether people were likely to accept onward referral.

> *You do still get that resistance, and I just say, "I'm not telling you you're dying. I'm not telling you you're actively dying. You've got an incurable disease, so palliative care is absolutely right to be in place, and I do think that they can help." Then, it might be that that time, you just let them have a think about it and then the next time, you say, "So what do you think about...?" And they do, most of the time, all come around. (HCP_04, ILD specialist nurse)*

> *I think it takes, on average, about two discussions. So, one where I address it and that that is an option just to help with symptom management and just holistically address their whole lot of symptoms because it isn't always just breathlessness. Often it takes two discussions and saying, "Listen, have you had a thought about it?" not that I want to push them, but, yes, just give them a bit of time to digest it, and most of the time they agree. (HCP_10, Respiratory consultant)*

Healthcare professionals favoured this 'staged discussion' approach where the topic was introduced slowly and gradually over a series of appointments. The difficulty of a staged approach is that patients might unexpectedly deteriorate prior to their next planned review which could mean missing out on valuable support from palliative care specialists and services in the community.

5.2.2 Challenging

Remote consultations, introduced because of the COVID-19 pandemic, presented many new challenges for healthcare professionals and all the interviews touched on this subject as it was a significant change in healthcare practice over the preceding 18 months. Healthcare professionals found it difficult to assess people with ILD over the phone and reported their unease about assessing symptoms such as breathlessness and weight loss. Relying on patient-reported symptoms rather than objective markers of deterioration such as weight loss, oxygen saturations and lung function, was felt to be less reliable and resulted in patients' deterioration being under-recognised.

> *I've found it really very, very difficult assessing people. And particularly thinking of, sort of, symptom control, palliative care needs, and trying to work out exactly how ill they were. (HCP_07, Respiratory consultant)*

> *It is really difficult to assess a breathless patient over the phone. Is that normal breathlessness? Is that breathlessness worse? It is really difficult. You will say to a patient, "Have you lost any weight?" "Oh, I don't think so pet, no." Then a week later they are admitted, and you think, "Oh my God they are emaciated." What they tell you on the phone and what is actually happening is very different. (HCP_05, Respiratory specialist nurse)*

These extracts emphasise the reliance that healthcare professionals place on objective markers of disease progression and the belief that many patients under-reported their symptoms. It is unknown whether this alleged under-reporting of symptoms was deliberate to avoid hospital review and potential admission or whether patients simply did not want to trouble a clinical team whom they perceived to be busy and overworked.

Healthcare professionals often relied on the presence of relatives in outpatient consultations to circumvent the *"uncomplaining" (HCP_07, Respiratory consultant)* nature of people with ILD. Remote consultations often prevented relatives being directly involved in consultations. Relatives offered additional, relevant information about how people with ILD were managing at home and helped to provide a whole picture of life for that person at home.

> *Sometimes, when you see somebody face-to-face, you can pick up on body language, and a relative sitting beside them, where you kind of go, "Actually, I think they might be struggling, I think there might be a role for that." (HCP_03, Respiratory consultant)*

The involvement of relatives in outpatient appointments was perceived to be important for holistic care. Remote consultations presented a barrier for relatives who would like to be

more involved in important discussions, especially when this included consideration of future care needs.

Respiratory clinicians avoided discussing palliative care over the telephone as this was deemed a sensitive subject. Instead, a further face-to-face appointment was often made to discuss the topic further; leading to increased workload in an already busy outpatient clinic and loss of continuity as the follow-up appointment might be with a different healthcare professional.

> *And I think, over the phone, myself, sometimes you can clearly recognise when someone is dying. And actually, having that conversation over the phone, it feels uncomfortable sometimes. That's a bit difficult. For example, we don't like to give lung cancer diagnosis over the phone, and we don't like to talk about palliation, end of life, it's very, very difficult over the phone. (HCP_03, Respiratory consultant)*

This extract also highlights the differences in how a diagnosis of ILD and lung cancer are perceived by healthcare professionals and patients. Both lung cancer and ILD can be terminal, with often similar life expectancies, and initial diagnoses will be shocking to patients and their relatives. Although clinicians endeavoured to arrange face to face appointments to speak to people about a diagnosis of lung cancer, people with ILD were frequently diagnosed over the phone.

Palliative care clinicians also struggled with conducting their consultations remotely. This was less about their own discomfort talking about sensitive topics over the phone, but more about missing the non-verbal cues which help to guide the pace and direction of conversations.

> *Yes, the non-verbal communication. If you've said something to test the waters a bit, you can see their reaction before they say anything. I think you get the more complete picture of how someone's responding to your conversation. (HCP_02, Palliative care doctor)*

This excerpt demonstrates the key difficulties with discussing sensitive topics remotely and opposes healthcare professionals' preference for gentle and gradual introduction of palliative care through staged discussions with patients. If healthcare professionals are unable to observe visual feedback during these conversations, there is concern that patients would not receive the information that they are seeking at an individual pace.

Healthcare professionals acknowledged that many patients and their relatives had misconceptions about the role of palliative care. People with ILD found the thought of

palliative care frightening and felt referral was a sign that healthcare professionals identified a significant deterioration in their clinical condition or that they were actively dying.

> *When I first mentioned it to one lady, she said "Oh, no." I did all my spiel, but she said, "It just seems so final." (HCP_04, ILD specialist nurse)*

> *Some patients will say, "I'm not having that," "Is that the Macmillan nurses?' or "That's what you get when you die" or "You go in one of them and you don't come out." (HCP_01, Palliative care specialist nurse)*

These misconceptions posed barriers which preventing some people with ILD being referred onto specialist services. However, healthcare professionals felt that the phrasing of the initial introduction to palliative care influenced patients accepting the referral.

> *it depends how you phrase your words. And I think palliative care; you don't necessarily- you can say 'palliative care', but you don't necessarily need to say it in those words. And you can, sort of, talk around it a little bit more. And I haven't really found many problems with it. (HCP_06, Respiratory consultant)*

Continuity of care was identified as another challenge which presented a barrier to onward referral to specialist palliative care. People with ILD were reviewed in the regional clinic, but this was often by a different clinician on each visit which affected continuity of care. Some consultations were over the phone and when palliative care issues were identified a face-to-face review was usually arranged. These face-to-face appointments might not always be with the same healthcare professional or might be a long time in the future, therefore disrupting valuable continuity of care and affecting the timing of referral to palliative care.

> *I bring people back. And what I tend to do is see if I can get them- Obviously, our waiting lists are so big at the moment, that we have to try and get them into clinic some other way. So, I bring them to see [ILD specialist nurses]. So, there are those limitations there as well, because I saw the patient but then wouldn't be seeing them on the follow-up. Or I would try to be involved, but not so directly. So, it's difficult, yes. (HCP_03, Respiratory consultant)*

Even when trying to maintain continuity of care through a mixture of face-to-face and remote consultations, it was hard for patients to remember who they were speaking to and which team the healthcare professional was associated with.

> *And even when I speak to them on the phone, a lot of them find that quite difficult because they get rung up by lots of different healthcare professionals and they don't really remember who is who. And they can't put a face to a name, "Who's that nice lady ringing up this time?" and it's really hard for them. (HCP_02, Palliative care doctor)*

These problems with continuity of care and involvement of numerous different healthcare professionals were evident prior to the COVID-19 pandemic, but the reduced face-to-face contact and shift to remote consultations had exacerbated this issue.

5.3 Planning for end of life care

The second category established from the healthcare professionals' data was named 'planning' (Figure 5-3). This category involved recognition that patients were deteriorating and the benefits of palliative care in enhancing patient experience. These included the additional time available during palliative care appointments, the focus on facilitating advance care planning discussions and helping patients to access community services. However, this category also covered healthcare professionals' observation that people with ILD had inequity of access to palliative care resources compared to people with advanced cancer.

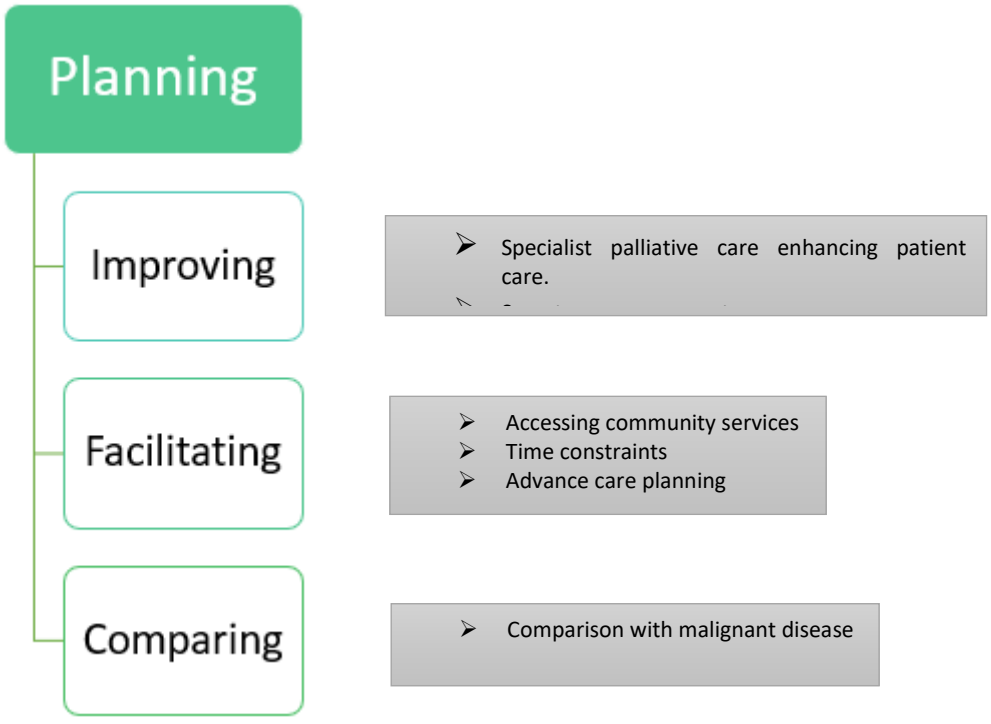

Figure 5-3 Constructing the category 'planning'.

5.3.1 Improving

Healthcare professionals identified multiple benefits for people with ILD who were known to palliative care services. These included introducing medications for symptom management, allowing more time in appointments, improved continuity of care and familiarity towards the

end of life, supporting patients emotional and physical needs and facilitating advance care planning.

> *I'll say to some patients, "Look, I can try and manage your symptoms here, but there are people that are far better at doing this than we are. Their job is to look at everything. Some of the things we don't look at are 'Is there equipment in your home that would make life so much easier?'" that we just don't get around to doing because we are full on with trying to sort other bits and pieces out about their disease. (HCP_04, ILD Specialist nurse)*

This quote highlights some of the difficulties faced by healthcare professionals with providing palliative care to patients within a respiratory clinic. Although many healthcare professionals felt confident providing palliative care interventions, time constraints and the focus on disease modifying treatments meant that they felt palliative care was better provided by specialist palliative care teams. It was seen as the role of palliative care teams to treat patients in a more holistic manner and facilitate support in the community through other professionals such as occupational Therapy.

> *the palliative care team, they should be working in conjunction with social work teams. That would involve an environmental visit and also management of breathlessness, management of pain control, if they have got any other issues. And other things which are required for end of life care. For example, appropriate occupational OT input – bedside toilet, all those things, you know. (HCP_09, Respiratory consultant)*

One of the main advantages of patients being seen by palliative care teams was enhanced emotional and practical support. This was possible due to reduced limitations on time in palliative care clinics, and the multi-disciplinary team who were available to review the patients.

> *giving more time for them to discuss in detail their breathlessness, so we can tease out whether it is all breathlessness, whether it is some anxiety breathlessness, and whether that anxiety breathlessness is proportionate or disproportionate helps us to decide which drugs to use. Then perhaps feed them into- we've got a palliative care OT who does some breathlessness management at the hospice. Perhaps just providing a bit more pastoral, emotional support as well, if we feel as though they are in the last six to twelve months of their life. (HCP_08, Palliative care consultant)*

Hospice services provided short inpatient assessment admissions which allowed respite for carers, improved symptom management, and familiarity with hospice staff and premises towards the end of life. Unfortunately, there was apparent geographical disparity in access to hospice services, which operated on a local basis and were unable to easily admit patients from outside their local area. A local hospice was affiliated with the regional ILD team and

therefore were referred and admitted patients with ILD more frequently than other hospice services. This resulted in upskilling of staff in the hospice who were more confident looking after patients with ILD towards the end of life.

> *So sometimes it is about, why don't we just get them in and let us see what we can do. So that is from a completely patient-centred, holistic approach. So, it is your physical, your spiritual, your psychologic, everything, and plus bringing all the different MDTs, because we have so much we can offer. Sometimes they need to just come in and feel the place, and then they are more willing to try different things as well. (HCP_01, Palliative care specialist nurse)*

> *I have put patients in touch with their local hospices for respite for the outpatient services, and often that's the kind of initial link between ending up in the hospice at a later stage of their illness. […] It's preferable for patients to go somewhere closer to home for most of the time. (HCP_02, Palliative care doctor)*

The two extracts demonstrate that a specific goal or outcome is not always specified in a hospice admission. Patients were referred for a variety of reasons including improving symptom management, psychological support, carer respite and end of life care. The calming hospice environment was appreciated by patients, and some expressed that they would like to return to the hospice for end of life care.

Accessing community palliative care services was more difficult, especially from the tertiary centre, where healthcare professionals did not have in depth knowledge of the services in every part of the region.

> *But certainly, the places where I've worked, I've known exactly who to ask, how to contact them, what added extras there might be available for the patients in that region. Whereas when it's been a patient somewhere where I haven't worked, it's been a bit more time-consuming to find out, or perhaps I've thought I've referred to the right person and it wasn't actually the correct person. (HCP_02, Palliative care doctor)*

Here a rotational palliative care doctor demonstrated that knowledge of the community services available in each local area was crucial to improving access to community services. The quote also highlights referrals were delayed by inadequate knowledge of local services, resulting in more work for healthcare professionals and patients waiting longer to be reviewed.

5.3.2 Facilitating

Palliative care has always been a significant part of the management of people with ILD and was previously provided in the main by respiratory clinicians. However, increasing outpatient

clinic capacity has led to difficulty in providing palliative care interventions and particularly advance care planning discussions in specialist ILD and general respiratory clinics.

