

A mixed-methods study of the Care Needs of individuals with idiopathic Pulmonary fibrosis and their carers—CaNoPy: a study protocol

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ABSTRACT

Introduction: Idiopathic pulmonary fibrosis (IPF) is a progressive, life-threatening illness of unknown aetiology, with no proven pharmacological treatments. There is a limited evidence base indicating that the disease negatively affects quality of life, leading to increased dependence, restrictions on daily activities and fatigue. However, there is a paucity of in-depth information on disease impact across its trajectory, particularly in relation to unmet needs, outcomes of importance to patients and the experiences of carers. Furthermore, little is known about the support and information needs of individuals and their carers, or at what point individual need should trigger a referral to palliative care services.

Methods and analysis: A mixed-methods study is proposed recruiting individuals with IPF at different stages of the disease and their carers from three respiratory centres in England and Wales. In-depth interviews will be undertaken with participants, adopting an Interpretative Phenomenological Analysis approach. The study will also use validated questionnaires to explore quality of life (EQ-5D), depression (Hospital Anxiety and Depression Scale), breathlessness (Borg dyspnoea scale) and cough (Leicester Cough Questionnaire, Cough Symptom Score).

Ethics and dissemination: Ethical approvals were gained in April 2012. Palliative care research is a developing field, but there has been limited focus on IPF. We anticipate that the results of the study will enable healthcare professionals to provide appropriate palliative care across the trajectory for individuals with the disease, and their carers, and we therefore aim to disseminate via relevant respiratory and palliative care journals and conferences. We will also support the lay representative involved in the project to disseminate the findings to patient groups.

INTRODUCTION

Idiopathic pulmonary fibrosis (IPF) is a progressive, life-limiting condition characterised by chronic inflammation and scarring,¹

ARTICLE SUMMARY

Article focus

- Idiopathic pulmonary fibrosis is a progressive, life-threatening illness with high symptom burden. However, there has been very limited research into patient perception of need, carer burden or patient/carer defined outcomes of importance in this population.
- A cross-sectional mixed-methods study is proposed to explore the experiences and needs of individuals and their carers across the illness trajectory of IPF.

Key messages

- The findings from this study should influence the care provided across the illness trajectory, particularly in terms of the information needs of individuals and carers at different stages of the disease, and identification of triggers for palliative care service involvement.
- The study will also determine outcomes of importance to patients which might influence clinical service evaluation and the design of future interventional studies in IPF.

Strengths and limitations of this study

- While this study is cross-sectional, rather than longitudinal, a large sample of patients at different stages of the disease, as well as their relatives, will be included.
- This multicentre study in England and Wales will also adopt a mixed-methods approach, including qualitative interviews and the use of validated questionnaires.

causing breathlessness and a dry cough in the individual.² The aetiology of IPF is unknown and the disease is progressive.³ The illness trajectory of IPF is variable and a study from the UK found that individuals lived with the disease for a median of 3 years before death,⁴ which is usually due to respiratory failure.² Identifying the prevalence of IPF is challenging as no mandatory

monitoring register exists, but the overall incidence in the UK is 7.44/100 000, with more men and older people affected.⁵ While anti-inflammatory, immunosuppressant and antifibrotic medications are prescribed for IPF;¹ no pharmacological treatments are proven to treat IPF,⁶ with the only significant treatment intervention being lung transplantation.

Lee *et al*⁷ describe a holistic approach to care for individuals with IPF, including disease-management (including medications), promoting education and self-management and symptom management. They further assert that palliative care should be fundamental and central to the management of IPF,⁷ which has been similarly encouraged in recent clinical guidance from the National Institute for Health and Care Excellence in the UK.⁸ Palliative care is defined by the WHO⁹ as:

an approach that improves the quality of life of patients and their families facing the problem associated with life-threatening illness, through the prevention and relief of suffering by means of early identification and impeccable assessment and treatment of pain and other problems, physical, psychosocial and spiritual

However, there is a paucity of research considering at what stage palliative care should be offered to individuals with IPF, and what care and support patients feel would benefit them.

This protocol therefore describes a proposed cross-sectional mixed-methods study (CaNoPy: *Care Needs of individuals with idiopathic Pulmonary fibrosis and their carers*) designed to investigate the needs and experiences of individuals with IPF, and their carers, across the illness trajectory.

