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CaNoPy: A mixed-methods study of the care needs of individuals with idiopathic pulmonary fibrosis and their carers. A study protocol.

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Care needs of individuals with idiopathic pulmonary fibrosis and their carers

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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 idiopathic pulmonary fibrosis. 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.

KEYWORDS

Idiopathic pulmonary fibrosis, carer, mixed-methods, Interpretative Phenomenological Analysis

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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-method study is proposed to explore the experiences and needs of individuals and their carers across the illness trajectory of idiopathic pulmonary fibrosis.

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 both 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 differences stages of the disease, and their relatives, will be included;
- This multi-centre study, in both England and Wales, will also adopt a mixed-methods approach, including qualitative interviews and the use of validated questionnaires.

INTRODUCTION

Idiopathic pulmonary fibrosis (IPF) is a progressive, life-limiting condition characterised by chronic inflammation and scarring,[1] 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 United Kingdom (UK) found that individuals lived with the disease for a median of three 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 per 100,000, with more men and older people affected.[5] While anti-inflammatory, immunosuppressant and anti-fibrotic medications are prescribed for IPF;[1] no pharmacologic 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 (NICE) in the UK.[8] Palliative care is defined by the World Health Organisation [9] 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 designed to investigate the needs and experiences of individuals with idiopathic pulmonary fibrosis, 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. The search identified several studies considering quality of life for individuals with IPF, but fewer studies used a qualitative approach to explore their experiences of the disease. A dearth of studies focussing on the experiences of carers/ family members is also noted.

Quality of life and IPF

The World Health Organisation [10] define quality of life 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 quality of life 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 quality of life of individuals with IPF. After a methodical literature search, Swigris et al. [11] identified only seven studies that assessed quality of life in a total of 512 adults with IPF. The included studies, which used the Medical Outcomes Study Short-Form 36-item tool (SF-36), World Health Organisation Quality of Life 100-item tool (WHO-QOL) or the St George's Respiratory Questionnaire (SGRQ), were conducted in Japan, the Netherlands, Brazil and the

United States of America (USA).[11] Health-related quality of life 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 quality of life.[11]

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

The small number of studies assessing the quality of life of people with IPF thus highlights significantly reduced outcomes, in particular in terms of physical health and sleep quality. However, no studies were identified that quantitatively measured relatives' or carers' quality of life when caring for an individual with this progressive, terminal disease. Nor have they

explored in detail the experiences underlying quality of life deterioration or patient/carer perceptions of interventions which might alter outcomes of importance. Of particular interest given the variable trajectory of the disease, is the identification of triggers for supportive and palliative interventions.

Qualitative experiences of IPF

Three studies were identified that explore the experiences of individuals with IPF using qualitative methods; although none of the studies discuss their methodological or philosophical approaches. Additionally, the papers do not discuss 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 quality of life measure by comparing the findings of the study to commonly-used global or respiratory tools. Dyspnoea and coughing were found to be distressing and impaired quality of life, 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 quality of life 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 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 the 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]

More 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 do not explicate their needs while caring for an individual with IPF, while Schoenheit et

al.'s [14] study included relatives but the authors make 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. Interrogating 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 both 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 (quality of life, anxiety and depression, and IPF symptoms) and in-depth interviews utilising Interpretive 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 proforma, which classifies individuals at different stages on the IPF trajectory and documents respiratory co-morbidities.

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

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.

Table one: participant group characteristics

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

Participants will be purposively sampled [16] to represent the four categories above, based on their FVC scores contained in their clinical notes, e.g. limited progressive, limited stable, extensive progressive, extensive stable. Congruent with the recommendations for Interpretative Phenomenological Analysis (IPA), the sample size for each group will be 6-10 individuals with IPF and 6-10 carers per homogenous 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 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.

Data collection

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

Co-morbidities and demographic data Demographic variables (age, marital status, location) and co-morbidities (in particular chronic obstructive pulmonary disease, pulmonary hypertension and lung cancer) of the 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 quality of life, anxiety and depression and symptoms of IPF. These questionnaires will enable the research team to observe whether quality of life, anxiety and depression change over time and how these correlate with dyspnoea and coughing.