> *"The ILD team are a fantastic team, but you have to understand that they have massive clinics, and they very much have to go off what is happening with the lungs and, like, 'What do we need to do with the lungs?' Of course, by recognising there are other things, that is what that team do, they pass it onto us, because they just have not got the time." (HCP_01, Palliative care specialist nurse)*

> *I don't get enough time in a busy respiratory clinic to really provide adequate palliative care. If I think that they are really just palliative, then they come to a palliative care clinic which I do once a week, and they get half an hour. (HCP_08, Palliative care consultant)*

Planning for end of life care has been primarily devolved to the palliative care team (from the tertiary centre) or specialist nurses in district general hospitals where access to specialist palliative care was reduced. Respiratory healthcare professionals viewed the role of specialist palliative care teams as managing patients' symptoms and facilitating end of life care planning with advance care planning discussions.

> *If we are going to it [advance care planning], it needs to be done properly, there is no point just doing it to say that we have done it. So, for us we feel that it is better placed to be done in the community, by either the day unit, or the district nursing team, because then they have got somebody who they can get to know, breakdown some of those barriers, because it is a difficult conversation to have with people. I think us doing it in a 10-minute appointment, is just not appropriate at all. (HCP_05, Respiratory specialist nurse)*

> *So, I'm referring her to Palliative Care because she wants to talk about preferred places of care, she wants to talk about advance care planning, she wants to write a will, she wants to plan her funeral. She wants to just put that in a box, forget about it and concentrate on living and getting a transplant, but she wants to have these conversations and Palliative Care is the right place to have those because I don't feel like that... We don't do very much with that bit. I'm okay with the 'symptoms' bit, but I have to admit I don't do a lot with advance care planning and things like that. (*HCP_04, ILD specialist nurse)

This quote demonstrates that although healthcare professionals often cited time constraints as the main reason for not addressing advance care planning in respiratory clinics, actually many lacked confidence in talking about this topic. In reality, even the palliative care specialists found this aspect of patient care difficult.

> *I do advanced care planning, but it can be quite hard over the phone when I have never met the person. So, I do wait until I get an indication that they are ready for that. But I do some advance care planning, I do talk about resuscitation. (HCP_01, Palliative care specialist nurse)*

Here the challenges of remote consultations are declared again, and the staged discussion approach favoured by healthcare professionals when introducing palliative care continues when exploring discussions about end of life care. Prior to the pandemic, specialist palliative care clinicians were present in the specialist ILD clinic and were able to review patients immediately after palliative care needs were identified. The same staged approach to end of life discussions was followed, but face-to-face review enabled healthcare professionals to develop their relationship with patients and relatives more easily, meaning that these conversations sometimes took place earlier.

There was recognition that patients with ILD were 'palliative from the time of diagnosis'. Respiratory clinicians at the tertiary centre felt that this was conveyed to patients and their families, and most people were fully aware of their poor prognosis. However, this view was challenged by a palliative care specialist who remarked that patients were often not fully aware of their prognosis when referred to palliative care and that this resulted in difficult conversations at this stage.

> *I think a lot of patients with ILD, and their carers, are not prepared that they could die from the ILD in the same way that someone who's been told, "You've got terminal cancer, it's stage four. The average patient lives for two months with this cancer." They don't have their heads around it in the same way, and I find the conversations a lot more difficult. (HCP_02, Palliative care doctor)*

Here the doctor reflected on the challenges of discussing end of life with patients with ILD compared to those with advanced cancer who had a clearer understanding of their likely prognosis. These challenges are not unique to people with ILD but were a consideration for people with many types of non-malignant disease. Advance care planning for people with ILD involved discussion about management of acute exacerbations and acknowledging that they may become rapidly more unwell.

> *I have found it quite difficult compared to doing [advance care planning] with patients with cancer, if I'm honest, for the reason that the patients seem to deteriorate so quickly, and seeing someone breathless is really, really scary. Whereas most patients with cancer tend to get more sleepy, and that tends to be less scary for families and carers. (HCP_02 Palliative care doctor)*

5.3.3 Comparing

Access to community services for patients with ILD was regarded as deficient compared to patients with lung cancer. There was a perception that community teams lacked the knowledge about ILD and were not as quick to respond to requests for home support services as they were for patients with a malignant diagnosis.

> *If you have got a patient who is a non-cancer diagnosis but has this awful quality of life, I find often I feel like you are banging your head against a brick wall sometimes to get them visits. If I refer for a support visit for somebody with interstitial lung […] I might be met with, "Well we will put them on for a telephone. We will ring them in the next four to six weeks." If I rang and said, "I have got a lung cancer patient who is the best supportive care, can you put them on for a support visit?" "Yes, no problem we will go out on Friday and see them." The difference is vast. It is huge. (HCP_05, Respiratory specialist nurse)*

> *[district nurses] often try and get me to do a home visit with them or on my own, because they feel a bit out of their comfort zone, so that can be quite difficult. I think they are probably not getting as good a deal as cancer patients with pain. (HCP_08, Palliative care consultant)*

> *But if they've got cancer, everything changes, everybody will want to go with them. So, if you've got lung cancer, I can get everybody involved. But if they have just got solely ILD, it's an absolute nightmare. But you can say that for any chronic condition. (HCP_11, Respiratory specialist nurse)*

Macmillan nurses were viewed at 'gatekeeping' services and often requested a GP or district nurse review to assess palliative care needs prior to agreeing to review the patient at home. This was frustrating when the referral was being made by experienced palliative care healthcare professionals who had already spent time speaking to the patient and assessing their palliative care needs.

> *And sometimes I've been asking for the patient to be seen by the specialist palliative care team in their community, and the answer has been no, they have to be seen by the district nurse first, and I've found that really frustrating. And I'm not blaming the district nurses. I just don't think that you wouldn't expect them to know all the same things about it. (HCP_02, Palliative care doctor)*

There was a perception that the lack of engagement from community services arose from inadequate experience and low confidence managing patients with ILD. Breathlessness was viewed as a complex and difficult symptom to manage, resulting in patients with ILD having reduced access to palliative care services in the community.

> *One of the things I encounter is that the community palliative care team feel less confident managing breathlessness than they do cancer patients with pain. I think they find it harder to know the sort of trajectory of decline in chronic lung disease. I*

> *find them often being a bit more hands off. […] I think that they don't get as much experience of that sort of patient generally. Those sorts of patients probably aren't discussed at palliative care MDTs as much as they perhaps should. They don't get admitted to hospices. They probably don't get as much input from the palliative care consultants in the community. (HCP_08, Palliative care consultant)*

> *I think because of ILD being a rarer disease, often the district nurses might not have the required knowledge to provide the basic palliative care for those patients in comparison to similar patient groups like patient with severe COPD. So, they might apply principles from the wrong patient group to the ILD patients, and it's not always particularly helpful. (HCP_02, Palliative care doctor)*

However, one factor described by many healthcare professionals was the staffing crisis in community district nursing and palliative care teams. Community resources were stretched, and this may have magnified the difference in care between patients with malignant and non-malignant diagnoses.

5.4 Supporting end of life care

The final category generated from the healthcare professionals' data was named 'supporting' (Figure 5-4). This category encompasses the role of healthcare professionals in supporting people with ILD at the end of life by recognising dying, managing symptoms and providing psychological support. Healthcare professionals reflected on symptom management at the end of life and how the rapid decline towards the end of life presented difficulties with providing end of life care and influenced location of death.

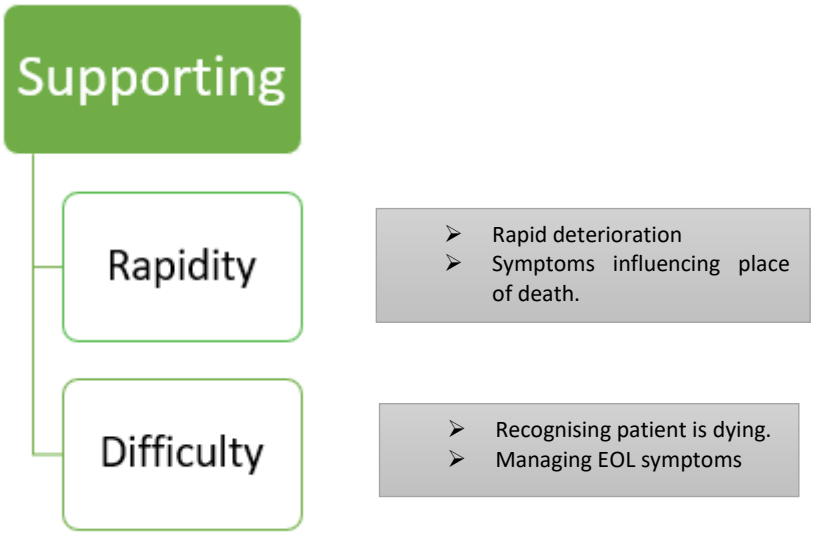

Figure 5-4 Constructing the category 'supporting'.

5.4.1 Rapidity

All healthcare professionals commented on the speed of deterioration in patients' symptoms in the last few weeks and days of life. Although healthcare professionals expected this sudden decline, they reported difficulty in predicting prognosis even when patients were in the last few weeks of life.

> *You know, one of those patients who, "How have you been?" "Oh yes, managing quite well. Walking three miles a day, doing things." You know, not the sort of person I would consider doing a palliative care referral to community. And then an acute exacerbation, and just has come in and died. (HCP_07, Respiratory consultant)*

> *I think it's that communication as well, especially with COVID, that you can't get the families in quick enough. The families don't know what's going on. It's a quick, rapid deterioration. They've come in with an infection and then, the next minute, they're dying. (HCP_11, Respiratory specialist nurse)*

These excerpts identify the speed of onset of an acute exacerbation which can be life-threatening and largely unexpected by the patient and their family. Some patients may never be referred to palliative care services as they are not felt to have reached 'that stage' in the illness journey, yet they can die unexpectedly from an acute exacerbation. Healthcare professionals found difficulty in explaining this risk to patients and their families.

> *He was with us for a week and, very unfortunately, despite all the treatment we could give him, he deteriorated very, very suddenly on ITU. And the time that we had to get palliative care involved and recognise that was incredibly short, particularly when our main focus in dealing with such an unwell patient was actually to get him the treatment that might actually reverse the condition. (HCP_03, Respiratory consultant)*

This quote highlights that the focus of care during an acute exacerbation is active treatment and reversal of the inflammatory process, however, this can sometimes come at the expense of involving palliative care teams for someone who may equally not recover from this acute deterioration. Some clinicians viewed involvement of specialist palliative care teams as unhelpful for the morale of patients who were acutely unwell, preferring to focus on the possibility of recovery from the acute illness.

The rapidity of symptom deterioration appeared to influence the place of death for people with ILD. Healthcare professionals agreed that many patients with ILD died in hospital, despite expressing a preference for dying at home. The general consensus was that the high

burden of symptoms and potentially high oxygen requirements forced patients to be admitted to a healthcare environment for end of life care even if this was not their preferred place of death.

> *I think that can be frightening as well, being at home, I think, especially when you have got the symptoms. […] I think some patients who we have looked after who have died would be hard work in the community, and distressing if they are not getting some extra medicine or the staff to come and take some pressure off the family, reassuring the family. (HCP_01, Palliative care specialist nurse)*

> *It's awful because it's not where they want to be, but there's only so much oxygen somebody can get from a concentrator a' home. I don't know how we get that better – because it needs to be better. (HCP_04, ILD specialist nurse)*

For many people with ILD, the anxiety of poorly managed symptoms in the community influenced the decision to remain in hospital. Healthcare professionals recognised that in some cases patients with ILD were very symptomatic towards the end of life and that this could not always be managed in the community. Healthcare professionals also viewed the use of high-flow oxygen as a barrier to considering end of life care in the community.

> *They get in and they're on such, you know, degrees of high-flow oxygen and so debilitated at the time that it is then quite tricky to get them home again. (HCP_07, Respiratory consultant)*

This quote suggests that some healthcare professionals felt more comfortable caring for people with ILD in a healthcare environment rather than planning discharge home due to the degree of perceived symptoms. Community carer support was limited and people with ILD remained 'stuck' in hospital due to lack of community resources.

> *I think the hindering thing is if they need care. If they need any increase in care, then they're stuck, just because there is no carer support service out there for the patch. That cannot be delivered. (HCP_10, Respiratory consultant)*

The speed with which people with ILD deteriorated towards the end of life was recognised as a challenge to providing end of life care in the community. Advance care planning was particularly difficult due to the potential for rapidly changing circumstances.

> *I've tried to come up with emergency healthcare plans, etc., to facilitate that, but my observation of people dying of ILD in hospices and hospitals is that they tend to deteriorate very quickly and be very acutely unwell, and that's something that is difficult to manage at home, whatever your condition is. So, I don't know how achievable it is to have end of life care at home with ILD and the availability of support that we have in this country at home. (HCP_02, Palliative care doctor)*

In this extract, a palliative care doctor explains the difficulty she finds with discussing advance care planning with patients given the uncertainty about managing their symptoms at home. Although most patients would prefer to die at home, many do not comprehend the likely acute deterioration in their symptoms at the end of life and frequently plans were not in place to manage this acute change in palliative care needs. Death in the community required recognition that a patient was dying, which was difficult for even the most experienced clinicians.

> *Hard, really hard, yes. I think it is probably easier in interstitial lung disease than it maybe is in COPD. But it is much harder than it is in lung cancer, yes. I feel I probably have as good a grasp as anybody and I still find it very hard. I think it just is generally hard. (HCP_08, Palliative care consultant)*

5.4.2 Difficulty

Healthcare professionals reported difficulty providing good end of life care for people with ILD. Management of breathlessness symptoms was particularly difficult, requiring the delicate balance of medications to relieve fear and anxiety and avoid excessive sedation.

> *I think extreme breathlessness can be quite hard to manage at end of life, without causing sedation. It is easy to sedate somebody, but to keep them alert and having some quality of life whilst managing severe breathlessness is really difficult. It is quite difficult to predict which patients will respond to medication and which medication. (HCP_08, Palliative care consultant)*

This excerpt from a palliative care consultant demonstrates that this was a widespread challenge experienced by even experienced palliative care clinicians. The major issue presented with uncertainty about medication effects was that often patients with ILD required immediate adjustments and escalation of medications which was almost impossible to achieve in the community.

Some remarked that patients with ILD often had poorly managed symptoms at the end of life and medications aimed at symptom control were less predictable and often less effective than in malignant disease.

