An exploratory aerodigestive swallowing study in the chronic respiratory disease,

Idiopathic Pulmonary Fibrosis.

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Abstract

Introduction: Dysphagia occurs in various respiratory conditions, leading to an elevated risk of pulmonary complications due to aspiration. In Idiopathic Pulmonary Fibrosis (IPF), reflux-associated aspiration and dysregulated lung microbiome are implicated in the disease pathophysiology, but there is limited research on swallowing dysfunction in IPF.

Aim: The overall aim of this research project is to explore swallowing in patients diagnosed with IPF including perceived changes in their ability to eat and drink.

Methods and results: Four studies were conducted using different research designs. The first study aimed to describe the perception of swallowing and oropharyngeal swallowing physiology and safety using the ten-item Eating Assessment Tool (EAT-10) and the Videofluoroscopy Swallow Study (VFSS) (n:10). The results showed an increased self-reporting of dysphagia symptoms and a range of swallowing dysfunction, including aspiration into an unprotected airway. The second study assessed swallowing safety and performance using the Water Swallow Test (WST) (n:33). The study identified signs of penetration and/or aspiration and indicated that swallow performance may be influenced by gender and age. IPF patients demonstrated lower swallow performance compared with age- and sex-matched individuals without dysphagia. The WST can be conducted online effectively in community and hospital clinics. The third study described IPF patients' perceptions of swallowing dysfunction, laryngopharyngeal reflux and laryngeal hypersensitivity symptoms using three validated questionnaires: EAT-10, Reflux Symptoms Index (RSI), and Newcastle Laryngeal Hypersensitivity Questionnaire (LHQ) (n:40). The study revealed that a considerable proportion of patients experience symptoms like swallowing difficulties, laryngopharyngeal reflux and discomfort in the larynx. The fourth study explored the lived eating and drinking experience for IPF patients using qualitative interviews (n:14). The interviews highlighted those patients who experienced eating and drinking changes impacting them physically, emotionally and socially. They employed various coping strategies and expressed a strong desire for knowledge about these changes in their lived experiences.

Conclusion: Swallowing dysfunction is a prevalent issue among patients diagnosed with IPF.

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Declaration

This thesis presents my original research work, which includes online and hospital work, interviews, assessments, and analysis of the results. However, there are some exceptions to the above, which are outlined below:

For the study described in Chapter 3, Dr. Rhys Jones designed the study and was responsible for participant recruitment. Additionally, a certified speech and language therapist conducted the Videofluoroscopy Swallow Studies at Freeman Hospital. Prof. Patterson rated all the VFSS studies. I did the analysis, interpretation of the findings, and writing the chapter.

Dedication

This thesis is dedicated to:

My mother (Norah Alkhamees)

My husband (Turki Alhumaidan)

My daughters (Alhanoof & Sumou)

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List of Abbreviations

APF: Action for Pulmonary Fibrosis

ASHA: American Speech-Language-Hearing Association

ATAQ-IPF: The Tool to Assess Quality of life in IPF

BLF: British Lung Foundation

BMI: Body Mass Index

CI: confidence interval

Cm: centimetre

COPD: Chronic Obstructive Pulmonary Disease

DA: Deglutition Apnoea

DLCO: Diffusing Capacity of the Lung for Carbon Monoxide

EAT-10: Eating Assessment Tool

EX/EX: expiration during both pre- and post-swallow phases

EX/IN: expiration during the pre-swallow phase and inspiration during the post-swallow phase

FEES Flexible Endoscopic Evaluation of Swallowing

FVC: Forced Vital Capacity

FHS: Functional Health Status

GORD: Gastro Oesophageal Reflux Disease

HR: Heart Rates

HRCT: High-Resolution Computed Tomography

HR-QoL: Health-Related Quality of Life

ILD: Interstitial Lung Disease

IN/EX: inspiration during the pre-swallow phase and expiration during the post-swallow phase

IN/IN: inspiration during both pre- and post-swallow phases

K-BILD: The King's Brief Interstitial Lung Disease

LLN: Lower Level of Normal

LPR: Laryngeal Pharyngeal Reflux

LTOT: Long-Term Oxygen Therapy

MBSImP: Modified Barium Swallow Study Impairment Profile

MBSS: Modified Barium Swallow Study

MDT: Multi-*Disciplinary* Team

MRC: Medical Research Council dyspnoea scale

Newcastle LHQ: Newcastle Laryngeal Hypersensitivity Questionnaire

NHS: National Health Services

NICE: National Institute for Health and Care Excellence

OSA: Obstructive Sleep Apnoea

PAS: Penetration Aspiration Scale

PIS: Patient Information Sheet

PROs: Patient-Reported Outcomes

RA: Rheumatoid Arthritis

RC: Respiratory Care

RCSLT: Royal College of Speech and Language Therapists

RR: Respiratory Rate

RSI: Reflux Symptom Index

RT: Respiratory Therapist

RVI: Royal Victoria infirmary

SA: Swallowing Apnoea

SAD: Swallow Apnoea Duration

SGRQ-I: The St Georges' Respiratory Questionnaire in IPF

SLT: Speech and Language Therapist

SPO_2: peripheral capillary oxygen saturation

TLCO: Total Lung Capacity

Tsp: teaspoon

UES: Upper Oesophageal Sphincter

UIP: Usual Interstitial Pneumonia

ULN: Upper Level of Normal

VFSS: Videofluoroscopy Swallow Study

WST: Water Swallow Test

Wv: weight per volume

Publications and presentations during my PhD project

Publications (papers) directly related to my PhD project:

1. Manuscript publication, November 2022: **Alamer A**, Jones R, Drinnan M, Simpson AJ, Griffin M, Patterson JM, Althuwaybi A, Ward C, Forrest IA. **Oropharyngeal swallowing physiology and safety in patients with Idiopathic Pulmonary Fibrosis: a consecutive descriptive case series**. BMC Pulm Med. 2022 Nov 17;22(1):422. doi: 10.1186/s12890-022-02232-3. PMID: 36384569; PMCID: PMC9670476.

2. Manuscript publication, March 2024: **Alamer AA**, Ward C, Forrest I, et al. Eating and drinking experience in patients with idiopathic pulmonary fibrosis: a qualitative study. BMJ Open 2024;14:e078608. doi:10.1136/ bmjopen-2023-078608.

Publications (papers) during my PhD project not directly related to my PhD project:

1. Brennan, M., McDonnell, M.J., Ward, C., **Alamer**, A. Duignan, N. & Rutherford, R.M. (2020). **Bronchiectasis in the Elderly—A Disease That Has Not Gone Away**. *Curr Geri Rep* **9**, 19–29. https://doi.org/10.1007/s13670-020-00315-9.

2. Althuwaybi, A., **Alamer, A.**, McDonnell, M.J., Brennan, M., Rutherford, R.M., Wilcox, M.D., Chater, P.I., Pearson, J.P., & Ward, C. (2021). **A narrative review of the potential role of microaspiration and a dysregulated aerodigestive microbiome in lung disease**. *Annals of Esophagus*. Annals of Esophagus; Vol 6 (March 25, 2023): Annals of Esophagus 2021.

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2. Poster presentation, Newcastle University Translational and clinical research institute Live (NUTCRI), June 2021: **Oropharyngeal swallowing pathophysiology in patients with idiopathic pulmonary fibrosis: A consecutive descriptive case series.**
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4. Poster presentation, The European Respiratory Society (ERS) International Congress, September 2023 (Milan): **"The eating and drinking experience in patients with IPF: a qualitative study". Amal Alamer**, Chris Ward, Ian Forrest, Michael Drinnan, Joanne M Patterson.

Chapter 1. Introduction, contextual background and thesis overview

1.1 Introduction

This chapter is an introductory chapter to my thesis. The introduction consists of three main sections. Firstly, the dynamics of swallowing physiology and coordination between breathing and swallowing. Secondly, dysphagia and assessment of the swallowing function. The last part focuses on the definition of Idiopathic Pulmonary Fibrosis (IPF), a description of it and swallowing in patients with IPF. The chapter concludes with an explanation of the purpose of the thesis and an overview of its contents.

1.2 Normal swallowing

Nutrition and hydration are crucial for sustaining good health, overall well-being and the preservation of life. The acts of eating and drinking are routine activities that we often take for granted. A normal swallow is a complex biomechanical process coordinated with breathing so that the airway remains protected when food, liquid or secretions pass through the pharynx. The swallowing process barely takes a second and involves a complex coordination of sensory-motor functions where more than 25 different muscles, mostly paired, in the mouth, pharynx, larynx and oesophagus, and six different cranial nerves are engaged in the act of swallowing (Jean, 2001). On average, healthy adults swallow 580 times a day in order to eat, drink and manage saliva (Matsuo and Palmer, 2008). Swallowing serves an important role not only in facilitating the intake of nutrients, but also in effectively managing the internal secretions produced by the upper and lower parts of the digestive system. These secretions include substances such as saliva and bile as well as nasal, tracheal and bronchial secretions (Speyer et al., 2022b). Swallowing is commonly subdivided into three consecutive phases, which are the oral, pharyngeal and oesophageal stages (Logemann, 1988).

1.2.1 The oral preparatory stage and oral stage

During the oral preparatory stage (voluntary stage) the liquids are taken into the mouth and retained either on the floor of the mouth or against the hard palate through upwards motion of the tongue. Solid food is ground down and mixed with saliva to form a soft aggregate ready for swallowing; this is known as a “bolus”. The bolus is shaped and sized using jaw rotatory and shearing forces. During the process of bolus formation, the posterior tongue is in contact with the soft palate, preventing the bolus from entering the pharynx before swallowing. Throughout this phase, the airways remain open. The duration of the oral preparatory stage can vary from two to 120 seconds, depending on the consistency of the bolus being prepared (Logemann, 1988; Matsuo and Palmer, 2008).

During the oral stage, the bolus is then transported to the oropharynx via the oral tongue. The tongue tip and blade squeeze upwards towards the hard palate, moving the bolus posteriorly. The lips and cheeks remain taut, to maintain oral pressure and prevent spillage anteriorly or loss of bolus to the cheeks. As the bolus reaches the back of the tongue, the soft palate is elevated to protect the nasopharynx from food entry and to close the airways superiorly (Logemann, 1988; Matsuo and Palmer, 2008). Respiration continues normally at this stage. Where the length of the oral preparatory stage is influenced by the consistency of the bolus, the oral stage should not exceed one second in duration (Logemann et al., 2008).

1.2.2 The pharyngeal stage

During the pharyngeal stage (involuntary stage), which averages one second, the soft palate remains elevated and contracted to prevent bolus regurgitation. The retraction of the base of the tongue is the major source of generating pressure. The pharyngeal wall muscles contract sequentially from the top to the bottom, squeezing the bolus downwards. The pharynx also shortens vertically to reduce the volume of the pharyngeal cavity. The larynx is pulled up, with the epiglottis flipping over to cover the entry of the respiratory passage. The upwards and anterior movement of the hyoid bone subsequently moves the epiglottis into a horizontal position, aiding airway protection. This anterior movement moves the larynx upwards and forwards, away from the direction of bolus flow. The cricopharyngeus relaxes and the upper oesophageal sphincter is opened by the combined action of the anterior and upwards laryngeal elevation, and passively by pressure from the arriving bolus (Logemann,

1988; Matsuo and Palmer, 2008; Im *et al.*, 2012; Ghannouchi *et al.*, 2016a). During this time, breathing temporarily pauses as a result of the protective mechanisms of the pharyngeal stage. The urge to swallow takes precedence over the urge to breathe, providing an additional layer of airway protection during the period of swallow apnoea (Matsuo and Palmer, 2008; Ghannouchi *et al.*, 2016a).

1.2.3 The oesophageal stage

During the oesophageal phase (involuntary phase) the lower oesophageal sphincter (LES) relaxes to allow the bolus to move to the stomach (Matsuo and Palmer, 2008; Ghannouchi *et al.*, 2016a). Oesophageal swallowing dysfunction is beyond the scope of this thesis and therefore not further described here.

1.2.4 Breathing and swallowing coordination

Respiration and swallowing are physiological processes that are closely interrelated in their neural control and demonstrate a highly coordinated partnership in their roles in basic survival. Breathing and swallowing are physiologically linked to maintain adequate gas exchange and hydration and prevent penetration and/or aspiration during swallowing (Martin-Harris et al., 2005). The coordination between breathing and swallowing interactions have been extensively established in scientific research. Breathing and swallowing do not occur simultaneously, as the act of swallowing briefly interrupts the normal respiratory cycle (Martin-Harris et al., 2003; Martin-Harris et al., 2005).

Swallowing is associated with a brief cessation of respiration, called swallowing apnoea (SA) or deglutition apnoea (DA) (Martin-Harris, 2006). Durations of swallow apnoea ranged from 0.50 second to 10.02 seconds, with extreme outliers in the older subjects (Martin-Harris et al., 2005). SA onset and duration vary based on gender, age and bolus viscosity differences (Hiss et al., 2004).

A normal swallow usually occurs in the beginning or middle of an exhalation at mid to low lung volume (Dozier *et al.*, 2006; Hopkins-Rossabi *et al.*, 2019; Curtis;Dakin and Troche, 2020). The timing of the SA onset is vital for the safe passage of the bolus, as the pharynx is the shared route for both food and air. Early and/or delayed SA onset during oropharyngeal swallowing predisposes the patient to laryngeal penetration or aspiration (Hiss et al., 2004; Hopkins-Rossabi et al., 2019).

1.2.5 Effect of age and normal swallowing

Successful ageing is a process of lifelong change and adaptation. It is well known that with advancing age the body experiences some physiological changes that may not be due to disease or illness but are due simply to the natural ageing process. Sarcopenia refers to an age-related, progressive and generalised condition affecting the skeletal muscle that leads to a loss of muscle strength and mass (Cruz-Jentoft et al., 2019). This loss of muscle can increase the risk of negative outcomes such as falls, fractures, physical disability and even mortality (Cruz-Jentoft et al., 2019; Cruz-Jentoft and Sayer, 2019). Studies indicate that skeletal muscle mass and strength may begin to decline as early as the fourth decade of life and that this decline occurs in a linear fashion (Walston, 2012). By the eighth decade of life, up to 50% of skeletal muscle mass may be lost (Walston, 2012). Sarcopenia may have severe consequences for older adults as it may result in a decline in strength and function, leading to negative health outcomes such as loss of function, disability and frailty (Xue et al., 2011; Dufour et al., 2013). Additionally, sarcopenia has been linked to various acute and chronic diseases, increased insulin resistance, fatigue, falls and even mortality. In particular, studies have found a strong association between sarcopenia and rheumatologic conditions, particularly rheumatoid arthritis (RA) in women (Walston, 2012).

Although the ageing process involves various changes in anatomy, physiology, psychology and function, which increase the likelihood of dysphagia in older adults, a healthy older adult's swallow is not inherently impaired (Humbert and Robbins, 2008). Presbyphagia is a term used to describe the natural changes in the swallowing mechanism that occur in healthy older adults as a result of ageing (McCoy and Varindani Desai, 2018). These changes can affect every stage of the swallowing process, leading to issues such as difficulty controlling and moving boluses, delays in initiating the pharyngeal swallow, ineffective pharyngeal clearance, impaired cricopharyngeal opening, and decreased effectiveness of secondary oesophageal peristalsis (Nogueira, 2015).

In the oral stage of swallowing, research has documented anatomical and physiological changes occurring as individuals age. According to a study by Logemann (1990), increasing age is associated with an increase in the fat-to-muscle ratio and connective tissue of the tongue, resulting in a reduction in tongue pressure. Furthermore, altered taste perception and decreased salivary flow (xerostomia) have been demonstrated to impair bolus control (Schiffman, 1993; Robbins;Bridges and

Taylor, 2006). In addition, older adults who have poor natural teeth often have difficulty preparing their food boluses, resulting in less control over the food lump in their mouth and longer chewing duration (Robbins;Bridges and Taylor, 2006).

Research by Cook et al. (1994) has shown that healthy older adults, with a mean age of 68 ±8 years, have a slightly but significantly longer oral transit time compared to younger adults, with a mean age of 28 ±7.5 years. The oral transit time was measured by the time required for the passage of the bolus through the mouth to the exit from the oral cavity. Specifically, the older group took 0.7 ±0.06 seconds for 5 ml of water and 0.6 ±0.02 seconds for 10 ml of water, while the younger group took 0.46 ±0.07 seconds for 5 ml of water and 0.44 ±0.06 seconds for 10 ml of water (Cook et al., 1994). This finding has been supported by more recent studies, including Leslie et al. (2005) and Yoshikawa et al. (2005).

In the pharyngeal stage of swallowing, previous studies have indicated that older adults tend to experience a delay in the onset of pharyngeal swallow during the swallowing process (Rosenbek et al., 1996; Dua et al., 1997; Logemann et al., 2000). A delayed pharyngeal swallow can be operationally defined as a time difference between the arrival of the bolus head at the posterior angle of the mandible and the onset of hyoid motion, where this onset of motion occurs later than expected (Logemann et al., 2000). Martin-Harris et al. (2007) study found that a delay in pharyngeal swallow was present in all age groups. However, healthy adults over the age of 50 years had a longer delay compared with younger adults. Specifically, the delay in pharyngeal swallow for older adults was approximately 220 milliseconds longer than that observed in younger adults (Martin-Harris et al., 2007).

Older adults experience a reduction in both range of motion and velocity of movement in the hyoid bone compared with younger adults (Logemann et al., 2000; Kang et al., 2010). Multiple suprahyoid muscles, including the geniohyoid muscle, work together to raise, protract and stabilise the hyoid bone during swallowing (Feng et al., 2013). The geniohyoid muscle connects the posterior aspect of the mandible in the midline to the anterior surface of the body of the hyoid bone and contracts to move the hyoid bone upwards and forwards, along with the mylohyoid, stylohyoid and anterior belly of the digastric muscles (Pearson Jr;Langmore and Zumwalt, 2011; Feng *et al.*, 2013). Age-related muscle loss or sarcopenia of these suprahyoid muscles may contribute to a

reduction in hyoid bone movement during ageing, leading to an increased risk of aspiration and/or penetration in older adults (Feng et al., 2013; Feng et al., 2014).

During the pharyngeal stage of swallowing, the retraction of the tongue base is one of the main driving forces that propels the bolus and is particularly crucial to ensure the smooth entry of the bolus into the pharynx (Feng;Zhang and Wang, 2023). Reduced retraction of the tongue base weakens its ability to clear food from the pharynx, which, in turn, leads to increased pharyngeal residue (Hosseini et al., 2019; Mehraban-Far et al., 2021).

With ageing, the amount of muscle fibres in the upper oesophageal sphincter (UES) decreases, and the impulses responsible for maintaining tension in the cricopharyngeal muscle also decline gradually (Shaker and Lang, 1994). The presence of a properly functioning UES is essential for performing a sufficient pharyngeal swallow and to avoid the backflow of oesophagus contents into the pharynx, known as esophagopharyngeal reflux (Nishikubo et al., 2015). According to Nishikubo et al. (ibid), inadequate reduction in UES pressure during swallowing, which is characterised by more than 5 mmHg of residual pressure, was found in 15.4% of the early elderly group (aged 60–69) and 30.4% of the late elderly group (aged 70–83), as measured by pharyngeal high-resolution manometry (Nishikubo et al., 2015). This indicates a lack of relaxation of the UES and delayed opening time, increasing the risk of pharyngeal residual and aspiration (Nishikubo *et al.*, 2015; Feng;Zhang and Wang, 2023). This observation is consistent with previous studies (Dejaeger *et al.*, 1994; Leonard;Kendall and McKenzie, 2004).

From a functional point of view, it has been observed that as individuals age, there is a general decline in both the mass and function of respiratory muscles among normal, healthy older adults (Roman;Rossiter and Casaburi, 2016). Previous research has indicated that with increasing age, the stiffness of the diaphragm and intercostal muscles tends to increase, leading to a decrease in chest wall compliance and elasticity (Kelly;McCarter and Barnwell, 1993; Kim and Sapienza, 2005). This alteration in the shape of the thoracic region results in a reduction in chest wall compliance, as evidenced by changes in spinal curvature, increased sternal curvature and thinning of chest wall muscles (Hochhegger et al., 2012). Consequently, the respiratory muscles have to exert more effort to sustain breathing. The considerable decline in pulmonary function and reduced chest wall compliance among older adults impairs their ability to

effectively clear respiratory residues, thereby increasing the risk of food entering the respiratory tract.

According to the study conducted by Hiss et al. (2004), it was found that older adults tend to initiate SA earlier in comparison with young adults. This observation indicates a potential decrease in neural reserves related to timing among older individuals. It is plausible to suggest that older adults may compensate for the slowed sequencing of swallowing events by initiating SA earlier (Hiss et al., 2004). This proactive approach ensures coordination between respiration and swallowing. The earlier onset of SA is likely a compensatory mechanism to counteract the neural and structural changes associated with ageing (Hiss et al., 2004). Furthermore, Leslie et al. (2005) found that older adults had a greater duration of swallow apnoea duration (SAD) compared with younger adults. They observed a significant positive correlation between age and the length of swallow apnoea. The mean duration of SAD for 5 ml water boluses was 0.74 seconds (±0.17) with a Pearson's correlation coefficient (r) of 0.433 and a p-value of 0.002. Similarly, for 5 ml yogurt boluses, the mean SAD duration was 0.73 seconds (±0.19) with a Pearson's correlation coefficient (r) of 0.367 and a p-value of 0.0232.

The extended duration of SAD in older adults may serve as a compensatory mechanism for various age-related changes such as longer transit times in the oropharynx and hypopharynx and delayed initiation of hyolaryngeal excursion (Robbins et al., 1992; Yoshikawa et al., 2005). One crucial mechanism to safeguard the airway against foreign material aspiration is vocal cord closure. It is essential to maintain airway protection while the bolus is moving through the pharynx, which means the time it takes for the bolus to pass should be shorter than the time required for vocal cord adduction (Ren et al., 1993). Reducing the safety margin between these two events increases the risk of aspiration (Leslie et al., 2005). This observation has also been documented in other studies (Hiss;Treole and Stuart, 2001; Wang *et al.*, 2015).

The respiratory pattern before and after swallowing was categorised into four distinct respiratory phase patterns: expiration during both pre- and post-swallow phases (EX/EX), expiration during the pre-swallow phase and inspiration during the post-swallow phase (EX/IN), inspiration during the pre-swallow phase and expiration during the post-swallow phase (IN/EX), and inspiration during both pre- and post-swallow phases (IN/IN) (Martin-Harris et al., 2005). The EX/EX pattern was identified as the

most prevalent among the four respiratory phase patterns and the remaining three patterns were considered less dominant (Hopkins-Rossabi et al., 2019). However, the frequency of the EX/EX pattern can vary depending on various factors, including bolus volume, consistency, gender and age (Hopkins-Rossabi et al., 2019). In the study conducted by Martin-Harris et al. (2005), it was observed that older adults were more likely to show non-dominant respiratory patterns such as IN/IN and EX/IN in comparison with younger adults. These patterns were captured using a nasal airflow sensor. The mean age for individuals exhibiting the predominant EX/EX pattern was 56 (±23) years. On the other hand, for the non-predominant patterns IN/IN and EX/IN, the mean age was 68 (±21) years (Martin-Harris et al., 2005). This finding has been further supported by a more recent study (Wang et al., 2015).

1.3 Oropharyngeal dysphagia

1.3.1 Definition

Dysphagia is not considered a specific disease diagnosis but rather a diagnosis based on symptoms, which can be caused by various underlying conditions. Dysphagia is commonly defined as the difficulty experienced in moving saliva, food and liquids from the mouth to the stomach (Logemann, 1998). Patients with dysphagia may encounter challenges in one or more aspects of the anatomical or physiological components involved in the oral, pharyngeal or oesophageal stages of swallowing (Logemann, 1995). Swallowing disorders can be found across all age groups and may manifest either suddenly or progressively deteriorate over time (Lazarus and Logemann, 1987).

Research indicates that around 8% of the global population is affected by dysphagia (Smith;Bryant and Hemsley, 2023). Moreover, the prevalence of dysphagia tends to be higher in specific populations. For instance, among older individuals residing in aged care facilities, the estimated prevalence of dysphagia is approximately 52.7% (Engh and Speyer, 2022).

Dysphagia frequently develops in patients with cerebral palsy or intellectual disability, stroke, Parkinson's or motor neurone disease, and following treatment for head and neck cancer (Dozier et al., 2006; Malhi, 2016).

Ghannouchi et al. (2016) conducted a systematic review exploring the swallowing function of patients with chronic respiratory diseases. Their study included 26 research

papers, with 11 focusing on obstructive sleep apnoea (OSA) and 15 on chronic obstructive pulmonary disease (COPD). The results of their review revealed that both OSA and COPD contribute to a higher occurrence of oropharyngeal dysphagia. These findings indicate that there is an increasing body of literature investigating the connection between chronic respiratory diseases and swallowing (Ghannouchi *et al.*, 2016a).

Common signs and symptoms of dysphagia include challenges with swallowing, coughing before, during or after swallowing, experiencing pain or discomfort during swallowing, regurgitation of food or liquids, recurring respiratory infections, the presence of a gurgly or wet voice, and unintentional weight loss (Lind, 2003).

1.3.2 Complications of dysphagia

The consequences of oropharyngeal dysphagia can be severe. Aspiration pneumonia, malnutrition and dehydration are frequently observed as the most common complications associated with dysphagia (Schindler;Ginocchio and Ruoppolo, 2008).

Impairment in swallowing efficacy can result in an imbalanced oral intake of food and liquids, which may lead to malnutrition and/or dehydration. Malnutrition is characterised as a clinical condition resulting from an imbalance of energy, protein and other nutrients, leading to measurable adverse effects on body composition, physical function and clinical outcomes (Tagliaferri et al., 2019).

The consequences of malnutrition extend to healthcare costs and health outcomes, including prolonged hospital stays, an increased susceptibility to infections, hindered recovery and higher mortality rates (Norman et al., 2008). The Council of Europe addressed a significant prevalence of undernutrition in hospitalised patients, emphasising dysphagia as a prominent factor contributing to malnutrition (Carrión et al., 2015).

A study conducted by Silvia Carrión et al. (2015) examined 1,662 consecutively hospitalised patients aged ≥70 years with acute diseases. The study assessed dysphagia using the volume-viscosity swallow test and nutritional status using the Mini Nutritional Assessment®. The findings revealed a prevalence of oropharyngeal dysphagia of 68.4% (95% CI 63.3–73.4) among patients with malnutrition, 41.7% (95%

CI 37.7–54.6) among patients at risk for malnutrition and only 14.7% (95% CI 8.9–20.5) among well-nourished older patients ($p < 0.001$).

In a study conducted by Susan Langmore et al. (1998), it was found that among patients who developed aspiration pneumonia, with an average age of 71 years, 81% of them had oropharyngeal dysphagia. The assessment of dysphagia was conducted using fluoroscopic swallow examination and *Flexible* Endoscopic Evaluation of Swallowing (FEES). The results showed that 58% of the patients aspirated liquid, 27% aspirated food and 50% aspirated secretions (Langmore et al., 1998). Aspiration pneumonia is one of the most serious complications related to swallowing dysfunction (Carrión et al., 2019).

1.3.3 Assessment of swallowing function

Swallowing is a complex process influenced by multiple factors, necessitating a comprehensive assessment approach that considers various dimensions to gain a complete understanding of the nature and severity of dysphagia. The identification of dysphagia can be established by utilising functional health status questionnaires to gather patient-reported swallowing data, or by conducting clinical screenings and assessments, as well as instrumental evaluations to examine objective swallowing characteristics.

1.3.4 Screening

Screening is widely acknowledged as the initial step in the management of dysphagia, as it serves the purpose of identifying patients who may be at risk of experiencing swallowing difficulties. The ideal screening tool should be efficient, non-invasive and reliable. The literature describes various types of screening methods, such as conducting trial swallows with different volumes of water or varying viscosities. Other screening approaches involve combining trial swallows with pulse oximetry, evaluating clinical signs (such as abnormal gag reflex, voice changes or voluntary cough), utilising cervical auscultation or considering relevant aspects of the patient's medical history (such as recurrent episodes of pneumonia or cough tests). Once patients are identified as being at risk, they should be referred for a comprehensive swallowing assessment (Kertscher et al., 2014; Speyer et al., 2022b). The details of swallow screening will be covered in Chapter 4.

1.3.4.1 Clinical assessment

Following the screening process, when a patient has been identified as being at risk for dysphagia, further assessment is necessary. A non-instrumental clinical assessment, performed by a dysphagia expert, is the subsequent step after screening. This clinical evaluation of swallowing is a subjective assessment that aims to identify potential causes of swallowing disorders, evaluate the possibility of aspiration and/or penetration, and determine the need for additional diagnostic tests, This evaluation is referred to as a “bedside examination” (Maccarini et al., 2007; Speyer et al., 2022b).