1) Quality of life (QOL): a validated, global health-related QOL tool will be used to evaluate quality of life 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 quality of life 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 (HADS) 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 review [21] found that dyspnoea assessment scales have not been validated for use in palliative care but 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-tem self-completion tool measuring physical, psychological and social quality of life in relation to living with a chronic cough, which demonstrated high specificity and sensitivity.[22] The Cough Symptom Score [23] 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 minutes, and this will occur before the interview to minimise the influence of topics discussed on questionnaire completion.

In-depth interviews IPA is as a qualitative psychological approach used to explore how people make sense of major events in their lives.[17] Three philosophical approaches influence IPA:[17] exploring the lived experience (phenomenology); interpretation of the phenomenon (hermeneutics); exploring the particular rather attempting generalise a group

(idiography). This methodology has previously been used successfully to explore palliative care issues, with both patients [24] and healthcare professionals.[25]

To enable access to detailed personal accounts of how participants experience IPF [17], the research team will utilise semi-structured interviews with people with the disease and their carers. The interviews will be conducted at a place and time convenient for the participants, either in their homes or a quiet clinic location, or over the telephone if preferred. It is anticipated that the interviews will last between 30 and 60 minutes, 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 participants' consent, interviews will be audio-recorded and transcribed verbatim.

An interview schedule will be used (see Table 2) while also enabling 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.

Table two: interview schedules

Individuals with idiopathic pulmonary fibrosis
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, 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 might need? Is there anything else you've 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've thought of that you would like to mention or discuss now?

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.

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% confidence interval (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 super-ordinate themes and themes within.

The four different groups of participants will be analysed separately with comparison made between the groups. The research group will confer on the analysis to ensure that there is agreement across the themes. 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.

ETHICS AND DISSEMINATION

Ethical considerations

The study was approved by the university and regional National Health Service Research Ethics Committee in Wales and governance was gained from the three hospital sites in April 2012. The Research Governance Frameworks for England and Wales [30,31] and guidelines from the National Patient Safety Agency [32] were followed when designing the study.

Participants will have a minimum of 24 hours to decide whether to take part in the study and the research team will ensure that 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 a member of the research team 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 Act [34] and data will be 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 People [36] and Involve [37], the study documentation was reviewed by a lay representative volunteer at the research centre hosting the study. The research 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' quality of life and experience across the

disease trajectory should promote complementarity [38] and deepen interpretations from the study. Yardley [39] asserts 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; 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, in particular 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' quality of life 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 both 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] Idiopathic pulmonary fibrosis is, as previously discussed, an under-researched disease. We anticipate that the results of this study will provide fundamental information considering the experiences and needs of individuals and their carers, both quantitatively and qualitatively, and will therefore be disseminated via relevant clinical and research journals and international conferences, encompassing both palliative care and respiratory specialities. The Chief Investigator and three of the Co-Investigators are Consultant Clinicians in palliative medicine and respiratory specialities, which will enable the planning and provision of appropriate palliative care services for both 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 idiopathic pulmonary fibrosis, with discussion of the limited previous research undertaken in this area in terms of quality of life 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 both 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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We wish to acknowledge the clinical teams who granted access and will support recruitment.

Competing interests:

None

Authors' contributions:

AB is the Chief Investigator, designed the study and reviewed the manuscript. CS contributed to the protocol, reviewed the manuscript and will collect and analyse data. JB prepared the manuscript. KH, BHG and RH are site Principal Investigators. GG designed the study. AN designed the study and reviewed the manuscript.

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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 idiopathic pulmonary fibrosis. 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.

KEYWORDS

Idiopathic pulmonary fibrosis, carer, mixed-methods, Interpretative Phenomenological Analysis

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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-method study is proposed to explore the experiences and needs of individuals and their carers across the illness trajectory of idiopathic pulmonary fibrosis.

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 both 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 differences stages of the disease, and their relatives, will be included;
- This multi-centre study, in both England and Wales, will also adopt a mixed-methods approach, including qualitative interviews and the use of validated questionnaires.