> *I think extreme breathlessness can be quite hard to manage at end of life, without causing sedation. It is easy to sedate somebody, but to keep them alert and having some quality of life whilst managing severe breathlessness is really difficult. It is quite difficult to predict which patients will respond to medication and which medication. (HCP_08, Palliative care consultant)*

> *I still don't think we're as good at treating breathlessness as we are at treating pain. I think that there are definitely more limitations on the management of breathlessness than there are on things to try for pain control. (HCP_02, Palliative care doctor)*

This second quote mentions the limitations of treatments aimed at managing breathlessness and hints at a comparison between end of life care in ILD and in malignant disease. When the symptoms experienced by people with ILD at end of life were compared to those with malignant disease, ILD patients were more alert, aware they were dying, anxious and symptomatic from breathlessness.

> *I think with ILD, there's just so much insight. There's already such a build-up for them, because they've lived with it for so long, and that, kind of, fear of what's going to happen. (HCP_06, Respiratory consultant)*

> *So, I think, especially with the rapidly progressive ones, getting in top of symptoms, and maybe the psychology around these symptoms, is incredibly difficult. Yes. So, I think she was actually symptomatic, which was unfortunate, but a reality, I think, in these particular cases. (HCP_03, Respiratory consultant)*

Here a respiratory consultant reflected that it was inevitable that some patients with ILD would remain symptomatic at the very end of life. The psychology surrounding breathlessness and associated fear of dying was an aspect of symptom management that was not frequently addressed. Healthcare professionals remarked on their difficulty and distress managing patient with uncontrolled symptoms towards the end of life.

> *it can be very distressing when you see how symptomatic they are and yet wakeful at the same time. (HCP_02, Palliative care doctor)*

> *I always find it very difficult managing patients at the end of life, particularly when they're acutely unwell. Basically, the only answer seems to be to have large doses of morphine-midazolam sometimes. (HCP_07, Respiratory consultant)*

The difficulty expressed by healthcare professionals was related to an appreciation of the massive symptom burden and fear experienced by patients. People with ILD generally remained alert towards the very end of life and this required healthcare professionals to actively engage with someone who is aware that they are dying. This compares to other patients who are often unconscious at the end of life, circumstances which were more familiar and comfortable for the treating clinician. Healthcare professionals explained the requirement for high doses of sedating medications to adequately manage patients' symptoms as another factor that contributed to discomfort providing end of life care.

Those healthcare professionals who worked in palliative care recalled patients who had died in a hospice and reported well controlled symptoms towards the end of life. Healthcare professionals based in hospitals were more likely to talk about patients with poorly controlled symptoms at the end of life, which may reflect their bias towards remembering these more distressing cases. In the following quote, a palliative care nurse reflected on achieving a peaceful death in a hospice environment and the contrast between this and a busy hospital ward.

> *So, it is nice, I think, when people can come away from that, they can come into a place where they can shut the door, you look in the garden, and it is beautiful. That is how you want people to die, peacefully. Not on a busy ward with acute patients. (HCP_01, Palliative care specialist nurse)*

This description of the calming hospice environment and its influence on achieving a peaceful death is a sharp contrast to the symptomatic, anxious patients described by hospital respiratory clinicians. The hospice is portrayed as providing a calm space for patients, on the other side of the door, a space for relaxing and reflection. The quote suggests that the hospice provides additional benefits of escapism from people's situation provided by the less medicalised, 'calmer' environment of a hospice and the importance of nature in distracting patients from symptoms. Unfortunately, an acute exacerbation of ILD and rapid decline in symptoms often led to people with ILD being admitted to hospital where the peaceful death reflected on in this statement may have been more difficult to realise. In reality, many people with ILD die in hospital, but efforts are made for this to be in a side room and as such a quieter, more peaceful environment.

Dying in the community relied on intensive district nursing input to support people and their families at the end of life. Healthcare professionals felt this support was limited by staffing issues in community teams leading to reduced capacity to adequately support people at home. Taking into account the difficulty described by experienced respiratory clinicians in predicting trajectory and managing breathless symptoms at the end of life, it is unsurprising that many felt that district nursing teams were unprepared for this role.

> *It relies upon the district nurses. The district nurses are the bedrock, at the end of the day, they're the ones at the coalface. Patients exacerbate so quickly, they take you by surprise. You think they're alright one minute, and then the next minute, they're dying. But we do rely upon the district nurses, and I don't feel they're educated enough on interstitial lung disease. (HCP_11, Respiratory specialist nurse)*

5.5 Chapter summary

This chapter has presented an analysis of the results from healthcare professionals who participated in the interviews for this research study. The three main categories presented, i) identifying, ii) planning and iii) supporting, relate to the role of healthcare professionals in the end of life care of people with ILD.

Healthcare professionals recognised uncertainty surrounding prognosis and problems identifying which patients would benefit from referral to specialist palliative care. Further challenges were recognised in the reliance on remote consultations and misconceptions that people with ILD held about palliative care. The benefits of palliative care for people with ILD were well recognised by healthcare professionals, especially the improvement in end of life care planning. The rapid deterioration and difficulty managing symptoms at the end of life presented particular challenges for healthcare professionals and also influenced location of death.

The next chapter (Chapter 6) synthesises the results from patient and bereaved relative data (Chapter 4) and healthcare professional data from this chapter. The discussion chapter then compares and contrasts the study findings with existing literature and highlights the new knowledge generated by this study.

Chapter 6 Discussion

The purpose of this chapter is to synthesise and summarise the primary findings from patients, bereaved relatives and healthcare professional participants. The study findings are compared to current literature, with discussion of how they relate to the wider body of literature. Reflections on the research process and the strengths and weaknesses of this study are presented. Finally, this chapter concludes with recommendations for future research and implications for practice.

6.1 Purpose of the study

The purpose of the current study was to explore the end of life care experience for people with ILD and examine potential barriers to accessing palliative care services. The study sought to answer the research questions from the viewpoint of people with ILD, bereaved relatives and healthcare professionals involved in the care of patients with ILD. Specific research questions were:

- What is the end of life experience of people dying with ILD?
- What factors influence patients' and relatives' satisfaction with end of life care?
- Are there any perceived barriers preventing access to palliative care services in the North East and Cumbria?

6.2 Synthesis of results and introduction of overarching categories

To present a unified synthesis of results, this section builds on the findings presented in Chapters 4 and 5 and synthesises these data in four overarching categories: i) acknowledging uncertainty, ii) assessing and organising support, iii) avoiding discussion about an uncertain future, iv) accelerating symptoms at the end of life. This section also situates the findings of this study within the current literature.

The following diagram (Figure 6-1) depicts the main categories that shape the patients' and bereaved relatives' illness journey (Chapter 4) and how the role of healthcare professionals (Chapter 5) correlate with these categories. This diagram also illustrates the key categories identified from both data sets and the relationship between the overarching categories identified from integration of the data.

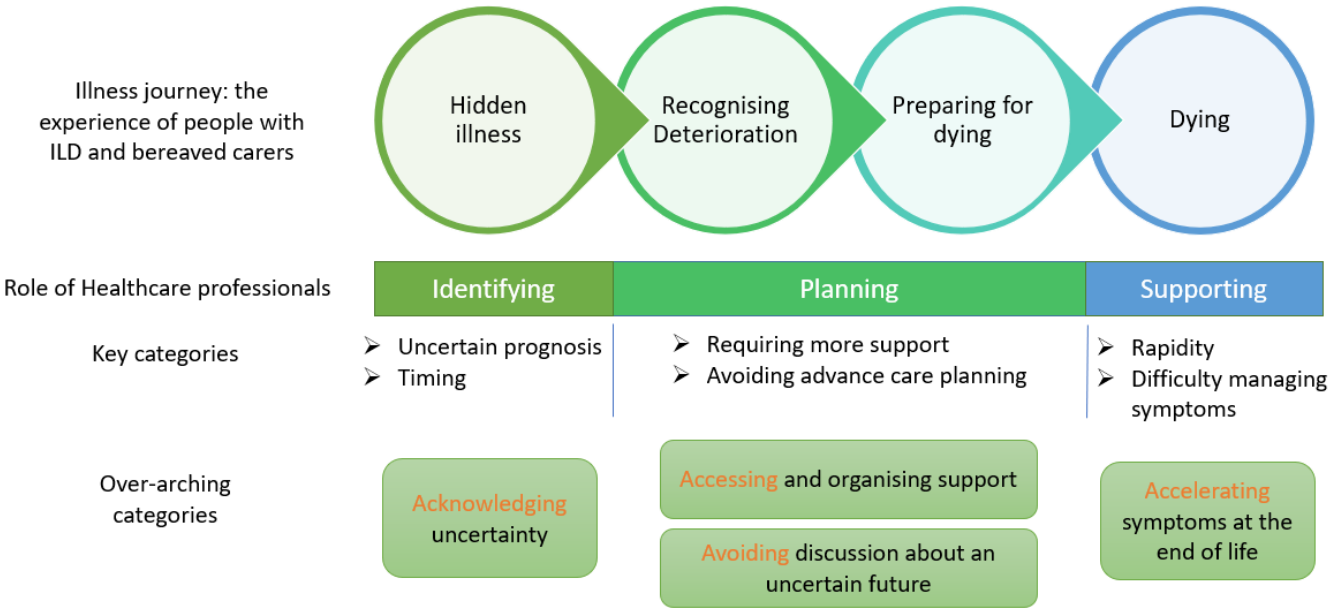

Figure 6-1 Synthesis of whole data set and development of overarching categories

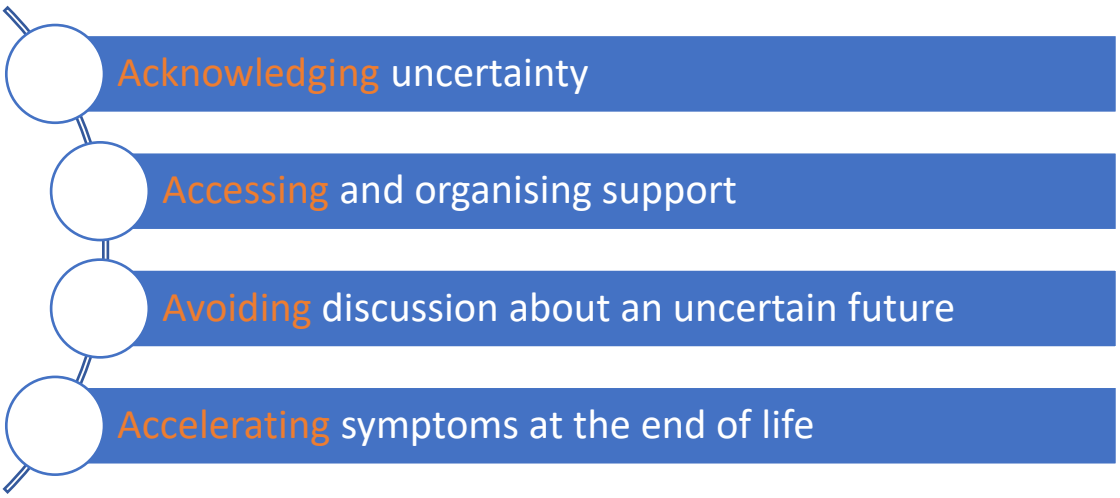

Figure 6-2 Final overarching categories generated from data synthesis and analysis.

The overarching categories (Figure 6-2) were identified from synthesis of the patient, bereaved relative and healthcare professional data. They represent the major abstract categories which emerged from both data sets and demonstrate a shared understanding of the experience of end of life care for people with ILD. The scope of each of these final overarching categories is defined in the following section and the relationship between the categories and the research question is explained.

6.2.1 Acknowledging uncertainty

'Acknowledging uncertainty' references the uncertainty and anxiety experienced by people living with ILD from the time of initial diagnosis and towards the end of life. The uncertainty surrounding disease progression and prognosis meant that both people with ILD and healthcare professionals avoided discussion about the future. This uncertainty was a source of significant anxiety and psychological distress for people with ILD and their families.

ILD was viewed as a 'hidden illness' within society and many participants experienced a protracted diagnostic period due to initial misdiagnosis and treatment for more common respiratory disease. Current literature has identified that people with ILD were often diagnosed late in the disease trajectory (Hoyer, Prior et al. 2019, van der Sar, Jones et al. 2021) and that misdiagnosis with alternative conditions was common, affecting over 50% of patients (Cosgrove, Bianchi et al. 2018). Delayed referral to specialist centres, often related to diagnostic delay and misdiagnosis, has been recognised as a source of additional worry and concern for people with ILD (Duck, Spencer et al. 2015). Following review at the specialist centre, some participants in the current study still had limited understanding of what to expect in the future due to the uncertain disease trajectory. This aligns with the findings of Bajwah et al (2013) who reported that people with ILD had a good understanding that their disease was terminal, but poor understanding of their individual prognosis or how their disease would manifest towards the end of life.

Prognostic uncertainty early in the illness journey weighed heavily on people with ILD and their relatives. This uncertainty was fuelled by diagnostic delay leading to significant importance being placed on review in the specialist ILD clinic. Prognostic uncertainty is common in chronic lung disease and has been associated with lower rates of implementation of palliative care (Coventry, Grande et al. 2005). Acknowledging the uncertainty surrounding disease progression and prognosis in ILD would help healthcare professionals to support patients in navigating their illness journey and allow open discussion about future fears and requirements. However, healthcare professionals recalled their difficulty with identifying an individual patients' prognosis which contributed to discomfort talking about prognosis in detail with patients and relatives. Wijsenbeek et al

(2022) highlighted that is common for clinicians to find communicating prognosis in IPF challenging due to the variable course of the disease, the risk of acute exacerbations and poor prognosis. The authors also emphasised the importance of not allowing this uncertainty to lead to "prognostic paralysis" preventing clinicians from having meaningful conversations with patients.