Literature review

A literature search was undertaken using MEDLINE, CINAHL and PubMed, with additional hand-searching of reference lists, to identify individuals' and carers' experiences of IPF and the impact of the disease on their quality of life (QOL). The search identified several studies considering QOL for individuals with IPF, but fewer studies used a qualitative approach to explore their experiences of the disease. A dearth of studies focusing on the experiences of carers/family members is also noted.

QOL and IPF

The WHO¹⁰ defines QOL as an individual's "perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns" (p.7). Furthermore, Swigris *et al*¹¹ assert that health-related QOL is an individual's "perception of the impact of health (in all its many facets) on his or her quality of life" (p.588). A systematic review by Swigris *et al*¹¹ revealed that a small number of studies consider the QOL of individuals with IPF. After a methodical

literature search, Swigris *et al*¹¹ identified only seven studies that assessed QOL in a total of 512 adults with IPF. The included studies, which used the Medical Outcomes Study Short-Form 36-item tool (SF-36), the WHO-QOL 100-item tool or the St George's Respiratory Questionnaire, were conducted in Japan, the Netherlands, Brazil and the USA.¹¹ Health-related QOL was found to be significantly lower than the general population in almost all domains, but particularly physical aspects such as respiratory symptoms, energy levels and degrees of independence.¹¹ Specifically, dyspnoea was associated with worse QOL.¹¹

Recent studies considering QOL have been identified since Swigris *et al*'s¹¹ systematic review. An American cross-sectional study of 41 adults with IPF assessed fatigue, sleep quality and QOL with the following validated tools: Pittsburgh sleep quality index, Epworth sleepiness scale and SF-36.¹² Participants in Krishnan *et al*'s¹² study reported significantly poorer sleep than the general population, and like Swigris *et al*¹¹ identified that QOL was significantly reduced in most domains, in particular physical aspects. Additionally, sleep quality was associated with reduced QOL, which included physical and emotional measures,¹² and the authors thus recommend interventions to improve sleep quality. Few studies have adopted longitudinal approaches when considering QOL in people with IPF. However, Tomioka *et al*¹³ adopted a cross-sectional and longitudinal approach, measuring QOL using SF-36 at baseline (n=46) and again at least 12 months later (n=32) for participants who had not died, but developed other major diseases or were lost to follow-up. At baseline, participants reported significantly reduced QOL compared with the general population, while QOL had worsened significantly longitudinally in terms of physical function and bodily pain.¹³

The small number of studies assessing the QOL of people with IPF thus highlights significantly reduced outcomes, particularly in terms of physical health and sleep quality. However, no studies were identified that quantitatively measured relatives' or carers' QOL when caring for an individual with this progressive terminal disease. Nor have any studies explored in detail the experiences underlying QOL deterioration or patient/carer perceptions of interventions which might alter outcomes of importance. Given the variable trajectory of the disease, of particular interest is the identification of triggers for supportive and palliative interventions.

Qualitative experiences of IPF

Three studies were identified that explored the experiences of individuals with IPF using qualitative methods, although none of the studies discussed their methodological or philosophical approaches. Additionally, the papers did not discuss the participants' disease stages.

Swigris *et al*² undertook focus groups or individual interviews with 20 adults living with IPF in the USA. The purpose of the study was to develop an IPF-sensitive

health-related QOL measure by comparing the findings of the study with commonly-used global or respiratory tools. Dyspnoea and coughing were found to be distressing and impaired QOL and medications for IPF caused significant side effects; sleep quality was affected; low energy or exhaustion affected daily activities; forward planning was necessary and employment was either impossible or for some necessary to pay for medical care.² Furthermore, participants were concerned about being a physical or financial burden; appreciation was expressed towards relatives; IPF led to decreased libido or inability to undertake sexual activity; social activities were limited and participants were fearful about their health and recognised their mortality.² The authors concluded that an IPF-specific QOL instrument is required as their participants' perspectives of the disease were not sufficiently reflected in generic tools.