INTRODUCTION

Idiopathic pulmonary fibrosis (IPF) is a progressive, life-limiting condition characterised by chronic inflammation and scarring,[1] 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 United Kingdom (UK) found that individuals lived with the disease for a median of three 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 per 100,000, with more men and older people affected.[5] While anti-inflammatory, immunosuppressant and anti-fibrotic medications are prescribed for IPF;[1] no pharmacologic 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 (NICE) in the UK.[8] Palliative care is defined by the World Health Organisation [9] 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 idiopathic pulmonary fibrosis, 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. The search identified several studies considering quality of life for individuals with IPF, but fewer studies used a qualitative approach to explore their experiences of the disease. A dearth of studies focussing on the experiences of carers/ family members is also noted.

Quality of life and IPF

The World Health Organisation [10] define quality of life 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 quality of life 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 quality of life of individuals with IPF. After a methodical literature search, Swigris et al. [11] identified only seven studies that assessed quality of life in a total of 512 adults with IPF. The included

studies, which used the Medical Outcomes Study Short-Form 36-item tool (SF-36), World Health Organisation Quality of Life 100-item tool (WHO-QOL) or the St George's Respiratory Questionnaire (SGRQ), were conducted in Japan, the Netherlands, Brazil and the United States of America (USA).[11] Health-related quality of life 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 quality of life.[11]

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

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

Qualitative experiences of IPF

Three studies were identified that explore the experiences of individuals with IPF using qualitative methods; although none of the studies discuss their methodological or philosophical approaches. Additionally, the papers do not discuss 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 quality of life measure by comparing the findings of the study to commonly-used global or respiratory tools. Dyspnoea and coughing were found to be distressing and impaired quality of life, 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 quality of life 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 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 the 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]

More 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 do not explicate their needs while caring for an individual with IPF, while Schoenheit et al.'s [14] study included relatives but the authors make 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. Interrogating 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 both 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 (quality of life, anxiety and depression, and IPF symptoms) and in-depth interviews utilising Interpretive 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 proforma, which classifies individuals at different stages on the IPF trajectory and documents respiratory co-morbidities.

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

Table one: participant group characteristics

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

Participants will be purposively sampled [16] to represent the four categories above, based on their FVC scores contained in their clinical notes, e.g. limited progressive, limited stable, extensive progressive, extensive stable. Congruent with the recommendations for Interpretative Phenomenological Analysis (IPA), the sample size for each group will be 6-10 individuals with IPF and 6-10 carers per homogenous 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 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.

Data collection

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

Co-morbidities and demographic data Demographic variables (age, marital status, location) and co-morbidities (in particular chronic obstructive pulmonary disease, pulmonary hypertension and lung cancer) of the 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 quality of life, anxiety and depression and symptoms of IPF. These questionnaires will enable the research team to observe whether quality of life, anxiety and depression change over time and how these correlate with dyspnoea and coughing.

1) Quality of life (QOL): a validated, global health-related QOL tool will be used to evaluate quality of life 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 quality of life 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 (HADS) 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 review [21] found that dyspnoea assessment scales have not been validated for use in palliative care but 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-tem self-completion tool measuring physical, psychological and social quality of life in relation to living with a chronic cough, which demonstrated high specificity and sensitivity.[22] The Cough Symptom Score [23] 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 minutes, 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 IPA:[17] 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 patients [24] and healthcare professionals.[25]

To enable access to detailed personal accounts of how participants experience IPF [17], the research team will utilise semi-structured interviews with people with the disease and their carers. The interviews will be conducted at a place and time convenient for the participants, either 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 minutes, 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 participants' consent, interviews will be audio-recorded and transcribed verbatim.

An interview schedule will be used (see Table 2) while also enabling 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.

Table two: interview schedules

Individuals with idiopathic pulmonary fibrosis
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, 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 might need? Is there anything else you've 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've thought of that you would like to mention or discuss now?

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% confidence interval (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 super-ordinate themes and themes within.

The four different groups of participants will be analysed separately with comparison 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 member second 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 study was approved by the university and 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. The Research Governance Frameworks for

England and Wales [30,31] and guidelines from the National Patient Safety Agency [32] were followed when designing the study.