Uncertainty about disease progression and prognosis presented a challenge for healthcare professionals when identifying the appropriate time to initiate discussions about palliative care. Prognostic uncertainty and unpredictable disease trajectory are factors that have been previously identified as barriers to timely involvement of palliative care services for people with other chronic respiratory diseases (Jabbarian, Zwakman et al. 2018, Mc Veigh, Reid et al. 2018). Appropriate timing for palliative care discussions in ILD has been considered in existing literature with authors in agreement that earlier palliative care involvement is beneficial to patient care (Lindell, Liang et al. 2015, Archibald, Bakal et al. 2020). However, identifying when to refer people with ILD to specialist palliative care teams remains a challenge. It is generally accepted that decisions about palliative care involvement should be individual to each patient and prompted by changes in health status (Sampson, Gill et al. 2015, Kim, Olive et al. 2020). In the current study, healthcare professionals responded to and relied on deterioration in patients' health status to prompt thoughts of onward referral to palliative care services. However, infrequent and remote consultations hindered healthcare professionals' ability to monitor patients and, in some circumstances, meant that the opportunity to discuss palliative care was missed. Healthcare professionals favoured staged discussions about palliative care over a series of outpatient appointments, viewing this approach as protecting patients from being frightened by sudden introduction of the subject. This staged approach is supported by qualitative research involving caregivers of people with advanced cancer who preferred to receive written resources supplemented by conversations that were ideally staged overtime (Collins, McLachlan et al. 2018).

The participants in this study advocated that earlier palliative care involvement for people with ILD was required to allow time for effective symptom management and planning end of life care. Earlier introduction of palliative care has been associated with an increased likelihood of people dying at home, both in cohort and population level studies (Seow, Sutradhar et al. 2021, Driller, Talseth-Palmer et al. 2022) and smaller studies involving patients with ILD (Kalluri, Claveria et al. 2018, Archibald, Bakal et al. 2020). These studies

suggest that when patients were involved with palliative care earlier in their disease trajectory and given the opportunity to participate in advance care planning, their end of life outcomes improved. Earlier palliative care involvement can only be achieved with acknowledgement of the uncertainty surrounding disease progression and openly discussing the uncertain prognosis faced by many people with ILD. Healthcare professionals and some patient and bereaved carer participants asserted the benefits of early involvement of palliative care services, including the holistic approach to symptom management, psychological support and facilitating advance care planning discussions. A significant benefit of palliative care involvement was improved continuity of care with regular consultations or visits and helping patients to access community services. This improved continuity of care was attributed to increased time and resources for more frequent (sometimes weekly) contact with palliative care specialists who regularly liaised with the ILD team. This finding is supported by qualitative research that found patients and families were most likely to experience continuity of care with the involvement of a small number of healthcare professionals, who provided multidisciplinary care and regularly updated all healthcare professionals who were involved (den Herder-van der Eerden, Hasselaar et al. 2017).

6.2.2 Accessing and organising support

There were several factors which led to difficulty for people with ILD organising and accessing support. Initially, many people with ILD had misconceptions about the role of palliative care and some were reluctant to be referred onto services that would have helped them to access support in the community. The misconception that palliative care is synonymous with end of life care is common, with many people identifying palliative care as caring for people in the final few days of life (McIlfatrick, Hasson et al. 2014). Geographical disparity in access to support services was evident from both sets of interview data. People with ILD were less likely to appreciate the geographical barriers placed on them, but healthcare professionals were able to compare the care received by individuals in different areas and identify this disparity in access to services. The inequality in access to palliative care and support services between rural and urban locations has been described in existing literature and is not unique to this study (Mc Veigh, Reid et al. 2017, Zou, Nouraie et al. 2019). These findings are supported by previous work by the North East and Cumbria ILD service that showed a clear centralisation of access to specialist palliative care around the

tertiary ILD centre and more urban locations (Kavanagh, Palmer et al. 2017, Wakefield, Palmer et al. 2019).

Healthcare professionals also highlighted the inequality between people with ILD accessing services compared to people with malignant diagnoses. The diagnosis of lung cancer appeared to have a protected status that ensured outpatient appointments, specialist nursing staff and access to services was preserved despite pressures on general respiratory services from the COVID-19 pandemic. Unfavourable comparisons between the care of patients with malignant and non-malignant disease are well established in existing literature. Exley et al (2005) highlighted that compared to people with cancer, those people dying of end-stage cardiorespiratory disease were less likely receive comprehensive information about their prognosis, to be aware that they were dying, or receive district nursing care. In more recent literature, comparisons between the care received by patients with chronic lung disease including ILD and lung cancer frequently highlighted deficiencies in the care received by patients with non-malignant diagnoses (Ahmadi, Wysham et al. 2016, Koyauchi, Suzuki et al. 2021). Koyauchi et al (2021) conducted an observational study in Japan that focused on end of life care and found significant inadequacies in the care received by patients with ILD compared with lung cancer. These discrepancies are also well established in relation to patients with other non-malignant conditions, who tend to be more unwell with low performance status and a poorer prognosis at the time of review by specialist palliative care (Lau, Meaney et al. 2021).

In the current study, healthcare professionals were frustrated by the difficulty they experienced arranging community support for patients with ILD compared to their patients with lung cancer. It was suggested that barriers to providing community support for ILD patients related to insufficient knowledge of the disease, underappreciation of the symptom burden experienced by patients and low confidence managing breathlessness symptoms, resulting in community teams feeling unconfident managing ILD patients. Staffing shortages in community Macmillan teams was also seen as a barrier preventing adequate support, meaning that patients with malignant diagnoses were given priority where resources were scarce.

A common viewpoint from patient and bereaved carer participants was disappointment and perceived abandonment by healthcare teams. This was highlighted in response to cancelled

and delayed appointments and the change to remote consultations which were precipitated by the COVID 19 pandemic. People with ILD and bereaved relatives did not view remote consultations as an adequate alternative for in-person appointments and there were examples of delayed palliative care referrals due to reliance on remote consultations. Many felt they had not been properly assessed unless they had a face-to-face appointment. Regular review by a palliative care specialist was also not regarded as a replacement to appointments with the ILD service and people with ILD expected their clinic appointments to continue with the same frequency. Although healthcare professionals preferred in-person appointments, the awareness that patients felt neglected and disappointed with services was not prominent in healthcare professional interviews. Remote consultations could be viewed as a way to counteract the effect of tertiary centralised services. However, there were inherent problems with remote consultations identified by healthcare professionals in this study most notably the difficulty identifying patients who would benefit from referral to specialist palliative care and discomfort talking about palliative care over the phone.

Recently published literature has considered remote consultations in view of the restrictions on healthcare provision due to COVID-19. Remote consultations in specialist palliative care were generally acceptable to patients and relatives, although the evidence was limited outside of patients with cancer (Jess, Timm et al. 2019, Sutherland, Stickland et al. 2020). Evidence in ILD was constrained to an international survey of healthcare professionals' opinions about the use of telehealth for monitoring patients with ILD (Althobiani, Alqahtani et al. 2021). Objective measures such as spirometry and oxygen saturation monitoring were felt to be the most useful for monitoring patients and identifying requirement for in-person appointment. A systematic review considering remote consultations in COPD found there was no overall benefit from remote monitoring in addition to usual care (Janjua, Carter et al. 2021). Further research is required to assess the utility of remote consultations for monitoring patients with ILD in the community. Results from the current study suggest that there is no substitute for in-person review from patients' and relatives' perspective; this also remains healthcare professionals' preference for patients with advanced disease.

This study was undertaken during the COVID-19 pandemic and invariably this influenced access to healthcare and organising support for people approaching the end of life. Several bereaved relative participants were unable to see their loved ones at the end of life due to hospital visiting policies and this appeared to adversely affect many aspects of their grief and

adjustment after their relative's death. Qualitative interview data from bereaved relatives and healthcare professionals has highlighted the tensions between hospital visiting polices and what both healthcare professionals and relatives felt was appropriate at the end of life (Hanna, Rapa et al. 2021, Hanna, Rapa et al. 2021). Access to community services, both medical and social, was limited during the pandemic and people with ILD felt isolated and abandoned. This was particularly difficult for those people who lived alone or who had been regularly accessing support services, such as hospice day unit facilities. There has been significant strain placed on both hospice and community palliative care services during the pandemic adding further demands to the already stretched workforce (Kates, Gerolamo et al. 2021, Hasson, Slater et al. 2022).

6.2.3 Avoiding discussion about an uncertain future

Avoiding discussion about the future was a significant issue identified in this research study. Despite the focus of the study being end of life care in ILD, interviews with patient participants often did not cover end of life care as people were reluctant to talk about the future. These views are common amongst patients with advanced respiratory disease. A systematic review identified that around half of patients with advanced COPD did not want information about end of life care as they felt this knowledge would cause distress (Momen, Hadfield et al. 2012). The majority of patient participants in the current study were known to palliative care services and had a general understanding that time was short. However, aside from resuscitation conversations, few had any appreciation of what was likely to happen towards the end of life. For some this was a conscious decision not to ask or explore this topic further, whereas others had not been given this opportunity. Many saw themselves as having no future and felt that discussion about the details of end of life care was too distressing or frightening.

People with ILD preferred to view themselves as 'living with ILD' rather than 'dying with ILD' and this distinction was evident from the interview transcripts. They avoided talking to healthcare professionals and family members about disease progression and dying, preferring to focus on living with ILD and taking each day as it comes. This attitude meant that many people with ILD did not discuss end of life care and relied on healthcare professionals to make the 'right decisions' for them at the end of life. Bereaved relatives also recalled how their relative had not wanted to discuss end of life plans. Conversely, some

relatives wanted the opportunity to discuss end of life care but felt unsupported and were unable to speak to anyone about their concerns. Bereaved relatives remarked that plans were not in place for end of life care in the community and that the situation changed too rapidly to allow for adequate plans to be put in place prior to death.

Low levels of advance care planning have been reported for people with ILD (Rajala, Lehto et al. 2016, Liang, Hoffman et al. 2017, Guo, Mann et al. 2020, Koyauchi, Suzuki et al. 2021). A systematic review concluded that advance care planning was surprisingly uncommon for patients with advanced respiratory disease, possibly due to the uncertain disease trajectory and 'ambivalence' of both patients and healthcare professionals (Jabbarian, Zwakman et al. 2018). Respiratory healthcare professionals in the current study all identified the importance of advance care planning but did not see this as part of their job role. They alluded to low confidence in discussing this area of patient care and time constraints as barriers preventing advance care planning conversations in respiratory outpatient appointments. Respiratory clinicians relied on specialist palliative care colleagues to explore advance care planning as part of the holistic care provided by this specialist team. In opposition to this view, survey data from respiratory healthcare professionals has suggested that incorporating a more defined role in palliative care was a desirable evolution of professional responsibility (Goodridge and Peters 2019) and that involvement of specialist palliative care was unlikely to add additional benefits to patients' management (Kim, Olive et al. 2020). The integration of specialist palliative care colleagues embedded within the North East and Cumbria ILD service may account for the expectation that all patients should be referred onto specialists for future planning conversations.

6.2.4 Accelerating symptoms at the end of life

People with ILD wanted to die peacefully, with their family present and well managed symptoms. Sadly, this was not the standard of end of life care reported by some of the bereaved relatives. Many people with ILD stated a preference to remain at home at the end of life. However, avoidance of advance care planning forced a reliance on healthcare professionals to make the right decisions at the right time. It was evident from bereaved relatives' experiences that this was frequently not possible as rapid deterioration in symptoms required an acute response, which mostly led to unplanned healthcare admission at the end of life. Healthcare professionals and bereaved relatives recognised the influence

of rapid deterioration and difficulty managing symptoms on location of death, resulting in most people with ILD dying in hospital. However, many of these deaths in healthcare environments were perceived as peaceful by relatives who accepted the challenges with providing the same level of support in the community.

It is well recognised that the majority of people with ILD die in an acute healthcare environment (Lindell, Liang et al. 2015, Rajala, Lehto et al. 2016, Higginson, Reilly et al. 2017, Zou, Nouraie et al. 2019, Cross, Ely et al. 2020). The current study identified that rapidly accelerating and overwhelming symptoms were often experienced by people with ILD towards the end of life. Existing literature confirms that the intensity of symptoms experienced by people with ILD significantly increase in the last six months of life and near to death (Rajala, Lehto et al. 2018). Rapid changes in patients' health and functional status frequently required admission to acute healthcare facilities. This occurred because plans were not in place for end of life care at home or community teams were unable to respond quickly enough to alleviate symptoms. Family members who care for older adults at the end of life at home have been described as "navigating a caregiving abyss" as they struggle to manage multiple roles and to access services and resources (Michaels, Chen et al. 2022). Previous work published by a Canadian ILD centre has advocated that early introduction of palliative care can facilitate patients with ILD dying at home (Archibald, Bakal et al. 2020). However, the palliative care bundle described in this study relied on community based ILD teams and provided a level of care that would be unachievable with the understaffed and stretched resources of the community palliative care teams in the North East and Cumbria.

Symptom control at the end of life was a challenge for even experienced healthcare professionals, who recounted their difficulty with managing symptoms of extreme breathlessness, reporting that often medications were inadequate, and patients were frightened at the end of life. These challenges related to two aspects of care: difficulty managing the physical symptoms of severe breathlessness and discomfort with the psychological manifestations of supporting an alert and orientated patient at the end of life. Healthcare professionals reported feeling more confident and experienced in managing symptoms of pain than severe breathlessness. This was in part due to the number of effective medications available for pain management, but also some underlying discomfort relating to the physical symptom of unrelenting breathlessness. The discomfort managing breathlessness was cited as a contributing factor to the difficulty engaging community teams

as well as a challenge facing experienced palliative care and respiratory clinicians when providing end of life care. Lunn et al (2019) reported that trainee respiratory clinicians felt overwhelmed by the difficulties described by patients with chronic lung disease and in some cases avoided discussing breathlessness as they felt there were no effective treatment options. Survey data from 130 Japanese respiratory physicians reported physician difficulty with providing end of life care for people with IPF compared to those with lung cancer (Akiyama, Fujisawa et al. 2020). However, responses indicated that it was prognostic uncertainty and difficulty communicating goals of care that caused the most difficulty for physicians, rather than difficulty managing patients' symptoms.

There appeared to be two types of death from ILD. One where people deteriorated quickly and unexpectedly, often resulting in changes to their preferred advance care planning decisions or resulted in relatives not being present at the end of life. The findings suggest that healthcare professionals' perceived discharge from hospital to be almost impossible at the end of life due to patients' overwhelming symptoms. These preconceptions may have limited discussions with dying patients about their preferred place of death. Alternatively, people deteriorated more slowly and dying was recognised and community or hospital teams were able to support the person and their family in their preferred place of care. The type of death was important in shaping the carers' memories, their grief and ability to move forwards with their own life. Important factors which influenced whether people with ILD died peacefully and as they wished included effective symptom control, experienced healthcare staff and the presence of family. The experience of healthcare professionals and their ability to respond rapidly to accelerating symptoms was important in reassuring relatives and adequately managing end of life symptoms.