Schoenheit *et al*¹⁴ undertook single in-depth interviews with 45 adults with IPF from the following five European countries: Spain, Italy, UK, France and Germany. Additionally, 18 relatives were present during the interviews, although the article provides little insight into their experiences. The authors used psychological techniques of asking participants to select images that express their feelings and asking them to recall what was said in a particular situation. The study also collated details of symptoms and revealed that dyspnoea was experienced by 68% of participants, 59% reported a cough and 28% reported fatigue.¹⁴ The majority of participants had experienced delayed diagnoses and criticised the care they received, while a minority of participants who were diagnosed promptly reported their care more positively.¹⁴ Both groups, however, reported rushed and insensitive diagnosis and a lack of available information to them about the disease. IPF was found to have a substantial impact on daily life in terms of reduced independence, difficulty in continuing relationships and struggling financially through being unable to work.¹⁴

Recently, Bajwah *et al*¹⁵ interviewed eight patients with IPF, four carers (related to different patients) and six healthcare professionals in the UK. They highlighted that patients and carers had limited understanding of the disease, which made it difficult to plan ahead, and that patients had not discussed end-of-life preferences.¹⁵ While patients and carers reported feeling satisfied with the care provided by the respiratory team, they also reported a lack of coordination between different healthcare professionals and teams.¹⁵

IPF thus has a broad negative impact on everyday life for the individual, particularly in terms of increased dependence on relatives, reduction in socialising, financial concerns, recognition of mortality and a dearth of information. Bajwah *et al*¹⁵ included a small sample of carers but did not explicate their needs while caring for an individual with IPF, while Schoenheit *et al*'s¹⁴ study included relatives but the authors made little reference to them in their paper. Therefore, additional studies are

required to understand carer experiences of IPF and what support they require to care for a relative with the condition. Exploring the experiences of carers and their needs is crucial in a condition that is terminal and will thus require a high level of support from those closest to the individual with IPF.

This protocol thus presents a study designed to explore the perspectives of the individual with IPF and their carer at different stages of the disease.

METHODS

Aim and objectives

Aim: The aim of this study is to explore the needs of individuals with IPF and their families across the illness trajectory.

Objectives

1. Identify changes in individuals' and carers' perceived palliative care needs over the progression of IPF in order to improve future service interventions.
2. Identify time points or triggers at which palliative care services might effectively be introduced.
3. Define the specific information needs of individuals and their carers.
4. Evaluate specifically the experiences and roles of the carer.

Methodology and methods

The uncertain nature of disease progression makes a longitudinal study difficult to achieve in a set time frame, and therefore a cross-sectional design with individuals at different stages of the IPF trajectory was chosen. To meet the aim and objectives of the study, a mixed-methods approach will be undertaken, encompassing the use of validated assessment tools (QOL, anxiety and depression and IPF symptoms) and in-depth interviews utilising Interpretative Phenomenological Analysis (IPA) methodology. Participants will be recruited and data collected from three National Health Service (NHS) respiratory centres, including two Health Boards within Wales and one NHS Trust in England.

Recruitment and sampling

Individuals with IPF and their carers (a person of their choice who contributes most to their care, or at an earlier disease stage provides emotional support) will be recruited from the three respiratory centres, where a member of the clinical team will provide them with information about the study. Eligibility for the study will be decided by the clinical team according to a study pro forma, which classifies individuals at different stages on the IPF trajectory and documents respiratory comorbidities.

The inclusion criteria for individuals will be a diagnosis of IPF and receiving medical care for IPF at one of the three centres, as well as the ability to give informed consent and to communicate sufficiently to take part in an interview. The inclusion criteria for carers include

caring for an individual with IPF in the study, as well as the ability to give informed consent and communicate adequately to be interviewed. The exclusion criteria for individuals with IPF and carers will be any factor that prevents communication or comprehension. A disease typology was generated by palliative and respiratory consultants who are part of the research team to classify four different stages of the disease. To provide an insight into the individuals', and thus the carers', needs across the disease trajectory, four groups of participants (see [table 1](#)) will be recruited, including people with

1. Limited disease: forced vital capacity (FVC) greater than 50% predicted and gas transfer (TLCO) greater than 40% predicted;
2. Extensive disease: FVC less than 50% or TLCO less than 40% predicted;
3. Progressive disease: a fall in either FVC greater than 10% or TLCO greater than 15% during the previous 12 months;
4. Stable disease: a fall of less than 10% in FVC or less than 15% in TLCO in the previous 12 months.

Participants will be purposively sampled¹⁶ to represent the four categories above, based on their FVC scores contained in their clinical notes, for example, limited progressive, limited stable, extensive progressive, extensive stable. Congruent with the recommendations for IPA, the sample size for each group will be 6–10 individuals with IPF and 6–10 carers per homogeneous group,¹⁷ to represent a perspective rather than a population. While the total sample size (n=48–80) is therefore large for the methodology, it is necessary to gain insight into the perspectives of the four groups of participants.