The clinical assessment of dysphagia encompasses multiple aspects, including a thorough review of the patient's medical history, conducting a physical examination, and gathering subjective information about the swallowing problem through patient complaints and descriptions. Expert clinicians also make clinical observations during the interview and examination process (Karkos et al., 2009; Scharitzer et al., 2017).

The comprehensive assessment of dysphagia entails evaluating various aspects, such as oral motor function, swallowing coordination, sensation, and the presence of dysphagia signs or symptoms (McAllister et al., 2016). This assessment involves observing the patient's oral intake, assessing cranial nerve function, evaluating swallowing manoeuvres and considering these findings to make decisions regarding oral feeding or alternative feeding routes and plans for rehabilitation (Maccarini et al., 2007; McAllister et al., 2016). Additionally, clinical indicators such as coughing, choking or voice changes that occur during swallowing are examined to further inform the assessment process (Speyer et al., 2022b).

1.3.4.2 Patient-reported outcome measures

Patient-reported outcome measures (PROMs) are frequently used to capture a patient's personal perspective or understanding of a medical condition and to assess the impact of treatment. These measures help evaluate the effectiveness of interventions based on the patient's subjective experience.

The assessment of dysphagia through self-evaluation encompasses two main components: functional health status (FHS) and health-related quality of life (HR-QoL). FHS focuses on the impact of the disease on specific functional aspects, while HR-QoL considers an individual's subjective perception of their health, considering social, functional and psychological factors (Speyer et al., 2022a). Despite being distinct

concepts, many dysphagia questionnaires combine FHS and HR-QoL to provide a comprehensive assessment of the condition's impact on individuals. However, this integration poses challenges in interpretation and makes it difficult to differentiate disease-related functioning from the patient's experience of disease-related quality of life (Speyer et al., 2022a; Speyer et al., 2022b).

Since the introduction of the first dysphagia-related PROMs in 1987, there has been a significant increase in their number (Andersen et al., 1987). This growth reflects the recognition of their importance in both clinical and research settings, highlighting the need for comprehensive assessments of dysphagia and its impact on individuals (Patel et al., 2017).

These measures include both disease-specific instruments targeting conditions such as achalasia, post-operative dysphagia from spine surgery, Parkinson's disease, multiple sclerosis, eosinophilic esophagitis or malignancy, as well as broader measures designed to assess a diverse range of individuals affected by dysphagia (Patel et al., 2017).

The Eating Assessment Tool (EAT-10), a validated questionnaire for self-reported dysphagia, is a widely used measure and was used in this thesis (see Chapters 3 and 5, also Appendix 7).

More details about PROMs are covered in Chapter 5.

1.3.4.3 Instrumental evaluation

The two most common instrumental diagnostic swallowing procedures available are FEES and the Videofluoroscopy Swallow Study (VFSS). They are considered the "gold standard" for objectively evaluating aspiration, including silent aspiration and other physiological issues during the pharyngeal phase of swallowing. These procedures have been validated and supported by evidence-based guidelines published by the American Speech-Language-Hearing Association and the Royal College of Speech and Language Therapists (RCSLT) (American Speech-Language-Hearing Association, 2004; Royal College of Speech Language Therapists, 2013; Wallace et al., 2020; Langmore et al., 2022).

The FEES, initially developed by Susan Langmore in 1986, involves the insertion of a thin and flexible nasoscope into the nasal passageway, which is directed towards the

laryngopharynx (Langmore;Kenneth and Olsen, 1988). Equipped with a light source, camera, monitor and recording device, the nasoscope allows for the visualisation of various aspects such as secretions, sensation, surface anatomy, mucosal abnormalities, bolus flow, airway protection, glottic closure and bolus location within the hypopharynx (Langmore;Kenneth and Olsen, 1988; Langmore, 2006). This examination method offers several advantages, including good patient tolerance, absence of radiation, repeatability, cost-effectiveness and use of mobile equipment for bedridden patients (Mozzanica et al., 2019). However, some patients may experience discomfort, gagging or vomiting during the procedure, with a low risk of epistaxis, reflex syncope, mucosal perforation, laryngospasm and vasovagal response (Nacci et al., 2016). However, FEES does have some limitations, including a "white out" period, inability to assess the oral phase of swallowing, inability to screen the oesophagus, discomfort associated with nasoscopy and time-consuming disinfection processes (Nacci et al., 2008).

The VFSS, also referred to as a Modified Barium Swallow Study (MBSS), is a radiographic examination that offers real-time visualisation of the functioning of the oral cavity, pharynx and upper oesophagus (Martin-Harris and Jones, 2008). The VFSS allows for the detection and assessment of aspiration, including its timing. It also helps clinicians to identify the physiological causes of aspiration, which can often be treated (Dodds;Logemann and Stewart, 1990). Moreover, the VFSS enables the observation of how swallowing physiology is influenced by various factors such as bolus volumes, textures and compensatory strategies (Martin-Harris and Jones, 2008). The VFSS typically uses x-rays to capture images or videos while administrating different bolus volumes and constituents (Belafsky and Kuhn, 2014). The VFSS was used in this thesis (Chapter 3). Further details about the VFSS are also covered in Chapter 3.

1.4 Interstitial lung diseases

1.4.1 Background

Interstitial lung disease (ILD) is an umbrella term for a large family of related diseases, though not all of them exclusively target the interstitium of the lungs. ILDs are characterised by inflammation or fibrosis occurring in the interstitial space of the lungs. This leads to impaired gas exchange, resulting in breathlessness, and in severe cases, respiratory failure and even death (Wijsenbeek;Suzuki and Maher, 2022).

There are over 200 different disorders that can cause ILD. These disorders range from extremely rare conditions like lymphangioleiomyomatosis to systemic conditions like systemic sclerosis or RA, where ILD is a common manifestation, to more prevalent diseases like IPF, which accounts for 1% of all deaths in the UK (Harari *et al.*, 2018; Wijsenbeek;Suzuki and Maher, 2022).

Patients with ILD usually present with symptoms of breathlessness, coughing and limited daily activity, and this may be associated with depression and anxiety, resulting in impaired quality of life (Raghu and Brown, 2004; Wallis and Spinks, 2015).

There are different types of ILD, and the classification of this disease is based on the causes, such as idiopathic, autoimmune-related, exposure-related (including iatrogenic), ILDs characterised by cysts or airspace filling, sarcoidosis, and orphan diseases (acute eosinophilic pneumonia, chronic eosinophilic pneumonia and malignant diseases-associated ILDs) (Wijsenbeek;Suzuki and Maher, 2022). The onset of ILD can vary from a gradual to an acute and severe onset that poses a risk to life. Diagnosis and classification of these diseases rely on a combination of clinical, radiological and occasionally pathological information (Wallis and Spinks, 2015; Wijsenbeek;Suzuki and Maher, 2022).

1.4.2 Idiopathic pulmonary fibrosis

1.4.2.1 Definition

IPF is a chronic and progressive ILD characterised by unknown causes. It is primarily defined by the progressive scarring and fibrosis of lung tissue, leading to irreversible loss of pulmonary function (Raghu et al., 2022). Typical symptoms of IPF include chronic exertional dyspnoea (shortness of breath during physical activity) and a persistent dry cough that can persist for months to years (Lederer and Martinez, 2018). Risk factors for IPF include older age, male sex, genetic susceptibility, lung microbiome and a history of cigarette smoking (Zaman and Lee, 2018).

IPF is the most common type of fibrotic ILD. In the United Kingdom, its estimated incidence is between 5.3 and 7.3 cases per 100,000 individuals per year, while in the United States, rates using the same classification system have been reported as ranging from 31.1 to 93.7 cases per 100,000 individuals per year (Hutchinson et al., 2015). The mortality rates associated with IPF have been increasing over time. In the

UK, the rates rose from 4.6 cases per 100,000 individuals per year in 2001 to 9.4 cases per 100,000 individuals per year in 2011, while in the United States, a similarly increasing trend was observed between 2000 and 2010, with rates escalating from 4.4 to 5.3 cases per 100,000 individuals per year (Hutchinson et al., 2014). IPF is more frequently diagnosed in the sixth or seventh decade of life and is uncommon below the age of 50 years (Raghu et al., 2014). The median survival for IPF patients is approximately three to five years from the time of diagnosis if left untreated (Glass et al., 2022).

IPF is considered a rare disease, and its worldwide incidence and prevalence vary. The estimated adjusted worldwide incidence and prevalence of IPF fall within the range of 0.09–1.30 and 0.33–4.51 per 10,000 individuals, respectively (Maher et al., 2021).

1.4.2.2 Clinical presentation

The initial clinical presentation of IPF is non-specific and includes symptoms such as exertional dyspnoea, dry cough, fatigue and a gradual decline in the ability to perform daily activities (Sgalla;Biffi and Richeldi, 2016). One of the early physical examination findings is the presence of bibasilar inspiratory "velcro-type" crackles on chest auscultation (Cottin and Cordier, 2012). Finger clubbing and hypoxemia at rest or during exertion are common physical findings (Sgalla;Biffi and Richeldi, 2016). As the disease progresses to its advanced stages, cyanosis and signs of right ventricular failure may develop. Ultimately, respiratory failure can occur, leading to death in these patients (King;Pardo and Selman, 2011).

1.4.2.3 Diagnosis

Pulmonary function tests are a non-invasive method in assessing the severity of IPF and monitoring the progression of the disease. In patients with suspected IPF, the results of pulmonary function tests show a restrictive pattern, characterised by decreased lung function parameters such as forced vital capacity (FVC), total lung capacity (TLC) and diffusing capacity of the lung for carbon monoxide (DLCO) (Lederer and Martinez, 2018). It is important to note that in the early stages of IPF, lung function parameters may appear normal or only minimally impaired (Alsomali et al., 2023). The degree of FVC and DLCO reduction correlates directly with the reduction in IPF survival rate. However, relying solely on baseline lung function as a predictor of mortality in IPF is not effective. Instead, composite scoring systems such as the Gender-Age-

Physiology (GAP) index provide better prognostic accuracy in assessing the progression and potential outcomes of IPF (Kolb and Collard, 2014).

Diagnosis is preferably made by specialists within a multi-*disciplinary* team MDT setting and access to high-resolution computed tomography (HRCT) imaging. HRCT is considered the gold standard for diagnosing IPF. HCRT of the lung findings in IPF is characterised by the radiographic pattern of usual interstitial pneumonia (UIP) (Raghu et al., 2022). The UIP pattern on radiology typically presents as subpleural shadowing with an apicobasal gradient, traction bronchiectasis, honeycombing appearance and traction bronchiolectasis. Ground-glass opacification and fine reticulation may also be observed in some cases (Raghu et al., 2018).

1.4.2.4 Management of IPF

The management of IPF presents significant challenges, primarily due to the incomplete comprehension of the disease's underlying pathological mechanisms. Furthermore, the presence of various comorbidities, such as cor pulmonale, pulmonary hypertension and pneumonia, adds to the complexity of managing IPF. According to the guidelines provided by the National Institute for Health and Care Excellence (NICE), pirfenidone and nintedanib are recommended anti-fibrotic medications for the treatment of IPF, but specifically for individuals whose FVC ranges from 50% to 80% of the predicted value (Fraser and Hoyles, 2016). These medications have shown efficacy in slowing down the decline of FVC in patients with IPF (Fraser and Hoyles, 2016). Additionally, they have been found to reduce the risk of acute exacerbations, which can occur at any stage of the disease (Alsomali et al., 2023).

In February 2023, the NICE guidelines were updated to expand the eligibility for nintedanib medication to include patients with an FVC above 80% of the predicted value. This change is expected to enhance treatment access for a broader population of individuals with IPF (NICE, 2023). It is acknowledged that the severity of symptoms or radiological findings at diagnosis may not necessarily correlate with the percentage predicted FVC (Arcadu et al., 2017). As a result, prescribing nintedanib should not be limited only to those with an FVC below 80% predicted.

IPF patients are provided with long-term oxygen therapy (LTOT), vaccinations, pulmonary rehabilitation and supportive care to assist them in managing daily activities and enhancing their quality of life. Lung transplantation is considered as a potential

treatment option for patients with advanced IPF who experience substantial decline in lung function and have a poor quality of life.

1.4.2.5 Swallowing and IPF

To date, swallowing and dysphagia has not been studied in IPF and this represents an important gap in knowledge. This may have translational significance in a pathophysiology where new treatments are very much needed. More details about swallowing in IPF is covered in the literature review section (3.2) in Chapter 3.

1.4.3 Key aims and objectives

The overall aim of this research project is to explore swallowing in patients diagnosed with IPF including perceived changes in their ability to eat and drink.

1.4.4 Objectives

1. Describe swallowing and oropharyngeal swallowing physiology and safety in patients with IPF using the gold standard instrumental assessment, the Videofluoroscopy Swallow Study (VFSS).
2. Assess swallowing safety and performance in patients with IPF using a simple screening Water Swallow Test (WST).
3. Measure symptoms of swallowing dysfunction, laryngeopharyngeal reflux and laryngeal hypersensitivity in IPF patients using validated questionnaires.
4. Explore the eating and drinking lived experience of patients with IPF using a qualitative research design.

1.4.5 Thesis overview

This thesis will follow a manuscript style for Chapters 3–6, as each chapter represents a different study. The study described in Chapter 3 was the first to be conducted as part of this thesis. Subsequently, the studies presented in Chapters 4–6 were conducted concurrently and not consecutively.

- Chapter 1: (this chapter): A general introduction, background, overall aim, objectives and thesis overview.
- Chapter 2: General material and methods.

- Chapter 3:Oropharyngeal swallowing physiology and safety: The Videofluoroscopy Swallow Study: A clinical perspective.
- Chapter 4: Swallowing safety and performance: The Water Swallow Test: A clinical perspective.
- Chapter 5: Swallowing and throat symptoms: The patients' reported assessment: A patients' perspective.
- Chapter 6: Eating and drinking experience: The qualitative study: A patients' perspective.
- Chapter 7: Final discussion and future recommendations.
- Chapter 8: COVID-19 impact and disruption.

Chapter 2. Material and methods

2.1 Chapter overview

This thesis includes four studies aiming to assess swallowing from different perspectives: clinical (Chapters 3 and 4) and patients' perspective (Chapters 5 and 6). The demographic data and analysis are common to more than one study. In this chapter, I will discuss the study designs, assessment tools and recruitment procedures. More specific details about the methods will be described in the subsequent chapters and frequently refer back to the key section in this chapter.

2.2 Study design

Given the exploratory nature of this research project, which focuses on investigating swallowing in patients with idiopathic pulmonary fibrosis (IPF), it is important to recognise that eating and drinking encompasses multiple dimensions of functioning, including physical, emotional and social aspects. In light of this complexity, I decided to employ diverse approaches and utilise both quantitative and qualitative methods in order to effectively address the research aim and objectives. This approach allows for a comprehensive exploration of the topic, as opposed to solely relying on a singular, in-depth methodology.

This thesis consists of four observational studies, using quantitative and qualitative methodologies. The quantitative data presented in this thesis are observational descriptive case series and observational cross-sectional data. The qualitative study is a descriptive study, using semi-structured interview data. More specific details about the study designs of the conducted studies are described in the subsequent chapters. (see Table 1).

Study	Design	Participants	Data analysis
Oropharyngeal swallowing physiology and safety: The Videofluoroscopy Swallow Study: A clinical perspective (Chapter 3)	Descriptive, case series	IPF patients (n:10)	Descriptive
Swallowing safety and performance: The Water Swallow Test: A clinical perspective (Chapter 4)	Observational, cross-sectional	IPF patients (n:33)	Descriptive and comparative statistics
Swallowing and throat symptoms: The patients' reported assessment: A patients' perspective (Chapter 5)	Observational, cross-sectional	IPF patients (n:40)	Descriptive statistics
Eating and drinking experience: The qualitative study: A patients' perspective (Chapter 6)	Descriptive, qualitative study using interview	IPF patients (n:14)	Descriptive qualitative

Table 1. An overview of the conducted studies
(n: number)

2.3 Recruitment

2.3.1 Eligibility criteria

The general inclusion and exclusion criteria for all studies are outlined below, while the detailed inclusion and exclusion criteria specific to each study is found in the corresponding methods section within each chapter.

2.3.1.1 Inclusion criteria

The studies included competent adults (over the age of 18) with a secure diagnosis of IPF, as defined by international consensus guidelines (Raghu et al., 2011; Raghu et al., 2018).

2.3.1.2 Exclusion criteria

- History of other lung diseases.
- History of pre-existing swallowing problem due to other causes such as: head and neck pathology excepting tonsillectomy/adenoidectomy, previous thoracic surgery, stroke, and neurological diagnosis that may be associated with dysphagia.
- Difficulty in understanding written or verbal information in English.
- Inability to follow instructions, for example: learning difficulties.
- Individuals with significant communication difficulties, cognitive impairment or memory difficulties such that informed consent could not be given.

2.4 Data collection

2.4.1 Data collection for Study 1 (Chapter 3)

Recruitment was undertaken from the Interstitial Lung Disease (ILD) clinic at the Royal Victoria Infirmary (RVI), Newcastle upon Tyne, between March 2014 and February 2015, before I started my PhD study. More details about the recruitment for this study are provided in Chapter 3.

2.4.2 Data collection for Studies 2, 3 and 4 (Chapters 4, 5 and 6)

Recruitment was undertaken remotely and face to face. This followed contact with pulmonary fibrosis support groups around the UK and local working with the ILD clinic at the RVI, Newcastle upon Tyne between January 2021 and November 2021.

2.4.2.1 Remote recruitment of patients using UK IPF Support groups (January 2021–June 2021).

Action for Pulmonary Fibrosis (APF) is a patient-led charity that was founded in 2013 by patients, family and pulmonary fibrosis specialists under the umbrella of the British Lung Foundation (BLF). APF aims to provide support to people with pulmonary fibrosis and their families, educate health professionals and the public about the disease, and fund research to attempt to identify improved treatment for the disease, in order to attempt to better understand the courses of the disease.

Patient recruitment started during the third UK COVID-19 lockdown (January 2021); face-to-face contact with IPF patients was not possible during that time, as they were considered clinically extremely vulnerable by the NHS, and had been advised to shield.

Following government advice due to COVID-19 measures, support groups were not meeting face to face, and in some areas virtual meetings were taking place instead. Therefore, approaching IPF patients from pulmonary fibrosis support groups virtually was the most feasible option available at that time. The aim was to recruit unselected participants with a diagnosis of IPF, without previous evidence of swallowing difficulty, and this point was emphasised by the researcher during the meeting with the support group representatives in order to avoid recruitment bias.

Step one: local support groups (January 2021–March 2021)

Recruitment initially targeted the local support group in the North East area:

The Northern Region Idiopathic Pulmonary Fibrosis Support Group leader was contacted by email and phone to arrange an online meeting. A Zoom meeting with the support group leader was held to discuss the study in detail, for example inclusion and exclusion criteria and the possibility of approaching group members virtually. A booklet with three sections was sent by email to the Northern Region IPF Support Group leader, including the following: 1. A study invitation letter, which had researcher contact details (email and phone number); 2. A research information sheet (see Appendix 1); 3. A certificate of consent for the patients (see Appendix 2). The booklet was circulated by the support group leader to the group members using the mailing list, which had patient and carer contact emails. Additionally, patients could also receive the study booklet by post, depending on their preference.

The South Tees Pulmonary Fibrosis Support Group leader was contacted by email and a research booklet was sent.

Step two: regional support groups (March 2021–June 2021)

Following positive experience of contacting my local patient support group I expanded this strategy to other parts of the UK to recruit further patients. Recruitment targeted regional pulmonary fibrosis support groups around the UK. The APF research officer was contacted by email, and a Zoom meeting was held to discuss the research and IPF patients' recruitment. The research officer was able to put me in contact with the

leaders of five active pulmonary fibrosis around the UK. A Zoom meeting to discuss the study was conducted with each group leader, and the research booklets were sent to them. Recruitment was conducted in order, starting with recruitment from one group, until no more members could be recruited, before moving on to the next group.

The list of regional pulmonary fibrosis support groups, ordered from the first recruited to the last recruited, is as follows:

1. South West Peninsula Pulmonary Fibrosis Support Group
2. Windsor Pulmonary Fibrosis Support Group
3. Swansea Pulmonary Fibrosis Support Group
4. Bolton Pulmonary Fibrosis Support Group

Step three: support groups in Northern Ireland (April 2021–July 2021)

Recruitment was also undertaken from Northern Ireland. The research officer for APFs approached the Northern Trust Pulmonary Fibrosis Support Group in Northern Ireland. A Zoom meeting with the support group leader was arranged, and the research booklet was sent by email to be distributed to all group members (see Table 2 and Figure 1).

Pulmonary fibrosis support group	**Number of IPF patients recruited**
Northern Region Idiopathic Pulmonary Fibrosis Support Group	11
South Tees Pulmonary Fibrosis Support Group	1
South West Peninsula Pulmonary Fibrosis Support Group	5
Windsor Pulmonary Fibrosis Support Group	1
Swansea Pulmonary Fibrosis Support Group	1
Bolton Pulmonary Fibrosis Support Group	0
Northern Trust Pulmonary Fibrosis Support Group in Northern Ireland	2
Total number of IPF patients recruited from pulmonary fibrosis support groups	**21**

Table 2. List of pulmonary fibrosis support groups and the number of IPF patients recruited from each group.

2.4.2.2 Interstitial lung disease (ILD) clinic at the RVI, face-to-face approach (August 2021–November 2021)

Face-to-face recruitment in Newcastle targeted the ILD clinic, which is a speciality clinic of the respiratory medicine department at the RVI. ILD is an umbrella term that encompasses a wide range of conditions affecting lung tissue, such as sarcoidosis, usual interstitial pneumonia and IPF.

The lists of ILD patients seen face to face in the ILD clinic were reviewed by the researcher. Patients diagnosed with IPF were identified after checking the MDT diagnosis letter located in the patient's electronic medical file. Potentially suitable IPF patients, who met the study inclusion criteria, were selected by the researcher and provided with a participant information sheet (PIS) while they were waiting in the waiting room. Additionally, the study was explained briefly to the IPF patients by the researcher while they were waiting in the waiting area. Patients were given 30 to 40 minutes to consider participation in the study. Those agreeing to participate in the study were invited to clinic C. The study was described in detail, with full written consent taken at the outset, before study activities commenced (see Figure 2).

2.4.2.3 Recruitment procedures

IPF support groups recruitment procedures

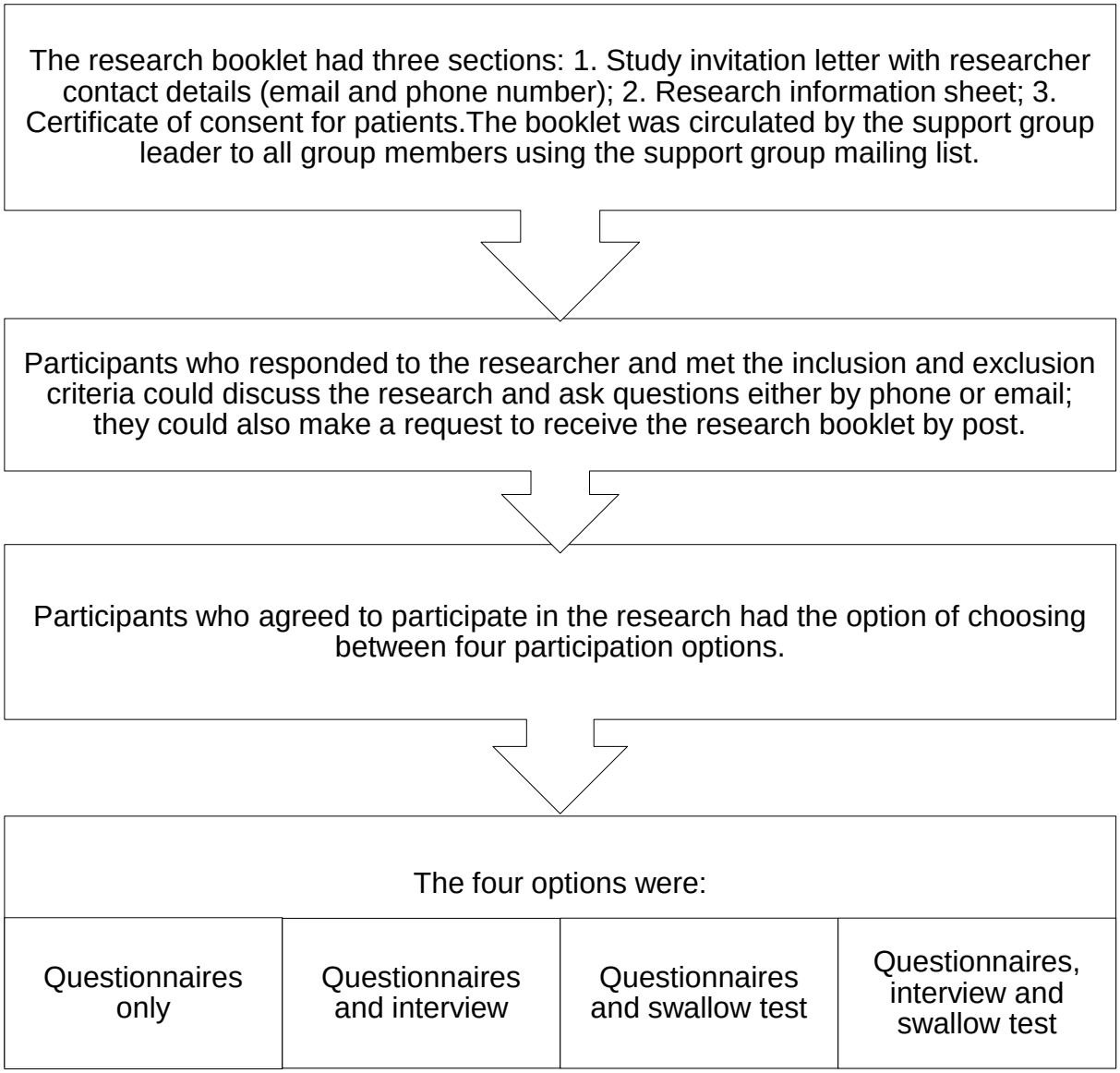

Figure 1. The recruitment procedure from IPF support groups.

ILD clinic recruitment procedures

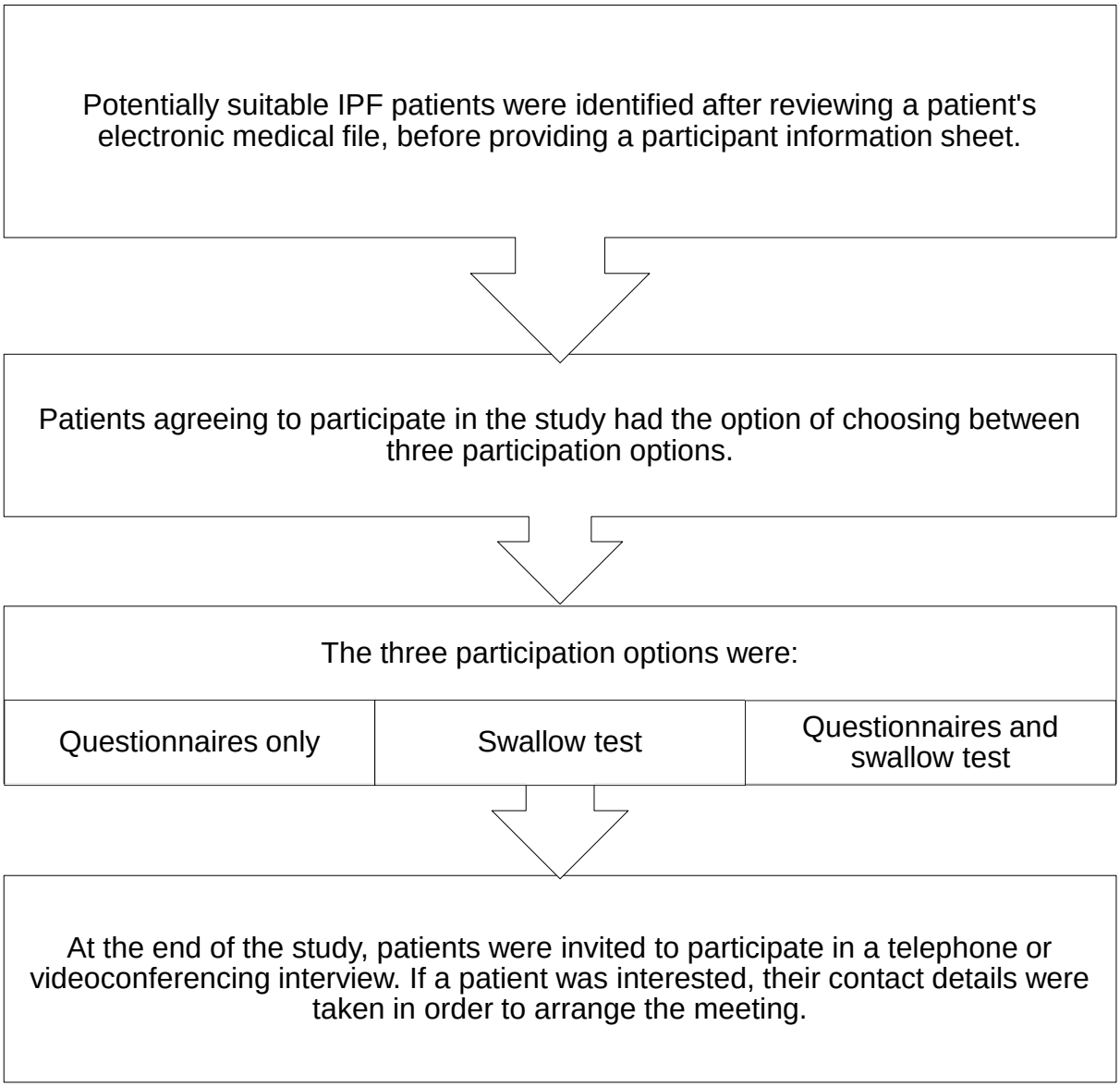

Figure 2. The recruitment procedure from the ILD clinic.