Data from the current study suggests that location of death was not overly important to people with ILD; instead, their priority was experiencing a peaceful death. Central to the concept of a peaceful death was that their symptoms were well controlled, where both physical and psychological symptoms of a patient are considered as well as supporting family members. A peaceful and satisfactory death would avoid a traumatic experience for patients and their relatives and ensure that lasting memories held by bereaved relatives were positive. Exley et al (2007)argued that 'home' is not merely about the physical space, but about the social and emotional relationships within. The findings of the current study align with this view, that family presence and emotional relationships were more important than

the physical location of death. Existing literature refers to the concept of a 'safe death', where symptoms are well managed, and relatives do not feel overwhelmed by the dying process (Rainsford, Phillips et al. 2018). Patients reported that dying in a safe place was more important than the location and trust and familiarity based on previous experience with healthcare teams was aligned with achieving a safe death (Rainsford, Phillips et al. 2018). A study conducted in Denmark indicated that patients and carers expressed distress by admission to an unfamiliar hospital at the end of life (Egerod, Kaldan et al. 2019). In the current study, although familiarity with healthcare professionals was seen to enhance the end of life experience, it was not felt to be an essential component.

A systematic review of literature regarding the quality of end of life care in ILD, conducted as part of this research project, identified that little was known about the appropriateness or safety of dying from ILD either at home or in an acute setting (Palmer, Kavanagh et al. 2021). This raises the questions of which factors support a safe and peaceful death for people with ILD and how can healthcare professionals improve adherence to this standard of care for all patients with ILD. In the current study, factors that influenced satisfaction with end of life care included well controlled symptoms, the presence of family, support from experienced healthcare professionals and open and honest communication. Figure 6-3 demonstrates the factors identified in this research project which contributed to people with ILD having a peaceful death.

Figure 6-3 Achieving a peaceful death for people with ILD.

6.3 Summary of key findings

The synthesis of results has presented and explained the overarching categories identified from the combined research data and compared these findings with existing literature. In this section these key findings will be summarised and related to the original research questions.

The end of life experience for people with ILD was very variable and influenced by multiple factors. The most notable factors were the speed of deterioration at the end of life and whether advance care planning discussions had been conducted and acted upon. The majority of people with ILD died in an acute healthcare setting due to rapid changes in their symptom burden in the last few weeks and days of life and few had sufficient community support to avoid hospital admission. However, the experience of end of life care in hospital was variable and satisfaction with care was related to experiencing a peaceful and expected death, rather than the location of death.

There were multiple barriers identified that prevented access to palliative care services in the North East and Cumbria. The uncertain prognosis and difficulty predicting sudden

deterioration in otherwise relatively well patients presented a challenge when identifying which patients should be referred to palliative care. Patient misconceptions about the role of palliative care were a further barrier, requiring a staged discussion approach which introduced further delays in referrals. However, the main barriers to accessing palliative care services were felt to be related to issues within community palliative care teams. These issues included inadequate resources, insufficient knowledge of ILD, prioritising patients with malignant diagnoses and poor communication between community, secondary and tertiary care. There was a definite geographical disparity in access to community services and local hospice care, with those patients living in more urban areas having improved access to services. This was more evident with a palliative care team linked to the specialist clinic, as patients who lived close-by were able to have in-person support more frequently.

Many of the findings of this study are not unique to patients with ILD. The rapid deterioration in symptoms experienced towards the end of life and the potential for improvement with treatment are common in other diseases with unpredictable disease trajectory. Patients with haematological malignancies share these traits with ILD. A Dutch study identified that people with haematological malignancy were more commonly admitted to hospital towards the end of life and more frequently died in hospital compared to patients with advanced solid tumour malignancy (Verhoef, de Nijs et al. 2020). The authors advocated the need for an integrated palliative care model for patients with haematological malignancy due to the uncertain disease trajectory to allow patients and clinicians to “hope for the best and prepare for the rest”.

Integrated palliative care models have also been suggested for patients with heart failure who follow an equally uncertain disease trajectory and are at risk of sudden, unexpected death (Atkinson, Hughes et al. 2022, Lee and Hwang 2022). In Cardiff, a co-specialty community clinic was established, which allowed patients to access both cardiology and palliative care specialists together, effectively overcoming referral barriers and delivering palliative input in a value-based manner (Atkinson, Hughes et al. 2022). The obvious benefit to patients from this service model is the early access to palliative care without waiting for clinicians or patients to recognise triggers and deterioration in their health status. However, there may be some patients where earlier discussion of palliative care may not be welcomed. The need for integrated care should be balanced with the often differing information requirements of people with advanced disease, as desire for prognostic

information varies between individuals and also for an given individual over time (Russell and Ward 2011).

6.4 Study strengths and limitations

This is the first study known to use a qualitative approach to explore the end of life experience of people with ILD from the viewpoint of patients, bereaved carers and healthcare professionals. The study led to uncovering of new knowledge that can be used in clinical practice to improve the end of life care for this patient group. There was previously an absence of published research considering symptom management and what factors influenced the quality of end of life care for people with ILD.

Constructivist grounded theory methodology was the most appropriate methodological approach to address the research question and was a strength of the study. The qualitative methodology allowed for open exploration of participants' experiences and the data generated provided a rich description of living with advanced ILD, preparing for dying and care in the final days and hours of life. The breadth of experiences illustrated in interviews supports understanding of the experience of end of life care for people with ILD. The constructivist grounded theory approach enabled exploration of new meanings related to facilitating quality end of life care and achieving a safe death for people with ILD.

The sampling approach used in this study was a further strength of the study design. Purposive sampling was used initially to ensure the sample was diverse in the type of ILD, exposure to specialist palliative care and geographical location. Subsequently, theoretical sampling was used to identify patients who lived in different geographical locations (rural/urban) to explore access to palliative care services and bereaved relatives of people who died in different healthcare environments, allowing further exploration of concepts that were emerging from data analysis. Socioeconomic data for the patient and bereaved relative participants shows that the study included participants from a wide socioeconomic background (index of multiple deprivation ranging from 1-10). However, there was very little ethnic diversity within the study participants, which likely reflects the ILD population in the North East and Cumbria.

Despite the sensitive and emotive nature of the topic, recruitment of an adequate number of patients and bereaved relatives was possible, something which has historically been a

challenge in healthcare research. This study demonstrated that, with sensitive and thoughtful research processes, very unwell people with advanced disease can be given the opportunity to participate in research. Adaptations to the study design due to COVID-19 restrictions meant that all patient and bereaved relative participants were interviewed remotely (either via telephone or video-conferencing platform). This was against the conventional in-person approach used for interviewing participants about sensitive topics, however this study exhibits that remote interviewing is acceptable to participants even when talking about challenging topics.

The study involved a sample size of 30 participants. The aim of constructivist grounded theory is to continue sampling until theoretical saturation is achieved. Recruitment was ended after 30 participants in part due to time constraints and because a judgment was made that the main categories showed depth in terms of their development and no new concepts were found from incoming data that were important to the developing theory. Although the sample size was relatively small, it was appropriate to the methodological design of constructivist grounded theory and was sufficient to answer the research question.

Recruitment was challenging and there are factors which meant that, despite using a purposive recruitment strategy, the people approached for participation in the study may not fully represent the experiences of people with ILD towards the end of their lives. One notable problem encountered was the difficulty identifying carers for many deceased people with ILD. This meant that most of the bereaved carer participants had lived with their relative (mostly spouse or partner) as this was the only address available to send recruitment information about the study. This led to exclusion of relatives who did not live with the deceased and more importantly little is known about the end of life care for those people with ILD who lived alone. Two of the people with ILD who were interviewed lived alone, both were proactive in discussing end of life care and preparing for the future but were worried about how they would manage towards the end of life.

Recruitment of bereaved relatives was particularly difficult with 58 participant information packs sent out to recruit the nine participants. The ‘opt in’ approach to recruitment was appropriate given the sensitive nature of the study topic, but this is likely to have contributed to the low recruitment rate from the bereaved carers who were approached. As bereavement and end of life care is an emotive topic, it may be that participants were more

likely to respond to the invitation letter if they had a particularly good or bad experience and the 'opt in' approach may have amplified this participant self-selection. The interviews undertaken with bereaved carers described diverse experiences of end of life care and many relatives who responded had a particular issue with their relatives' care which they wanted to highlight.

The focus of this research study was to understand the end of life experience of people with ILD, and the expectation was that discussing dying would be the main component of the patient and bereaved relative interviews. However, patients primarily talked about the challenges of living with ILD and the burden of symptoms and actually very little of their interview data related to dying. Despite consenting to be involved in a research study about end of life care, many people with ILD had not considered their end of life wishes or were reluctant to talk about this aspect of their illness journey. People with ILD considered themselves as 'living with progressive ILD' rather than 'dying with progressive ILD'. This subtle difference in the emphasis of patient interviews led to the development of the first two main categories, 'hidden illness' and 'recognising deterioration', which concentrate on living with ILD and are constructed from the data derived from these interviews. The main category 'dying: the important bit' was largely constructed from data from bereaved relative interviews, as patients were unable to discuss dying in detail as this was not their lived experience. This reliance on others to 'tell the story' of the person dying is a limitation seen in all end of life research and acceptance of bereaved relatives' accounts of dying has been widely adopted as a satisfactory approach.

This study focused on the specific clinical model used within the North East and Cumbria and it could be argued that as a result the findings may not be generalisable to patients within other care models. However, the study included participants who accessed healthcare in a variety of different environments (e.g., accident and emergency, tertiary ILD centre, district general hospital, hospice, community palliative care services) suggesting that the issues identified are generalisable to patients within different healthcare settings. One aspect that is missing from the study is the voice of primary and community services who deliver the majority of palliative care in the community setting. The study did not set out to recruit healthcare professionals from primary and community teams, but their experiences would have enriched the data generated.

My role as a respiratory doctor meant that I had in-depth clinical knowledge of ILD; having worked within both the regional ILD service and in a local hospice with specialist palliative care colleagues. Consideration was taken to avoid recruitment of people with ILD or bereaved relatives of people with whom I had significant clinical involvement through these job roles. However, I had a close working relationship with some of the healthcare professionals interviewed for the study. My professional background could have resulted in bias in relation to the conduction of the interviews and the data analysis. However, this was monitored by members of the supervision team who read through the interview transcripts and triangulated the data coding process. I aimed to prevent bias influencing the research process by engaging in reflexivity (3.6.10), a process that required critical self-reflection of the research experience. The study was undertaken to contribute to clinical knowledge and improve practice; therefore, it could be argued that my expertise in the condition and area of practice permitted a deeper understanding of a complex phenomenon and comprehension of its clinical relevance. Participants may have opened up to me in different ways due to my knowledge of ILD and the sense of shared understanding of the condition. My expertise and background in clinical medicine should be regarded as both a strength and limitation in relation to this study.

6.5 Reflections

I began my research journey by exploring my own ontological and epistemological beliefs as this area of philosophy is not something I had previously considered. I found this deviation into research philosophy enlightening as my training in clinical medicine firmly pushes my beliefs towards realist ontology, a stance which I have previously accepted without appreciating other perspectives. I examined different types of qualitative research methodology and identified constructivist grounded theory as an appropriate methodology for this study. The exploratory and inductive style of constructivist grounded theory is appropriate when little is known about the research topic with the aim to generate new knowledge in the form of theory (Mills 2015). I studied the different grounded theory approaches and decided to use constructivist grounded theory (Charmaz 2014) as the methodological approach for this study, as this aligned with my ontological/epistemological perspective of critical realism.

From the start of this research project, I was acutely aware of how my role as a respiratory doctor working within both the tertiary ILD service and local hospice would influence data generation. The questions I asked in interviews will have been influenced by my previous experiences of end of life care in ILD and caring for patients who were dying of this condition in hospital and hospice environment. I now recognise that my interpretation of the results was also influenced by my background in clinical respiratory medicine, which will have influenced the language used to describe codes and categories and the formulation of final overarching categories. The research findings presented here are my interpretation of the data generated from interviewing key stakeholders and should not be viewed as representative of the complete end of life experience for people with ILD.

I found the interpretive nature of constructivist grounded theory intimidating to begin with as I am more confident working in a world where there is a 'correct way' of approaching things and a single correct interpretation of results. I felt I needed someone to verify that my codes were 'correct' and that I was approaching the methodology in the correct way as my understanding that robust coding would aid formation of core categories and identification of overarching categories. I struggled with the non-linear direction of the research analysis, expecting it to progress through stages of initial coding, focussed coding, category formation and finally allowing identification of overarching categories. Instead, I was faced with moving back-and-forth between coding and constructing categories and at times wondered if I would be able to cohesively write-up the findings. However, the process of writing my results chapter aided my analysis and even at this stage I found that categories increased and diminished in importance as I worked on generating a unified understanding of the phenomenon.

To avoid confusion about my dual roles as a clinician and researcher, patients or bereaved relatives who I had significant involvement with were ineligible for recruitment into the study. However, despite this precaution, there were patients who identified me as being part of the regional ILD service and as such may have been less willing to discuss dissatisfaction with their care from this service. There were times when I had difficulty maintaining the researcher-participant relationship and not allowing the interview to change focus into a therapeutic doctor-patient encounter. To try to avoid this situation I informed participants prior to the interview that I would only ask questions related to the research interview as I was interested in their story, but that I would be able to answer any further

questions they might have following the interview. I found this approach was helpful and meant that I was able to balance the feeling of wanting to help as a clinician with the impartial researcher. For example, there were people for whom I chased up palliative care referral or suggested bereavement support services in this discussion after the interview.

One aspect of the research process which I found particularly challenging was conducting remote interviews about such a sensitive subject. When planning the concept of the research project I had intended to conduct all interview in-person, but this approach had to be adapted in view of the COVID-19 pandemic. Instead, I found myself grappling with videoconferencing technology and recording interviews over the phone which felt much less personal. All patient and bereaved relative interviews were conducted remotely; for some participants this was preferrable as they felt they would have been more upset speaking to me in person. However, for most there were times when I would have preferred to be present in the room, especially when someone was upset, to offer comfort and interpret their body language about whether to continue the interview. Alongside my research project, I was adapting to holding outpatient clinic conversations over the phone, which I also found difficult, and shared the same concerns and discomfort as the healthcare professional participants about assessing patients and talking about palliative care over the phone.