Potential participants will be provided with a participant information sheet, reply letter and stamped addressed envelope by a member of the clinical team in the respiratory clinic, and requested to return the reply slip to the research team if they are happy to be contacted to take part in the study. Willing participants will then be telephoned by a researcher and an interview will be arranged at a time and place convenient for them.

Table 1 Participant group characteristics

| Participant group and characteristics | Limited disease | Extensive disease |
|---------------------------------------|--|--|
| Progressive disease | 6–10 individuals with IPF | 6–10 individuals with IPF |
| Stable disease | 6–10 carers 6–10 individuals with IPF | 6–10 carers 6–10 individuals with IPF |
| | 6–10 carers | 6–10 carers n=48–80 |

IPF, idiopathic pulmonary fibrosis.

Data collection

Three data collection methods will be used in this study: recording of demographic and comorbidities data, questionnaires and in-depth interviews.

Comorbidities and demographic data: Demographic variables (age, marital status, location) and comorbidities (in particular chronic obstructive pulmonary disease, pulmonary hypertension and lung cancer) of individuals with IPF will be recorded by clinicians at the clinic on a case report form.

Questionnaires: Prior to the in-depth interview, individuals with IPF will be requested to complete a booklet of questionnaires covering QOL, anxiety and depression and symptoms of IPF. These questionnaires will enable the research team to observe whether QOL, anxiety and depression change over time and how these correlate with dyspnoea and coughing.

1. QOL: a validated, global health-related QOL tool will be used to evaluate QOL in the form of the EQ-5D, which encompasses five questions on mobility, self-care, usual activities, pain/discomfort and anxiety/depression.¹⁸ Swigris *et al*¹⁹ designed and tested a QOL assessment tool specifically for IPF (ATAQ-IPF), but no other studies were identified that use this tool, and therefore we opted for a more generic but well-validated tool.
2. Anxiety and depression: the validated Hospital Anxiety and Depression Scale includes 14 questions and has been used widely across patient populations and found to be of high specificity and sensitivity.²⁰
3. Breathlessness: a systematic review²¹ found that dyspnoea assessment scales have not been validated for use in palliative care, but it also identified that the Borg dyspnoea scale, measuring severity of breathlessness on a numerical scale, appeared the most appropriate for use with this population.
4. Cough: the Leicester Cough Questionnaire is a 19-item self-completion tool measuring the physical, psychological and social QOL in relation to living with a chronic cough, which demonstrated high specificity and sensitivity.²² The Cough Symptom Score²³ measures the severity of the cough on a visual analogue scale.

The researcher will assist participants to complete the questionnaires as required, which should take around 20 min, and this will occur before the interview to minimise the influence of topics discussed on questionnaire completion.

In-depth interviews: IPA is a qualitative psychological approach used to explore how people make sense of major events in their lives.¹⁷ Three philosophical approaches influence IPA¹⁷: exploring the lived experience (phenomenology); interpretation of the phenomenon (hermeneutics); exploring the particular rather than attempting to generalise a group (idiography). This methodology has previously been used successfully to explore palliative care issues, with both patients²⁴ and healthcare professionals.²⁵

To enable access to detailed personal accounts of how participants experience IPF,¹⁷ the research team will utilise semistructured interviews with people with the disease and their carers. The interviews will be conducted at a place and time convenient for the participants, in their homes or a quiet clinic location or over the telephone, if preferred. One researcher will conduct the interviews across all sites. It is anticipated that the interviews will last between 30 and 60 min, with the interviewer terminating the discussion if they become concerned that the participant is unwell or fatigued. We aim to interview individuals with IPF and carers separately, as is common in qualitative studies with both parties^{26 27} and recommended by Smith *et al.*¹⁷ If so, relatives will be interviewed first to allow individuals with IPF to have a break between completing the questionnaire and being interviewed. However, participants will be interviewed together if they prefer, which Cavers *et al.*²⁸ allowed in their qualitative study due to their participants with glioma struggling at times with communication. With the participants' consent, interviews will be audio-recorded and transcribed verbatim.