Recruitments for the study population

The total number of patients who participated in this project and the type of participation are presented in Table 3.

	Recruitment methods	Questionnaires	WST	Interview
IPF1	Support group	✓	✓	✓
IPF2	Support group	✓	✓	✓
IPF3	Support group	✓	✓	✓
IPF4	Support group	✓	X	✓
IPF5	Support group	✓	✓	✓
IPF6	Support group	✓	✓	✓
IPF7	Support group	✓	X	X
IPF8	Support group	✓	X	X
IPF9	Support group	✓	X	X
IPF10	Support group	✓	✓	✓
IPF11	Support group	✓	X	X
IPF12	Support group	✓	✓	✓
IPF13	Support group	✓	✓	✓
IPF14	Support group	✓	✓	✓
IPF15	Support group	✓	X	X
IPF16	Support group	✓	✓	✓
IPF17	Support group	✓	✓	X
IPF18	Support group	✓	✓	X
IPF19	Support group	✓	✓	X
IPF20	Support group	✓	✓	✓
IPF21	Support group	✓	X	X
IPF22	ILD clinic	✓	✓	X
IPF23	ILD clinic	✓	✓	✓
IPF24	ILD clinic	✓	✓	X
IPF25	ILD clinic	✓	✓	X
IPF26	ILD clinic	✓	✓	X
IPF27	ILD clinic	✓	✓	X
IPF28	ILD clinic	✓	✓	X
IPF29	ILD clinic	✓	✓	X
IPF30	ILD clinic	✓	✓	✓
IPF31	ILD clinic	✓	✓	X
IPF32	ILD clinic	✓	✓	X
IPF33	ILD clinic	✓	✓	X
IPF34	ILD clinic	✓	✓	X
IPF35	ILD clinic	✓	✓	X
IPF36	ILD clinic	✓	✓	X
IPF37	ILD clinic	✓	✓	X
IPF38	ILD clinic	✓	✓	X
IPF39	ILD clinic	✓	✓	X
IPF40	ILD clinic	✓	✓	X
IPF41	ILD clinic	X	X	✓

Table 3. Recruitments for the study population
IPF: idiopathic pulmonary fibrosis, WST: Water Swallow Test

2.5 Sample size

2.5.1 Sample size for the quantitative studies

The analysis of data was expected to be mainly descriptive. As a result, the recruitment goal of 30 participants was chosen to be practical and aligned with the ability to generate robust descriptive analyses.

2.5.2 Sample size for the qualitative study

The patients were recruited using purposive sampling methods. The number of interviews conducted was informed by the concept of "information power". Full details about the methods used for sampling and recruiting patients for the qualitative study are described in detail in Chapter 6.

2.6 Involvement of Patients in Study Design

Patient and public involvement (PPI) played a pivotal role in the design phase of this study, particularly in ensuring the relevance and accessibility of study materials to patients with IPF. Two representatives from Northern Region IPF support group actively participated in piloting the participants' information sheet. This involvement not only enriched the content of the information sheet but also provided invaluable insights into the preferences and needs of the target population. The piloting process involved soliciting feedback from the patient representatives regarding the clarity, comprehensibility, and relevance of the information presented in the participants' information sheet. Their perspectives were instrumental in identifying areas for improvement and refining the language to ensure it was understandable to individuals with varying levels of literacy and familiarity with medical terminology.

2.7 Consent

A consent form was signed before study activities started. The patients from the support groups received the consent form via post or email. If post was used, the consent form was accompanied by a stamped addressed envelope for the return of the signed consent to the researcher. The patients from ILD clinics received the consent form in the clinic before the study started. In the consent form, the patient had

the option to consent to participate in only the questionnaires, in the questionnaires and interview, in the questionnaires and swallow test, or to participate in the whole study (questionnaires, interview and swallow test).

The rights of individuals to change their mind about participation in the study and/or withdraw without giving a reason were respected. Patients could withdraw their consent at any time. If they did so, their personal data was destroyed as soon as possible. If the patient withdrew during the interview, the partial transcript was discarded. If a patient decided to withdraw after the interview had been completed and wished their transcript to be destroyed, this was done. Consent forms of such patients were retained but were marked as a patient who had withdrawn.

Finally, patients were asked whether they would like to receive a summary of the findings of the research once the study was over. Those who wanted to receive this information were asked how they would like to receive it. Their preferred means of contact was retained separately from the consent forms and solely for the purposes of sharing the study results.

2.7.1 Reimbursement

In recognition of their participation and as a gesture of gratitude, participants in this thesis study were offered vouchers as a token of appreciation.

- A £30 online shopping voucher was given as a thank you for participation in the questionnaires only.
- A £50 online shopping voucher was given as a thank you for participation in the questionnaires and interview.
- A £50 online shopping voucher was given as a thank you for participation in the questionnaires and swallow test.
- A £70 online shopping voucher was given as a thank you for participation in the whole study (questionnaires, interview and swallow test).

2.8 Analysis

Full details about the descriptive, comparative, quantitative and qualitative analysis are described in detail in each subsequent chapter.

2.8.1 The assessment tools

- Videofluoroscopy Swallow Study (VFSS) (Chapter 3).
- Water Swallow Test (WST) (Chapter 4).
- Eating Assessment Tool-10 (EAT-10) (Chapters 3 and 5).
- Reflux Symptoms Index (RSI) (Chapter 5).
- Newcastle Laryngeal Hypersensitivity Questionnaires (LHQ) (Chapter 5).
- Semi-structured interviews (Chapter 6).

Chapter 3. Oropharyngeal swallowing physiology and safety: The Videofluoroscopy Swallow Study: A clinical perspective

3.1 Introduction

This is the first results chapter of my thesis. In this study, swallowing was assessed from the clinical perspective using the gold standard instrumental assessment, the Videofluoroscopy Swallow Study (VFSS). Elements of the work described in this chapter have been previously presented as a poster presentation in the European Respiratory Society (ERS) International Congress 2020 (Alamer et al., 2020) (see Appendix 3), an oral presentation at the British Thoracic Society (BTS), winter meeting, 2021 (Alamer et al., 2021) (see Appendix 4) and published in a peer-reviewed paper (Alamer et al., 2022a) (see Appendix 5).

3.2 Literature review

To date, rare investigations of IPF and aspiration have been limited to considering gastro-oesophageal reflux disease, in which gastric content refluxes up into the oesophagus, with the potential to pass into the airway via the pharynx (microaspiration) (Wang et al., 2018). Concerns about the potential role of gastro-oesophageal reflux-associated aspiration in IPF pathophysiology have been sufficient to prompt an influential multicentre pilot trial of fundoplication (Raghu et al., 2006a). A dysregulated lung microbiome has also been implicated in high-profile studies (Spagnolo et al., 2019).

Patients with chronic respiratory diseases are at risk of oropharyngeal swallowing pathophysiology and may go unreported (Garand et al., 2018; Lin and Shune, 2020). Scarpel et al. (2021) conducted a study on patients with asthma to investigate the oral and pharyngeal phases of swallowing using VFSS. The study included 135 participants and found that patients with both mild and severe asthma exhibited disorganisation of the food bolus in the oral phase, including instability of its positioning, escape in the oral cavity, post-swallow oral residue and atypical tongue movement. Similarly, in the pharyngeal phase, both groups experienced late onset of the pharyngeal phase and pharyngeal residue (Scarpel et al., 2021). However, patients with severe asthma had a higher occurrence of multiple swallows, laryngeal penetration and aspiration compared with the mild asthma group (Scarpel et al., 2021).

In patients with chronic obstructive pulmonary disease (COPD), studies revealed abnormal swallowing physiology, such as reduced laryngeal elevation, early glottis closure and an increase in the laryngeal protective closure timing during swallowing (Mokhlesi et al., 2002; Cassiani et al., 2015). Cvejic et al. (2011) reported aspiration in 25% and penetration in 12.5% of patients with stable COPD (n:20), which was observed during VFSS. One explanation for the presence of oropharyngeal dysphagia in COPD is an alteration in the respiratory-swallow cycle (Shaker et al., 1992). COPD patients are more likely to swallow during inhalation or inhale after swallowing (Shaker et al., 1992; Gross et al., 2009). The negative pressure of inhalation has the potential to draw food or liquid into the airway, increasing the risk of aspiration (Gross et al., 2009). Moreover, restricted lung expansion will result in the swallow occurring at low lung volume, potentially limiting the positive subglottic pressure generated during swallowing. Decreased subglottic pressure when swallowing has been found to alter the swallowing physiology by slowing the bolus transit time, prolonging pharyngeal contraction duration and consequently increasing post-swallow pharyngeal residue (Gross et al., 2006; Gross et al., 2009).

The parenchymal lung scarring, ageing and hypoxaemia seen in IPF patients may act to disrupt respiratory-swallow coordination, leading to dysfunction. To my knowledge, there are no studies investigating swallowing dysfunction in people with IPF.

3.2.1 Aim

The aim of this preliminary study is to describe the perception of swallowing and oropharyngeal swallowing physiology and safety in patients with IPF.

3.2.2 Study objectives

1. To measure IPF patients' perception of swallowing difficulty.
2. To explore oropharyngeal swallowing physiology in IPF patients.
3. To assess swallowing safety (i.e. airway invasion) in IPF patients.

3.3 Specific materials and methods

3.3.1 Ethical approval

The study was approved by the Health Research Authority (HRA), North-West Preston Research Ethics Committee, REC reference 14/NW/1056 (see Appendix 6).

3.3.2 Study design

This is a descriptive, consecutive case series study design.

3.3.3 Specific eligibility criteria

In addition to the core inclusions and exclusions criteria described in the general methods (2.3.1.1 and 2.3.1.2).

3.3.3.1Inclusion criteria

- The study included competent adults (over the age of 18) with a secure diagnosis of IPF, as defined by international consensus guidelines (Raghu et al., 2011). Patients' recruitment started in March 2014 and ended in February 2015; therefore, the 2011 clinical practice guidelines for IPF diagnosis were followed.

3.3.3.2 Exclusion criteria

- Pregnancy
- Gastrointestinal disease, excepting controlled reflux symptoms
- In-patients

3.3.4 Recruitment

Consecutive outpatients diagnosed with IPF were enrolled from the interstitial lung disease (ILD) clinic at the Royal Victoria Infirmary (RVI), Newcastle upon Tyne, between March 2014 and February 2015. This ILD clinic regularly reviews outpatients with IPF, and the recruitment for this study was conducted within this clinic population. Patients who met the study criteria were approached by either a respiratory consultant or the lead investigator of the study. A verbal explanation of the study was provided, accompanied by a patient information sheet (PIS). Following a minimum waiting period

of 72 hours, the patients were contacted, and if they expressed willingness to participate, a study appointment was offered. Prior to the study assessment, written informed consent was obtained from the patients using a printed consent form.

3.3.5 Protocol

The study protocol comprised of two main components: firstly, patients were asked to complete a validated swallow questionnaire. This questionnaire was provided to them either through postal delivery or during their visit to the research site. Secondly, the patients underwent VFSS at the Freeman Hospital in Newcastle upon Tyne. Patients' demographics including age, gender and smoking history were recorded. In addition, medical research council dyspnoea scale (MRC), body mass index (BMI), and pulmonary function tests including forced vital capacity (FVC) (% of predicted), transfer factor of lung for carbon monoxide (TLCO) (%) of predicted were collected from patients' medical records.

3.3.5.1Swallowing questionnaire (Objective one)

The Eating Assessment Tool (EAT-10) is a quick (taking approximately two minutes to complete), self-administered and widely used validated questionnaire, which can be used to assess dysphagia symptoms (Belafsky et al., 2008). It has been used previously with patients with chronic respiratory disease such as COPD (Garand et al., 2018; Lindh et al., 2021). It consists of ten questions regarding swallowing difficulty. Each question is scored on a five-point Likert scale from 0 (no problem) to 4 (severe problem). The total EAT-10 score is calculated by adding up the scores across the ten statements (highest score = 40). A total score of 3 or more indicates swallowing difficulty (Belafsky et al., 2008) (see Appendix 7: The Eating Assessment Tool: EAT-10).

3.3.5.2 Swallowing assessment (Objectives two and three)

The Videofluoroscopy Swallow Study (VFSS)

Swallowing was assessed by a VFSS. The VFSS is a radiographic examination using fluoroscopy to capture and record real-time bolus flow throughout all stages of swallowing (Martin-Harris and Jones, 2008). (Further details about VFSS are described in Chapter 1).

A speech and language therapist (SLT) and a radiologist performed the VFSS examinations. The patients were seated in an upright position. The examination typically included testing different bolus volumes and constituents. Test boluses were thin liquid, paste (custard) and biscuit mixed with the radiopaque barium sulphate product E-Z-PAQUE. Liquids were administered first to avoid confounding the results due to remaining residue in the pharynx after ingesting solid consistencies. A penny was taped to the subject's chin during the swallowing study. The circular shape of the penny minimises the impact of head rotation and the known diameter of the coin allows for calibration of pixels per centimetre (cm) and thus calculation of areas and displacement on VF. All of the VFSS studies were recorded and were copied onto DVDs.

The consistencies and quantities were as follows:

- Thin liquid contrasts were mixed to a viscosity of 40% weight per volume (w/v). One container of E-Z-PAQUE (177 gram powder) was mixed with 100 ml water and shaken.
- Paste/custard contrast was mixed with 40% w/v E-Z-PAQUE powder.

<u>VFSS protocol</u>

On lateral/mid-sagittal view, to make judgements about oral cavity, pharyngeal cavity, larynx and cervical oesophagus (Martin-Harris and Jones, 2008).

(Bolus trials were presented in the precise order listed below):

1. 5 ml thin (two trials, teaspoon (tsp) or measured cup): the first 5 ml was not counted in the analysis, and only used for training purposes.

The instruction given to the patient was: "Please hold this in your mouth until asked to swallow."

2. 20 ml thin (one trial, tsp or measured cup).

The instruction given to the patient was: "Please try to take the whole amount and hold it in your mouth until I ask you to swallow."

3. 5 ml pudding (custard) (one trial, tsp).

The instruction given to the patient was: "Swallow when you are ready."

4. ½ biscuit – coated with 3 ml (1/2 tsp) paste contrast (one trial).

The instruction given to the patient was: “Chew this and swallow when you feel comfortable and ready to swallow.”

On anterior-posterior (coronal) view, useful to identify symmetry of bolus flow, pharyngeal wall contraction and oesophageal stage of swallowing (Martin-Harris and Jones, 2008).

1. 10 ml thin (one trial, tsp or measured cup).

The instruction given to the patient was: “Slightly raise your chin (but keep your head neutral, not tucked or extended), and hold this in your mouth until asked to swallow.”

VFSSs analysis

The VFSS studies were rated by an SLT trained in VFSS interpretation, certified in standardised Modified Barium Swallow Impairment Profile (MBSImP) using two validated scales:

1. The standardised MBSImP, a tool used to evaluate swallowing efficiency that measures 17 physiological components of adult swallowing mechanism using ordinal scaling. Ratings of 0 to 2, 3 or 4 points per component, with each score representing a unique observation of either structural movement, bolus flow or both from the VFSS (Martin-Harris *et al.*, 2008; Martin-Harris;Humphries and Garand, 2017). It covers three functional domains of the swallow: oral (0–22), pharyngeal (0–29) and oesophageal (0–4). The oesophageal domain was not reported, as it is beyond the scope of this thesis. The Overall impression (OI) score is the worst and the most impaired score for all bolus amounts and consistencies (Martin-Harris et al., 2008). The oral impairment score and pharyngeal impairment score were calculated by summing the OI scores. Scores were interpreted according to a clinically validated classification system (Beall et al., 2020). Figure 3. shows one example of a five-point scaled physiological component (Martin-Harris et al., 2008, p. 18). Table 4 presents the three functional domains across 17 physiological swallowing impairments in the MBSImP (Martin-Harris et al., 2008, p. 25).
2. The Penetration Aspiration Scale (PAS) is a tool used to evaluate swallowing safety, ranging in value from 1 to 8, recording the presence of laryngeal penetration/sub-glottic aspiration (1 = no airway invasion, 2–5 = penetration, 6–8 = aspiration) (Rosenbek et al., 1996; Robbins et al., 1999) (see Table 5). Penetration is defined as the passage of food or fluid into the airway just above the

level of the vocal cords, whereas aspiration is defined as the passage of food or fluid below the level of the vocal cord (Allen et al., 2010).

Higher scores for both tools indicate poorer swallowing.

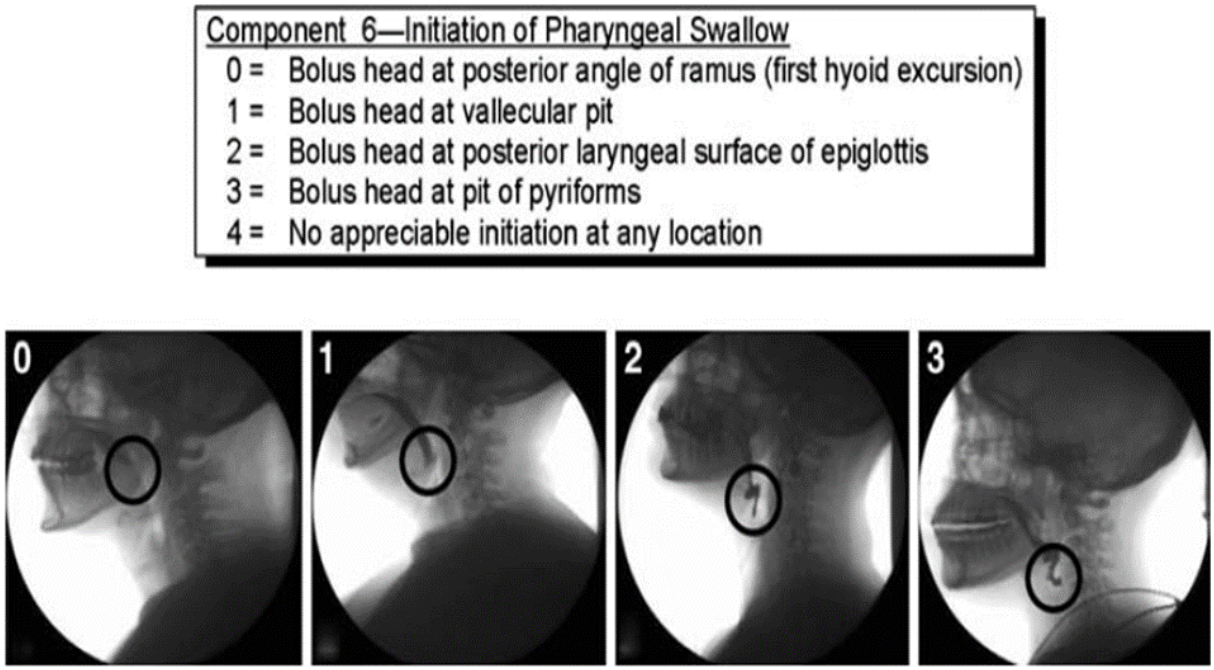

Figure 3. Operational definitions of the Initiation of Pharyngeal Swallow (IPS) component scores (Martin-Harris et al., 2008, p. 18).

Domain	Physiological Swallowing Components
Oral Domain	1.Lip closure (Lip C)
	2.Hold position/Tongue control during bolus hold (HP)
	3.Bolus preparation/mastication (BP)
	4.Bolus transport/lingual motion (BT)
	5.Oral residue (OR)
	6.Initiation of pharyngeal swallow (IPS)
Pharyngeal Domain	7.Soft palate elevation (SPE)
	8.Laryngeal elevation (LE)
	9.Anterior hyoid excursion/movement
	10.Epiglottic movement (EM)
	11.Laryngeal vestibular closure (LC)
	12.Pharyngeal stripping wave (PSW)
	13.Pharyngeal contraction (PC)
	14.Pharyngoesophageal segment opening (PESO)
	15.Tongue base retraction (TBR)
	16.Pharyngeal residue (PR)
Oesophageal Domain	17.Oesophageal clearance in upright position (EC)

Table 4. The three functional domains across 17 physiological swallowing impairments in the Modified Barium Swallow Impairment Profile (MBSImP)
The 17 components are grouped across three functional domains of swallowing, each component contributing uniquely to judgements of overall swallowing impairment. The oral domain includes components related to oral containment, oral tongue motility and oral bolus clearance. The pharyngeal domain comprises components related to airway protection and pharyngeal bolus clearance. The oesophageal domain includes one component related to oesophageal bolus clearance (Martin-Harris et al., 2008, p. 25).

PA Scale Score	Description
1	Material does not enter the airway
2	Material enters the airway, remains above the vocal folds and is ejected from the airway
3	Material enters the airway, remains above the vocal folds and is not ejected from the airway
4	Material enters the airway, contacts the vocal folds and is ejected from the airway
5	Material enters the airway, contacts the vocal folds and is not ejected from the airway
6	Material enters the airway, passes below the vocal folds and is ejected into the larynx or out of the airway
7	Material enters the airway, passes below the vocal folds and is not ejected from the trachea despite effort
8	Material enters the airway, passes below the vocal folds and no effort is made to eject

Table 5. The eight-point Penetration Aspiration Scale
Scoring summary for the Penetration Aspiration (PA) Scale: 1 = no airway invasion, 2–5 = penetration, 6–8 = aspiration (Rosenbek et al., 1996, p. 94).

3.3.6 Data management

The VFSS video recordings (DVDs) were securely stored on the Newcastle Hospitals' Picture Archiving and Communication System (PACS).

3.3.7 Analysis

All continuous variables were presented as medians and the range (minimum–maximum). Categorical variables were presented using frequencies and percentages. Data from the EAT-10 was analysed against the published cut-off value for normal EAT-10. All descriptive analyses and graphic presentations in this chapter were performed using Minitab statistical software (version 21, Minitab Inc., State College, PA, USA) and Microsoft Excel 2016 (Microsoft Corporation, Washington, DC, USA).

3.4 Results

A total of 14 patients (ten males and four females), median age: 68.0 years, ([min–max] 51–82 years) were recruited for the study. Out of the total of 14 patients, ten of them (seven males and three females), median age: 63.0 years, ([min–max] 51–77 years) underwent the VFSS. See Figure 4 below.

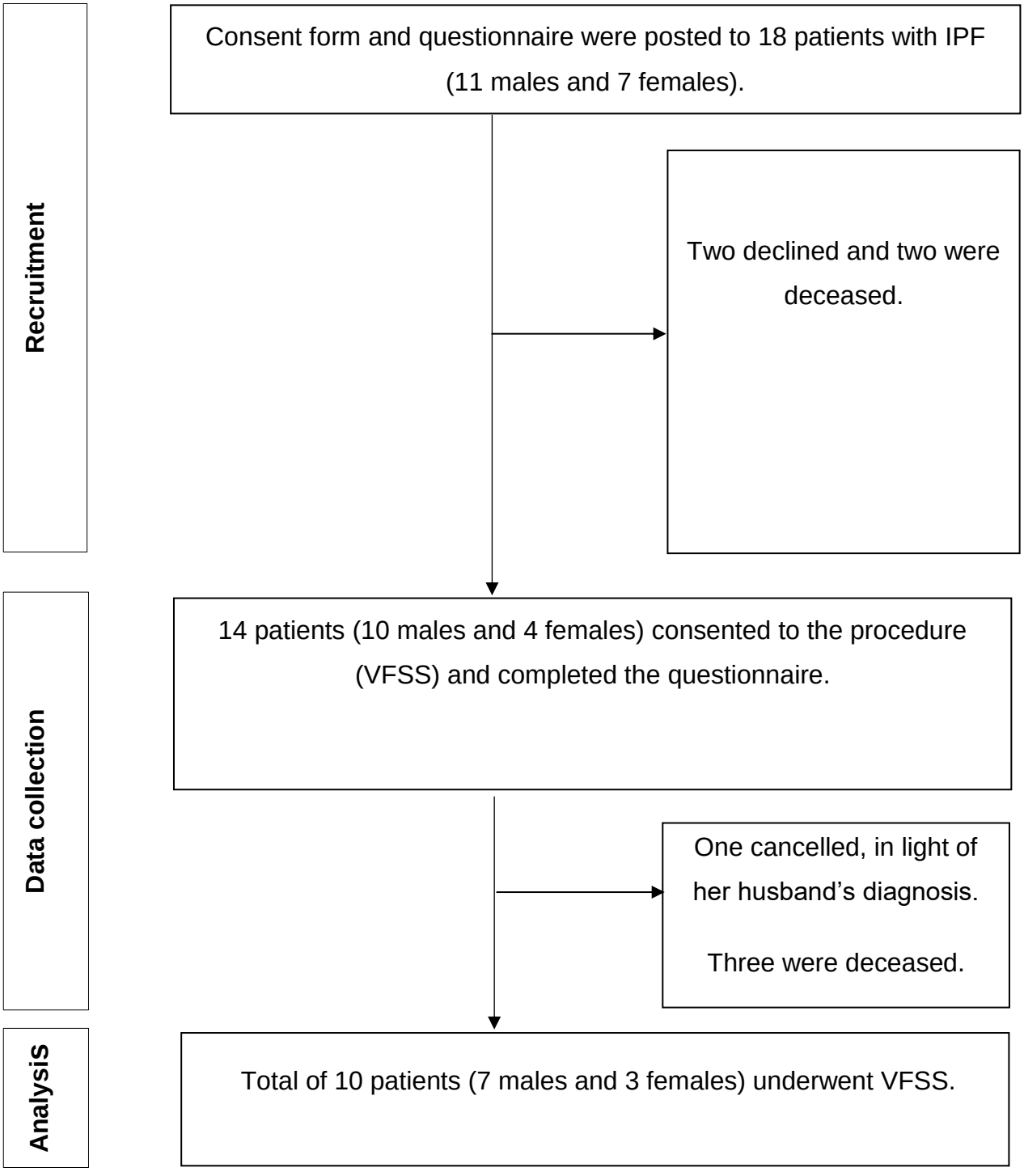

Figure 4. Consort flow diagram for the patients' recruitment for the study.

3.4.1 Patients' demographics and clinical characteristics

Patients' demographics and clinical characteristics are presented in Table 6. The majority of the patients were male (10/14, 71.4%) and ex-smokers (9/14, 64.3%). The median BMI was 25.3 kg/m^2 ([min–max] 19.3–45.6). The median MRC was 2 ([min–max] 1–4). The median FVC percentage of predicted was 70% ([min–max] 50%–92%). The median TLCO percentage of predicted was 62% ([min–max] 27%–78%).

Factor	Level	Value
N		14
Age in years, median (minimum-maximum)		68 (51-82)
Sex	Female	4 (4/14)
	Male	10 (10/14)
Smoking status	Current-smoker	3 (21.4%)
	Ex-smoker	9 (64.3%)
	Non-smoker	2 (14.3%)
BMI, kg/m^2, median (minimum-maximum)		25.3 (19.3-45.6)
MRC Dyspnoea Scale, median (minimum-maximum)		2 (1-4)
FVC% of predicted, median (minimum-maximum)		70%(50%-92%)
TLCO% of predicted, median (minimum-maximum)		62(27-78%)

Table 6. Median (min-max) or (%) for the patients' demographics and clinical characteristics
N: number; BMI: Body mass index; MRC: Medical Research Council dyspnoea scale; FVC: Forced Vital Capacity; TLCO: Transfer factor of Lung for Carbon Monoxide.

3.4.2 EAT-10: self-reported swallowing symptoms

A total of 14 IPF patients completed the EAT-10 questionnaire. The total median EAT-10 score was 0, ([min–max] 0–25) for all patients. Scores above the cut-off of <3 for

normal swallowing were recorded in four patients, with values of 25, 15, 14 and 13 (Belafsky et al., 2008). All data are presented in Figure 5.