Some healthcare professional interviews were held in-person as they were with colleagues who I worked with on a weekly basis. As a researcher I found these interviews much more comfortable, but this could also have been because these were people who I knew well, and we were discussing their jobs rather than deeply personal experiences. McConnell-Henry et al (2010) highlighted that when a researcher and participant have a pre-existing relationship that the stages of rapport building are rapidly accelerated leading to the generation of data that is rich in breadth and depth. I agree with this statement as I found interviews with colleagues yielded rich data and that this was enhanced by my prior relationship with these participants. When interviewing my colleagues, I was aware of the risk of “conceptual blindness” where my own thoughts and feelings about the field might influence the tone or direction of the interview (Coar and Sim 2006). I sought to address this issue by openly acknowledging my presuppositions in the interview and encouraging participants to explain their own experiences comprehensively.

Finally, this seems an appropriate place to reflect on the impact of the COVID 19 pandemic on both the undertaking of this research project and the results. People with ILD were classed as 'clinically extremely vulnerable' and as such have spent long periods of time isolating at home during the two-year span of this research project. This obviously increased feelings of loss and social isolation which were evident from the patient and bereaved carer interviews. The impact of the pandemic on services meant that many people with ILD felt abandoned by their clinical teams, waiting long periods of time for follow-up appointments that were often replaced by remote consultations. Palliative care services, such as day hospices, closed during the national lockdown and there was reduced availability of community teams such as Macmillan and district nursing teams. The inequality between people with ILD and lung cancer was highlighted by the re-deployment of specialist respiratory nurses to look after COVID patients in hospital, whereas lung cancer specialist nurses continued with their usual roles as there were two-week rule targets to observe. It could be argued that the results of this study are less relevant to the usual experience of people with ILD as the COVID 19 pandemic has affected their care in such diverse ways. However, I would assert that the pandemic has exacerbated and highlighted the already existing healthcare inequalities affecting people with ILD. As we remain a long way from returning to normal healthcare provision following the pandemic, the results of this study remain relevant for future service planning and ensuring provision of quality end of life care for people with ILD.

6.6 Clinical implications and recommendations

This section examines the contribution of the research to the field of end of life care in ILD and how it may influence clinical practice. This study has enhanced understanding of the end of life priorities of people with ILD and their families. The recommendations are presented in three sections: i) organisation of specialist palliative care services for people with ILD, ii) supporting advance care planning discussions, iii) understanding markers of quality in end of life care.

6.6.1 Organisation of specialist palliative care services for people with ILD

Recommendations from this study focus on how to best deliver good local palliative care within the tertiary care models frequently used for organisation of ILD services. This hinges on good communication between tertiary ILD teams and local palliative care services. Healthcare professionals should review existing services with the aim to facilitate improved referral and communication with local and community palliative care services. Recognition that patients are deteriorating is paramount, unfortunately the unpredictable disease trajectory observed in ILD is a major challenge to timely involvement of local palliative care services. Decision aid tools can be effective to help clinicians identify patients with palliative care needs and prompt onward referral to specialist palliative care (Sharp, Lamb et al. 2018). A collaborative MDT meeting focused on identifying patients' palliative care needs can enhance communication with community teams and facilitate appropriate referrals (Barratt, Morales et al. 2018).

The findings of this study support the integration of specialist palliative care and ILD services to overcome referral barriers and improve communication with local and community palliative care services. There are numerous studies that describe the benefits of integration of specialist palliative care within oncology services, and there are several different models used to approach this integration (Mathews, Hannon et al. 2021). Evidence supporting early integrated palliative care mainly comes from randomised controlled trials conducted in stand-alone clinics (Zimmermann, Swami et al. 2014, Maltoni, Scarpi et al. 2016, Temel, Greer et al. 2017). Although this is suggested as the optimal model for early integration of palliative care, stand-alone clinics tend to be orientated around tertiary services and therefore this approach does not address the geographical disparities identified in this study. Palliative care embedded within specialist outpatient services has been known to reduce referral barriers due to the geographical proximity meaning easier access to palliative care advice and reducing stigma attached to this referral (Hui, Hannon et al. 2018). Alternatively, early integration of home-based palliative care (i.e., GP/MacMillan nursing teams in the UK) could be arranged for patients. However, this also has drawbacks the main being discontinuity of care as a result of lack of communication structure and central coordination (Dhollander, De Vleminck et al. 2019). In ILD, several papers have discussed the benefits of integrated care in increasing advance care planning conversations, reducing healthcare utilisation at the end of life and supporting patients to die in their preferred location

(Bajwah, Ross et al. 2015, Barratt, Morales et al. 2018, Kalluri, Claveria et al. 2018, Archibald, Bakal et al. 2020). However, there is no prescriptive way to approach integration of services and therefore it is recommended that ILD teams work with existing palliative care services to agree a model that fits within and enhances existing service structures.

6.6.2 Supporting advance care planning discussions

This study identified that people with ILD are often reluctant to discuss advance care planning, thus creating a further obstacle to facilitating local palliative care for patients. This thesis has described the factors that interplay and influence people with ILD and their families' satisfaction with end of life care. These factors will vary for every patient, and may change over time for a given individual, meaning that advance care planning discussions remain important to establish priorities for each patient and their family. However, as the findings of this study denote, advance care planning discussions should be led by the patients' request for information and may be quite limited for people with ILD as many do not want to talk about the future in detail. This ambivalence towards proactive discussion of the future has been highlighted in people with chronic lung disease and is thought to be related to unpredictable disease trajectory and insufficient knowledge of the severity of disease (Jabbarian, Zwakman et al. 2018). Patients in the current study frequently preferred to trust that clinicians will make the right decisions for them at the right time based on their prior discussions and stated preferences, rather than discussing the details of end of life care. On this basis, the findings of this study support initial advance care planning discussion as part of an integrated clinic and should be undertaken by any member of the team. Many patients may not want to discuss advance care planning, but should be aware of DNAR decisions etc, and for those who want to have more lengthy discussions a referral to a specialist palliative care team integrated within the service is appropriate.

6.6.3 Understanding markers of quality in end of life care

This study identified that location of death was not as important to people with ILD as achieving a 'peaceful death'. The assumption that dying at home is automatically superior to dying in a hospital is outdated (Hoare, Antunes et al. 2022). This study has identified that home may not be the preferred or most appropriate place of care in which to manage people's physical and psychological symptoms at the end of life. These findings encourage a

change in the view of using location of death as a surrogate marker of good end of life care and a move towards measuring quality based on achievement of good symptom control. This is a superior marker of the quality of end of life care but is more difficult to measure especially in population based and retrospective studies. International policy-makers and researchers have recognised the need for robust measures of the quality of end of life care (Lind, Adolfsson et al. 2015, House of Commons Health Committee 2016, National End Of Life Care Intelligence Network 2016), but there is no consensus about end of life care indicators relevant to all patients in all healthcare settings (Virdun, Luckett et al. 2018). Finding the best ways to review the quality of end of life care is important to be able to benchmark services and ensure that quality is achieved throughout large geographical areas rather than for small pockets within services.

6.6.4 Future research opportunities

The study has highlighted several areas where current evidence is limited, providing a number of opportunities for further research. Based on the study recommendations above, a follow-on project could explore patient satisfaction and acceptance of the integration of ILD and specialist palliative care services within the tertiary centre or develop, validate and test an intervention to support advance care planning conversations within ILD consultations. Further research examining the markers of quality in end of life care in ILD, particularly symptom control at the end of life, in relation to place of death would also help to answer questions raised in this thesis.

Determining the experience of community teams looking after patients with ILD at the end of life would be an informative and valuable topic for future research. This current study identified barriers to accessing community palliative care resources, however, the findings were based on the perspective of healthcare professionals working in tertiary and secondary care. Community teams may identify different barriers that prevent access to palliative care resources and this additional information would build a more complete picture of the issues surrounding providing quality end of life care for people with ILD. It should be noted that community teams will only see a small number of patients with ILD, consequently, to make this research viable, inclusion of other non-malignant respiratory diseases may be necessary.

6.7 Conclusion

This study is the first to critically examine the end of life care of people with ILD from the perspective of patients, bereaved relatives and healthcare professionals. It addresses a significant gap in the existing research knowledge; specifically challenges surrounding symptom control at the end of life and whether dying in an acute setting is considered acceptable by people with ILD and their families. These findings constitute important new knowledge to inform clinical practice in relation to end of life care in ILD. They help make sense of the experience of dying from ILD and factors that influence quality and safety of end of life care. The findings bring a new appreciation of the illness journey experienced by people with ILD and healthcare professionals' roles at distinct points throughout this illness journey. This knowledge creates an opportunity for healthcare professionals to offer appropriate and timely interventions to support people with ILD in navigating their illness journey.

The core categories identified from the study findings were 'navigating the uncertain illness journey with ILD' (from patients' and relatives' perspectives) and the 'roles of healthcare professionals' (from healthcare professionals' perspective). When data were combined four further overarching categories were identified which answered the research questions from the viewpoint of all the key stakeholders. These overarching categories were: i) acknowledging uncertainty, ii) accessing and organising support, iii) avoiding discussion about an uncertain future, iv) accelerating symptoms at the end of life.

The primary research objective was to increase understanding of the end of life care experience of people with ILD. The findings of this study indicate that this experience was variable and dependent on numerous factors. The overarching category 'accelerating symptoms towards the end of life' examined the challenges faced by people with ILD in the final few days and hours of life. It was established that the majority of people with ILD suffered rapidly deteriorating symptoms towards the end of life, which influenced the burden of symptoms suffered, the presence of family at the end of life and location of death. A significant finding were the challenges reported by healthcare professionals in managing severe breathlessness at the end of life. This was in part due to the perceived limitation of available medications to adequately manage the physical symptoms, but also due to

discomfort with supporting the psychological aspects of fear and anxiety that were associated with patients' awareness of dying.

An additional aspect of the main research objective was evaluating what factors influenced patients' and relatives' satisfaction with end of life care. The study identified that people with ILD wanted a 'peaceful death'. The idea of a peaceful death was explored and elements that contributed to this concept were outlined in Figure 6-3. Central to this concept were that symptoms were well controlled, and that death was anticipated. As discussed above, end of life care was variable; there were examples of patients who achieved a peaceful death and also those where end of life care was unsatisfactory. There were some circumstances where symptom management at the end of life was inadequate, due to inexperienced healthcare professionals, poor advance care planning or unexpected rapid deterioration. Dying was frequently not recognised by healthcare professionals and therefore death was unanticipated by patients and relatives. However, for those patients who were recognised as dying, the majority had a peaceful death, with well controlled symptoms and family present at the end of life.

A secondary research objective considered perceived barriers preventing access to specialist palliative care services. This study identified several apparent barriers that precluded timely referral of patients to specialist palliative care services, these are summarised in Table 11.

Patient related barriers	Healthcare professional and healthcare system related barriers
➢ Misconceptions about palliative care ➢ Avoiding advance care planning discussions ➢ Uncertain prognosis - expecting more time	➢ Prognostic uncertainty – unexpected deterioration ➢ Limited time to address palliative care in respiratory clinics ➢ Remote consultations – missing 'triggers' for referral ➢ Lack of community palliative care resources ➢ Priority given to patients with malignant diagnoses ➢ Geographical disparity in access to services

Table 11 Perceived barriers preventing patient involvement with specialist palliative care services.

The overarching category 'avoiding discussion about an uncertain future' highlighted that both patients and healthcare professionals avoided discussion about end of life care which

limited opportunities for engagement with advance care planning conversations. From a patient perspective, misconceptions about palliative care, uncertainty about prognosis and expecting more time, contributed to avoiding discussion about the future and a reluctance to accept palliative care referral. The overarching category 'acknowledging uncertainty' demonstrated the requirement for open and honest conversations between patients and healthcare professionals, recognising that the illness journey with ILD is unpredictable. This direct approach would potentially overcome some of the patient related barriers preventing engagement with palliative care services.

The overarching category 'accessing and organising support' identified crucial healthcare system related barriers influencing patients access to palliative care services including limited community palliative care resources and geographical disparity. One of the most concerning aspects was the widespread impression that community teams prioritised patients with malignant diagnoses and that patients with ILD were seen as a lower priority despite their significant symptom burden and very poor prognosis. It was suggested that this was due to limited knowledge of ILD in the community and reduced confidence in the management of symptoms of breathlessness.

This study contributed to the current literature considering end of life care in ILD, which largely focused on establishing palliative care needs and reviewing location of death. The findings are important because they address a gap in the existing literature surrounding the quality of end of life care for people with ILD. The study identified that people with ILD prioritise a peaceful death over the location of death and established factors that contributed to achieving a peaceful death. This information should be used to transform healthcare policy, by developing regional guidelines and integrating existing networks, to ensure that all patients with ILD have access to the same quality of care to allow a peaceful death.

A significant finding from this research study was the rapid and frequently unexpected deterioration in patients' symptoms at the end of life. This was the biggest challenge to providing good end of life care, as often patients' symptoms deteriorated too quickly for community support to be arranged, leading to acute hospital admission at the end of life. As discussed previously, the location of death was less important to people with ILD than rapid response to symptom management. Advance care planning conversations need to

acknowledge the uncertainty about disease trajectory and potential for rapidly escalating symptoms at the end of life. This would allow for people with ILD and relatives to be realistic about end of life care and shift the focus of future care planning discussions towards achieving a peaceful death rather than an emphasis on location of death. Assessing the quality of end of life care should move away from the location of death and focus on patients perceived safety and satisfaction with end of life care.

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Appendix A: HRA approval letter

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Dr Evelyn Palmer
Clinical research fellow in respiratory medicine
Newcastle Hospitals NHS Trust
Population health sciences institute
Newcastle University
Newcastle upon Tyne
NE1 7RU

Email: approvals@hra.nhs.uk
HCRW.approvals@wales.nhs.uk

16 November 2020

Dear Dr Palmer

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**Critically examining the experience of end of life care for people with interstitial lung disease: views of patients, families and healthcare professionals.**
IRAS project ID:	**276064**
Protocol number:	**09527**
REC reference:	**20/WM/0267**
Sponsor	**The Newcastle upon Tyne NHS Foundation Trust**

I am pleased to confirm that **HRA and Health and Care Research Wales (HCRW) Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report

(including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **276064**. Please quote this on all correspondence.

Yours sincerely,
Rekha Keshvara

Approvals Manager

Email: approvals@hra.nhs.uk

Copy to: *Ms Elaine Chapman, The Newcastle upon Tyne Hospitals NHS Foundation Trust*

Appendix B: Participant facing documents

i. Invitation letter

Newcastle Interstitial Lung Disease Service
Respiratory Department
Royal Victoria Infirmary
Newcastle upon Tyne, NE1 4LP
Date

Recipient name
Street address,
City, Postcode.