An interview schedule will be used (see box 1) which will also enable participants to influence the agenda and discuss topics pertinent to them.¹⁷ The interview process is dynamic and iterative, and so the schedule will be reviewed after the first few interviews to assess whether alterations are necessary based on interviewee priorities.

Data analysis

While the quantitative and qualitative data will be analysed separately using appropriate methods, a complementary analysis of both data sets will seek to define key points or triggers for palliative care involvement. This will enable the identification of key components of participants' experiences of the IPF trajectory and clarify what possible interventions could be of benefit to patients and carers.

Quantitative: The quantitative data will be analysed using SPSS by a member of the research team who is a statistician.

1. Descriptive statistics will be used to present the questionnaire data in graphic format and questionnaire-specific methodologies will be employed.
2. Categorical data will be presented as proportions with a 95% CI and continuous data as means with a 95% CI. The limited size of the data set means that the analysis will be exploratory.

Qualitative: IPA data analysis involves considering each case (participant) in turn and systematically interpreting how participants have interpreted their experience, before a narrative account of each case is developed.¹⁷ A six-step approach to data analysis is recommended by Smith *et al.*¹⁷

1. Reading and rereading: listening to the interview and reading the transcript to familiarise oneself with the data and ensure that the participant is the focus of the analysis.

2. Initial noting: reading the transcript and noting anything important, including what is said (descriptive), the context of this (linguistic) and identifying patterns in the data and what these mean (conceptual).
3. Developing emerging themes: turning the notes into themes by summarising what is important in the transcript.
4. Connecting themes: this involves mapping how the emergent themes fit together.
5. Moving to the next case: repeating the process with each case, ensuring that each case is treated individually by trying to bracket out the findings from previous cases.
6. Patterns across cases: examining the cases for connections, considering how themes from one case feature in another and which themes are the strongest—redefining themes is common at this stage. The result should be superordinate themes and themes within.

The four different groups of participants will be analysed separately, with comparisons made between the groups. The data will be primarily analysed by the researcher responsible for data collection. Tong *et al.*²⁹ recommend research triangulation to promote a deeper understanding of the phenomenon, and therefore 10% of the data will be double coded for agreement by a second member of the research team. Additionally, the research team will confer on the analysis to ensure that there is agreement across the themes.

ETHICS AND DISSEMINATION

Ethical considerations

The Research Governance Frameworks for England and Wales^{30 31} and guidelines from the National Patient Safety Agency³² were followed when designing the study.

Participants will have a minimum of 24 h to decide whether to take part in the study, and the research team will ensure that the participants are fully aware of the details of the research prior to collecting written informed consent. Informed consent, which is central to ethical research,³⁰ will be taken by the researcher conducting the interview who is experienced at doing so, or by a member of the clinical team who has undertaken appropriate Good Clinical Practice (research) training. The research team will ensure that all participants have the capacity to consent in line with the Mental Capacity Act.³³ All data will be kept strictly confidential according to the principles of the Data Protection Act³⁴ and stored safely in the research unit.

There is growing impetus to include patients and the public in health and social care research as members of the research team, rather than solely as participants, which Tischler *et al.*³⁵ argue encourages the research to be relevant to patients. Therefore, in line with guidance from Involving People³⁶ and Involve,³⁷ the study documentation was reviewed by a lay representative volunteer at the research centre hosting the study. The research

**Box 1** Interview schedules*Individuals with idiopathic pulmonary fibrosis (IPF)*

Diagnosis

1. What symptoms were you experiencing when you were first diagnosed with IPF? What made you seek medical attention?
2. When and how did you get diagnosed with IPF?
3. Had you heard about the condition before? If yes, what did you know about it?
4. What information were you given about your illness? How useful did you find this information?
5. Did you seek out other information on IPF? If so, what and how useful was it?

Living with IPF

6. How does your illness affect you? How has it impacted on your quality of life?
7. How have you been coping with or managing your illness?
8. Which services have you been receiving?
9. What do you think about the support that you have been receiving from health professionals?
10. Are there any gaps in the care that you have been receiving? What else could be done to help you?

The future

11. What is your understanding of how your illness will progress? Do you feel you have enough information about this? What else would you like to know?
 12. Do you anticipate the need for more help later on? What kind of help do you think you might need?
- Is there anything else you have thought of that you would like to mention or discuss now?