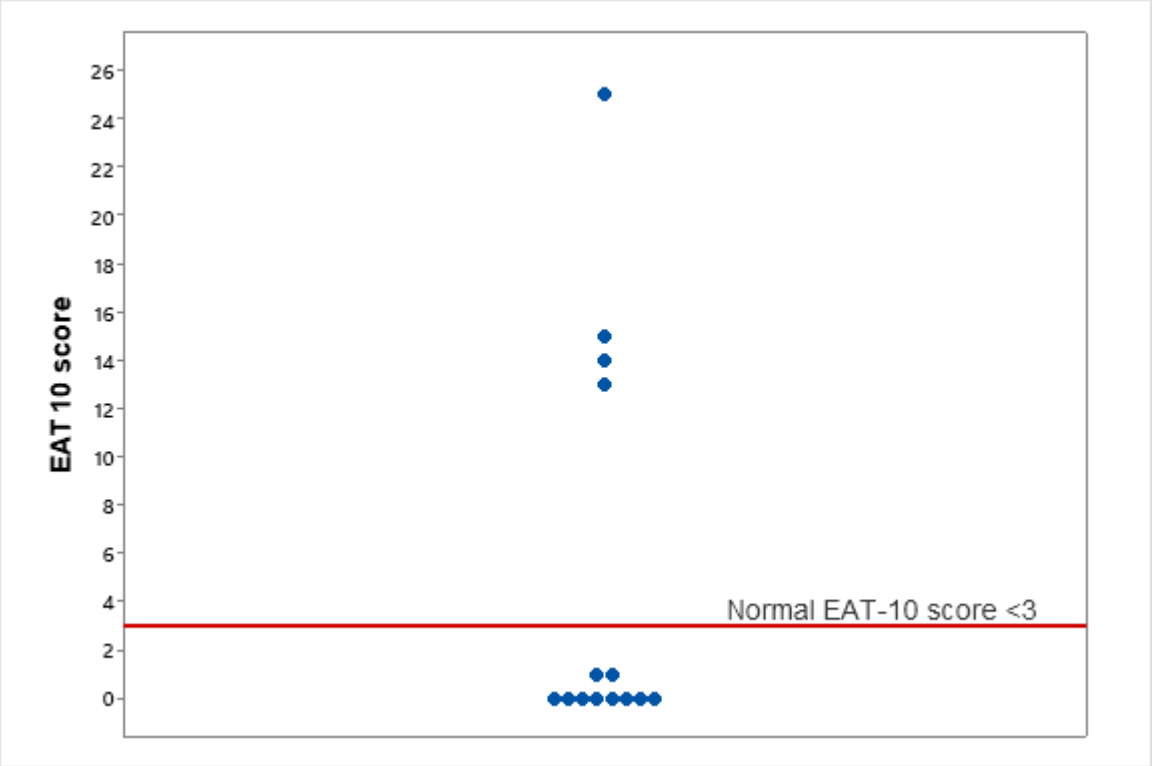

Figure 5. EAT-10 scores for all IPF patients (n:14).
EAT-10 score (y-axis) for patients. The red line shows the upper score for EAT-10. A score of ≥ 3 indicates an abnormal EAT-10 score.

3.4.3 The Videofluoroscopy Swallow Study (VFSS)

All data are presented in Table 7. Median MBSImp for oral impairment was 5 ([min–max] 3–7) and pharyngeal impairment was 4 ([min–max] 1–14), indicating overall mild alteration to swallowing. Patient 1 scored 14 in pharyngeal impairment, indicating mild/moderate impairment (Beall et al., 2020).

Patients	Oral domain							Pharyngeal domain										PAS	EAT -10
	Lip closure	Tongue control	Bolus preparation	Bolus transport	Oral residue	Initiation of pharyngeal swallow	Oral impairment	Soft palate elevation	Laryngeal elevation	Anterior hyoid excursion/movement	Epiglottic movement	Laryngeal vestibular closure	Pharyngeal stripping wave	Pharyngeal contraction	Pharyngoesophageal segment opening	Tongue base retraction	Pharyngeal residue	Pharyngeal impairment		
1	0	1	0	0	0	3	4	0	1	1	1	2	1	3	1	2	2	14	8	25
2	0	2	0	0	2	3	7	0	1	1	1	2	1	0	1	2	2	11	2	0
3	0	1	0	0	2	3	6	0	1	0	0	1	0	0	0	2	2	6	1	0
4	0	2	0	0	2	3	7	0	1	0	1	0	0		1	0	2	5	2	1
5	0	0	0	0	2	3	5	0	0	0	0	0	1	0	1	0	2	4	1	0
6	0	2	0	0	2	3	7	0	0	0	0	0	0	0	0	0	2	2	1	0
7	0	2	0	0	0	3	5	0	1	0	0	0	0	0	0	0	2	3	1	14
8	0	0	0	0	0	3	3	0	1	0	0	1	0	0	0	0	2	4	2	0
9	0		0	0	2	2	4	0	0	1	0	0	1		0	0	0	2	1	0
10	0	2	0	0	0	2	4	0	0	0	0	1	0		0	0	0	1	1	13

Table 7. Videofluoroscopy Swallow Study (VFSS) and Eating Assessment Tool-10 (EAT-10) results

Results for all patients relating to the 17 physiological components showing the Overall impression (OI) score, oral and pharyngeal impairment scores, PAS and EAT-10. EAT-10, Eating Assessment Tool: Green cells indicate no dysfunction, yellow cells mild dysfunction, orange cells moderate dysfunction, red cells indicate severe dysfunction and white blank cells denote missing data.

3.4.3.1 Swallow physiology: the MBS impairment profile

The MBSImP scores suggested no to mild swallowing impairment during oral and pharyngeal stages (Beall et al., 2020).

Lip closure, bolus preparation and transport, and soft palate elevation were uniformly normal. However, all patients had evidence of oral residue, six had a reduction in tongue control and eight patients had a late initiation of pharyngeal swallow.

For the pharyngeal stage of swallowing, five patients had evidence of incomplete laryngeal closure and one patient had a bilateral bulging of both pharyngeal walls. Eight patients had a reduction in tongue base retraction. Nine patients had evidence of post-swallow pharyngeal residue.

3.4.3.1 Swallow safety: the Penetration/Aspiration Scale

On PAS, 3/10 patients (Patients 2, 4 and 8) had airway penetration. Only Patient 1 aspirated liquid without a cough response; this patient had reduced laryngeal elevation and incomplete laryngeal vestibular closure, resulting in the residue lying below the true vocal cords without a response to eject the aspirated liquid (see Figure 6).

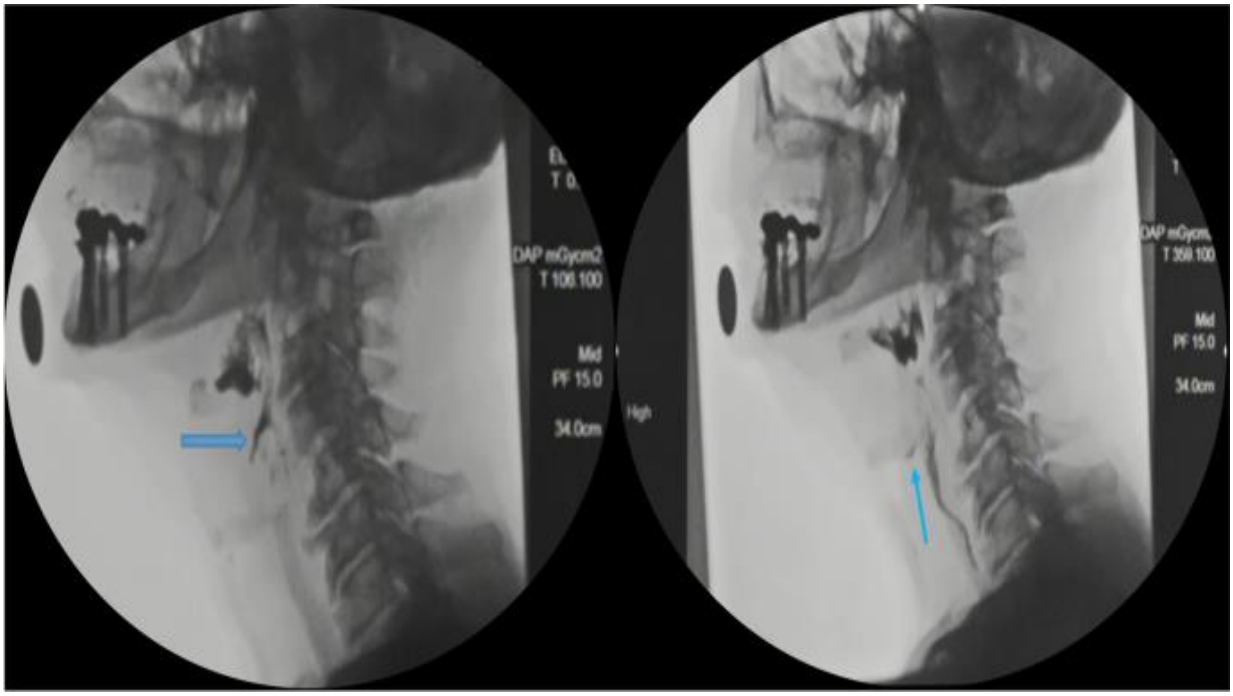

Figure 6. Oblique sagittal views during swallow of 20 ml thin liquid showing (left) penetration of barium into the larynx and (right) tracheal aspiration with contrast medium below the region of penetration.

3.5 Discussion

To my knowledge, dysphagia has not been described in patients with IPF. The purpose of this study was to describe the perception of swallowing difficulty, oropharyngeal swallowing physiology and safety in an unselected small sample of patients with a secure, mixed disciplinary diagnosis of IPF, without previous evidence of swallowing difficulty.

3.5.1 Self-reported swallowing symptoms

The widely used patient-reported symptoms of swallowing impairment, the EAT-10 tool, was used to assess patients' perception of swallowing difficulty. In this study, the median EAT-10 score was 0, with a range of 0–25. Four out of 14 patients (29%) had markedly raised total EAT-10 scores, with values of 25, 15, 14 and 13 (Belafsky et al., 2008). Considering the application of EAT-10 in chronic respiratory diseases, although nothing has been found in IPF, a recent study in 30 patients with acute exacerbations of COPD showed that 67% of patients had a raised EAT-10 score, compared with 23% of patients with cardiac disease (Lindh et al., 2021). A prior

study found that 44% of patients with stable COPD scored ≥3 in EAT-10 compared with 19% of healthy control (Garand et al., 2018).

In this study, subjective dysphagia symptoms reported by the EAT-10 tool appeared unrelated to the nature and severity of the oropharyngeal swallow impairment observed during VFSS. Patient 1, who demonstrated aspiration on videofluoroscopy, had the highest EAT-10 score of 25, but patients who scored 13 and 14 in the EAT-10 tool had relatively normal swallow physiology detected during VFSS (Table 4). These exploratory findings in a limited number of patients are consistent with previous studies in COPD, which has previously shown a weak association of EAT-10 with the swallowing impairments identified in VFSS (Garand et al., 2018). Further studies are therefore indicated in patients with IPF, within whom it is important to identify patients who may have difficulty swallowing regardless of whether aspiration is present. The EAT-10 tool helps to extend understanding about broad aspects of swallowing, which includes patient-centred social and emotional information, not captured by objective instrumental tests.

3.5.2 Swallow physiology and safety

Videofluoroscopy studies demonstrated a range of physiology in the ten patients studied. The swallow from laryngeal elevation onwards was consistently and highly disrupted in Patients 1 and 2; these two patients had an abnormal physiology according to the MBSImP classification system (Beall et al., 2020). However, Patient 2 had no airway invasion, despite objectively having the worst physiology of all, suggesting that a high score on MBSImP may not correspond to an unsafe swallow. Higher scores on some components of MBSImP may be regarded as “normal”; for example, the bolus may enter the pharynx before swallow is initiated even in healthy individuals and some features may be part of a healthy ageing profile (Dua et al., 1997). In PAS, Patients 2, 4 and 8 had airway penetration; we also noted that Patients 4 and 8 had relatively normal physiology according to MBSImP. In the scarce literature, normal older swallowers sometimes have scores of 2 and 3 in PAS and our findings may therefore represent the combined effects of both normal ageing and IPF pathophysiology and require further study (Robbins et al., 1999). There is no available evidence in the literature of which I am aware

comparing the prevalence of swallowing impairments in IPF patients with the general population of similar age, with no known neurological disorders.

Parenchymal lung scarring and hypoxaemia may disrupt the complex coordination of normal swallowing and breathing function and, in principle, dysphagia may contribute to a complex dysregulated aerodigestive homeostasis in patients with IPF. The true incidence of dysphagia in IPF is unknown, but oral dysbiosis has been linked with a range of lung diseases including pneumonia, COPD and lung cancer (Pathak et al., 2021). The oral cavity has been shown to be a source of diverse bacteria and it is of interest that this can include non-gastric reservoirs of *Helicobacter pylori* (Kashyap et al., 2020), which has been associated with a more severe disease phenotype, higher mortality and lung function decline in people with IPF (Bennett et al., 2014).

As fibrotic changes progress and lung function declines in IPF, the affected lung may be expected to become more susceptible to external challenges such as aspiration. Non-sterile aspiration, related to dysphagia and unprotected by cough, therefore represents a candidate source of complex lung injury and microbiome dysregulation. Aspiration is noted to be a trigger of threatening acute exacerbations (Kamiya and Panlaqui, 2021). Acute exacerbations in IPF are of high concern, as they represent the most common cause of death in IPF. Just under half of deaths in IPF are preceded by an acute exacerbation, and the median survival after such an exacerbation is approximately three to four months (Collard et al., 2016).

3.5.3 Limitations

This prospective consecutive descriptive case series is a novel contribution to the literature because, to my knowledge, this is the first to describe the perception of swallowing difficulty, oropharyngeal swallowing physiology and safety. This research design does not require large sample sizes or sophisticated research methods, meaning that it can be conducted with limited resources and with low cost (Kooistra et al., 2009). It is worth noting that this study was exploratory in nature, so the small sample size was sufficient to identify and justify the need for further investigation in this particular group. However, it is important to acknowledge certain limitations of this research design, including the absence of a comparison group and

the potential for bias. Moreover, it is important to mention that inter-rater reliability, which measures agreement between subjective ratings, was not assessed in this study. The primary rater for the VFSS studies was a certified SLT who had undergone training in the MBSImP and PAS used in the study. While having multiple raters and assessing inter-rater reliability would have been ideal, this may not have been practical or within the scope of this study's resources. Nonetheless, the presence of a respiratory therapist (RT) during the rating process provides an additional layer of observation and potential input to enhance the reliability of the ratings.

3.5.4 Implications for future research

Further exploration is needed to establish the association between dysphagia and IPF, and the clinical significance of such a link. It would be of interest for further studies to assess if the prevalence of dysphagia in IPF is above that expected in a population of a similar age and to compare these IPF findings with other patient groups with comorbidities that could increase the risk of swallowing problems/ aspiration (e.g. frailty).

This experience indicated that such studies are possible in patients with IPF but also underline that these are challenging. Of 18 patients approached in this study, five had already died and three deaths occurred in the 14 consenting patients before VFSS could be performed. In other settings, simple bedside screening tests are clinically informative in identifying the risk for aspiration and/or penetration, and together with selected patient-reported outcome measures, such approaches may be useful in frail patients (Patterson *et al.*, 2009; Regan;Lawson and De Aguiar, 2017). Safe approaches to augmented personalised therapy in selected patients, including speech and language intervention, could be rapidly implemented given the established model of mixed disciplinary care in IPF, if dysphagia is confirmed in further studies.

These preliminary results discussed in this chapter inspired me to conduct several research studies to explore swallowing in patients with IPF, discussed in the following chapters (4, 5 and 6).

Chapter 4. Swallowing safety and performance: The Water Swallow Test: A clinical perspective

4.1 Introduction

This is the second results chapter of my thesis. In this study, swallowing was assessed from the clinical perspective using a simple screening method: the Water Swallow Test (WST). Elements of the work described in this chapter have been previously presented as a poster presentation in the British Thoracic Society (BTS), winter meeting, 2022 (Alamer et al., 2022b) (see Appendix 8).

4.2 Literature review

Clinically, patients who are at risk of swallowing dysfunction (dysphagia) and aspiration are typically assessed using a swallow screening test. Patients with an increased risk of dysphagia subsequently receive further, more in-depth, swallowing assessments using instrumentation (Baijens et al., 2021). Clinical screening approaches used to assess dysphagia usually involve trial swallows (often called a WST) where patients swallow various amounts of water or a variety of liquids with a range of viscosities (Bours et al., 2009). In addition, dysphagia screening tests also involve taking a detailed medical history, observing clinical signs associated with dysphagia, e.g. weak cough, voice changes or drooling and may include other examinations, e.g. cervical auscultation and pulse oximetry (Speyer et al., 2022b). In order for a swallowing screening test to be useful it must be valid, reliable, non-invasive, simple to administer and the skills need to be easily transferred to other healthcare providers (Baijens et al., 2021). A WST fulfils these criteria (Brodsky et al., 2016).

The WST is a cost-effective and time-efficient (15–20 minutes) assessment used in clinical or community settings by trained healthcare providers (Logemann;Veis and Colangelo, 1999). During the test, patients are given a standardised amount of water to swallow. There are three different types of volume presentation in WSTs that are commonly used: single sips, consecutive sips and a progressive challenge in volume (Brodsky et al., 2016). All three variations evaluate how the patient

responds to swallowing water. To determine the presence or absence of penetration (when the bolus remains above the vocal cords) and/or aspiration (when the bolus passes through the vocal cords), criteria such as the patient's airway response (coughing, choking or throat clearing), changes in voice quality or the inability to complete the test are considered (Brodsky et al., 2016). A failed WST may indicate the need for further evaluation with instrumental assessments like the VFSS) or the FEES (Speyer et al., 2022b).

The single sip WST requires patients to swallow a specific amount of water, typically ranging from 1–5 to 6–20 ml (Kidd et al., 1993; McCullough et al., 2005; Wakasugi et al., 2008; Momosaki et al., 2013). In the consecutive sips WST, patients are instructed to continuously drink larger total volumes of water, usually around 90–100 ml, without pausing (Suiter and Leder, 2008; Patterson et al., 2011; Zhou et al., 2011). The third type of WST involves gradually increasing the volume of water until clinical signs of aspiration occur, such as coughing, throat clearing, post-swallow wet voice quality or reaching the maximum volume specified in the protocol (Smithard et al., 1998; Hey et al., 2013; Hassan and Aboloyoun, 2014).

Reliability testing of WSTs

Previous studies have examined the clinical effectiveness of the WST in screening for aspiration or penetration, often comparing it to the gold standard reference tests of VFSS or FEES (Brodsky et al., 2016). These studies have been conducted on diverse patient cohorts with various diseases. The findings indicate that when small volumes of water (approximately 1–5 ml) are used, the presence of coughing, throat clearing or wet voice quality after swallowing accurately indicates aspiration and/or penetration, with a sensitivity of 71% (95% CI, 63%–78%) and specificity of 90% (95% CI, 86%–93%) (Kidd et al., 1993; McCullough et al., 2005; Momosaki et al., 2013). However, the false-negative rate of the WST increases when only small volumes are used (Brodsky et al., 2016). In contrast, when patients consume consecutive sips from larger volumes (90–100 ml) without exhibiting any clinical signs of aspiration (e.g. coughing, throat clearing, post-swallow wet voice quality), the test accurately rules out aspiration (Brodsky et al., 2016). The reported sensitivity in this scenario is 91% (95% CI, 89%–93%), while the specificity is 53% (95% CI, 51%–55%) (Suiter and Leder, 2008; Patterson et al., 2011; Zhou et al.,

2011). It should be noted that the use of larger volumes of water may raise the risk of airway penetration and trigger airway responses such as coughing and throat clearing, potentially leading to an increase in false-positive results (Dozier et al., 2006). Based on these findings, combining the single and consecutive sips WST may improve the accuracy of clinical screening for aspiration.

The 100 ml WST

The 100 ml WST is a type of consecutive sips WST where patients are instructed to drink 100 ml of water as quickly and comfortably as possible while the timing and number of swallows are recorded (Patterson et al., 2009). The 100 ml WST provides valuable clinical information regarding swallowing performance and safety. This includes assessing signs of penetration and/or aspiration as well as measuring swallow volume (millilitres per swallow), swallow capacity (millilitres per second) and speed (time per swallow) (Patterson et al., 2009). Previous research has established normative values for swallow performance measures in the 100 ml WST based on a study involving 181 healthy individuals (90 males, median age 47.4 years [range 18.9–87.6], and 91 females, median age 55.4 years [range 18.9–91.3]) (Hughes and Wiles, 1996).

WSTs in patients with chronic lung diseases

In a study involving stable chronic obstructive pulmonary disease (COPD) patients with an average age of 71 ±7.7 years, WST was used to screen for swallowing dysfunction (Lindh et al., 2017). The researchers found that 49% of the patients showed signs of penetration and/or aspiration (Lindh et al., 2017). Another recent study conducted by Lindh et al. (2021) focused on COPD patients experiencing exacerbations, with an average age of 75 ±7.7 years. In this study, the WST was employed to screen for swallowing dysfunction, identifying abnormal swallowing when the swallowing capacity was ≤10 ml/second and coughing or a wet voice occurred during or after swallowing. The results showed that 80% of the patients had abnormal WST results, and an additional 70% had reduced swallowing capacity of ≤10 ml/second. These findings suggest that the WST may be a valuable screening tool for detecting swallowing dysfunction in COPD patients. However,

further research is needed to directly compare the WST with VFSS and/or FEES in this specific population for validation purposes.

WSTs in patients with idiopathic pulmonary fibrosis (IPF)

To the best of my knowledge, this study represents the first attempt to screen swallowing dysfunction in patients with IPF, as previous studies have primarily focused on patients with COPD. While COPD and IPF share certain clinical characteristics and are both chronic respiratory conditions, they differ significantly in terms of their underlying pathophysiology, treatment approaches and prognosis.

4.2.1 Aim

The aim of this study is to assess swallowing safety and performance (volume, capacity and speed) in patients with IPF using a simple screening WST.

4.2.2 Study objectives

- To explore whether WSTs can be conducted in patients with IPF via a videoconference call.
- To assess swallowing safety by identifying clinical signs of penetration and/or aspiration in subjects with IPF using two different WST conditions: single sips (5 ml and 10 ml) and consecutive sips (100 ml).
- To report baseline data of swallowing performance measures for the 100 ml WST in patients with IPF.
- To compare swallowing performance for the consecutive sips 100 ml WST in patients with IPF against published healthy non-dysphagic volunteers.

4.3 Specific materials and methods

4.3.1 Ethical approval

The study was approved by the 211628_Non-substantial amendment 3 (NSA-3) of the North East-North Tyneside2 Research Ethics Committee REC reference 18/NE/0037 (see Appendix 9: approval letter).

4.3.2 Study design

This is an observational, cross-sectional study design.

4.3.3 Specific eligibility criteria

In addition to the core inclusions and exclusions criteria described in the general methods (sections 2.3.1.1 and 2.3.1.2).

4.3.3.1 Exclusion criteria

- No access to video call program (for example, FaceTime, Zoom, Skype) for participants recruited from pulmonary fibrosis groups.

4.3.4 Recruitment

Recruitment was carried out using pulmonary fibrosis support groups and the interstitial lung disease (ILD) clinic for patients with IPF as previously described in (section 2.4.2). Patients with IPF recruited from the pulmonary support groups were all required to have access to a video call programme (FaceTime, Zoom, Skype). Prior to the study assessment, written informed consent was obtained from the patients using a printed consent form. Patients recruited from the pulmonary support groups were also required to have a partner, carer or friend living with them to serve as an extra precautionary measure against potential swallowing safety concerns.

4.3.5 Protocol

4.3.5.1Remote-based swallow testing

The WST was conducted remotely via videoconference call (Zoom Video Communications, Inc., 2021 or FaceTime). Patients were provided with two small measured cups (see Figure 7) and one large measured cup (see Figure 8) by post for administering water during the test.

Figure 7. Small measured cup.

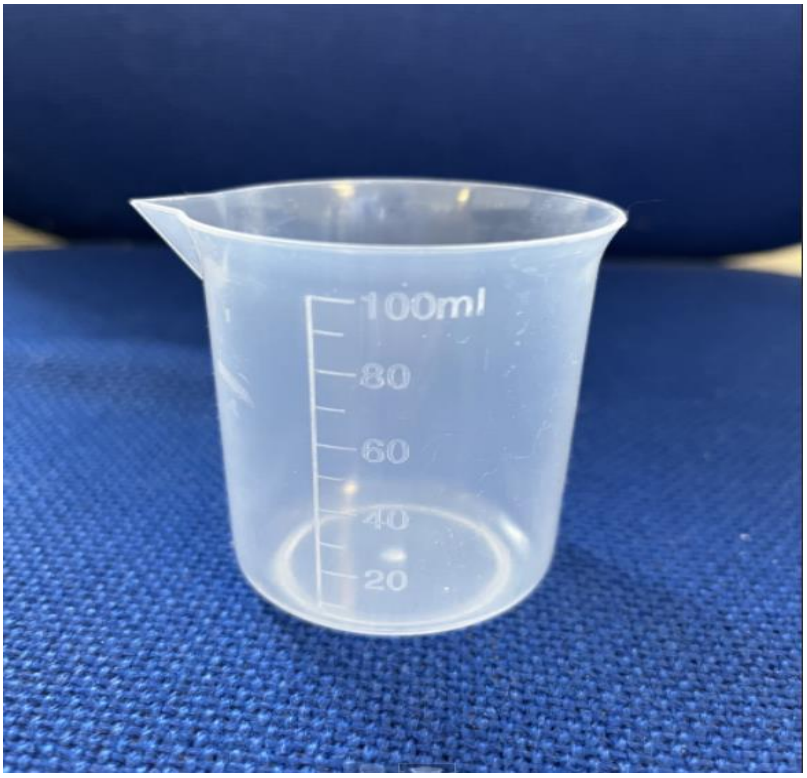

Figure 8. Large measured cup.

Patients' demographics including age, gender and smoking history were recorded. Patients were asked to record their heart rates (HR) and peripheral capillary oxygen saturation (SpO_2) if they had access to pulse oximetry at home. The respiratory rates (RR) of patients were counted by the researcher, through observation of chest movements. These measurements were collected both before and after the WST to enable continuous monitoring of the patients and identify any potential changes.

WST process

Patients were asked to measure 5 and 10 ml of tap water (room temperature) into the two small measuring cups and 100 ml of tap water (room temperature) into the large, calibrated cup. The amount of water measured out by the patients was verified by the researcher during the video conferencing call. Patients were asked to sit in an upright position with their shoulders turned to enable viewing on the video call while holding the cup so that the researcher was able to see the water in the cup. Patients were asked to say "aaaa" to identify the potential presence of a wet/gurgly voice and voice quality prior to and following each drink.

Instructions:

Single sips:

- For training purposes only, patients were asked to drink 5 ml from the small measuring cup in one single swallow that was not counted as part of the analysis.

  The following steps were included in the analysis:

- From the small measuring cup, patients drank 5 ml of water in one single swallow.

- From the small measuring cup, patients drank 10 ml of water in one single swallow.

Consecutive sips:

- From the large measuring cup, patients drank 100 ml of water as quickly and as comfortably as possible. The number of swallows and the time taken for patients to finish the water were recorded.

To evaluate swallowing performance, I used a stopwatch to time the test, starting from when the water reached the patient's bottom lip to when the larynx came to rest following the last swallow, which was usually accompanied by other signals, such as exhalation, phonation or opening of the mouth. The number of swallows was counted simultaneously by me by observing the upward movement of the patient's Adam's apple (one elevation equals one swallow). The volume of water swallowed was recorded and if the patient was unable to complete the task, the residual water remaining in the measured cup was recorded. Following each WST, the patient's RR, HR and SpO_2 were recorded (see Appendix 10: Water Swallow Test evaluation form).

4. 3.5.2 Clinic-based swallow testing

Prior to the WST, patients' demographics including age, gender and smoking history were recorded. In addition, the Medical Research Council dyspnoea scale (MRC), body mass index (BMI) and pulmonary function tests including forced vital capacity

(FVC) (% of predicted) and transfer factor of lung for carbon monoxide (TLCO) (% of predicted) were collected from patients' medical records.

The RR was counted by observing the patient's chest movement. The HR and SpO_2 readings were measured by pulse oximetry. The WST was conducted face-to-face at the clinic, using a very similar protocol to that used for remote assessments, as reviewed above. Patients were provided with two small (Figure 7) and one large (Figure 8) measuring cups to administer the water test in the clinic; this used the same cups used in the remote assessments of water swallowing.