Participants will have a minimum of 24 hours to decide whether to take part in the study and the research team will ensure that 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 Act [34] and data will be 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 People [36] and Involve [37], the study documentation was reviewed by a lay representative volunteer at the research centre hosting the study. The research 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' quality of life and experience across the disease trajectory should promote complementarity [38] and deepen interpretations from the study. Yardley [39] asserts 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; 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, in particular 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' quality of life 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 both 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] Idiopathic pulmonary fibrosis is, as previously discussed, an under-researched disease. We anticipate that the results of this study will provide fundamental information considering the experiences and needs of individuals and their carers, both quantitatively and qualitatively, and will therefore be disseminated via relevant clinical and research journals and international conferences, encompassing both palliative care and respiratory specialities. The Chief Investigator and three of the Co-Investigators are Consultant Clinicians in palliative medicine and respiratory specialities, which will enable the planning and provision of appropriate palliative care services for both 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 idiopathic pulmonary fibrosis, with discussion of the limited previous research undertaken in this area in terms of quality of life 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 both 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.

Acknowledgment:

We wish to acknowledge the clinical teams who granted access and will support recruitment.

Competing interests:

None

Authors' contributions:

AB is the Chief Investigator, designed the study and reviewed the manuscript. CS contributed to the protocol, reviewed the manuscript and will collect and analyse data. JB prepared the manuscript. KH, BHG and RH are site Principal Investigators. GG designed the study. AN designed the study and reviewed the manuscript.

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Full title:

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

Short title:

Care needs of individuals with idiopathic pulmonary fibrosis and their carers

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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 idiopathic pulmonary fibrosis. 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.

KEYWORDS

Idiopathic pulmonary fibrosis, carer, mixed-methods, Interpretative Phenomenological Analysis

WORD COUNT

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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-method study is proposed to explore the experiences and needs of individuals and their carers across the illness trajectory of idiopathic pulmonary fibrosis.

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 both 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 differences stages of the disease, and their relatives, will be included;
- This multi-centre study, in both England and Wales, will also adopt a mixed-methods approach, including qualitative interviews and the use of validated questionnaires.

INTRODUCTION

Idiopathic pulmonary fibrosis (IPF) is a progressive, life-limiting condition characterised by chronic inflammation and scarring,[1] 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 United Kingdom (UK) found that individuals lived with the disease for a median of three 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 per 100,000, with more men and older people affected.[5] While anti-inflammatory, immunosuppressant and anti-fibrotic medications are prescribed for IPF;[1] no pharmacologic 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 (NICE) in the UK.[8] Palliative care is defined by the World Health Organisation [9] 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 idiopathic pulmonary fibrosis, 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. The search identified several studies considering quality of life for individuals with IPF, but fewer studies used a qualitative approach to explore their experiences of the disease. A dearth of studies focussing on the experiences of carers/ family members is also noted.

Quality of life and IPF

The World Health Organisation [10] define quality of life 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 quality of life 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 quality of life of individuals with IPF. After a methodical literature search, Swigris et al. [11] identified only seven studies that assessed quality of life in a total of 512 adults with IPF. The included studies, which used the Medical Outcomes Study Short-Form 36-item tool (SF-36), World Health Organisation Quality of Life 100-item tool (WHO-QOL) or the St George's

Respiratory Questionnaire (SGRQ), were conducted in Japan, the Netherlands, Brazil and the United States of America (USA).[11] Health-related quality of life 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 quality of life.[11]

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

The small number of studies assessing the quality of life of people with IPF thus highlights significantly reduced outcomes, in particular in terms of physical health and sleep quality. However, no studies were identified that quantitatively measured relatives' or carers' quality

of life when caring for an individual with this progressive, terminal disease. Nor have they explored in detail the experiences underlying quality of life deterioration or patient/carer perceptions of interventions which might alter outcomes of importance. Of particular interest given the variable trajectory of the disease, is the identification of triggers for supportive and palliative interventions.