Dear Recipient name,

Critically examining the experience of end of life care for people with interstitial lung disease: views of patients, families and healthcare professionals

We are writing to invite you to participate in the above research study which is being undertaken by Newcastle Interstitial Lung Disease service in collaboration with Marie Curie. The study aims to develop a better understanding of the experiences of care towards the end of life for patients with interstitial lung disease, including idiopathic pulmonary fibrosis (IPF), within the North East and Cumbria. We hope to use the information from this project to improve care for patients with these lung conditions.

The project involves interviews with patients who have interstitial lung disease, relatives of people who have died from the disease and also members of the Respiratory teams throughout the region. We will be interviewing people who live in different areas of the North East and Cumbria who have a wide range of experiences.

There is an information leaflet about the study included with this letter. Please read this information carefully before deciding whether you would like to take part and do not hesitate to get in touch if you have any questions. If you would like to participate in this study, please send your reply either via email (e.l.palmer2@newcastle.ac.uk) or post (in the prepaid envelope enclosed) and the lead researcher will contact you to arrange a convenient time and place for an interview.

Thank you in advance for your help.

Yours sincerely,

The Newcastle Hospitals Interstitial Lung Disease Service

ii. Participant information leaflet (patients)

Critically examining the experience of end of life care for people with interstitial lung disease: views of patients, families and healthcare professionals

INFORMATION SHEET FOR PATIENTS

First of all, let us express our regrets that you have become so unwell with your lung condition. We would like to invite you to take part in a research study which aims to improve the care for patients with interstitial lung disease. Before you decide whether you would like to be involved, it is important that you understand why the research is being done and what it will involve. Please take your time to read the following information carefully and discuss it with friends, relatives and staff at the Royal Victoria Infirmary clinic if you wish. Ask us anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part. Thank you for reading this information leaflet.

What is the purpose of the study?

The study is looking at the experiences of end of life care for people with interstitial lung disease (ILD). We know that people with interstitial lung disease can have a large number of symptoms, particularly breathlessness, which are often difficult to manage. Previous research has shown that people with ILD often do not have access to the same level of services as people with other significant conditions such as lung cancer. The study seeks to increase our knowledge of end of life care for people with ILD in the North East and Cumbria and identify things that we are doing well and areas for improvement.

Why have I been chosen?

You have been invited to participate in this research project because unfortunately your interstitial lung disease has progressed to a relatively advanced stage. We would like to have the opportunity to talk to you in confidence about the care and support services that you have experienced in the management of the disease so far, and to understand your thoughts and expectations for the future.

The principal objective of this project is to understand the level of services and care currently provided over in the last year of life for patients with ILD across the region, to share examples of good practice and suggest improvements where necessary.

Do I have to take part?

We understand that it may be difficult for you to talk about these topics and it is up to you to decide whether or not you would like to take part in this study. If you do decide to take part you will be free to withdraw at any time and without giving a reason. Your decision about whether to be involved in the study will not have any effect on your ongoing health care or treatment from the ILD service.

What will happen if I take part?

The lead researcher, Dr Eve Palmer, will contact you to arrange a convenient time and method to speak to you. You will be sent a consent form which will need to be returned prior to the interview either by post (prepaid envelope) or email. Due to coronavirus restrictions the interview will be conducted either by phone or videoconferencing. Zoom is the method of videoconferencing used in this study and is secure and easy to use. If you prefer to have the interview through Zoom, you will be sent instructions and a link via email which will allow you to connect easily to this software. Usually this interview would take one hour and be recorded using a digital recording device. There are no set questions, but there are some topic areas that we would like to ask you about. You will be given information about the topics that will be covered during the interview prior to taking part so that you can decide if these are topics that you are happy to talk about. We would like you to explain your experience in your own words and therefore we will talk about what you think is relevant and important to you.

We understand that this is likely to be a difficult topic for you to discuss, if you become upset during the interview and no longer wish to continue, you can stop the interview and recording at any time. If you wish you can have a relative or carer present, they will also need to complete a consent form.

What are the possible benefits of taking part?

The study aims to increase the knowledge concerning end of life care for people with interstitial lung disease throughout the North East and Cumbria. By taking part you will be helping us to improve this care for others in the future. You may also personally find it helpful to talk to someone about your experience of living with interstitial lung disease.

What are the possible disadvantages of taking part?

You may find it difficult or upsetting to talk about topics to do with end of life care. If you find that the interview causes you distress the recording can be stopped at any time. If you are distressed following the interview or wish to talk about any of the topics in further detail, please contact the ILD specialist nurses (Clare Donaldson and Lyndsay Langlands 0191 282 6803) who will arrange support through the ILD clinic or your own GP.

Will my participation in the study be kept confidential?

All of the recorded information from the interview will be typed into an anonymised document and the original recordings destroyed. This information will then be only identified using a unique code and all of your personal information (your name and address etc) will be removed from the written document so that you cannot be recognised from it. You will not be identified in any report or publication.

All of the information shared during the interview will be kept confidential, except in the event of a significant disclosure which poses a risk to you or other people. We would usually inform your GP of your participation in the study in case you require any further support from them following the interview. We will only inform your GP with your consent.

What will happen if I decide to stop being in the study at any point?

You are able to withdraw from the study at any time and without needing to give a reason. If this is before your interview, then no information about you will be entered into the study. You will be able to withdraw from the study after your conversation with the researcher. However, it will only be possible to remove your data from the study for 14 days after your interview as once the recording has been typed and anonymised, it may not be possible to identify your data to remove it from the study results.

What will happen to the information collected in the study?

Information collected for this study will be stored on password protected files at Newcastle University. This means that Newcastle University is responsible for looking after your information and using it properly. We will use your name, telephone number and address to contact you about the research study. Individuals at Newcastle University may look at your interview data to check the accuracy of the research study. The only people at Newcastle University who will have access to information that identifies you will be people who need to contact you to arrange the interview or audit the data collection process.

You can find out more about how Newcastle University uses your information at https://www.ncl.ac.uk/data.protection/dataprotectionpolicy/privacynotice/ and by contacting Newcastle University's Data Protection Officer (Maureen Wilkinson, rec-man@ncl.ac.uk).

More information about how the NHS manages research participant information can be found on the following website: https://www.hra.nhs.uk/information-about-patients/

What will happen to the results of the research study?

The results of the study will be written up by Dr Eve Palmer as part of her MD thesis which she is undertaking through Newcastle University. The information from this study will directly influence the future care of patients with interstitial lung disease in the North East and Cumbria. We aim to publish the findings so that other organisations in the UK can also benefit from this information.

Who is organising and funding the study?

The research is funded by Marie Curie and charitable funds from the Royal Victoria Infirmary ILD department.

Have patients and the public been involved in the study design?

The study is supported by patient and carer support groups in both the North East and Teesside who have identified improving end of life care as being a priority for patients. A patient and public representative panel has been formed which includes people with interstitial lung disease, relatives and bereaved relatives. The panel members have advised on all the information that is given to participants. The questions that are asked during the research conversation have been reviewed by the panel members to make sure that they are relevant and will not cause undue distress.

Who should I contact if I have a question or complaint?

The lead researcher who will be conducting the interviews is Dr Eve Palmer. This project is supervised by a team from Newcastle University. If you have any questions about the project please contact Dr Palmer directly. If you have any problems or complaints about the study please direct these to Dr Shelina Visram who is a member of the supervisory team.

Researcher: Dr Eve Palmer e.l.palmer2@newcastle.ac.uk Telephone: 07835409082

Project supervisor: Dr Shelina Visram shelina.visram@newcastle.ac.uk

If you prefer to raise your concerns with someone not involved in your care you can contact the Patient Advise and Liason Service (PALS). This service is confidential and can be contacted on the Freephone: 0800 032 0202.

Alternatively, if you wish to make a formal complaint then you can contact the Patient Relations Department through any of the details below:

Telephone: 0191 223 1382 or 0191 223 1454 Email: patient.relations@nuth.nhs.uk

Address: Patient Relations Department, The Newcastle upon Tyne NHS Foundation Trust, The Freeman Hospital, Newcastle upon Tyne, NE7 7DN

iii. Participant information leaflet (relatives)

Critically examining the experience of end of life care for people with interstitial lung disease: views of patients, families and healthcare professionals

INFORMATION SHEET FOR RELATIVES

First of all, please let us express our sincere condolences for your recent loss. We would like to invite you to take part in a research study which aims to improve the end of life care for people with interstitial lung disease. Before you decide whether you would like to be involved, it is important that you understand why the research is being done and what it will involve. Please take your time to read the following information carefully and discuss it with friends, relatives and staff at the Royal Victoria Infirmary clinic if you wish. Ask us anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part. Thank you for reading this information leaflet.

What is the purpose of the study?

The study is looking at the experiences of end of life care for people with interstitial lung disease (ILD). We know that people with ILD can have a large number of symptoms, particularly breathlessness, which are often difficult to manage. Previous research has shown that people with ILD often do not have access to the same level of services as people with other significant conditions such as lung cancer.

The study seeks to increase our knowledge of end of life care for people with ILD in the North East and Cumbria and identify things that we are doing well and areas for improvement.

Why have I been chosen?

You have been invited to participate in this study as you are a close relative or carer of a person who has sadly died from ILD. We would like the opportunity to talk to you in confidence about the final days and weeks of their illness and their experience of dying from this illness.

The principal objective of this project is to understand the level of services and care currently provided over in the last year of life for people with ILD across the region, to share examples of good practice and suggest improvements where necessary.

Do I have to take part?

We know that it may be too upsetting for you to talk about these topics. We understand this may be very difficult for you to talk about and it is up to you to decide whether or not you would like to take part in this study. If you feel unable to participate now, but that you would like to be involved in the future then please contact us when you feel it is a more appropriate time for you. If you do decide to participate, you will be free to withdraw at any time and without reason.

What will happen if I take part?

The lead researcher, Dr Eve Palmer, will contact you to arrange a convenient time and method to speak to you. You will be sent a consent form which will need to be returned prior to the interview either by post (prepaid envelope) or email. Due to coronavirus restrictions the interview will be conducted either by phone or videoconferencing. Zoom is the method of videoconferencing used in this study and is secure and easy to use. If you prefer to have the interview through Zoom, you will be sent instructions and a link via email which will allow you to connect easily to this software. Usually, this interview would take one hour and be recorded using a digital recording device. There are no set questions, but there are some topic areas that we would like to ask you about. You will be given information about the topics that will be covered during the interview prior to taking part so that you can decide if these are topics that you are happy to talk about. We would like you to explain your relative's experience in your own words and therefore we will talk about what you think is relevant and important. We understand that this is likely to be a difficult topic to discuss, and if you become upset and no longer wish to continue, you can stop the interview and recording at any time.

What are the possible benefits of taking part?

The study aims to increase the knowledge about end of life care for people with interstitial lung disease throughout the North East and Cumbria. By taking part you will be helping us to improve this care for others in the future. You may also personally find it helpful to talk to someone about the death of your family member or friend.

What are the possible disadvantages of taking part?

You may find it difficult or upsetting to talk about the final days and weeks of your relative's or close friend's life. Talking about this subject may also leave you with new feelings or questions about their death. If you would like further support following the interview then please contact Jacquie Leaman, who is a member of the bereavement team at Marie Curie hospice on 0191 219 1035 or jacquie.leaman@mariecurie.org.uk.

Will my participation in the study be kept confidential?

All of the recorded information from the interview will be typed into an anonymised document and the original recordings destroyed. This information will then be only identified using a unique code and all of your personal information (your name and address etc) will be removed from the written document so that you cannot be recognised from it. You will not be identified in any report or publication.

All of the information shared during the interview will be kept confidential, except in the event of a significant disclosure which poses a risk to you or other people. We would usually inform your GP of your participation in the study in case you require any further support from them following the interview. We will only inform your GP with your consent.

What will happen if I decide to stop being in the study at any point?

You are able to withdraw from the study at any time and without needing to give a reason. If this is before your interview, then no information about you will be entered into the study. You will be able to withdraw from the study after your interview. However, it will only be possible to remove your data from the study for 14 days after your interview as once the recording has been typed up and anonymised, it may not be possible to identify your data to remove it from the study results.

What will happen to the information collected in the study?

Information collected for this study will be stored on password protected files at Newcastle University. This means that Newcastle University is responsible for looking after your information and using it properly. We will use your name, telephone number and address to contact you about the research study. The only people at Newcastle University who will have access to information that identifies you will be people who need to contact you to arrange the interview or audit the data collection process.

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More information about how the NHS manages research participant information can be found on the following website: https://www.hra.nhs.uk/information-about-patients/

What will happen to the results of the research study?

The results of the study will be written up by Dr Eve Palmer as part of her doctoral thesis which she is undertaking at Newcastle University. The information will directly influence the future care of patients with ILD in the North East and Cumbria. We aim to publish the research so that organisations in other areas of the UK can also benefit from the findings.

Who is organising and funding the study?

The research is funded by Marie Curie and charitable funds from the Royal Victoria Infirmary ILD department.

Have patients and the public been involved in the study design?

The study is supported by patient and carer support groups in the North East who have identified improving end of life care as a priority for patients. A patient and public representative panel has been formed which includes people with ILD, relatives and bereaved relatives. The panel members have advised on all participant information. The questions that are asked during the research conversation have also been reviewed to make sure that they are relevant and will not cause undue distress.

Who should I contact if I have a question or complaint?

The lead researcher who will be conducting the interviews is Dr Eve Palmer. This project is supervised by a team from Newcastle University. If you have any questions about the project please contact Dr Palmer directly. If you have any problems or complaints about the study please direct these to Dr Shelina Visram who is a member of the supervisory team.

Researcher: Dr Eve Palmer e.l.palmer2@newcastle.ac.uk Telephone: 07835409082
Project supervisor: Dr Shelina Visram shelina.visram@newcastle.ac.uk

If you prefer to raise your concerns with someone not involved in your care you can contact the Patient Advise and Liaison Service (PALS). This service is confidential and can be contacted on the Freephone: 0800 032 0202.