WST process

I measured 5 and 10 ml of bottled water (room temperature) into the two small measuring cups and 100ml of bottled water (room temperature) into the large measuring cup. Patients were seated in an upright position. Patients were asked to say "aaaa" to identify the presence of a wet/gurgly voice and voice quality prior to and following each drink.

Instructions:

Single sips:

- From the small measuring cup, patients drank 5 ml of water in one single swallow. This was for training purposes and was not part of the analysis.

  The following steps were included in the analysis:

- From the small measuring cup, patients drank 5 ml of water in one single swallow.

- From the small measuring cup, patients drank 10 ml of water in one single swallow.

Consecutive sips:

- Using the large measuring cup, patients drank 100 ml of water as quickly and as comfortably as they possibly could. The number of swallows and the time taken for patients to finish the water were recorded.

Using a stopwatch, the timing was started when the water reached the patient's bottom lip, finishing when the larynx came to rest following the last swallow, which was usually accompanied by other signals, such as exhalation, phonation or opening of the mouth. The number of swallows was counted simultaneously by the researcher by palpating the thyroid cartilage for laryngeal elevation. The volume of water swallowed was recorded, and if the patient was unable to complete the task, the residual water was measured by syringe, using a minimum scale of 1 ml. Following the WST, the RR, HR and SpO_2 measurements were taken (see Appendix 10: Water Swallow Test evaluation form).

4.3.5.3 Clinical end point/response

Data was divided according to the criteria for the presence or absence of penetration and/or aspiration as described by (Suiter and Leder, 2008). Patients who were unable to complete the WST were identified by:

- Inability to drink the whole amount of water.
- Coughing or choking following the swallowing of the water.
- Post-swallow wet voice quality.

4.3.5.4 Swallowing performance parameters for the 100 ml WST

- Swallow volume (ml per swallow) was defined as the volume of water the patient drank in one swallow and it was calculated by dividing the total volume the patient drank by the total number of swallows recorded by the researcher for that patient. A high swallow volume score indicates a better swallowing performance.

- Swallow capacity (ml per second) was defined as the ability of the subject to swallow a specific amount of water in a given time frame and was calculated as the volume of water swallowed in millilitres divided by the time it took to swallow the water. A high swallow capacity score indicates a better swallowing performance.

- Swallow speed (time per swallow) was defined as the period of time it took the patient to complete a single swallow and was calculated as the total number of

swallows divided by the total time taken. A low swallow speed suggests better swallowing performance.

Patients who choked when swallowing in either the single sip (5 and 10 ml) or consecutive sips (100 ml) were asked to stop immediately. This was recorded as failure to complete the WST regardless of whether they had finished drinking the water or not.

4.3.6 Analysis

Continuous variables were presented using medians, range (minimum–maximum) and the 25th and the 75th percentile (Q1–Q3). Categorical variables were presented using frequencies and percentages. Analysis was largely descriptive. Where necessary, non-parametric tests were used because of the sample size (n: 33). The Mann–Whitney U test was used to compare whether differences in the swallowing performance measures were present in the two independent groups (males and females).

Bivariate correlation coefficients were calculated to measure the relationships between continuous variables. Pearson's product moment correlation coefficient (r) was used when data was normally distributed, and Spearman's rank correlation coefficient (rho) was used when extreme values were present. Measures of correlation range from −1 to 1 (0 for no correlation, 1 for a perfect positive correlation and −1 for a perfect negative correlation) (Mukaka, 2012). Data were considered statistically significant with a p-value of <0.05.

Data on swallowing performance for the consecutive sips WST (100 ml) including swallow volume and capacity were analysed against previously published data from a healthy control population, matching for age and gender where appropriate (Hughes and Wiles, 1996). In Hughes and Wiles (ibid), swallow volume measures were reported in medians and the 25th and the 75th percentile (Q1–Q3); however, the swallow capacity measures were reported in means and ±(SD). The Lower Level Normal (LLN) was used as the cut-off value for abnormality for swallow capacity in IPF. The LLN was taken as the lower limit of the 95% confidence interval of the normative data. All statistical analyses and graphic presentations in this chapter were performed using Minitab statistical software (version 21, Minitab Inc., State

College, PA, USA) and Microsoft Excel 2016 (Microsoft Corporation, Washington, DC, USA).

4.4 Results

A total of 34 patients with IPF were recruited to the study. Fourteen patients were recruited from support groups and 20 patients were recruited from ILD clinics, where the WST was completed remotely or face-to-face during the clinical visit, respectively. One patient who presented at the clinic was excluded from the study due to a history of tongue cancer.

4.4.1 Patients' demographics and clinical characteristics

Patients' demographics and clinical characteristics are presented in Table 8. Thirty-three patients with IPF completed the WST and were included in the analysis. The median age of patients was 72.0 years ([min–max] 54–92). Most patients were male (23/33, 69.7%) and non-smokers (22/33, 67%). The clinical characteristics for the patients recruited from the ILD clinic (n: 19) were as follows (median, min–max): BMI (27.7 kg/m^2, 22.9–40.0), MRC (3, 2–4), FVC% of predicted (69%, 24–83) and TLCO% of predicted (39%,18–76).

Factor	Level	Value
N		33
Age in years, median (minimum–maximum)		72 (54-92)
Sex	Female	10 (30%)
	Male	23 (70%)
Smoking status	Current smoker	1 (3%)
	Ex-smoker	10 (30%)
	Non-smoker	22 (67%)
N		19
BMI, kg/m^2, median (minimum–maximum)		27.7 (22.9–40.0)
MRC dyspnoea scale, median (minimum–maximum)		3 (2–4)
FVC% of predicted, median (minimum–maximum)		69% (24–83)
TLCO% of predicted, median (minimum–maximum)		39% (18–76)

Table 8. Median (min-max) or (%) for the patients' demographic and clinical characteristics.
N: number; BMI: body mass index; MRC: Medical Research Council dyspnoea scale; FVC% of predicted: forced vital capacity % of predicted; TLCO% of predicted: transfer factor of lung for carbon monoxide % of predicted.

Factor	Pre	Post
N	33	33
RR (breath/minute) median (minimum–maximum)	22 (16–32)	20 (16–34)
HR (beat/minute) median (minimum–maximum)	77 (62–103)	77 (64–101)
SpO_2 (%) median (minimum–maximum)	95% (91%–99%)	96% (91%–99%)

Table 9. Median (min-max) or (%) of the clinical characteristics pre- and post- the 100 millimetres (ml) WST.
N: number; RR: respiratory rate; HR: heart rate; SpO2: peripheral capillary oxygen saturation.

4.4.2 Study findings

4.4.2.1To explore whether WSTs can be conducted over a videoconference call with IPF patients

Patients recruited from IPF pulmonary fibrosis support groups completed the WST remotely via videoconference call (Zoom or FaceTime). The number of IPF patients who were able to complete the WST over a videoconference call is presented in Figure 9.

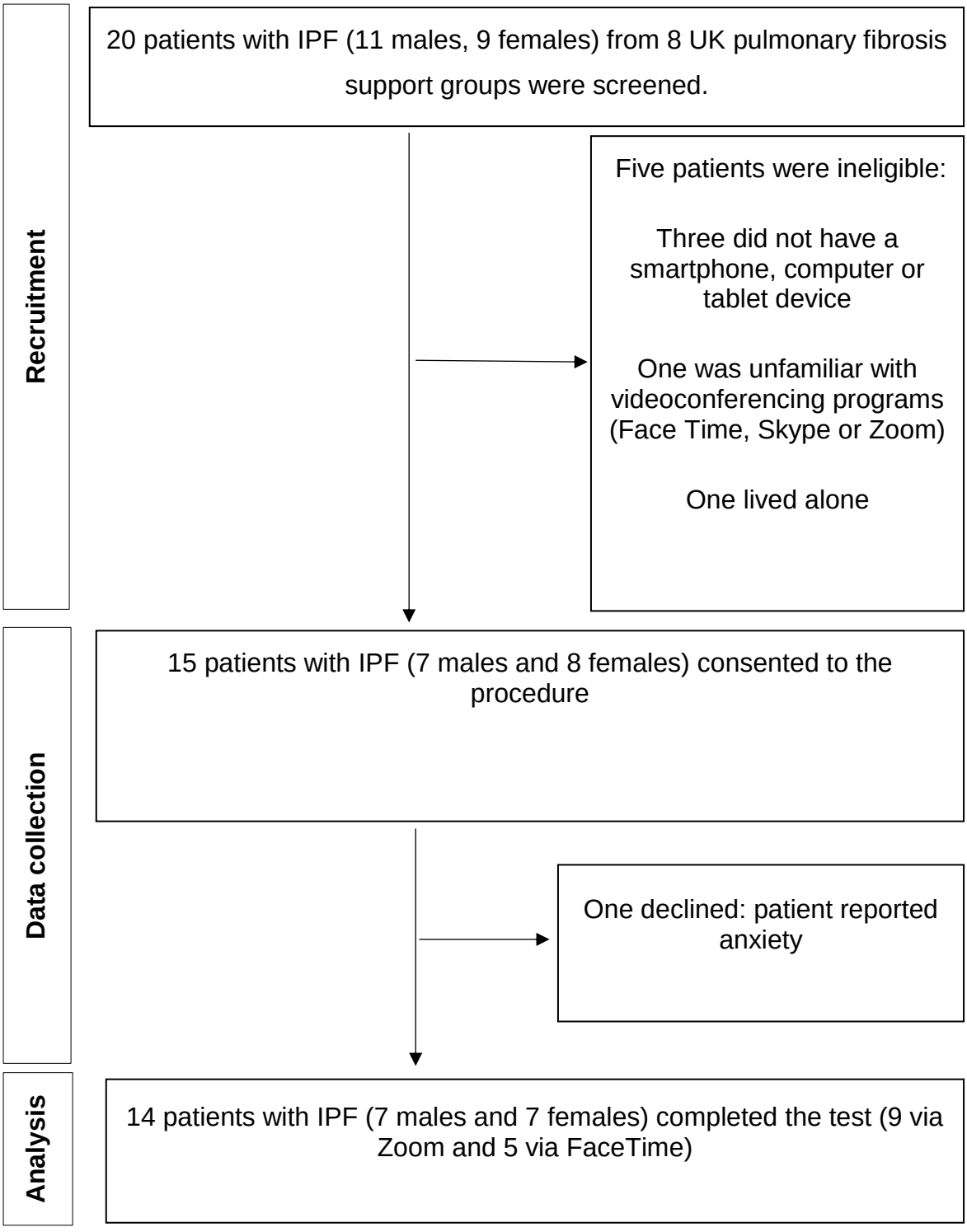

Figure 9. Consort flow diagram for support groups patients, recruitment study.

4.4.2.1 The clinical signs of penetration or aspiration in subjects with IPF using remote testing and clinic assessment using two different WST conditions: single sips (5 ml and 10 ml) and consecutive sips (100 ml)

All patients with IPF were able to complete both the single sips (5 ml) and consecutive sips (100 ml) WST protocol. Six patients (18%: 4 males, 2 females) with an age range of 61 to 92 years failed the WST and presented with signs of penetration and/or aspiration. There were no signs of choking during any of the assessments (see Figure 10).

Findings:

- One patient (3%: female), aged 70 years old, coughed and had a wet voice quality after drinking 10 ml during the single sip WST, and then completed the 100 ml consecutive sips WST.
- Four patients (12%; 3 males, 1 female) with an age range of 61 to 92 years old, coughed following the 100 ml consecutive sips WST.
- One patient (3%: male), aged 70 years old, coughed and had a wet voice quality following the 100 ml consecutive sips WST.
- None of the 33 patients had either an airway response or a wet voice quality following the 5 ml single sip WST.

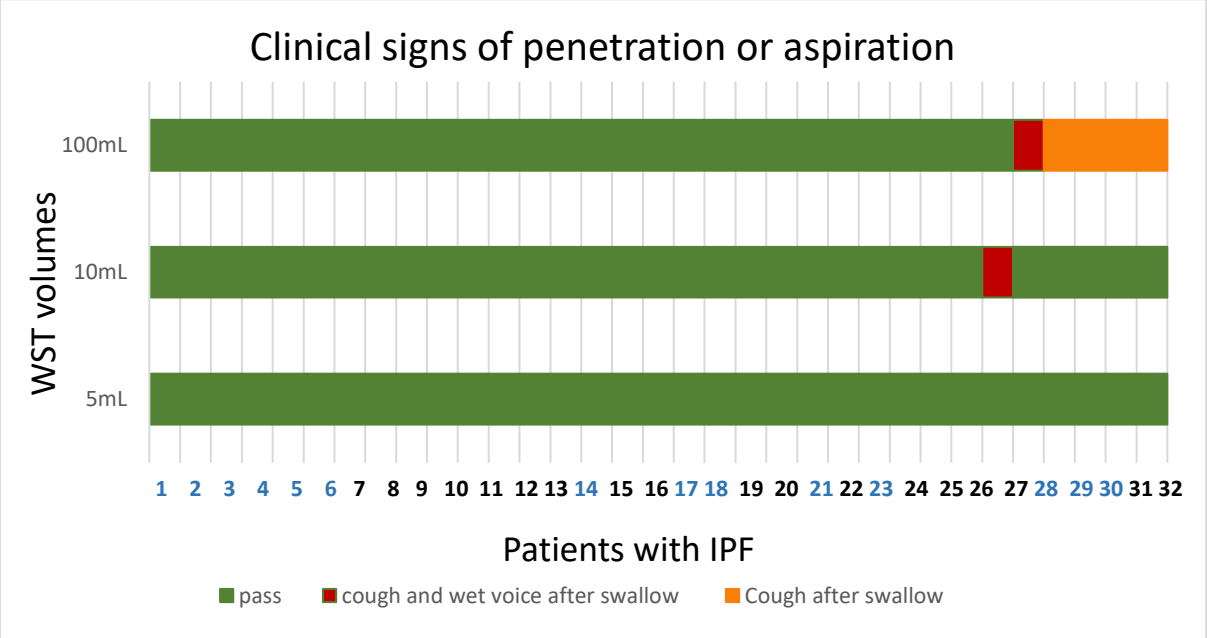

Figure 10. Clinical signs of penetration and/or aspiration in patients with IPF.
Blue numbers on the x-axis denote patients recruited remotely from pulmonary fibrosis support groups and black numbers represent patients recruited face-to-face from the ILD clinic. Green

cells indicate a test completed safely, i.e. patients had no airway response nor a wet voice quality after swallowing. Orange cells indicate failed tests where patients had an airway response (cough or throat clearance) after swallowing. Red cells indicate failed tests where the patients had both an airway response (cough or throat clearance) and wet voice quality after swallowing.

4.4.2.3 Baseline data and comparison of swallowing performance for the 100ml WST in patients based on gender and age

A summary of the swallowing performance measure scores for IPF patients is presented in Table 10. All 33 patients with IPF were able to complete the 100 ml WST. One patient coughed and had a wet voice quality, and four patients coughed following swallowing the 100 ml of water (Figure 10).

During the analysis, it became clear that among the three performance measures, swallow speed was the least represented of the level of impairment and did not effectively capture differences between patients. To illustrate, consider two patients with a swallow speed of 1 second: one completes the task in 12 seconds with 12 swallows, while the other completes it in 6 seconds with six swallows. Due to the considerable amount of data and variables involved, as well as the aforementioned reasons, swallow speed was excluded from the analysis.

Factor	**Value**
N	33
Volume in (ml/swallow), median (Q1–Q3) Mean ±(SD)	16.6 (12.5–20.0) 17±6.7
Capacity in (ml/second), median (Q1–Q3) Mean ±(SD)	10.6 (7.6–17.6) 12.5±6.6

Table 10. Median (Q1 and Q3) and mean ±(SD) swallowing performance parameters for all patients from the 100 millimetres (ml) WST.
N: number; Q1: 25th percentile; Q3: 75th percentile; SD: standard deviation.

Swallowing performance and gender

Generally, males had higher swallowing volume and capacity than females (see Figures 11 and 12). Males had a significantly higher swallow volume than females, with a mean rank difference of 8.2 ml ($p = .006$); however, there was no significant gender effect on swallow capacity ($p > .05$).

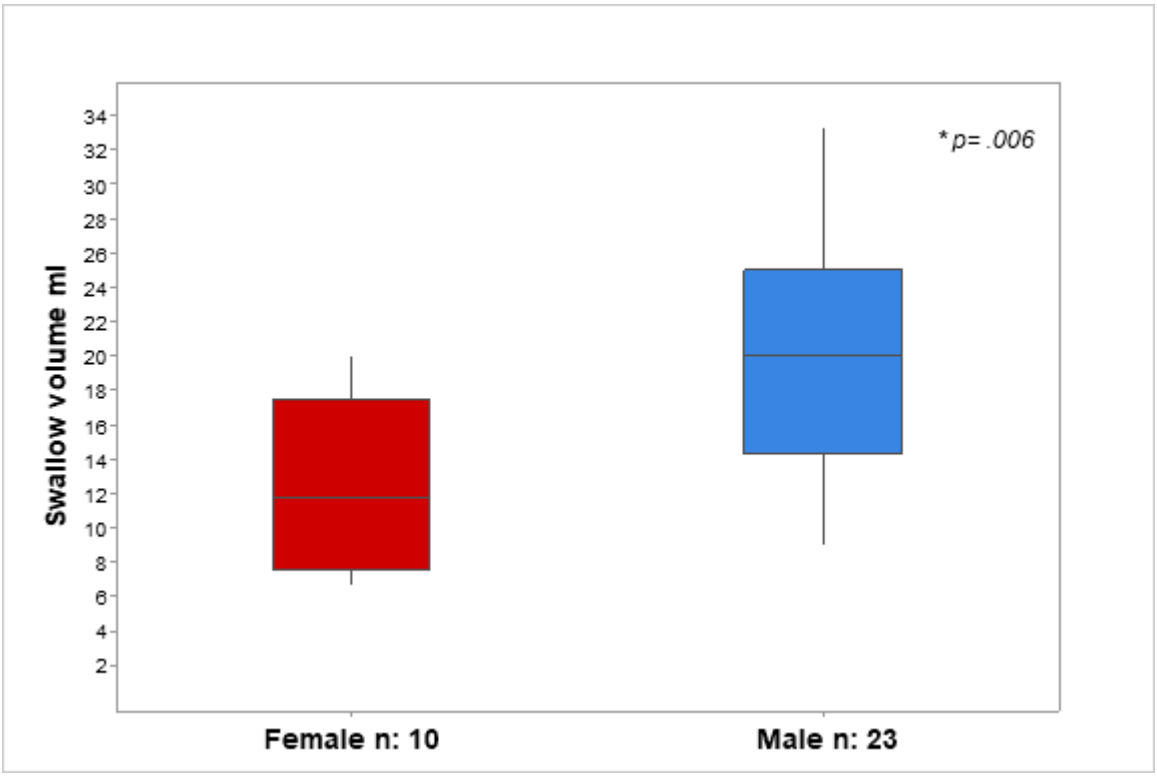

Figure 11. Median (Q1-Q3) swallow volume scores in millilitres (ml) grouped by sex. Significance was set at * p-value <.05. A higher score indicates a better swallow performance.

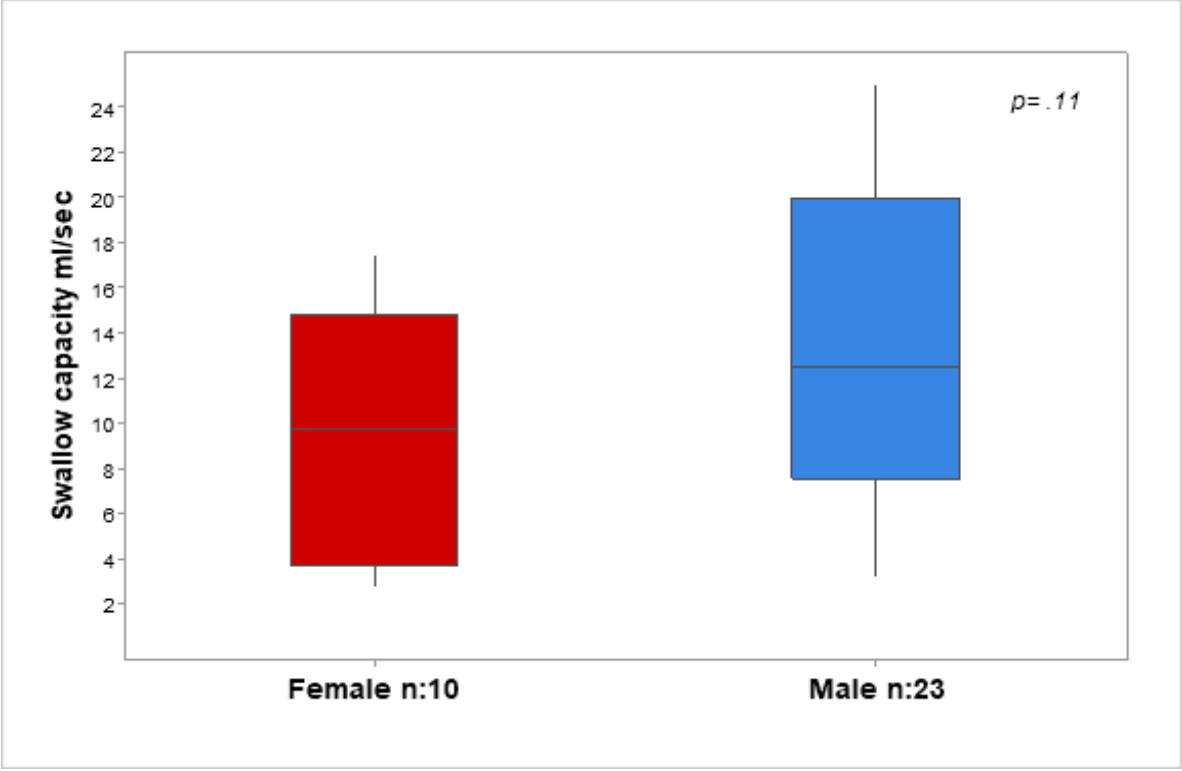

Figure 12. Median (Q1-Q3) swallow capacity scores in millilitres per second (ml/sec) grouped by sex.

Significance was set at p-value < .05. A higher score indicates a better swallow performance.

Swallowing performance based on age

The median age of patients was 72.0 years ([min–max] 54–92). The relationship between swallowing performance measures and age is depicted in Figures 13 and 14. There was a non-significant weak negative correlation between age and swallow volume (r = −.187, p-value = .20) and swallow capacity (r = −.285, p-value = .11), indicating that as age increases, the swallow volume and capacity tends to decrease.

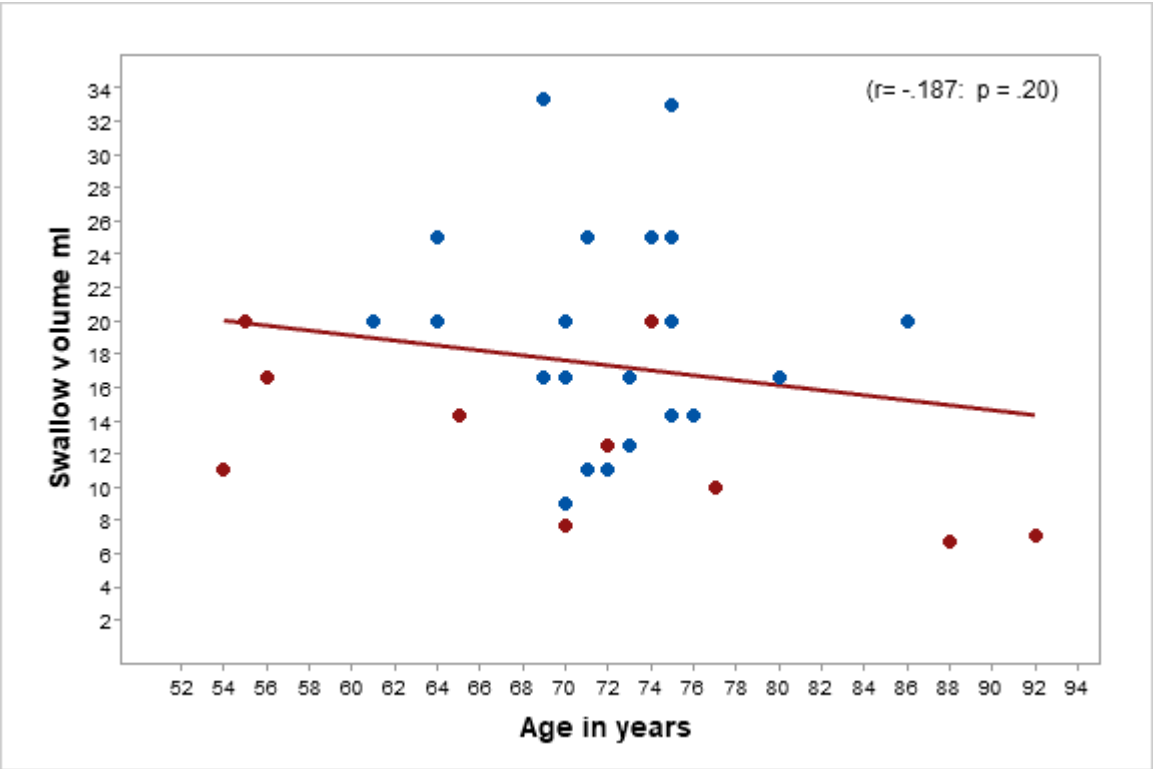

Figure 13. Correlation between age (years) and swallow volume scores (millilitres).
Red dots denote females (n=10) and blue dots males (n=23). (r: −.187, p-value: .20).

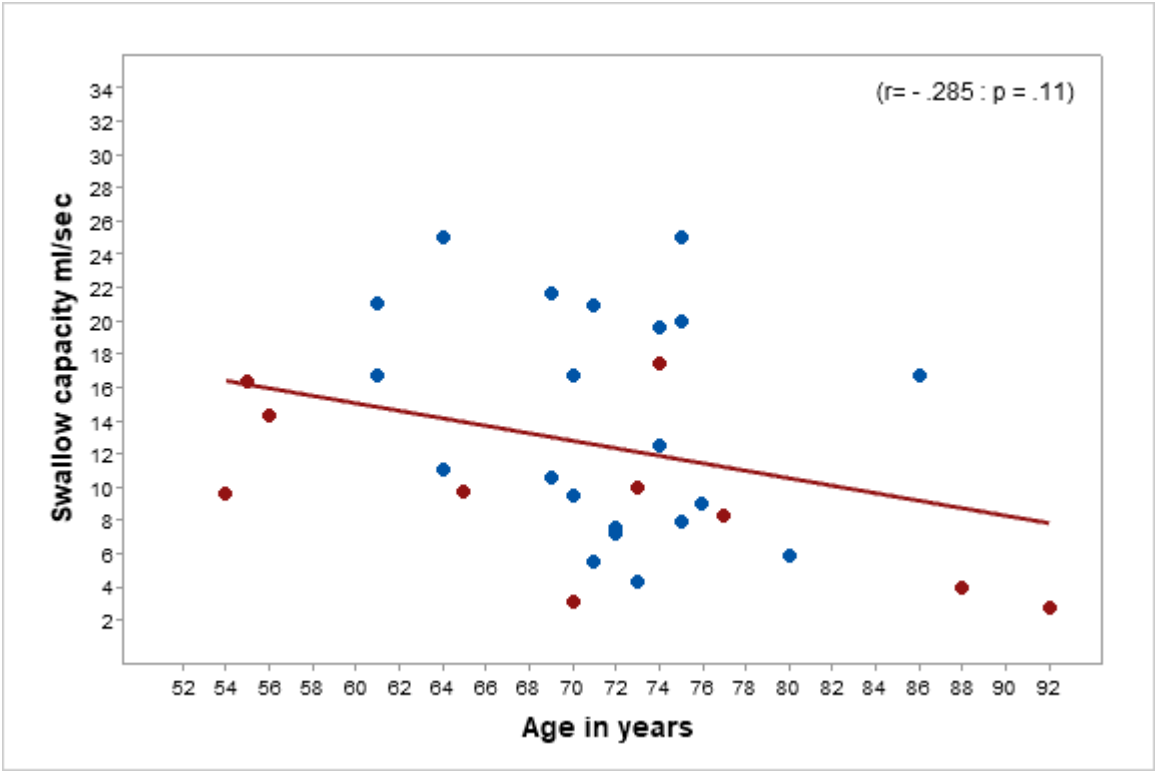

Figure 14. Correlation between age (years) and swallow capacity scores (millilitres/second). Red dots denote females (n=10) and blue dots males (n=23). (r = −.285: p-value: 0.108).

Comparison of baseline swallowing performance data following the 100 ml WST in IPF patients with published normative data

Swallow volume in ml

Generally, the swallow volume was lower in the IPF patients when compared with the published data from healthy control population, matching for age and sex (Hughes and Wiles, 1996). Eighteen of the IPF patients had a swallow volume below the 25th percentile of the swallow volume for the normative data, where lower scores indicate a decline in swallowing performance. All patients that failed the WST and had signs of penetration and/or aspiration had swallow volume scores below the 25th percentile of the normative data (see Figure 15).