Carers

Diagnosis

1. When and how did you first learn about (patient's name) illness?
2. What symptoms was (name) experiencing when they were first diagnosed with IPF? What made them seek medical attention?
3. Had you heard about the condition before? If yes, what did you know about it?
4. What information were you given about the illness? How useful did you find this information?
5. Did you seek out other information on IPF? If so, what and how useful was it?

Living with IPF

6. How does (name) illness affect them?
7. How have they been coping with these changes/managing their illness?
8. How does (name) illness affect you? How has it impacted on your quality of life?
9. How have you been coping with these changes?
10. Have you been receiving any professional support or assistance?
11. What do you think about the support that you and (name) have been receiving from health professionals?
12. Are there any gaps in the care that (name) has been receiving? What else could be done to help you both?

The future

13. What is your understanding of how (name) illness will progress? Do you feel you have enough information about this? What else would you like to know?
 14. Do you anticipate the need for more help later on? If so, what kind of help do you think might need?
- Is there anything else you have thought of that you would like to mention or discuss now?

centre has a substantial model of consumer involvement and the nominated study volunteer will be involved at all stages of the study and will attend regular meetings as a member of the research team.

Validity and reliability/rigour

Greene *et al*³⁸ argue that mixed-methods studies enable triangulation of results, thus increasing confidence in the findings of the research. Thus, utilising in-depth interviews and multiple questionnaires to explore participants' QOL and experience across the disease trajectory should promote complementarity³⁸ and deepen interpretations from the study. Yardley³⁹ asserts the characteristics of 'good' qualitative research.

1. Sensitivity to context: the thorough literature review for this study promotes sensitivity, which is supported by the clinical and research expertise of the research team;

2. Commitment and rigour and transparency and coherence: encouraged through the proposed systematic and sufficient sampling, experienced qualitative researchers collecting data and a multidisciplinary team of researchers analysing the data systematically;
3. Impact and importance: the objectives of the study are to generate evidence that can be translated into clinical practice, particularly in relation to the information and palliative service needs of individuals with IPF and their carers.

Furthermore, we aim to promote validity in the use of validated assessment tools with high specificity and sensitivity.

Limitations

One limitation of this study is the cross-sectional rather than longitudinal design. However, as previously

discussed, a longitudinal design is extremely challenging and resource intensive due to the progressive and unpredictable nature of IPF. We believe that the chosen cross-sectional design will provide representative data in an efficient and inclusive manner. Another limitation is the use of questionnaires that have not been specifically validated for use with this clinical population. Therefore, we have pragmatically selected tools that have been used successfully with similar groups, are not too onerous for participants to complete and provide a broad perspective of participants' QOL and provide insight into the impact of IPF symptoms on everyday life.

Dissemination

Palliative care research is a developing discipline with significant methodological challenges. It frequently aims to assess complex interventions in heterogeneous, vulnerable populations. Successful outcomes depend on robust methodological approaches which are complementary and which engage multidisciplinary researchers.^{40–41} Identifying key points of intervention and outcomes of importance to patients are essential to the development of well-designed pragmatic trials and the implementation of efficient, patient-focused clinical services.

There is increasing focus on ensuring that palliative care services are available to and accessed by individuals with non-malignant diseases—with emphasis on need, not diagnosis.⁴² IPF is, as previously discussed, an under-researched disease. We anticipate that the results of this study will provide fundamental information considering the quantitative and qualitative experiences and needs of individuals and their carers, and will therefore be disseminated via relevant clinical and research journals and international conferences, encompassing both palliative care and respiratory specialties. The Chief Investigator and three of the Co-Investigators are Consultant Clinicians in palliative medicine and respiratory specialties, which will enable the planning and provision of appropriate palliative care services for individuals with IPF and their carers across the illness trajectory. Furthermore, the lay representative involved with the project will be supported to disseminate the results to relevant patient groups.

This paper has explored the incidence and symptoms of IPF, with discussion of the limited previous research undertaken in this area in terms of QOL or experience of the disease. A paucity of research considering the experience and needs of carers was also identified. This protocol has presented a planned multicentre mixed-methods study in England and Wales with people at different stages of IPF and their carers, utilising validated questionnaires and in-depth interviews. The results of the study may help healthcare professionals to plan and implement appropriate palliative care services for people with IPF and appropriate support for their carers.