Qualitative experiences of IPF

Three studies were identified that explore the experiences of individuals with IPF using qualitative methods; although none of the studies discuss their methodological or philosophical approaches. Additionally, the papers do not discuss 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 quality of life measure by comparing the findings of the study to commonly-used global or respiratory tools. Dyspnoea and coughing were found to be distressing and impaired quality of life, 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 quality of life 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 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 the 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]

More 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 do not explicate their needs while caring for an individual with IPF, while Schoenheit et al.'s [14] study included relatives but the authors make 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. Interrogating 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 both 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 (quality of life, anxiety and depression, and IPF symptoms) and in-depth interviews utilising Interpretive 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 proforma, which classifies individuals at different stages on the IPF trajectory and documents respiratory co-morbidities.

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

Table one: participant group characteristics

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

Participants will be purposively sampled [16] to represent the four categories above, based on their FVC scores contained in their clinical notes, e.g. limited progressive, limited stable, extensive progressive, extensive stable. Congruent with the recommendations for Interpretative Phenomenological Analysis (IPA), the sample size for each group will be 6-10

individuals with IPF and 6-10 carers per homogenous 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 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.

Data collection

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

Co-morbidities and demographic data Demographic variables (age, marital status, location) and co-morbidities (in particular chronic obstructive pulmonary disease, pulmonary hypertension and lung cancer) of the 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 quality of life, anxiety and depression and symptoms of IPF. These questionnaires will enable the research team to observe whether quality of life, anxiety and depression change over time and how these correlate with dyspnoea and coughing.

1) Quality of life (QOL): a validated, global health-related QOL tool will be used to evaluate quality of life 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 quality of life 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 (HADS) 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 review [21] found that dyspnoea assessment scales have not been validated for use in palliative care but 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-tem self-completion tool measuring physical, psychological and social quality of life in relation to living with a chronic cough, which demonstrated high specificity and sensitivity.[22] The Cough Symptom Score [23] 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 minutes, and this will occur before the interview to minimise the influence of topics discussed on questionnaire completion.

In-depth interviews IPA is ~~as~~ a qualitative psychological approach used to explore how people make sense of major events in their lives.[17] Three philosophical approaches influence IPA:[17] 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 patients [24] and healthcare professionals.[25]

To enable access to detailed personal accounts of how participants experience IPF [17], the research team will utilise semi-structured interviews with people with the disease and their carers. The interviews will be conducted at a place and time convenient for the participants, either 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 minutes, 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 participants' consent, interviews will be audio-recorded and transcribed verbatim.

An interview schedule will be used (see Table 2) while also enabling 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.

Table two: interview schedules

Individuals with idiopathic pulmonary fibrosis
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, 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 might need? Is there anything else you've 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've thought of that you would like to mention or discuss now?

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% confidence interval (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 super-ordinate themes and themes within.

The four different groups of participants will be analysed separately with comparison 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 member second 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 study was approved by the university and ~~regional~~ 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. The Research Governance Frameworks

for England and Wales [30,31] and guidelines from the National Patient Safety Agency [32] were followed when designing the study.

Participants will have a minimum of 24 hours to decide whether to take part in the study and the research team will ensure that 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 Act [34] and data will be 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 People [36] and Involve [37], the study documentation was reviewed by a lay representative volunteer at the research centre hosting the study. The research 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' quality of life and experience across the disease trajectory should promote complementarity [38] and deepen interpretations from the study. Yardley [39] asserts 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; 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, in particular 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' quality of life 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 both 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] Idiopathic pulmonary fibrosis is, as previously discussed, an under-researched disease. We anticipate that the results of this study will provide fundamental information considering the experiences and needs of individuals and their carers, both quantitatively and qualitatively, and will therefore be disseminated via relevant clinical and research journals and international conferences, encompassing both palliative care and respiratory specialities. The Chief Investigator and three of the Co-Investigators are Consultant Clinicians in palliative medicine and respiratory specialities, which will enable the planning and provision of appropriate palliative care services for both 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 idiopathic pulmonary fibrosis, with discussion of the limited previous research undertaken in this area in terms of quality of life 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 both 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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Competing interests:

None

Authors' contributions:

AB is the Chief Investigator, designed the study and reviewed the manuscript. CS contributed to the protocol, reviewed the manuscript and will collect and analyse data. JB prepared the manuscript. KH, BHG and RH are site Principal Investigators. GG designed the study. AN designed the study and reviewed the manuscript.

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