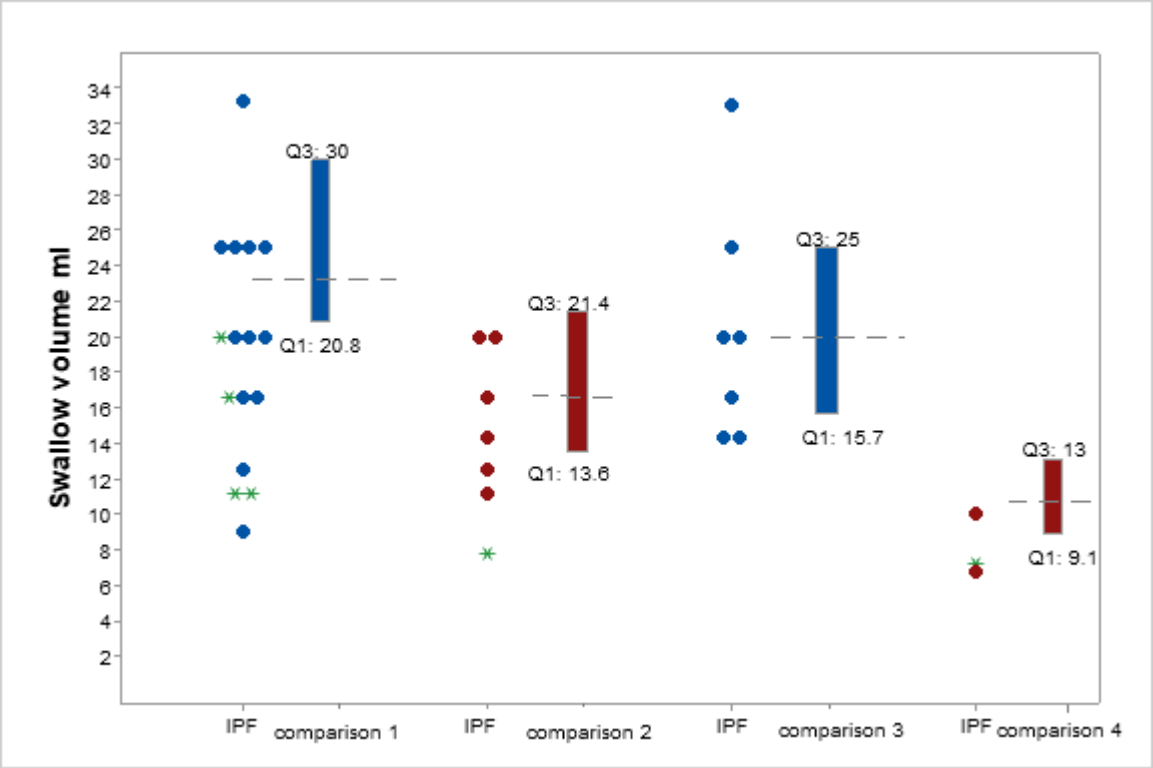

Figure 15. Median and 25th and 75th percentile swallow volume scores (ml) for IPF patients (n:33) and published normative healthy subjects (n:181).
Group comparisons are based on the subgroups described in (Hughes and Wiles, 1996). Comparison 1: male < 75 years (n: 26); comparison 2: female < 75 years (n:35); comparison 3: male > 75 years (n:12); comparison 4: female > 75 years (n: 10). Blue dots denote males and red dots females (both IPF). Blue rectangles denote males (healthy normative group), and red rectangles females (healthy normative group). Dashed lines denote medians for the healthy normative group and green asterisks denote patients who were unable to complete the WST and presented with signs of penetration and/or aspiration.

Swallow capacity in ml/second

Swallow capacity in IPF patients was compared to the published data from the healthy control population, matching for age and sex using means and LLNs (Hughes and Wiles, 1996). A total of 15% of IPF patients had a swallow capacity lower than the LLN of the swallow capacity for the normative data, which may indicate a decline in swallowing performance (see Figure 16).

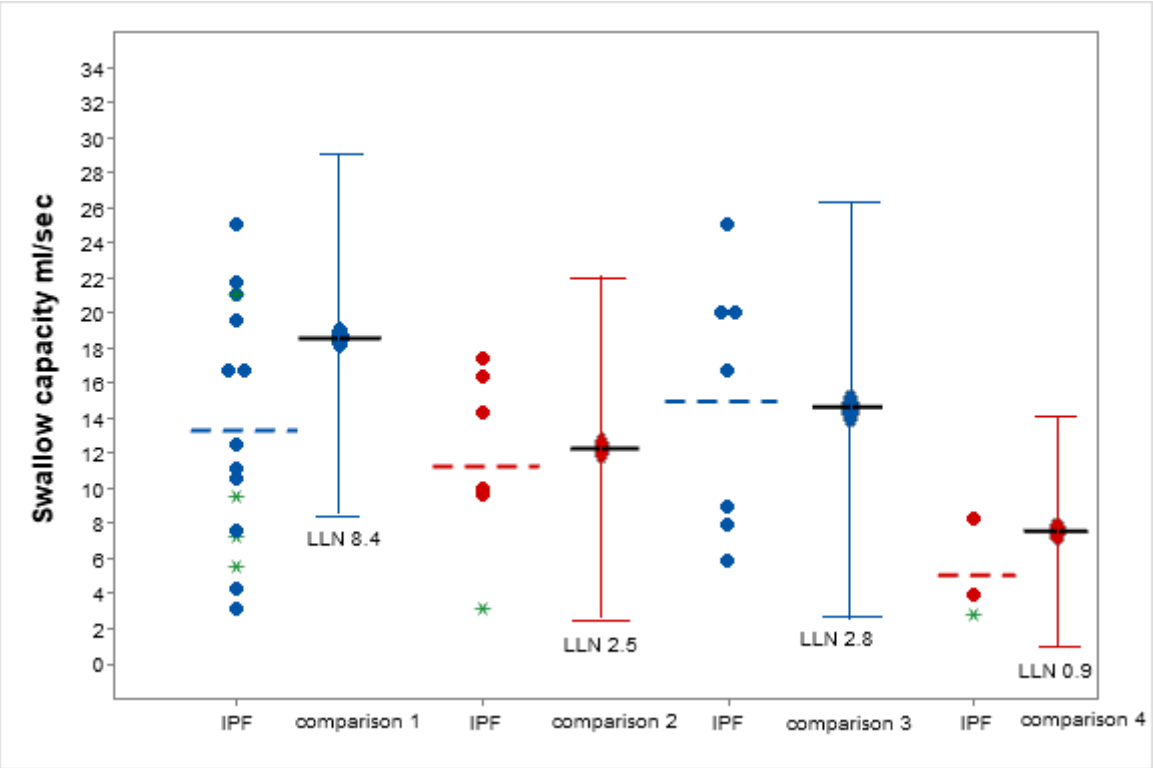

Figure 16. Swallow capacity in millilitres per second (ml/sec) in IPF patients (n:33) and published normative healthy subjects (n:181).
Group comparisons are based on the subgroups described in (Hughes and Wiles, 1996). Comparison 1: male < 75 years (n: 26); comparison 2: female <75 years (n:35); comparison 3: male > 75 years (n:12); comparison 4: female >75 years (n: 10). Blue dots denote males and red dots females (both IPF). Blue interval plots denote males (healthy normative group), and red interval plots females (healthy normative group). Dashed lines denote means for the IPF patients. Solid black lines donate means for the healthy normative group. Green asterisks denote patients who were unable to complete the WST and presented with signs of penetration and/or aspiration. The LLN was taken as the lower limit of the 95% confidence interval of the normative data (Hughes and Wiles, 1996).

4.5 Discussion

The aim of this chapter was to assess swallowing safety and performance in patients with IPF using a simple screening WST. This is the first study to screen for swallowing dysfunction in patients with IPF using a WST. Furthermore, here we have demonstrated novel findings that a WST can be performed online in the community and hospital clinics. Clinical signs of penetration and/or aspiration were assessed, and measures of swallow performance (volume and capacity) were analysed and compared against healthy non-dysphagic published data, while investigating the effects of gender and age. The key findings presented here are: 1) WSTs were successfully completed by IPF patients, including via videoconference calls (Zoom or FaceTime); 2) signs of penetration and/or aspiration were identified using the WST; 3) there was a signal that swallowing performance was influenced by gender and age; and 4) IPF patients had a lower swallowing performance when compared with age- and sex-matched, non-dysphagic published data. These findings indicate that, in principle, IPF patients may suffer from dysphagia and that the WST may be a useful clinical screening tool to identify IPF patients at risk of swallowing dysfunction. This might identify patients who are appropriate for further definitive investigation of swallowing physiology and allow earlier diagnosis and treatment of a previously unidentified treatable trait in people with IPF.

4.5.1 Study objective 1: exploring whether WSTs can be conducted over a videoconference call with IPF patients

In this study, for the first time, the WST was conducted remotely via videoconference call (Zoom or FaceTime) in IPF patients recruited directly from pulmonary fibrosis support groups. The recruitment phase commenced during the third UK COVID-19 lockdown in January 2021 when it was not possible to have face-to-face interactions with individuals suffering from IPF. This was due to the heightened vulnerability of IPF patients, as recognised by the NHS, which advised them to shield. Moreover, face-to-face WST screenings involving swallowing assessments can generate aerosols and droplets when patients cough or clear their throats. To tackle the challenges posed by the lockdown and the need to protect high-risk patients, virtual WST emerged as a viable research tool. Not only did this approach prove to be effective for this study, but it also holds the potential for future

clinical benefits by minimising the requirement for in-person contact (Ward et al., 2014; Borders et al., 2021; Miles et al., 2021; Ward et al., 2022).

Existing studies have indicated that remote swallow screening procedures offer several advantages, including increased flexibility and accessibility for patients. By eliminating barriers such as travel and time commitments, remote screenings enable patients to participate more easily and facilitate early detection of swallowing dysfunction (Burns et al., 2019; Ward et al., 2021). In this study, a total of 14 patients were recruited from support groups to participate in the remote WST sessions. Among them, nine patients successfully completed the WST using Zoom, while five utilised FaceTime for the remote assessments. However, six patients were unable to undergo the tests for various reasons. Among these six individuals, three did not possess the required electronic devices, and one person was unfamiliar with remote communication methods.

Furthermore, this study found that conducting the WST remotely was able to identify three patients who exhibited symptoms of penetration and/or aspiration, such as coughing, throat clearing and voice changes after swallowing. While this is the first study of its kind focusing on IPF patients, similar findings have been reported in individuals with movement disorders. Borders et al. (2021) conducted a study to assess the reliability of the timed water swallow test (TWST), the test of masticating and swallowing solids (TOMASS) and observations of oral intake performed remotely in patients with movement disorders who were referred for dysphagia evaluation. Seven researchers evaluated the tasks and reported high levels of reliability across all measures. Specifically, the TWST outcome measures, including total swallow time, number of swallows and amount of water consumed, showed "good" to "excellent" interrater reliability (ICC > .80). Furthermore, the clinical signs of penetration and/or aspiration demonstrated "excellent" reliability with over 92% agreement (Borders et al. 2021). It is worth noting that while these results are promising, Borders et al. (ibid) only compared outcome measures in a single testing situation (videoconferencing platform) without directly comparing them to face-to-face testing.

In a more recent study conducted by Karlsson et al. (2023), they examined the reliability of TWST and TOMASS both in face-to-face clinical visits and remotely

among patients with various clinical conditions. The study revealed a 76% agreement in TWST screening outcomes between face-to-face and remote assessments. Additionally, there were significant positive associations observed for the number of swallows and total swallow time ($\tau = 0.46$, $p < 0.001$, and $\tau = 0.58$, $p < 0.001$, respectively). Regarding the clinical signs of penetration and/or aspiration, there was a 78% agreement in voice changes ($\chi^2 = 55$, $p < 0.001$), while the agreement for coughing was not statistically significant ($\chi^2 = 0.94$, $p = 0.33$) between face-to-face and remote testing. These findings suggest that virtual testing for assessing swallowing dysfunction may be a viable alternative to face-to-face assessments (Karlsson et al., 2023). However, further research is necessary to determine how virtual testing for swallowing dysfunction compares to in-person assessments and to establish its suitability for routine use in community settings.

The American Speech-Language-Hearing Association (ASHA) has provided guidelines for patients who may benefit from telemedicine, which involves virtual testing (American Speech-Language-Hearing Association, 2021). These guidelines consider factors such as physical ability, cognitive functioning, speech intelligibility, language function, hearing and visual abilities, and access to and proficiency in using technology. In this study, three patients were deemed ineligible during the screening process due to their lack of access to the necessary electronic devices (smartphone, tablet or computer) required for remote assessments.

While technology and handheld devices have seen significant advancements, there may still be a "technology gap" that could limit engagement among certain patient groups. However, as technology continues to progress, the issue of access to electronic devices may become less prevalent. Nevertheless, it is important to make accommodations for these patients to ensure their engagement and optimal clinical management (Barney et al., 2020).

A consideration in the study was the inclusion of a caregiver during the online testing to ensure safety in case of an emergency and to provide support in the event of an emotional response (Sharma et al., 2013). Consequently, one participant was deemed ineligible after the screening process as they were living alone and did not have a caregiver present during the online testing.

4.5.2 Study objective 2: clinical signs of penetration and/or aspiration in patients with IPF using two different WST conditions: single (5 and 10 ml) and consecutive sips (100 ml)

In the current cohort of patients, a total of six patients (18%) experienced coughing and/or voice changes following the administration of the WST. However, it is challenging to determine whether these symptoms are a result of aspiration and/or penetration of the water or if they are attributed to a chronic cough, which is frequently reported in patients with IPF (Van Manen et al., 2016). Previous studies conducted on patients with COPD have shown a potential risk of cricopharyngeal dysfunction, leading to bolus retention in the piriform sinus (Stein et al., 1990). Another recent study focusing on hospitalised COPD patients found that four patients who exhibited clear signs of penetration and/or aspiration were unable to complete the 150 ml WST (Lindh et al., 2021). These findings suggest that the WST could be a useful tool to identify clinical signs of aspiration and/or penetration, but further investigation is needed to determine if these symptoms are directly caused by aspiration and/or penetration or if they are related to chronic cough. This subgroup of patients may represent a distinct minority subset who require additional definitive tests to evaluate their swallowing physiology.

4.5.3 Study objective 3: baseline swallowing performance data comparing age and gender using the 100 ml WST in patients with IPF

4.5.3.1 Gender

IPF is a serious lung condition that is typically more prevalent in males, who make up approximately 79% of all IPF cases (Jo et al., 2017). This gender disparity was also observed in the present study, where 70% of the cohort consisted of male participants.

Despite the higher prevalence of IPF in males, our data revealed that males had a higher swallow capacity and significantly higher swallow volume compared with females. These findings align with previously published normative data in healthy non-dysphagic adults, which also demonstrated higher swallow volume and capacity in males compared with females (Hughes and Wiles, 1996; Sarve;Krishnamurthy and Balasubramanium, 2021). The anatomical differences,

specifically the larger oral and pharyngeal cavity volume in males, likely contribute to their ability to swallow a greater volume of water (Alves et al., 2007).

However, only one study has explored the impact of gender on swallowing function in respiratory disease patients. Lindh et al. (2017) investigated the effect of gender on dysphagia burden (the number of swallow symptoms) in stable COPD patients. In contrast to this study, they reported no significant differences between gender and dysphagia burden. It is important to note that this study did not assess swallowing performance measures between males and females in COPD patients (Lindh et al., 2017).

4.5.3.2 Age

As individuals age, there is a general deterioration in physiological functioning throughout the body, which often leads to an increased burden of diseases. IPF is considered an age-related disease, as highlighted by the BTS national registry, where the average age of presentation was reported as 74 years (Joshi and Nagji, 2022). This age range aligns with the median age of 72 years observed in the current study.

In regard to swallowing performance, previous researches have shown age-related changes in various aspects, such as the time it takes for a swallow response (Namasivayam-MacDonald;Barbon and Steele, 2018), prolonged transition time between the oral and pharyngeal stages of swallowing (Rosenbek et al., 1996; Dua et al., 1997; Logemann et al., 2000), extended hyolaryngeal excursion (Feng et al., 2013), reduced pharyngeal pressure generation and loss of muscle mass, that impact upon swallowing (Feng;Zhang and Wang, 2023).

Similarly, an association was observed between advancing age and an increase in the duration of each swallow, along with a decrease in swallow volume and capacity. These findings suggest that alterations in swallowing performance measures may be linked to age-related changes in healthy swallowing. The term "presbyphagia" is used to describe age-related modifications in the mechanisms of swallowing, which can disrupt the pattern of deglutition and make older adults more susceptible to dysphagia. Although not confirmed in the present study, there is a possibility that the prolonged duration of each swallow observed in IPF patients may serve as an

age-related compensatory mechanism to mitigate the risk of aspiration (Sarve;Krishnamurthy and Balasubramanium, 2021). Regardless of the underlying cause, it is evident that IPF is more prevalent among an ageing population. Detecting swallowing dysfunction at an earlier stage may facilitate prompt diagnosis and treatment for these individuals.

4.5.4 Study objective 4: comparing baseline data of swallowing performance for the 100 ml WST in IPF participants against published normative data

Overall, it appears that IPF patients exhibit poorer swallowing performance compared with a non-dysphagic group mentioned in a published study (Hughes and Wiles, 1996). Previous research conducted on the COPD population used a capacity index of 10 ml/sec as the lower normal level and a time of less than or equal to 15 seconds to complete drinking 150 ml of water (WST) as the upper limit of normal (Lindh et al., 2017; Lindh et al., 2021). The findings indicated that 35.3% of stable COPD patients, 2% of patients with cardiac disease and 70% of patients with COPD exacerbation took longer than 15 seconds to complete drinking the water, exceeding the upper limit of normal. In this study, the protocol involved drinking 100 ml of water, so theoretically, with a capacity index of 10 ml/sec, the expected time to complete drinking the water would be 10 seconds. Based on this calculation, approximately 45% of IPF patients took longer than the upper limit of normal (10 seconds).

4.5.5 Strengths and limitations

This prospective, cross-sectional study is the first of its kind to assess swallowing dysfunction in patients with IPF using a simple swallow screen test (WST) both remotely and in-person. Clinical swallow screens are important in this population - and this test can be easily conducted by clinicians with minimal training. However, it is important to acknowledge that the study does have its limitations: (1) The timed nature of the 100 ml WST puts patients under pressure to consume the water quickly, which may not accurately reflect their typical drinking habits or their performance in everyday situations. The instructions provided to participants involve consuming the water as quickly and comfortably as possible, but the interpretation

of "comfortable" can vary from person to person. (2) The test was conducted in a single setting for each patient, either online for participants recruited from support groups or in-person at the ILD clinic. No evaluation was performed to compare the agreement between the two settings. It is important to highlight that this study is exploratory, and the objectives did not involve comparing outcomes between the two settings. Nevertheless, measures were taken to maintain consistency across settings and to minimise variation in the testing environment, such as using identical instructions and standardised measuring cups. (3) Due to COVID-19 restrictions, it was not possible to conduct a reference test using either FEES or VFSS to validate the study's findings. (4) Patients were selected through convenience sampling, which resulted in the exclusion of certain individuals who might have been suitable for the study. This exclusion could be due to factors such as lacking the necessary digital equipment or skills, as well as issues related to technology accessibility for patients recruited from support groups. Furthermore, the study did not consider the severity of IPF as an inclusion criterion, even though it could have potentially influenced the swallowing performance measures and the resulting outcomes. However, considering the circumstances of conducting the study during the COVID-19 pandemic, the chosen convenience sampling method was considered appropriate to fulfil the study's objectives.

4.6 Conclusion

This chapter indicates that an identifiable minority of people with IPF may be at risk for dysphagia and that the WST, including remote testing, is a practical candidate to identify patients who require or might benefit from more definitive investigation. This could represent a previously unidentified treatable trait present in some people with IPF where treatment options are very limited.

Chapter 5. Swallowing and throat symptoms: The patients' reported assessment: A patients' perspective

5.1 Introduction

This is the third results chapter of my thesis. In this study, swallowing was assessed from the patients' perspective using validated questionnaires. Elements of the work described in this chapter have been presented as oral presentation in the European Respiratory Society (ERS) International Congress, Milan 2023 (Alamer *et al.*, 2023b) (see Appendix 11).

5.2 Literature review

Clinical evaluation of swallowing and the use of instrumental assessments are key for identifying and characterising swallowing impairments. Instrumental assessments are particularly valuable, as they have the ability to identify symptoms that may affect the patient's health, such as silent aspiration.

However, it is important to note that swallowing impairment is just one aspect of the broader experience of eating and drinking. While it is significant, it does not fully capture the emotional, psychosocial and physical impact that swallowing difficulties can have on an individual's life. The perception of the patient plays a crucial role in understanding the extent to which the impairment restricts their daily lives. Self-reported methods, including interviews, surveys and questionnaires, are effective means to capture this valuable information and gain insight into the personal impact of swallowing difficulties on individuals' lives (Nolan and Birring, 2022).

Patient-reported outcome (PRO) measures refer to reports on a patient's health condition that come directly from the patient themselves, without interpretation by a clinician or other individuals (US Department of Health, 2006). PRO measures play an important role in patient-centred healthcare, as they promote effective communication, active patient engagement and increased confidence in managing one's health (Porter et al., 2016).

These measures allow patients to express their perceptions regarding various aspects such as health status, symptoms and quality of life (QoL). By collecting PRO data, insights may be gained that patients may not spontaneously share, thereby giving voice to their experiences within the complex landscape of their illness (Swigris and Fairclough, 2012). The integration of PRO measures into routine clinical practice is steadily advancing (Field;Holmes and Newell, 2019).

In the past, PRO measures utilised in research on interstitial lung disease (ILD) and idiopathic pulmonary fibrosis (IPF) were originally designed for other chronic respiratory conditions; however, more recently, there has been a development of several PRO measures specifically tailored for ILD or IPF. These relatively newly developed PRO measures primarily focus on assessing health-related quality of life (HR-QoL), such as The King's Brief interstitial lung disease (K-BILD), the St George's respiratory questionnaire in IPF (SGRQ-I) and a tool to assess quality of life in IPF (ATAQ-IPF) (Swigris *et al.*, 2010; Yorke;Jones and Swigris, 2010; Patel *et al.*, 2012; Nolan and Birring, 2022).

PRO measures for swallowing, reflux and throat symptoms

In the previous chapters (Chapters 3 and 4), my focus was on evaluating swallowing in patients with IPF using clinical assessments such as the Videofluroscopy Swallow Study (VFSS) and the Water Swallow Test (WST). The VFSS results revealed that three out of ten IPF patients experienced airway penetration, and one patient aspirated liquid without displaying a cough response. The WST showed that 18% of IPF patients failed the test and displayed signs of penetration and/or aspiration, such as coughing, throat clearing and/or a wet voice. Considering that coughing is a common occurrence in IPF patients, regardless of whether there is penetration and/or aspiration, it can be attributed to various factors such as reflux, IPF symptoms, cough-variant asthma, upper airway cough syndrome, medications or laryngeal hypersensitivity (Vigeland;Hughes and Horton, 2017).

Gastroesophageal reflux disease (GORD), characterised by the backflow of gastric acid and other stomach contents into the oesophagus, is a common condition associated with cough (Vigeland;Hughes and Horton, 2017). Among patients diagnosed with IPF, GORD is highly prevalent (Jones et al., 2018). Research

findings indicate that approximately 80–90% of IPF patients show evidence of GORD based on oesophageal pH monitoring, despite only 25–50% of them reporting typical reflux symptoms (Raghu et al., 2006b; Fahim et al., 2011). Another study observed elevated levels of bile acids and pepsin in bronchoalveolar lavage (BAL) fluid and saliva of IPF patients, which serve as recognised markers of GORD (Savarino et al., 2013).

Laryngopharyngeal reflux (LPR), also known as extraoesophageal reflux, refers to the backwards flow of gastric contents into the oesophagus, larynx and/or pharynx, without necessarily causing typical symptoms of heartburn or esophagitis. Signs and symptoms of LPR encompass: chronic cough, hoarseness, vocal fatigue, excessive throat clearing, globus pharyngeus, postnasal drip and dysphagia (Belafsky;Postma and Koufman, 2002). A study was conducted to investigate the association between IPF and LPR by utilising 24-hour laryngopharyngeal pH monitoring. The study included 44 patients diagnosed with IPF and 30 healthy individuals as the control group. The findings revealed that the proportion of IPF patients with pH values greater than 7.5 (indicating alkaline reflux) was higher when compared with the control group. It is noteworthy that the normal pH range in the lower airway is 7.0–7.5. This indicates that both alkaline reflux (pH > 7.5) and acidic reflux (pH < 6.5) were observed in patients with IPF, indicating the potential presence of laryngopharyngeal reflux in this population (Su et al., 2021).

Laryngeal Hypersensitivity Syndrome refers to a condition characterised by sensory dysfunction in the larynx resulting from abnormalities in the laryngeal adductor reflexes. These reflexes can lead to various symptoms, including coughing, laryngospasm, dysphonia (voice disorders), paradoxical movement of the vocal folds during breathing and discomfort while swallowing (McCabe and Altman, 2012).

Considering the factors associated with cough and upper airway symptoms mentioned above, I chose to evaluate extraoesophageal symptoms and laryngeal hypersensitivity in IPF patients, in addition to assessing swallowing-related outcomes. While GORD has been previously studied in IPF, my thesis focuses on the upper airway. Hence, I chose to investigate extraoesophageal reflux. In this study, I used specific patient-reported measures to assess swallowing, laryngopharyngeal reflux and laryngeal hypersensitivity in IPF patients.

5.2.1 Aim

To describe IPF patients' perceptions of swallowing dysfunction, laryngopharyngeal reflux and laryngeal hypersensitivity symptoms.

5.3 Specific materials and methods

5.3.1 Ethical approvals

The study was approved by the 211628_Non-substantial amendment 3 (NSA-3) of the North East-North Tyneside2 Research Ethics Committee REC reference 18/NE/0037 (see Appendix 9: approval letter).

5.3.2 Study design

This is an observational, cross-sectional study design.

5.3.3 Specific eligibility criteria

In addition to the core inclusions and exclusions criteria described in the general methods (sections 2.3.1.1 and 2.3.1.2).

5.3.3.1 Inclusion criteria

- Ability to understand and complete the questionnaires.

5.3.4 Recruitment

Recruitment was undertaken through the pulmonary fibrosis support groups and the regional ILD clinic for patients with IPF, described in detail in section 2.4.2. Once recruited, questionnaires were sent to the patients from the support groups via email. The questionnaires were provided either as an online questionnaire link using the Jisc Online Survey tool or as a PDF file attachment. Alternatively, for patients who preferred a physical copy, the questionnaires were sent through the post. The method of delivery depended on the individual patients' preferences. Patients were provided with a stamped addressed envelope to return the questionnaire to the research team. An additional questionnaire including questions about the patients'

demographics (age, gender and smoking history) was also sent to the patients using the same method as they requested.

Patients recruited from the ILD clinic completed the same questionnaires as those recruited from the pulmonary fibrosis support groups, with my support if needed during their clinical visit. Furthermore, patients' demographics, including age, gender and smoking history were recorded. In addition, measurements such as the Medical Research Council dyspnoea scale (MRC), body mass index (BMI) and pulmonary function tests including forced vital capacity (FVC) (% of predicted), and transfer factor of lung for carbon monoxide (TLCO) (% of predicted) were collected from patients' medical records.

5.3.5 Protocol

Self-reported questionnaires were used to assess the IPF patients' perception of swallowing (Eating Assessment Tool-10 [EAT-10]), extraoesophageal reflux (Reflux symptoms Index [RSI]) and laryngeal hyspersensitivity (Newcastle Laryngeal Hypersensitivity Questionnaire [LHQ]).

5.3.5.1 Swallowing questionnaire: EAT-10

In Chapter 3 of this thesis, the EAT-10 questionnaire was used. The description and details of the questionnaire can be found in that chapter, specifically in section 3.3.5.1. (see also Appendix 7: The Eating Assessment Tool: EAT-10).

5.3.5.2 Reflux questionnaire: RSI

The RSI is a brief validated questionnaire designed to assess laryngeal symptoms associated with laryngopharyngeal reflux. It is easily administered and highly reproducible**.** The RSI correlates well with the voice handicap index (VHI), which is a validated tool for assessing laryngopharyngeal reflux. The questionnaire consists of nine items, and each item is scored on a scale of 0–5, where a score of 0 indicates the absence of LPR symptoms and a score of 5 indicates severe symptoms. The total RSI score ranges from 0 to 45, with a score higher than 13 indicating the possibility of clinically significant LPR (Belafsky;Postma and Koufman, 2002). The

RSI has been previously utilised in studies involving IPF patients (Jones et al., 2018). (see Appendix 12: The Reflux Symptoms Index: RSI).