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Contributors AB is the Chief Investigator. He designed the study and reviewed the manuscript. CS contributed to the protocol, reviewed the manuscript and will collect and analyse the data. JB prepared the manuscript. KH, BH-G and RH are the site Principal Investigators. GG designed the study. AN designed the study and reviewed the manuscript. All authors have read and approved the final version of the manuscript.

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Competing interests None.

Ethics approval The study was approved by the university and the South East Wales National Health Service Research Ethics Committee in Wales (reference 12/WA/0109) and governance was gained from the three hospital sites in April 2012.

Provenance and peer review Not commissioned; internally peer reviewed.

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REFERENCES

1. Raghu G, Weycker D, Edelsberg J, *et al.* Incidence and prevalence of idiopathic pulmonary fibrosis. *Am J Respir Crit Care Med* 2006;174:810–16.
2. Swigris J, Stewart A, Gould M, *et al.* Patients' perspectives on how idiopathic pulmonary fibrosis affects the quality of their lives. *Health Qual Life Outcomes* 2005;3:61–9.
3. Selman M, King T, Pardo A. Idiopathic pulmonary fibrosis: prevailing and evolving hypotheses about its pathogenesis and implications for therapy. *Ann Intern Med* 2001;134:136–51.
4. Gribbin J, Hubbard R, Le Jeune I, *et al.* Incidence and mortality of idiopathic pulmonary fibrosis and sarcoidosis in the UK. *Thorax* 2006;61:980–5.
5. Navaratnam V, Fleming K, West J, *et al.* The rising incidence of idiopathic pulmonary fibrosis in the UK. *Thorax* 2011;66:462–7.
6. Raghu G, Collard H, Egan J, *et al.*, on behalf of the ATS/ERS/JRS/ALAT Committee on Idiopathic Pulmonary Fibrosis. An official ATS/ERS/JRS/ALAT statement: idiopathic pulmonary fibrosis: evidence-based guidelines for diagnosis and management. *Am J Respir Crit Care Med* 2011;183:788–824.
7. Lee J, McLaughlin S, Collard H. Comprehensive care of the patient with idiopathic pulmonary fibrosis. *Curr Opin Pulm Med* 2011;17:348–54.
8. National Institute for Health and Care Excellence. Idiopathic pulmonary fibrosis: diagnosis and management of suspected idiopathic pulmonary fibrosis. 2013. <http://www.nice.org.uk/nicemedia/live/14183/64124/64124.pdf>
9. World Health Organisation. WHO Definition of Palliative Care. 2012. <http://www.who.int/cancer/palliative/definition/en/>
10. World Health Organisation. WHOQOL Measuring Quality of Life. 1997. http://www.who.int/mental_health/media/68.pdf
11. Swigris J, Kuschner W, Jacobs S, *et al.* Health-related quality of life in patients with idiopathic pulmonary fibrosis: a systematic review. *Thorax* 2005;60:558–94.
12. Krishnan V, McCormack M, Mathai S, *et al.* Sleep quality and health-related quality of life in idiopathic pulmonary disease. *Chest* 2008;134:693–8.
13. Tomioka H, Imanaka K, Hashimoto K, *et al.* Health-related quality of life in patients with idiopathic pulmonary fibrosis—cross-sectional and longitudinal study. *Intern Med* 2007;46:1533–42.
14. Schoenheit G, Becattelli I, Cohen A. Living with idiopathic pulmonary fibrosis: an in-depth qualitative survey of European patients. *Chron Respir Dis* 2011;8:225–31.
15. Bajwah S, Koffman J, Higginson I, *et al.* 'I wish I knew more...' the end-of-life planning and information needs for end-stage fibrotic interstitial lung disease: views of patients, carers and health professionals. *BMJ Support Palliat Care* 2013;3:84–90.
16. Patton M. *Qualitative research and evaluation methods*. 3rd edn. London: Sage, 2002.