Alternatively, if you wish to make a formal complaint then you can contact the Patient Relations Department through any of the details below:

Telephone: 0191 223 1382 or 0191 223 1454 Email: patient.relations@nuth.nhs.uk

Address: Patient Relations Department, The Newcastle upon Tyne NHS Foundation Trust, The Freeman Hospital, Newcastle upon Tyne, NE7 7DN

iv. Participant information leaflet (healthcare professionals)

Critically examining the experience of end of life care for people with interstitial lung disease: views of patients, families and healthcare professionals

INFORMATION SHEET FOR HEALTHCARE PROFESSIONALS

You are being invited to take part in a research study. Before you decide whether you would like to be involved, please read the following information so that you understand why the research is being done and what it will involve. If you have any questions about the study, contact details for the research team are at the bottom of this information sheet. Thank you for reading this information leaflet.

What is the purpose of the study?

The study is looking at the experiences of end of life care for people with interstitial lung disease (ILD). Previous research in this area has identified that people with ILD can have significant symptoms, particularly breathlessness, and that they often do not have access to the same level of services as people with other significant conditions such as lung cancer. The study seeks to increase our knowledge of the patient experience of end of life care in the North East and Cumbria and identify things that we are doing well and areas for improvement.

Why have I been chosen?

You have been invited to participate in this study because you work with patients who have ILD. We would like the opportunity to talk to you in confidence about how you feel about palliative care and the services available to patients with ILD in your area and any problems you have experienced with accessing services. We would also like to understand local referral pathways to palliative care services.

The principal objective of this project is to understand the level of services and care currently provided over in the last year of life to this patient group across the region, to share examples of good practice and suggest improvements where necessary.

Do I have to take part?

It is up to you to decide whether you would like to take part in this study. If you do decide to participate, you will be free to withdraw at any time and without reason. Declining to participate will not affect your employment or career progression in any way.

What will happen if I take part?

The lead researcher, Dr Eve Palmer, will contact you to arrange a convenient time and method to speak to you. You will be sent a consent form which will need to be returned prior to the interview either by post or email. Due to coronavirus restrictions, the interviews will be conducted either over telephone or videoconferencing (Zoom). Usually this interview would take up to an hour and be recorded using a digital recording device. There are no set questions, but there are some topic areas that we would like to discuss. You will be given information about the topics that will be covered during the interview prior to taking part so that you can decide if these are topics that you are happy to talk about. We would like you to explain your experience in your own words and therefore we will talk about what you think is relevant and important to your patients.

Will the content of the interview be kept confidential?

All the recorded information from the interview will be typed into an anonymised document and the original recordings destroyed. This information will then be only identified using a unique code and all your personal information (your name, place of work etc) will be removed from the written document so that you cannot be recognised from it. You will not be identified in any report or publication.

All of the information shared during the interview will be kept confidential, except in the event of a significant disclosure which poses a risk to you or other people. You will be reminded at the start of the interview not to discuss your experience in a way that could identify patients. If there is a breach of patient confidentiality during the interview, the section of the interview transcript will be deleted and that information will not be used in the final analysis.

What will happen if I decide to stop being in the study at any point?

You can withdraw from the study at any time and without needing to give a reason. If this is before your interview, then no information about you will be entered into the study. You will be able to withdraw from the study after your interview. However, it will only be possible to remove your data from the study for 14 days after your interview as once the recording has been typed up and anonymised, it may not be possible to identify your data to remove it from the study results.

What will happen to the information collected in the study?

Information collected for this study will be stored on password protected files at Newcastle University. This means that Newcastle University is responsible for looking after your information and using it properly. We will use your name, telephone number and address to contact you about the research study. The only people at Newcastle University who will have access to information that identifies you will be people who need to contact you to arrange the interview or audit the data collection process.

You can find out more about how Newcastle University uses your information at https://www.ncl.ac.uk/data.protection/dataprotectionpolicy/privacynotice/ and by contacting Newcastle University's Data Protection Officer (Maureen Wilkinson, rec-man@ncl.ac.uk).

What will happen to the results of the research study?

The results of the study will be written up by Dr Eve Palmer as part of her doctoral thesis which she is undertaking at Newcastle University. Anonymous quotes from interviews will be used in the thesis, research reports and publications. The information will directly influence the future care of patients with ILD in the North East and Cumbria. We aim to publish the research so that organisations in other areas of the UK can also benefit from the findings.

Who is organising and funding the study?

The research is funded by Marie Curie and charitable funds from the Royal Victoria Infirmary ILD department.

Have patients and the public been involved in the study design?

The study is supported by patient and carer support groups in the North East who have identified improving end of life care as a priority for patients. A patient and public representative panel has been formed which includes people with ILD, relatives and bereaved relatives. The panel members have advised on all participant information.

Who should I contact if I have a question or complaint?

The lead researcher who will be conducting the interviews is Dr Eve Palmer. This project is supervised by a team from Newcastle University. If you have any questions about the project please contact Dr Palmer directly. If you have any problems or complaints about the study please direct these to Dr Shelina Visram who is a member of the supervisory team.

Researcher: Dr Eve Palmer e.l.palmer2@newcastle.ac.uk 07835409082
Project supervisor: Dr Shelina Visram shelina.visram@newcastle.ac.uk

Appendix B: Participant facing documents

v. Consent form

CONSENT FORM – PATIENT / RELATIVE INTERVIEWS

Critically examining the experience of end of life care for people with interstitial lung disease: views of patients, families and healthcare professionals.

Participant Identification Number:

		Please initial
1.	I confirm that I have read the information sheet dated October 2020 v1.1 I have had the opportunity to consider the information and ask questions. I am happy with the answers to any questions I have asked	☐
2.	I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, and without my medical care or legal rights being affected	☐
3.	I understand that if I wish to withdraw from the study, the data collected up to this point will be included in the analysis of the research project unless I withdraw within 14 days of the interview	☐
4.	I agree to the interview being audio recorded, and that anonymous quotations may be used in written documents, presentations or scientific papers. I understand that I will not be identified by name in these documents to maintain my anonymity	☐
5.	I understand that the data will be anonymously transcribed and securely stored by the research team. I understand that my involvement will remain confidential	☐
6.	I understand that data collected during the study may be looked at by the research team or individuals from regulatory authorities or from the NHS Trust where my care is delivered, where it is relevant to my taking part in this research.	☐
7.	I agree to my General Practitioner being informed of my participation in the study	☐
8.	I agree to take part in the above study.	☐

Name of Participant	Date	Signature
_____	_____	_____
Name of Researcher	Date	Signature

Patient / Relative Interview Consent Form
V1.1 23/10/2020
IRAS ID: 276064

Appendix C: Interview schedules

vi. Interview schedule: patient participants

Interview Topic Guide: Patient Participants

I am interested in learning about the experience of people living with interstitial lung disease and hearing their thoughts and feelings about care towards the end of their lives. I hope that understanding your experience and your priorities will help to better address the needs and concerns of people with interstitial lung disease. The conversation is open-ended; there are no specific questions to ask you, but there are broad topics that I would like to cover. These topics are listed below with some questions which may be asked if they are relevant to your experience.

The conversation is likely to last thirty minutes to an hour, but if at any time you feel uncomfortable continuing, please let me know.

- Tell me about your lung disease and how it has affected your life?
  - When were you diagnosed and what led to this diagnosis?
  - Can you tell me about the symptoms that you experience?
  - What treatment have you had for this disease?
  - How does having interstitial lung disease affect your life?

- Tell me about our involvement with anyone from specialist palliative care services?
  - Who have you seen from palliative care teams? Where did you see them and who made the referral?
  - What are your thoughts about palliative care?
  - Have there been any problems with accessing services?

- Tell me about any discussions you have had about care towards the end of your life (this might be with healthcare providers or family/friends)?
  - Have you had any discussions about your future care?
  - Do you have worries or concerns about what will happen towards the end of your life?
  - Do you have any ideas about where you would want to be cared for at the end of your life? What has influenced this choice?
  - What do you think will be your main priorities when you are approaching the end of your life?

vii. Interview schedule: bereaved relative participants

Interview Topic Guide: Bereaved relative participants

I am interested in learning about the experience of people who have lost relatives to interstitial lung disease and hearing their thoughts and feelings about care towards the end of their life. I hope that understanding your experience and your priorities will help to better address the needs and concerns of people who are dying from this disease. The conversation is open-ended; there are no specific questions to ask you, but there are broad topics that the interview would like to cover. These topics are listed below with some questions which may be asked if they are relevant to your experience.

The interview is likely to last up to an hour, but you can stop the conversation at any time if you feel uncomfortable.

- Tell me about your relative and how their lung disease affected them?
  - When were they diagnosed and what led to this diagnosis?
  - How did the disease develop over time and what symptoms did they experience?
  - How did having interstitial lung disease affect their life?

- Tell me about their involvement with palliative care services?
  - Did they have any involvement with palliative care services?
  - Who did they see from palliative care teams? Where did they see them and who made the referral?
  - What were their thoughts about the palliative care?
  - Did they have any problems with accessing services?

- Can you tell me about what happened when they died?
  - Was their death expected?
  - Where did they die and what influenced this? Did they say where they wanted to die and was this where they died?
  - Do you have any worries or concerns about their death or aspects of their care?

viii. Interview schedule: healthcare professional participants

Interview Topic Guide: Healthcare professionals

There are no specific questions which I need to ask during this interview, the aim is to understand your experience of providing end of life care to patients with interstitial lung disease and therefore the interview is led by what you feel are the most important areas to discuss. However, this is an example of the type of questions which may be asked during the interview.

- Tell me about your job role in relation to patients with ILD?
  - Are there any barriers/facilitators which help or prevent you providing palliative care for these patients?
  - What local palliative care services are available for your patients?
  - How do you find your interaction with the Newcastle ILD team?

- Can you tell me about any patients who you have looked after at the end of life?
  - Was their death expected or unexpected?
  - What therapeutic interventions were needed towards the end of life?
  - How did you feel after their death?

- Where do your patients with ILD die?
  - What influences this?
  - Did they say where they wanted to die and was this where they died?

Appendix D: Geographical location of participants

Appendix E: Which factors influence the quality of end-of-life care in interstitial lung disease?

Review Article

PALLIATIVE MEDICINE

Palliative Medicine
2022, Vol. 36(2) 237–253


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Which factors influence the quality of end-of-life care in interstitial lung disease? A systematic review with narrative synthesis

Evelyn Palmer[1,2,3], Emily Kavanagh[2], Shelina Visram[3], Anne-Marie Bourke[1,2], Ian Forrest[1] and Catherine Exley[3]

Abstract

Background: People dying from interstitial lung disease experience considerable symptoms and commonly die in an acute healthcare environment. However, there is limited understanding about the quality of their end-of-life care.

Aim: To synthesise evidence about end-of-life care in interstitial lung disease and identify factors that influence quality of care.

Design: Systematic literature review and narrative synthesis. The review protocol was prospectively registered with PROSPERO (CRD42020203197).

Data sources: Five electronic healthcare databases were searched (Medline, Embase, PubMed, Scopus and Web of Science) from January 1996 to February 2021. Studies were included if they focussed on the end-of-life care or death of patients with interstitial lung disease. Quality was assessed using the Critical Appraisal Skills Programme checklist for the relevant study design.

Results: A total of 4088 articles were identified by initial searches. Twenty-four met the inclusion criteria, providing evidence from 300,736 individuals across eight countries. Most patients with interstitial lung disease died in hospital, with some subjected to a high burden of investigations or life-prolonging treatments. Low levels of involvement with palliative care services and advance care planning contributed to the trend of patients dying in acute environments. This review identified a paucity of research that addressed symptom management in the last few days or weeks of life.

Conclusions: There is inadequate knowledge regarding the most appropriate location for end-of-life care for people with interstitial lung disease. Early palliative care involvement can improve accordance with end-of-life care wishes. Future research should consider symptom management at the end-of-life and association with location of death.

Keywords

Interstitial lung disease, palliative care, end-of-life care, death, systematic review

What is already known about the topic?

- The majority of patients with interstitial lung disease die in hospital.
- Patients with interstitial lung disease experience a high symptom burden which escalates towards the end-of-life.
- The uncertain disease trajectory in interstitial lung disease contributes to late referral to palliative care services.

What this paper adds?

- This study confirms low levels of referrals to specialist palliative care services and engagement with advance care planning amongst patients with interstitial lung disease.
- When preferred place of death is ascertained, many patients wish to die at home. However, the majority of patients with interstitial lung disease die in hospital.
- There is a paucity of research that considers symptom control at the end-of-life in this patient group.

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Appendix F: When should palliative care be introduced for people with progressive fibrotic interstitial lung disease?

Review Article

PALLIATIVE MEDICINE

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SAGE

When should palliative care be introduced for people with progressive fibrotic interstitial lung disease? A meta-ethnography of the experiences of people with end-stage interstitial lung disease and their family carers

Evelyn Palmer[1,2,3], Emily Kavanagh[2], Shelina Visram[3], Anne-Marie Bourke[1,2], Ian Forrest[1] and Catherine Exley[3]

Abstract

Background: Little is currently known about the perspectives of people with interstitial lung disease and their carers in relation to the timing of palliative care conversations.

Aim: To establish patients' and carers' views on palliative care in interstitial lung disease and identify an optimum time to introduce the concept of palliative care.

Design: Meta-ethnography of qualitative evidence. The review protocol was prospectively registered with PROSPERO (CRD42021243179).

Data sources: Five electronic healthcare databases were searched (Medline, Embase, CINAHL, Scopus and Web of Science) from 1st January 1996 to 31st March 2022. Studies were included that used qualitative methodology and included patients' or carers' perspectives on living with end-stage disease or palliative care. Quality was assessed using the Critical Appraisal Skills Programme checklist.

Results: About 1779 articles were identified by initial searches. Twelve met the inclusion criteria, providing evidence from 266 individuals across five countries. Three stages were identified in the illness journey of a person with interstitial lung disease: (1) Information seeking, (2) Grief and adjustment, (3) Fear of the future. Palliative care involvement was believed to be most appropriate in the latter two stages and should be prompted by changes in patients' health such as respiratory infections, onset of new symptoms, hospital admission, decline in physical function and initiation of oxygen.

Conclusions: Patients and carers prefer referral to palliative care services to be prompted by changes in health status. Future research should focus on supporting timely recognition of changes in patients' health status and how to respond in a community setting.

Keywords

Interstitial lung disease, palliative care, meta-ethnography, qualitative research

What is already known about the topic?

- Despite the poor prognosis associated with progressive fibrotic lung disease, many people are not known to palliative care services or are referred late in the disease process.
- The experience of people with interstitial lung disease and their carers towards the end-of-life is poorly researched and it remains unclear when is the most appropriate time to involve palliative care services.

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