5.3.5.3 Laryngeal hypersensitivity questionnaire: Newcastle LHQ

The Newcastle LHQ is a fully validated, simple, self-administered questionnaire that takes approximately five minutes to complete. It consists of 14 items focusing on abnormal laryngeal sensation across three distinct domains: obstruction (sensations of throat obstruction), pain/thermal (altered sensory experience of pain and/or temperature sensation in the laryngeal area) and irritation (abnormal sensation of throat irritation). The Newcastle LHQ has a seven-point Likert response scale, where 1 is the worst score and 7 is the best. The total LHQ scores are calculated by adding together the domain scores, with a total score ranging from 3 to 21. Each domain score is derived by dividing the total scores from items within a specific domain by the number of items in the domain, which ranges from one to seven, where lower scores suggest a greater impairment of the larynx (Vertigan;Bone and Gibson, 2014). (see Appendix 13: The Newcastle Laryngeal Hypersensitivity Questionnaires: Newcastle LHQ).

5.3.6 Analysis

Continuous variables were presented using means, medians, standard deviation and range (minimum–maximum). Categorical variables were presented using frequencies and percentages. Analysis was largely descriptive. Data from the EAT-10 and RSI were analysed against the published cut-off value for normal EAT-10 and RSI scores (Belafsky;Postma and Koufman, 2002; Belafsky *et al.*, 2008).

The Newcastle LHQ questionnaire was analysed with reference to published data from healthy control populations (n:15) (Vertigan;Bone and Gibson, 2014). Lower Level Normal (LLN) was used as the cut-off value for abnormality for Newcastle LHQ scores, as lower scores suggest greater impairment. The LLN was taken as the lower limit of the 95% confidence interval (CI) of the normative data. All statistical analyses and graphic presentations in this chapter were performed using Minitab statistical software (version 21, Minitab Inc., State College, PA, USA) and Microsoft Excel 2016 (Microsoft Corporation, Washington, DC, USA).

5.4 Results

A total of 42 IPF patients (22 from the support groups and 20 from the ILD clinic) were recruited and completed the three questionnaires (see Figure 17 below).

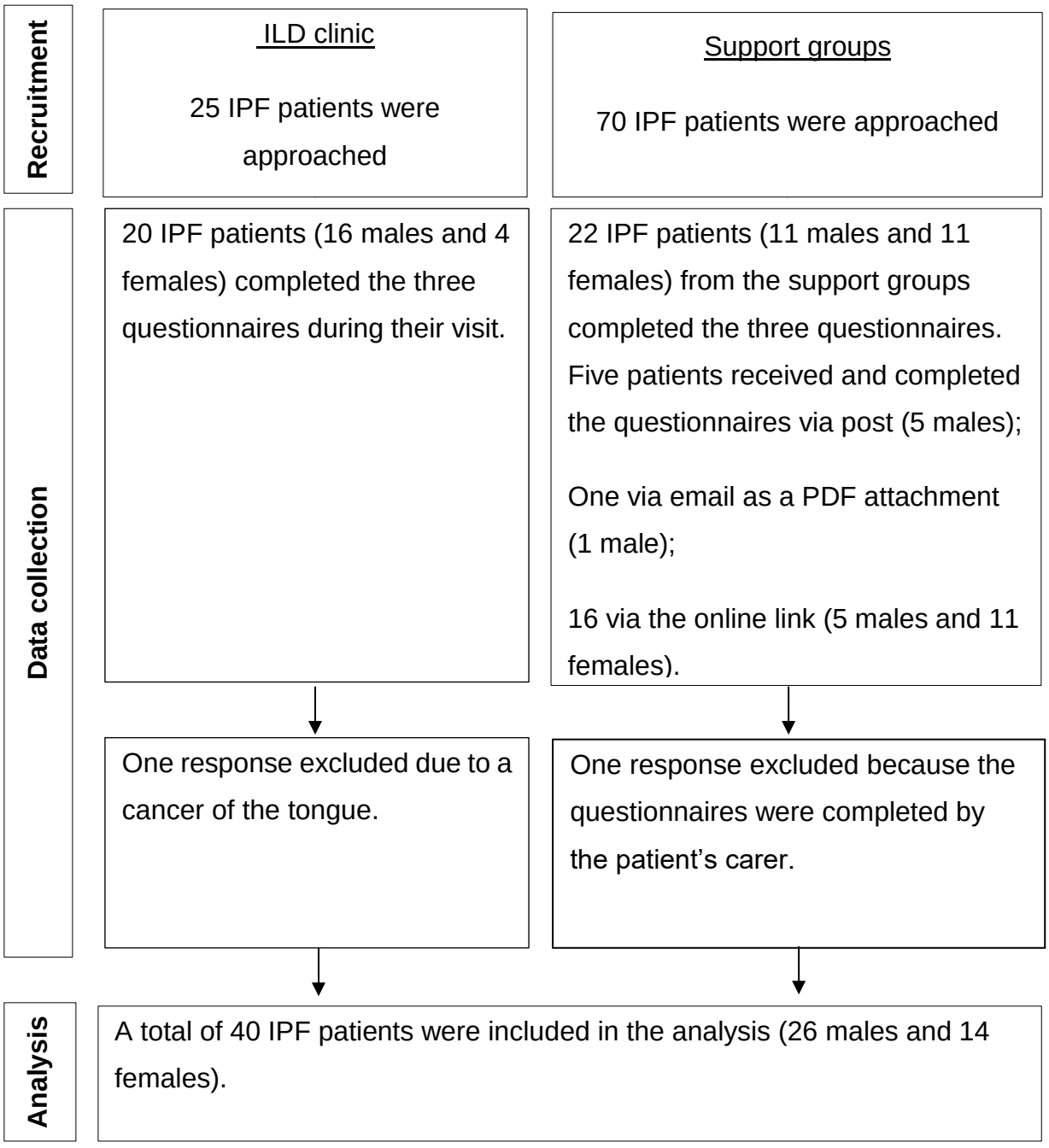

Figure 17. Consort flow diagram for IPF patients' recruitment.

5.4.1 Patients' demographics and clinical characteristics

Patients' demographics and clinical characteristics are presented in Table 11. A total of 40 IPF patients were included in the analysis. The median age of patients was 71.0 years ([min–max] 52–92). Most patients were males (26/40, 65%) and non-smokers (25/40, 62.5%). The patients recruited from the ILD clinic (n: 26) were as follows (median, min–max): BMI (27.0 kg/m^2, 21.7–40.0), MRC (3, 2–5), FVC% of predicted (66.5%, 34–83) and TLCO% of predicted (39%,36–51).

Factor	**Level**	**Value**
N		40
Age in years, median (minimum-maximum)		71 (52-92)
Gender	Female	14 (35%)
	Male	26 (65%)
Smoking	Non-smoker	25 (62.5%)
	Current smoker	2 (5%)
	Ex-smoker	13 (32.5%)
N		26
BMI, kg/m^2, median (minimum-maximum)		27 (21.7-40)
MRC dyspnoea scale, median (minimum-maximum)		3 (2-5)
FVC% of predicted, median (minimum-maximum)		66.5% (34-83)
TLCO% of predicted, median (minimum-maximum)		39% (36-51)

Table 11. Median (min-max) or (%) for the patients' demographic and clinical characteristics. N: number; BMI: Body mass index; MRC: Medical Research Council dyspnoea scale; FVC: Forced vital capacity; TLCO: Transfer factor of lung for carbon monoxide.

5.4.2 Self-reported swallowing symptoms: The EAT-10

The total mean score for the EAT-10 for all patients (n:40) was 5.9 ±6.5, with a median score of 4 and a range from 0 to 20. A total of 57% of patients had swallowing difficulty, which was evidenced by a total EAT-10 score of ≥3 (Belafsky et al., 2008) (see Figure 18 below).

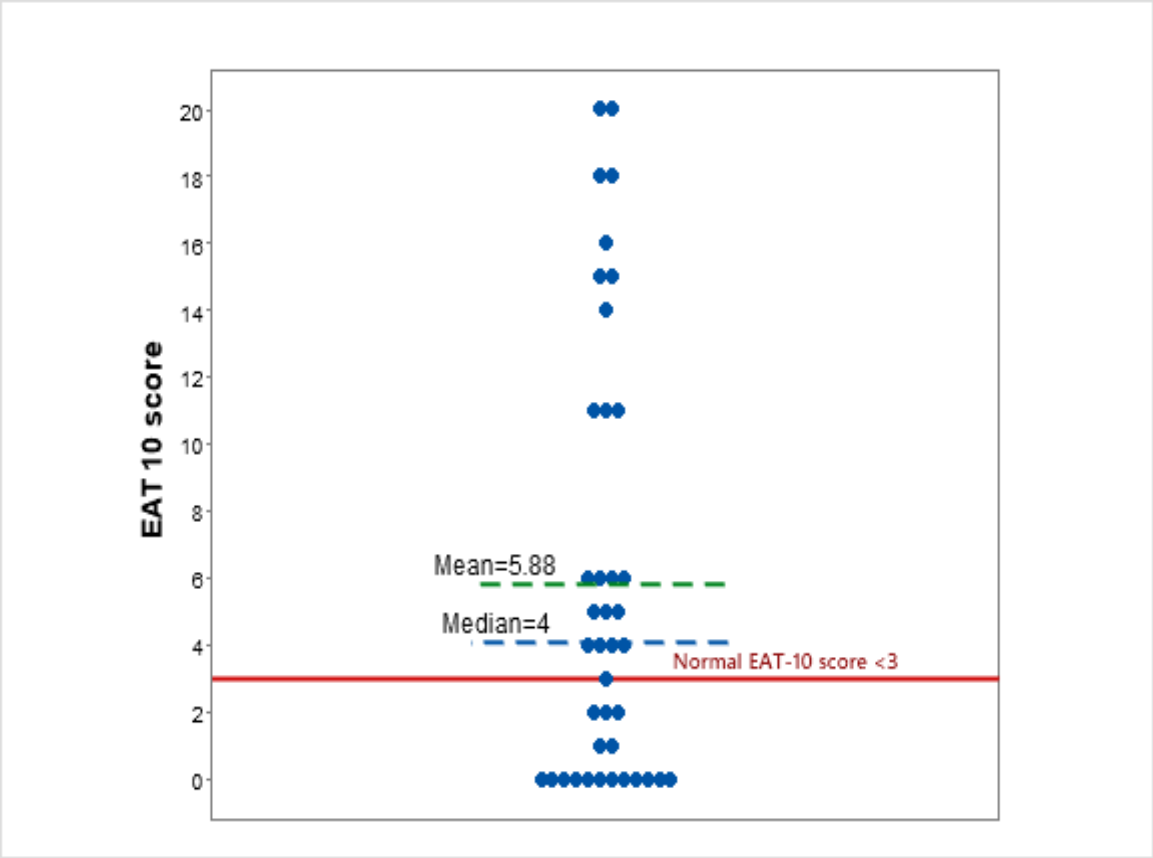

Figure 18. The EAT-10 scores for all IPF patients (n:40).
EAT-10 score (y-axis) for patients. The red line shows the upper score for EAT-10: a score of ≥ 3 indicates an abnormal EAT-10 score. The blue and green dashed lines denote the median and mean total EAT-10 score for all patients, respectively.

5.4.3 Self-reported reflux symptoms: The RSI

The mean score for the RSI for all patients (n:40) was 15.6 ±8.3, with a median of 15 and a range from 2 to 31. Twenty-four patients had an RSI score of ≥13, suggesting the presence of laryngopharyngeal reflux disease (Belafsky;Postma and Koufman, 2002) (see Figure 19 below).

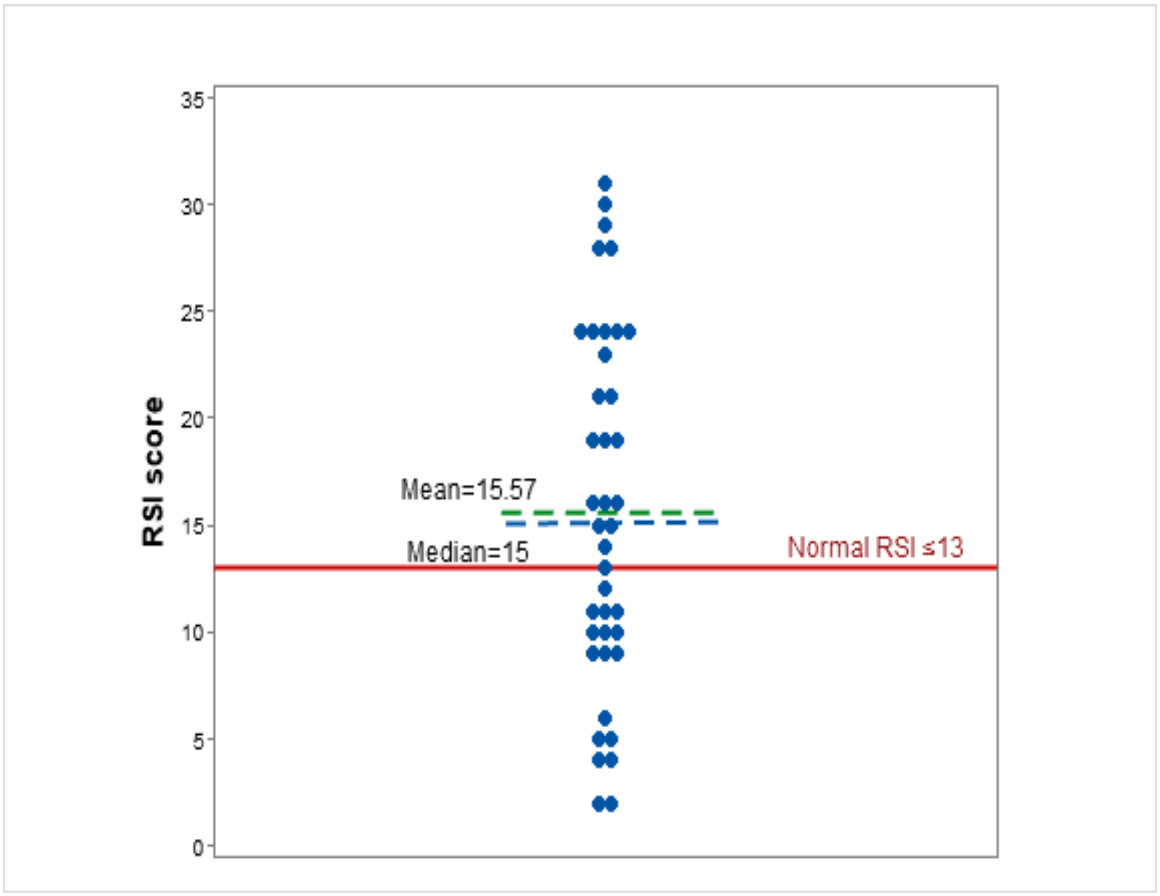

Figure 19. RSI scores for all patients (n:40).
RSI score (y-axis) for patients. The red line denotes the upper limit of normal, where a score of ≥13 indicate an abnormal RSI score. The blue and green dashed lines denote the median and mean RSI scores, respectively.

5.4.4 Self-reported throat symptoms: The Newcastle LHQ

The Newcastle LHQ total score was calculated by adding together the following domain scores from the questionnaire: obstructive, pain/thermal and irritation. The mean ±(SD) for the Newcastle LHQ total score for all patients (n=40) was 15.78 ±3.2, with a median score of 16 and a range from 9.48 to 21. Twenty-five IPF patients had a total Newcastle LHQ score that was below the lower level of the 95% CI of the published normal Newcastle LHQ score (n:15) (mean: 19.2 ±0.7) (17.8–20.6: 95% CI), suggesting the presence of laryngeal symptoms (Vertigan;Bone and Gibson, 2014). (see Figure 20 below).

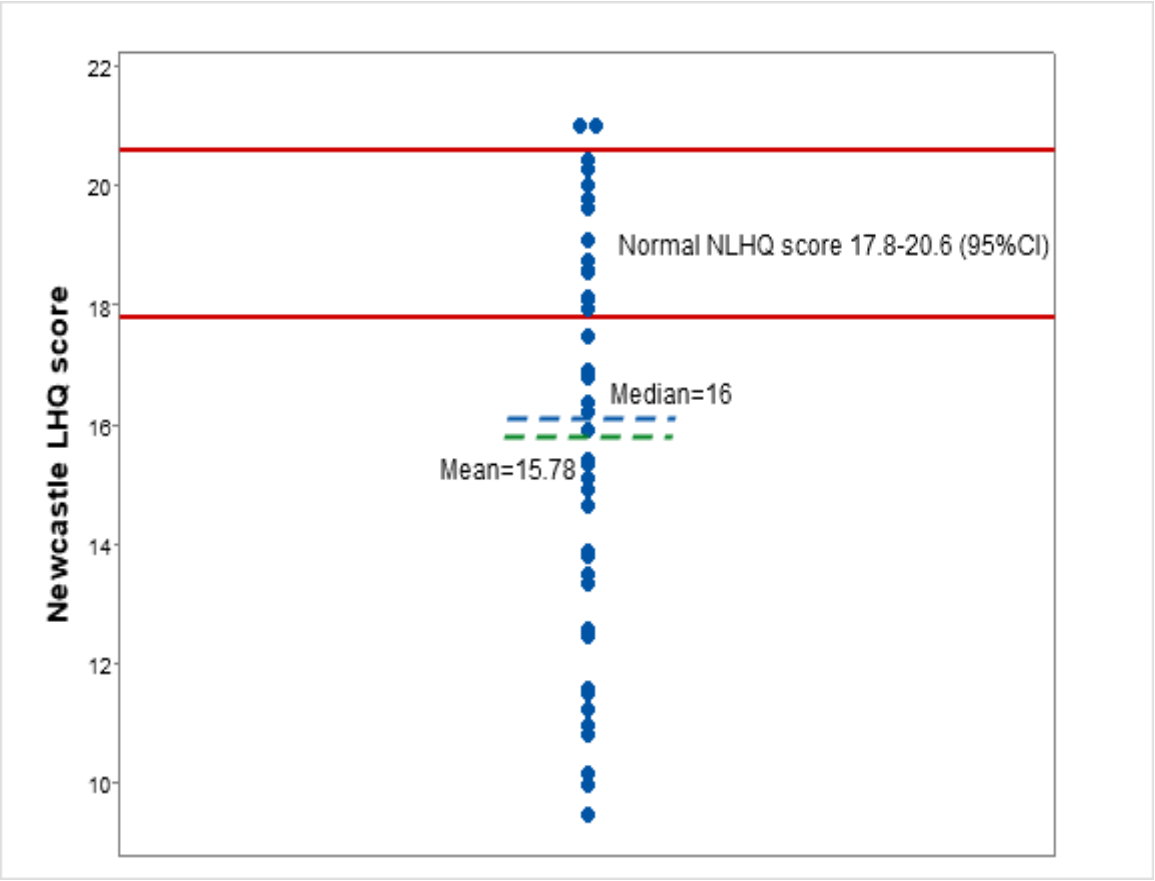

Figure 20. Newcastle LHQ total scores for all patients (n: 40).
The red lines denote the upper and lower level of the 95% CI of the published normal LHQ scores (n: 15), normal NLHQ scores 95% CI (17.8–20.6). Abnormal scores are those below the lower threshold of 17.8. The blue and green dashed lines denote the median and mean value for all patients, respectively.

5.5 Discussion

The aim of this chapter was to describe IPF patients' perceptions of swallowing dysfunction, laryngopharyngeal reflux and laryngeal hypersensitivity symptoms. Three established and validated questionnaires (EAT-10, RSI and Newcastle LHQ) were used. The key findings from this chapter are that self-reported symptoms of swallowing difficulty, laryngopharyngeal reflux and laryngeal hypersensitivity were higher in patients with IPF when compared with the general population.

In a similar manner to previously published studies utilising the EAT-10 and RSI questionnaires, patients here were able to complete the questionnaires easily during their clinical visits to the ILD clinic and remotely through a PDF or an online link attached to an email, evidenced by 100% compliance in completing the questionnaire. In contrast, IPF patients in this study reported that completing the Newcastle LHQ was time-intensive and the scoring system was confusing. This finding is consistent with a previous study by Hull et al. (2019), where patients also expressed that the Newcastle LHQ took a significant amount of time to complete, which somewhat limited its practical use in everyday clinical practice (Hull et al., 2020).

This study identified an increased self-reporting of dysphagia symptoms in IPF patients when measuring their condition using the EAT-10 questionnaire. The findings revealed a median score of 4, with a range from 0 to 20. A total of 57% of IPF patients perceived they had swallowing difficulty, evidenced by a total EAT-10 score of $\geq$3. (Belafsky et al., 2008).

While the study did not specifically involve IPF patients, previous research using a cut-off score of 9 on the EAT-10 questionnaire demonstrated its ability to accurately predict aspiration risk in patients with stable chronic obstructive pulmonary disease (COPD) when compared with FEES. The sensitivity and specificity were reported as 92% and 78%, respectively (Regan;Lawson and De Aguiar, 2017).

A more detailed analysis of the EAT-10 questionnaire revealed that the most reported problems were "coughing when I eat" and "when I swallow food, it sticks in my throat" with mean scores of 1.23 (±1.81) and 0.85 (±1), respectively. Patients with IPF commonly present clinically with a cough regardless of whether they have penetration and/or aspiration, therefore reducing the sensitivity of a "cough" as a useful indicator for bolus penetration or aspiration (Vigeland;Hughes and Horton, 2017). The exact

mechanism of cough in IPF is likely to be multifaceted, but potential causes include IPF pathophysiology, increased airway sensitivity to cough triggers, pharmacotherapy, postnasal drip, upper airway cough syndrome, reflux disease, laryngeal penetration and/or aspiration (Vigeland;Hughes and Horton, 2017). Further work is required to ascertain if cough can be a reliable marker of dysphagia in IPF, which to my knowledge has not been previously studied.

Furthermore, the EAT-10 questionnaire also identified that the second most prominent item for patients was “when I swallow food, it sticks in my throat”, suggesting that patients with IPF experience bolus retention in the pharynx. Although beyond the scope of this study, the frequency and extent of food sticking in the pharynx for IPF patients has not been studied. There may be other associated reasons for swallowing changes in IPF patients, such as age-related changes in swallowing mechanics (presbyphagia) (McCoy and Varindani Desai, 2018). For example, the striated muscles of the oesophagus may reduce in mass or coordination while increasing their dysfunction and fatigability, which would align with previous studies demonstrating dysfunction of respiratory and skeletal muscle in a similar fashion (Roman;Rossiter and Casaburi, 2016). Furthermore, bolus retention in IPF patients may be due to xerostomia (dry mouth), which may be caused by inhaled corticosteroid (ICS), antibiotics or oxygen (Lindh et al., 2021). Regardless of the cause, these findings indicate that EAT-10 may be useful in identifying specific aspects of patients’ swallowing symptoms. Earlier identification with the use of self-reported questionnaires such as EAT-10 may help identify patients’ symptoms before more severe clinical presentations.

In this study, it was found that the mean RSI score in IPF patients was 15.58 (±8.32) and the median score was 15. The range of scores observed in the study varied from 2 to 31. These findings align with a previous study on the same population of 36 IPF patients, which showed that even patients on proton pump inhibitor (PPI) medication had persistently abnormal RSI scores. In that study, the median RSI score for patients on PPI was 11 (ranging from 0 to 32), while for patients not on PPI, the median score was 10 (ranging from 0 to 39) (Jones et al., 2018).

The RSI is a validated questionnaire that focuses on symptoms related to extraoesophageal reflux. The original validation study of the RSI compared 25 patients with confirmed laryngopharyngeal reflux (LPR) to 25 age and gender-matched

controls. The study found that the LPR patients had a significantly higher mean RSI score compared to the controls (21.2 vs. 11.6; $p < 0.001$) (Belafsky;Postma and Koufman, 2002).

In clinical practice, the RSI is commonly used to assess the risk of laryngopharyngeal reflux in patients who present with symptoms such as cough and throat discomfort. Early diagnosis and treatment of laryngopharyngeal reflux may lead to improved clinical outcomes and enhance the daily activities of affected patients (Su et al., 2021).

There is a scarcity of literature addressing laryngeal symptoms in patients with IPF. This study revealed that among the IPF patients, 25 individuals had Newcastle LHQ scores below the lower limit of the 95% CI of the normal published Newcastle LHQ. Only thirteen patients had Newcastle LHQ scores within the 95% CI (17.8-20.6) of the normal published Newcastle LHQ score. A lower NLHQ score indicates a higher degree of laryngeal impairment.

These findings align with the results of a validity study conducted on the Newcastle LHQ (Vertigan;Bone and Gibson, 2014). The study involved 97 participants, including 82 patients from four clinical groups: chronic refractory cough (n=49), paradoxical vocal fold movement (n=22), globus pharyngeus (n=8) and muscle tension dysphonia (n=3), as well as 15 healthy controls. The Newcastle LHQ questionnaire demonstrated significant discriminant validity, as indicated by a mean difference of 5.5 in scores between patients with laryngeal disorders and healthy controls. Patients with laryngeal hypersensitivity exhibited similarly abnormal scores (Vertigan;Bone and Gibson, 2014). Furthermore, the Newcastle LHQ demonstrated improvement following behavioural speech pathology intervention, with an average reduction in LHQ score of 2.3 (Vertigan;Bone and Gibson, 2014).

To my knowledge, this study appears to be the first of its kind to use the Newcastle LHQ in the IPF population. Understanding patients' perceptions of their own laryngeal sensations could aid clinical teams in assessing the level of laryngeal discomfort experienced by IPF patients.

5.5.1 Limitations

The study acknowledged some limitations, including the use of convenience sampling, as mentioned in section 4.5.5. Due to COVID restrictions, the EAT-10 questionnaire

used in the study were not compared with reference instrumental tests of swallowing, so no assumption can be made regarding its ability to detect swallowing impairment. The study primarily relied on questionnaires to investigate the patients' perspective on swallowing, laryngopharyngeal reflux, and laryngeal discomfort. While this approach was practical and allowed for the identification of trends based on the results, it is important to note that relying solely on questionnaires may not have fully captured the perspectives of IPF patients. To address this concern, the next chapter will tackle the issue by presenting the patients' perspective using qualitative research methods.

5.5.2 Conclusion

Recently, there has been a shift among researchers in the field of IPF towards considering a broader context, which includes incorporating patient-reported measures. This approach is highly relevant to clinicians, as it provides a more accurate reflection of the daily concerns faced by both the patients and their caregivers. As a result, IPF-specific questionnaires have been developed to capture these important aspects.

The findings of this study reveal that a significant number of IPF patients report symptoms such as difficulty in swallowing, laryngopharyngeal reflux and laryngeal discomfort. These results shed light on the prevalence of these symptoms within the IPF population. However, in order to gain a more comprehensive understanding, further research is necessary to thoroughly explore and comprehend these symptoms in this vulnerable group of patients.

Chapter 6. Eating and drinking experience: The qualitative study: A patients' perspective

6.1 Introduction

This is the last results chapter of my thesis. In the previous chapters, swallowing was assessed from different perspectives:

(1) The clinical perspective using the gold standard instrumental assessment (Chapter 3), in which the Videofluoroscopy Swallow Study (VFSS) demonstrated a wide range of findings. Some patients were "normal", but others were not, with a range of impairments in oropharyngeal swallow physiology and safety. (2) The clinical perspective using a screening Water Swallow Test (WST) (Chapter 4); this demonstrated some patients with normal swallowing but also that some patients showed signs of penetration and/or aspiration observed during WST. (3) The patients' perspectives using validated questionnaires (Chapter 5); EAT-10 scores were higher than the normal cut-off value (< 3) in these groups of patients.

This chapter reports on a study seeking to explore idiopathic pulmonary fibrosis (IPF)-related eating and drinking challenges and gain a comprehensive picture of the disease's impact on the eating and drinking experience taken from the patients' perspective. A qualitative research design was used to provide a deeper understanding of the lived experience of living with IPF. Elements of the work described in this chapter have been presented as poster presentation in the European Respiratory Society (ERS) International Congress, Milan 2023 (Alamer *et al.*, 2023a)(see appendix 14), and published in a peer review journal (Alamer *et al.*, 2024)(see appendix 15).

6.2 Literature review

Several qualitative research studies have been published on patients' experiences with IPF, including their perceptions of treatments, pulmonary rehabilitation, palliative care and the COVID-19 pandemic, and their experience with the disease in general (Bridges *et al.*, 2015; Holland *et al.*, 2015; Overgaard *et al.*, 2016; Lindell *et al.*, 2017; Ryan and Meskell, 2022a).