17. Smith J, Flowers P, Larkin M. *Interpretative phenomenological analysis: theory, method and research*. Los Angeles: Sage, 2009:1–232.
18. Rabin R, de Charro F. EQ-5D: a measure of health status from the EuroQol Group. *Ann Med* 2001;33:337–43.
19. Swigris J, Wilson S, Green K, *et al*. Development of the ATAQ-IPF: a tool to assess quality of life in IPF. *Health Qual Life Outcomes* 2010;8:77–85.
20. Bjelland I, Dahl A, Haug T, *et al*. The validity of the hospital anxiety and depression scale: an updated literature review. *J Psychosom Res* 2002;52:69–77.
21. Dorman S, Byrne A, Edwards A. Which measurement scales should we use to measure breathlessness in palliative care? A systematic review. *Palliat Med* 2007;21:177–91.
22. Biring S, Prudon B, Carr A, *et al*. Development of a symptom specific health status measure for patients with chronic cough: Leicester Cough Questionnaire (LCQ). *Thorax* 2003;58:339–43.
23. Jones R, Hilldrup S, Hope-Gill B, *et al*. Mechanical induction of cough in Idiopathic Pulmonary Fibrosis. *Cough* 2011;7:2–12.
24. Seamark D, Blake S, Seamark C, *et al*. Living with severe chronic obstructive pulmonary disease (COPD): perceptions of patients and their carers: an interpretative phenomenological analysis. *Palliat Med* 2004;18:619–25.
25. Taubert M, Noble S, Nelson A. What challenges good palliative care provision out-of-hours? A qualitative interview study of out-of-hours general practitioners. *Palliat Med* 2011;1:13–18.
26. Orrevall Y, Tishelman C, Permert J. Home parenteral nutrition: a qualitative interview study of the experiences of advanced cancer patients and their families. *Clin Nutr* 2005;24:961–70.
27. White Y, Grenyer B. The biopsychosocial impact of end-stage renal disease: the experience of dialysis patients and their partners. *J Adv Nurs* 1999;30:1312–29.
28. Cavers D, Hacking B, Erridge S, *et al*. Adjustment and support needs of glioma patients and their relatives: serial interviews. *Psychooncology* 2012;1299–1305. doi: 10.1002/pon.3136
29. Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care* 2007;19:349–57.
30. Department of Health. *Research Governance Framework for Health and Social Care*. 2nd edn. 2005. http://www.dh.gov.uk/prod_consum_dh/groups/dh_digitalassets/@dh/@en/documents/digitalasset/dh_4122427.pdf
31. Welsh Assembly Government. *Research Governance Framework for Health and Social Care in Wales*. 2nd edn. 2009. <http://cymru.gov.uk/topics/health/research/word/publications/researchgovernance/?lang=en>
32. National Patient Safety Agency. Information Sheets and Consent Forms: Guidance for Researchers and Reviewers. 2011. <http://www.nres.nhs.uk/EasySiteWeb/GatewayLink.aspx?allid=338>.
33. United Kingdom. Mental Capacity Act 2005. 2005. http://www.opsi.gov.uk/acts/acts2005/ukpga_20050009_en_1.
34. United Kingdom. Data Protection Act 1998. 1998. http://www.opsi.gov.uk/acts/acts1998/ukpga_19980029_en_1
35. Tischler V, D'Silva K, Cheetham A, *et al*. Involving patients in research: the challenge of patient-centredness. *Int J Soc Psychiatry* 2010;56:623–33.
36. Moore B, Porteous C, Simon N. Pathways for involving people in social care and health research in Wales: a signposting document. 2007. <http://www.involvingpeople.org.uk/>
37. Vale C, Fitzgibbon J, Hanley B, *et al*. Public involvement in clinical trials: supplement to the briefing notes for researchers. 2012. <http://www.invo.org.uk/wp-content/uploads/2012/04/INVOLVEpublicinvolvementinclinicaltrialsBriefingnotes2012.pdf>
38. Greene J, Caracelli V, Graham W. Toward a conceptual framework for mixed-method evaluation designs. *Educ Eval Policy Anal* 1989;11:255–74.
39. Yardley L. Dilemmas in qualitative health research. *Psychol Health* 2000;15:215–28.
40. Byrne A, Hood K, Griffiths G, *et al*. Design and conduct of research and clinical trials. In: Walsh D, Caraceni A, Fainsinger R, Foley K, Glare P, Goh C, Lloyd-Williams M, Nunez Olarte J, Radbruch L, eds. *Palliative medicine*. Philadelphia: Saunders Elsevier, 2009:148–56.
41. Kaasa S, Hjermstad M, Loge J. Methodological and structural challenges in palliative care research: how have we fared in the last decades? *Palliat Med* 2006;20:727–34.
42. Fallon M, Foley P. Rising to the challenge of palliative care for non-malignant disease. *Palliat Med* 2012;26:99–100.

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