Overgaard et al. (2016) and Swigris et al. (2005) conducted studies exploring the experiences of patients with IPF. Overgaard et al. (2016) found that patients and caregivers faced challenges in obtaining a correct diagnosis and accessing reliable information about the disease. The study also identified various negative impacts of IPF on patients' quality of life (QoL), such as physical limitations, emotional distress and social isolation. Patients used coping strategies such as focusing on the positive aspects of life and seeking support from family and friends (Overgaard et al., 2016). Swigris et al. (2005) found that IPF patients experienced limitations in physical activity, sleep disturbances, fatigue, financial insecurity and fear of death, all of which negatively impacted their QoL. Notably, these studies did not examine issues related to eating and drinking (Swigris et al., 2005).

Aside from IPF, only a few qualitative studies on eating and drinking issues have been published in patients with chronic respiratory disease (Odencrants;Ehnfors and Grobe, 2005; Lin and Shune, 2022). Fourteen patients with chronic obstructive pulmonary disease (COPD) were interviewed (Lin and Shune, 2022). Coughing and shortness of breath were the most common symptoms experienced by COPD patients during oral intake, associated with anxiety, fear and embarrassment (Lin and Shune, 2022). In addition, they developed strategies to manage their oral intake, including ways of modifying the way they ate; for example, eating soft food, chewing well and taking small, regulated sips (Lin and Shune, 2022). These findings support a previous study which used a short survey to understand the experience of COPD patients with dyspnoea during swallowing (n=133) (Hoit et al., 2011). This found that 74% of subjects reported breathing discomfort when eating or drinking and many had modified the way they ate and drank and the way they breathed (Hoit et al., 2011).

There has been limited research on the nutritional needs and dietary restrictions of patients with IPF, which has demonstrated that IPF patients have suffered from several nutritional abnormalities including weight loss, muscle loss, loss of appetite and vitamin D deficiency, which may negatively affect the disease prognosis and their QoL (Faverio et al., 2020). To date I am not aware of research regarding the potential for swallowing difficulty in patients with IPF.

I published a study which measured the IPF patients' perception of swallowing difficulty using validated questionnaires (EAT-10) (Alamer et al., 2022a). Of these patients, 29% (4/10) exceeded the EAT-10 normal cut-off of < 3, and had markedly raised total

scores, with values of 25, 15, 14 and 13, indicating swallowing difficulty (Alamer *et al.*, 2022a). This topic has not been examined extensively, and to my knowledge there is no research providing an in-depth exploration on the eating and drinking experiences of IPF patients using qualitative research methods.

6.2.1 Aim

The aim of this study is to explore the lived eating and drinking experience of patients with IPF.

6.2.2 Study objectives

1. To understand the perceived eating and drinking changes experienced by patients with IPF.
2. To understand the impact of eating and drinking changes on patients with IPF.
3. To explore any coping strategies for and adjustments for these changes.

6.3 Specific methodology and methods

6.3.1 Philosophical assumptions

Multiple philosophical and theoretical traditions were explored to provide the foundation for the design of this study. Firstly, it is important to articulate the underlying philosophical assumptions that underpin the research. The term “ontology” relates to the nature of reality or being; what it is that we think we can know. This study follows the ontological position of relativism: reality is subjective, and there are multiple realities. Relativism “is an ontological position that conceptualises reality as the product of human action and interaction and doesn’t subscribe to the notion of a singular reality that exists independent of human practices, reality and truth are contingent, local and multiple” (Clarke and Braun, 2021, p. 173). Data was obtained from the perspectives of individuals with IPF disease, whose views and understandings are subjective and influenced by previous interactions, none of which are more or less valuable to explore. We gain subjective meanings, constructions and multiple complex realities (Allison and Pomeroy, 2000). Therefore, the ontological position adopted in this study was relativist.

While ontology relates to how knowledge exists, epistemology relates to the nature, justification and limitations of knowledge: what we think it is possible to know and how

we can know it (Clarke and Braun, 2021). This study takes the epistemological position of constructionism, which proposes that these realities are constrained by society. Constructionism “is founded on the premise that the research practices produce rather than reveal evidence” (Clarke and Braun, 2021, p. 178). In studying the eating and drinking experience of IPF patients from a constructivist epistemological position, the researcher would focus on understanding how patients construct their own experiences, beliefs and attitudes towards eating and drinking. The researcher would explore how patients' interpretations, values and cultural backgrounds shape their understanding of these experiences, as well as how social interactions and contextual factors influence their eating and drinking behaviours. Therefore, the epistemological position adopted in this study was constructionism.

An interview design using semi-structured interviews was employed in this study in order to gain a broader understanding of the eating and drinking experience in patients diagnosed with IPF. Semi-structured interviews provide an opportunity to ask the interviewee questions about particular aspects of his/her life or experiences, with topics and questions predefined in advance, to trigger and encourage the interviewee to talk (Silverman, 2020; Shelton;Philbin and Ramanadhan, 2022).

Although focus group data collection can achieve the same goal, the discussion may have included sensitive details about their experiences with this disease. This may include the effects of the disease on their eating and drinking experiences from a physical, social and emotional standpoint; therefore, a semi-structured interview was used to collect data in this study. The one-to-one interview allows for a more comprehensive picture of the disease and protects the privacy of patients (Fylan, 2005).

6.3.2 Study design

This is a descriptive qualitative study to gain a deeper understanding of IPF patients’ eating and drinking experiences from their perspective, using semi-structured interviews.

6.3.3 Eligibility criteria

The core inclusion and exclusion criteria mentioned in sections 2.3.1.1 and 2.3.1.2.

6.3.4 Participants

6.3.4.1 Sampling procedure

Patients with IPF were recruited based on the purposive sampling method using the maximum variation sampling approach.

Purposive sampling is a type of non-probability sampling method that is commonly used in qualitative research, in which a researcher deliberately chooses the sample with specific characteristics that are relevant to the research question. In other words, the researchers pick the participants “on purpose” (Etikan;Musa and Alkassim, 2016).

There are a range of different approaches to carrying out the purposive sampling method that all involve using the researchers’ judgement to choose participants that can best answer their research question (Palinkas et al., 2015). The maximum variation sampling approach is a sampling approach in which researchers attempt to collect data from as diverse a sample as possible about a certain topic. This involves identifying specific selection criteria that were considered of particular importance when constructing a sample of maximal variation (Patton, 2002; Bryman, 2016; Patterson and Dawson, 2017).

In this study, I chose the maximum variation sampling approach to ensure heterogeneity in the sample as well as to elicit diverse views and experiences. In the first few interviews, the sample was based on age (≤ 65 years vs. > 65 years) and sex (male or female), because eating and drinking can vary by age and gender (Chapter 1: Effect of age and normal swallowing, section 1.2.5). After an initial two or three interviews, the selection criteria were discussed with the supervisory team. Living status (alone or with someone) appeared to have an influence on eating and drinking experiences; therefore, it was included as a selection criterion for maximum variation.

Recruitment for the interviews continued until the study’s criteria for maximum variation sampling were met (i.e. at least one participant in each of the maximum variation categories). Table 12 contains a list of the participants interviewed according to the sampling categories with the maximum variation sampling strategy.

Demographics	Parameters	No. of participants
Age	≤ 65 years	4
	> 65 years	10
Gender	Male	8
	Female	6
Living status	Living alone	4
	Living with someone	10

Table 12. Maximum variation sampling categories for the participants interviewed.

6.3.4.2 Information power

The number of interviews conducted was informed by the concept of "information power". Information power is an alternative approach to "data saturation" (Malterud;Siersma and Guassora, 2016; Mott *et al.*, 2022; Turner *et al.*, 2022), which relates to no new information or data being derived during an iterative and systematic data analysis process, or when the researchers conclude that they have “heard it all” (LaDonna;Artino Jr and Balmer, 2021; Sarfo *et al.*, 2021). This may seem to be an easy concept to follow, but it can be difficult to determine when (or if) saturation is reached because there is considerable confusion about what data saturation means (LaDonna;Artino Jr and Balmer, 2021). Indeed, researchers have shown that indices of saturation in qualitative interview-based studies are often inadequately described, and authors focus on the number of participants in order to convince readers (or themselves) that a sufficiently large sample was recruited to achieve their study aim. Instead, many qualitative researchers have shifted to describing quality findings as sufficient, which covers both analytical sufficiency and data sufficiency (Malterud;Siersma and Guassora, 2016; LaDonna;Artino Jr and Balmer, 2021).

In light of the limitations of the data saturation concept, information power is considered a more appropriate measure of evaluation sufficiency (LaDonna;Artino Jr and Balmer, 2021). The information power indicates that the more information the sample holds that is relevant to the actual study, the lower the number of participants needed (Malterud;Siersma and Guassora, 2016; Beck *et al.*, 2022). The use of information power to determine whether qualitative findings are sufficient involves assessing five items: (1) study aim; (2) sample specificity; (3) use of established theory; (4) quality of dialogue; and (5) analysis strategy (Malterud;Siersma and Guassora, 2016; Turner *et al.*, 2022).

In this study, these five items – (1) focused aim and objectives (i.e. to understand eating and drinking experience of IPF patients), (2) tight sample specificity (i.e. using purposive sampling and maximum variation sampling approach), (3) high-quality interview dialogue, (4) established theory and (5) focused analysis strategy – together with an iterative analysis of the results and discussion with supervisory team were sufficient to inform the decision on information power and when to stop recruitment (Malterud;Siersma and Guassora, 2016; Beck *et al.*, 2022). More details about pilot interviews are described in section 6.3.6.3.

6.3.5 Recruitment

Recruitment for the semi-structured interviews was undertaken through pulmonary fibrosis support groups and the regional interstitial lung disease (ILD) clinic at the Royal Victoria Infirmary (RVI), Newcastle upon Tyne, from February 2021 to September 2021. Semi-structured interviews were conducted via telephone or virtually via videoconferencing call platforms (Zoom or FaceTime), depending on the participant's preference, due to COVID-19 restrictive measures (Varma et al., 2021). More details about the recruitment of patients are described in section 2.4.2.

6.3.6 Data collection

In qualitative research, interviews are the most common method of collecting data, and conducting them face-to-face is considered the "gold standard" (Novick, 2008; Creswell and Poth, 2016; Roberts;Pavlakis and Richards, 2021). However, meeting participants in person in the COVID-19 era has been complicated by the constraints of social distancing and the prioritisation of researchers' and participants' safety, particularly with vulnerable populations (Roberts;Pavlakis and Richards, 2021). This is particularly an issue with people with IPF, who are considered a high-risk group. Conducting interviews virtually or via telephone may provide researchers with the opportunity to gain insight into the context of the research topic, while also safeguarding participants and researchers from the risk of potential exposure to infections including COVID-19 (Lobe;Morgan and Hoffman, 2020).

6.3.6.1Telephone interviews

Previously, telephone interviews have been criticised for their potential to adversely affect the quality and richness of the collected data for several reasons, including

difficulty in establishing rapport with the participants, preventing the researcher from making observations and responding to emotional and visual cues (Azad et al., 2021). Furthermore, there may be an increased risk of misunderstandings and inability to know when to ask probing questions or discuss sensitive topics (Novick, 2008). However, a recent body of literature indicates that telephone interviews generate the same amount of data richness as face-to-face interviews, and although such interviews are more conversational and detailed, they do not clearly lead to differences in interview data richness (Johnson;Scheitle and Ecklund, 2021). Telephone interviews increase anonymity and reduce distraction, thus providing an opportunity to discuss topics that may be sensitive in nature. In addition, they offer flexibility regarding the timing and location of the interview (Azad et al., 2021).

6.3.6.2 Virtual interview

The participants were offered a choice to be interviewed virtually using videoconferencing call platforms such as Zoom and FaceTime.

These platforms allow two or more people located at different locations to communicate in real time by using audio and video images (Keen;Lomeli-Rodriguez and Joffe, 2022). Researchers and participants connect to the platforms using a computer, smartphone or tablet via a wireless or hardwired internet connection (Keen;Lomeli-Rodriguez and Joffe, 2022).

For qualitative research, virtual interviews offer several benefits, similarly to telephone interviews. In the study conducted by Gray et al. (2020), participants reported that virtual interviews are convenient, easy to use, accessible, cost effective and time saving. Some participants report feeling comfortable discussing personal topics in their own space and appreciate being able to see their interviewer when discussing a sensitive topic, whereas others find it negatively affects rapport (Gray et al., 2020). Deakin and Wakefield (2014) suggested several emails be exchanged ahead of the videoconference interview as a way to help build rapport (Deakin and Wakefield, 2014).

6.3.6.3 The interview

The interview topic guide

A semi-structured interview topic guide was devised to elicit detailed responses from each patient. The interview topic guide followed existing guidelines for the design of qualitative interviews (Pope;Mays and Popay, 2006; Bryman, 2016). It includes guidance on possible areas for the interviewer to probe that are relevant to the research questions and themes from the existing literature. The interviewer used the topic guide flexibly, so that each patient was able to raise and discuss issues in a manner and order that felt natural to them. In addition, the interviewer could also ensure that all areas of interest were discussed. It was developed following a comprehensive review of relevant medical literature (Schoenheit;Becattelli and Cohen, 2011; Overgaard *et al.*, 2016; Russell;Ripamonti and Vancheri, 2016; Ryan and Meskell, 2022b). Input from healthcare practitioners with expertise in IPF and discussion with the supervisory team was also included. The interview (and guide) began with a few questions designed to ease participants into the discussion (for example, questions about themselves, their circumstances and their diagnosis). The interviewer asked open-ended questions about each area of interest in the guide, and in addition, a range of sub-questions and prompts were developed to explore the issues in more depth. When necessary, questions were rephrased to aid a participant's understanding.

The topic guide consists of five sections (see Appendix 16: Semi-structured interview topic guide initial interviews).

1. Introduction to the interview: "thanked and welcomed the patients, and briefly described the study".

2. Demographics: "gathered some basic information about the patients such as: age, years since diagnosis, etc.".

3. This was an introductory question: "encouraged patients to discuss their experience with IPF in general".

4. Main interview: "focused on eating and drinking: open-ended questions covered the following topics (physical changes to eating, diet, enjoyment, lifestyle, adaptations and advice)".

5. Conclusion: “offered patients the opportunity to add other issues relating to the eating and drinking experience that were not covered during the interview”.

Pilot interview topic guide

Prior to conducting the patient interviews, the topic guide was piloted in two online interviews using a videoconferencing call platform (Zoom) with a speech and language therapist and a respiratory consultant in December 2020. As a novice interviewer, I found that this step was helpful for pacing the interview and time-keeping, while using a conversational tone and making necessary adjustments to the areas that needed improvement (Patterson and Dawson, 2017; DeJonckheere and Vaughn, 2019). The pilot interviews lasted approximately 40 to 50 minutes and were audio recorded. The interviewees provided some helpful suggestions to improve the quality of the interview.

After analysing the pilot interview audio recordings, the interview guide was revised by adding a sentence to the introductory section (“You may see me writing while we are in discussion – this is only to help me prepare for further discussion”) and at the end of the interview (“At the end you mentioned …… Is there anything you feel we have not covered in the interview about your experience of eating and drinking since your diagnosis that you would like to add now?”).

The interview process

At the beginning of the interview, I thanked patients for their time, demonstrated warm and open body language, checked the camera and microphone, and reminded them about their right to pause or end the interview at any time (Keen;Lomeli-Rodriguez and Joffe, 2022).

Interviewing is an iterative process in nature; data collection and analysis are conducted simultaneously, as in all qualitative research. Thus, as the interview progresses, the interview guide can be modified. This is because, as new issues arise, they will be added and discussed in subsequent interviews, making sure sufficient depth is achieved (Bryman, 2016; DeJonckheere and Vaughn, 2019).

The interview guide was modified after conducting four interviews with IPF patients (see Appendix 17: Semi-structured interview topic guide final interviews). Sub-questions in each area of the guide were removed to avoid leading questions. The warm-up questions – for example, about demographics and disease diagnosis and

prognosis – were shortened to allow more time to explore eating and drinking experiences, and to prevent patients from becoming tired or fatigued due to spending a long time talking. Furthermore, more probing, open questions were added to encourage patients to talk, and to explore topics that were introduced by patients in previous interviews such as: “Can you tell me a bit more about that?” and “What else?”.

Notes were taken during the interviews with patients for later reflection (see Appendix 18: example of note taking during interview). The patients were interviewed while they were in their homes. On occasion, another family member was present to assist with the technology, but they were not involved during the interview. Patients’ interviews lasted between 40 and 50 minutes. Patients had the opportunity, at the end of their interview, to raise any topic that they felt was important that had not been covered by the questions.

6.3.7 The importance of reflexivity

Reflexivity is an important aspect of qualitative research, especially in the context of qualitative interviews (Symon and Cassell, 2012, pp. 72-89). It refers to the process of considering the researcher’s own biases, perspectives and assumptions, their own professional role, and preconceived notions and how these may influence the research methodology, data collection, analysis and outcomes (Richards and Emslie, 2000; Symon and Cassell, 2012; Kristensen and Ravn, 2015). By engaging in reflexive practices, researchers can increase validity, credibility and transparency and can better appreciate the complexity of the experiences and perspectives of the interviewees (Kristensen and Ravn, 2015).

Reflexivity practice in this study was covered in two sections, 6.3.7.1 and 6.3.7.2.

6.3.7.1Reflection on the interviews

In the first few interviews, the patients were asked about their demographics as part of the interview topic guide, and one of the questions was, “Can you remember when you were diagnosed with IPF?”, but this question led to an extended discussion, in which patients narrated their whole journey from symptoms onset to diagnosis with IPF. In addition to the previously mentioned question regarding IPF diagnosis, I struggled with the introductory question, “Can we start off by you describing how IPF affects your life right now?”, as the patients started talking about their IPF experience from different

angles including difficulty in carrying out daily activities, breathlessness and emotional changes. This is quite understandable since it is a difficult and long journey with a serious lifelong illness. However, these questions were meant to ease the discussion and help the patients feel more relaxed. As a result, in the first few interviews, I felt I was slow in pace and the interview time was lengthy. In IPF, fatigue was reported to be one of the most burdensome symptoms, and tiredness was reported in up to 95% of patients (Kahlmann;Moor and Wijsenbeek, 2020). Therefore, it is very critical not to overwhelm them and consume their energy with a long discussion. With experience, I learned to keep time. I began with introductory questions, but once the patients seemed relaxed, the questions related to eating and drinking were addressed more quickly; for example, I said, “Can I stop you for a moment, we will have a time to talk about this later”.

The transcripts for the first interviews revealed that I was using leading questions such as “Any symptoms like shortness of breath during or after the mealtime?”, “Because you have mentioned the toast and the dry food, that you feel like it tickles, have you made any changes to the type of diet you have? For example, softer food, mixing food with something to make it softer?” and “Do you think this is because of the antifibrotic medication side effect?” or yes/no questions such as “Do you feel that something might go the wrong way when you eat or drink?” and “Do you cough when you eat or drink?”. I was using the interview topic question-by-question to direct the interview rather than allowing the patient’s answer to guide the process. However, the intended aim of the topic guide was to remain focused on the area of interest, not to guide the discussion. Furthermore, I had difficulty in controlling the discussion and refocusing the patients on eating and drinking experiences when they started talking about their IPF experiences in general. I then met with my supervisory team, which helped in addressing the challenges with the data collection. They advised me on how to steer the discussion and smoothly transition to the next question. Additionally, they advised me on how to probe more, for example, “Could we for a moment go back when you have said you are taking a longer time to eat? Can you tell me more about it?”, “Can you remember the last time that happened when you are eating?” and “Can you explain more?”.

During the interviews, I established trust by speaking warmly and listening attentively, nodding to indicate my engagement. Despite the absence of visual cues during phone interviews, I maintained a compassionate tone and acknowledged patients' emotions.

Verbal cues, such as affirmations and paraphrasing, were utilised during video-conferencing interviews to demonstrate my understanding and support. Through empathy and active listening, I strived to create a safe and supportive space for patients to share their experiences.

As the interviews progressed, I became more resilient in steering the discussion and better at persuading patients to give more information on the topics being discussed. In addition, I was able to build a positive and friendly rapport and the patients felt comfortable while being interviewed. Below is a quote from an IPF patient as an example of the positive relationship during the interview.

"We have to have you, Amal, around for a meal" (Steve, IPF patient).

6.3.7.2 Self-reflexivity (positionality)

Self-reflexivity (positionality) refers to the researcher's location in terms of their own experience, identities, beliefs and values and how these shape their understanding and interpretation of the qualitative data (Corlett and Mavin, 2018). Considering positionality in qualitative research helps researchers to be more aware of the potential biases that may exist in their research and to reflexively consider their role in shaping the research process and outcome (Corlett and Mavin, 2018). This can also help to enhance the validity and reliability of the findings, as the researcher is able to bring greater self-awareness and critical reflection to their work (Johnson, 1997).

This thesis involved four studies that aimed to assess swallowing from different dimensions. Since those studies were conducted concurrently and not consecutively, I could not avoid coming to the qualitative research topic without prior knowledge that may have shaped my expectations and assumptions, which may in turn have influenced the questions I asked and the way I interpreted the data. However, being aware of this potential impact provided me with the opportunity to engage in reflexivity and to critically reflect on my own role in shaping the research process and outcome (McGhee;Marland and Atkinson, 2007).

My professional background as a respiratory therapist (RT) had an influence on this study. The respiratory care (RC) profession is an allied health profession that focuses on the assessment and treatment of patients who suffer from cardiopulmonary system

abnormalities, under the direction of a medical professional (Alotaibi, 2015; Kacmarek;Stoller and Heuer, 2019).

In my role as an RT, I performed a variety of tasks, including patient assessment, airway management, oxygen and aerosol therapy, bronchial hygiene therapy, mechanical ventilation management in critical care units and emergency departments, pulmonary function testing, pulmonary rehabilitation, and several other modalities (Kacmarek;Stoller and Heuer, 2019).

Therefore, In the first few interviews, my RT identity was the lens through which I viewed, probed and guided the discussion. I focused mainly on breathing issues that patients experienced when eating or drinking, such as cough, breathlessness and choking, rather than the eating and drinking experience in general.

I conducted role-play interviews with three experienced qualitative researchers to practice interviewing skills and minimise professional background power effects. The goal was to see how well the interview was flowing and to keep the focus on asking questions and probing more, and not to lead patients to my expectations (Bryman, 2016).

In addition, the semi-structured interviews were conducted in English, my second language, so I may have had a challenge understanding some of the colloquial English terms. This may have prompted patients to use more euphemistic words and phrases. However, this challenge led me to ask more follow-up questions to clarify the patients' responses and ensure I had a full understanding, which might be allowed for a deeper and richer understanding of their experience.

6.3.8 Ethical considerations

6.3.8.1 Ethical approval

The study was approved by the 211628_Non-substantial amendment 3 (NSA-3) of the North East-North Tyneside2 Research Ethics Committee REC reference 18/NE/0037 (see Appendix 9: approval letter).

6.3.8.2 Consent

In addition to the consent process described in the general methods chapter (section 2.6), at the start of the telephone or video interview, I read each step of the consent document to the participant and asked them to confirm whether they understood and agreed to this part of the process. Responses were audio recorded as evidence of consent.

6.3.8.3 Assessment and management risk

Participating in a qualitative interview may have emotional consequences for interviewees. I am a qualified RT experienced in working with people with a respiratory condition. If the patients did not wish to answer any question during the interview, this was respected. They were also be made aware that they were free to ask for the recording device to be stopped or the interview to be paused or ceased.

6.3.8.4 Data management

Interviews were recorded using a password-encrypted audio recording device (Sony ICD-TX650 Slim Digital PCM/MP3 Stereo Voice Recorder) or videoconferencing call platform (Zoom). The audio or video files were transferred as soon as possible from the encrypted recording device to be held on password-protected drives at Newcastle University. The original recordings were kept during the analysis period to allow a return to the data if needed for checking purposes.

The verbatim interviews were transcribed by a professional transcription company and client non-disclosure agreement forms were signed by this company before any interviews were sent to them. The interview transcripts were coded for confidentiality and anonymity before analysis and kept in secure, locked storage that was only accessible to the research team.

In the transcripts, any details specific to the patients' demographics were removed: for example, country of origin, name of the doctor treating them and other names mentioned during the conversation. Pseudonyms were used instead of the real names of the patients for the purpose of presenting these quotations.

6.3.9 Analysis

The data analysis was conducted using the inductive thematic analysis analyst-driven approach described by Braun and Clarke (2006 and 2021), who suggested the six-phase analysis method that was followed in this study (Braun and Clarke, 2006; Clarke and Braun, 2021). Thematic analysis described by Braun and Clarke (2012) is a method of analysing qualitative data that emphasises content and meaning to define and explore topics within a dataset. It is helpful in assessing and understanding the underlying themes and narratives within data. Moreover, it allows the researcher to look beyond the superficial layer of data to explore the impact of context and meanings(Braun and Clarke, 2012). Using thematic analysis, results are displayed using different levels of main overarching themes and subthemes within them. Themes are defined as capturing "something important about the data in relation to the research question and represents some level of patterned response or meaning within the data set" (Braun and Clarke, 2006, p. 82). On the other hand, subthemes are defined as "themes-within-a-theme, they can be useful for giving structure to a particularly large and complex theme, and also for demonstrating the hierarchy of meaning within the data" (Braun and Clarke, 2006, p. 92).

This study used thematic analysis as the analysis method for qualitative data, since it provided a better fit for the research question, identifying commonalities in experience and perception among participants about a common phenomenon (IPF disease). Using thematic analysis, the researcher was able to draw themes across all 14 interview transcripts for IPF patients on a semantic and explicit level.

An ongoing and iterative analysis of the transcripts was conducted. After each interview, I reviewed the recording and reflected on my responses and the patient's responses. This was done to inform the direction of the next interviews to ensure sufficient depth was reached.

The phases of thematic analysis process are outlined in Table 13 below.

Phase	Description of the phase
Familiarise myself with the data.	Transcribe the interview data. Initially read and re-read the transcripts. Highlight any interesting and important text in relation to the research topic and question. Document any initial thoughts.
Generate initial codes.	Code transcripts line by line and also in chunks of meaningful text.
Search for themes.	Group the codes into potential themes.
Review themes.	Check if the themes work in relation to the coded extracts and the entire data set. Generate a thematic "map" of the analysis.
Define and name the themes.	Refine the specifics of each theme and the overall story the analysis tells. Generate clear definitions and names for each theme.
Produce the report.	Final analysis. Write up the analysis using examples from the data for each theme.

Table 13. Summary of the six-phase thematic analysis approach described by (Braun and Clarke, 2006, p. 87)

The six-phase process of analysis of the transcripts was iterative and reflective, with a substantial overlap between the six phases with an attitude of inquiry and interpretation.

To ensure the analytical rigour of the results, I read and coded all the transcripts for the first two interviews. I discussed the initial coding scheme with a member of the supervisory team, who also coded the transcripts. Additionally, common codes and themes were discussed and approved by the supervisory team to ensure an accurate reflection of the data and to help guide the process and validity of the analysis.

The analysis in this chapter was performed using the qualitative data analysis software package NVivo R1.6 (QSR international). NVivo R1.6 was used to organise the codes, develop memos and create mind maps.

To ensure the quality of the thematic analysis, I followed the 15-point checklist criteria below for good thematic analysis (Braun and Clarke, 2006, p. 96).(see Table 14).

Process	Number	Criteria
Transcription	1	The data have been transcribed to an appropriate level of detail, and the transcripts have been checked against the tapes for "accuracy"
Coding	2	Each item has been given equal attention in the coding process
	3	The coding process has been thorough, inclusive and comprehensive
	4	All relevant extracts for each theme have been collated
	5	Themes have been checked against one another and against the original dataset
	6	Themes are internally coherent, consistent and distinctive
Analysis	7	Data have been analysed, interpreted and made sense of
	8	The analysis and the data match one another
	9	The analysis tells a convincing and well-organised story about the data and the topic
	10	A good balance between analytic narrative and illustrative extracts is provided
Overall	11	Enough time has been allocated to complete all phases of the analysis adequately
Written report	12	The assumptions about and the specific approach to the thematic analysis are clearly explicated
	13	There is a good fit between what the researcher claims to do and what he/she shows he/she has done
	14	The language and concepts used in the report are consistent with the epistemological position of the analysis
	15	The researcher is positioned as active in the research process (i.e. themes do not just emerge)

Table 14. The 15-point checklist of criteria for good thematic analysis

6.4 Results

Forty-two patients were invited to participate. Twenty-two patients did not respond to the invitation; 15 IPF patients (nine males and six females) were interviewed. Of those that consented to be interviewed, ten were recruited from the IPF support groups and five from the ILD clinic. Patients were interviewed via telephone calls (n: 5), Zoom video calls (n: 7) and FaceTime video calls (n: 3). One interview transcript was excluded from the analysis as during the interview, the patient reported a history of tongue cancer, making him ineligible for the study. See Figure 21 below.

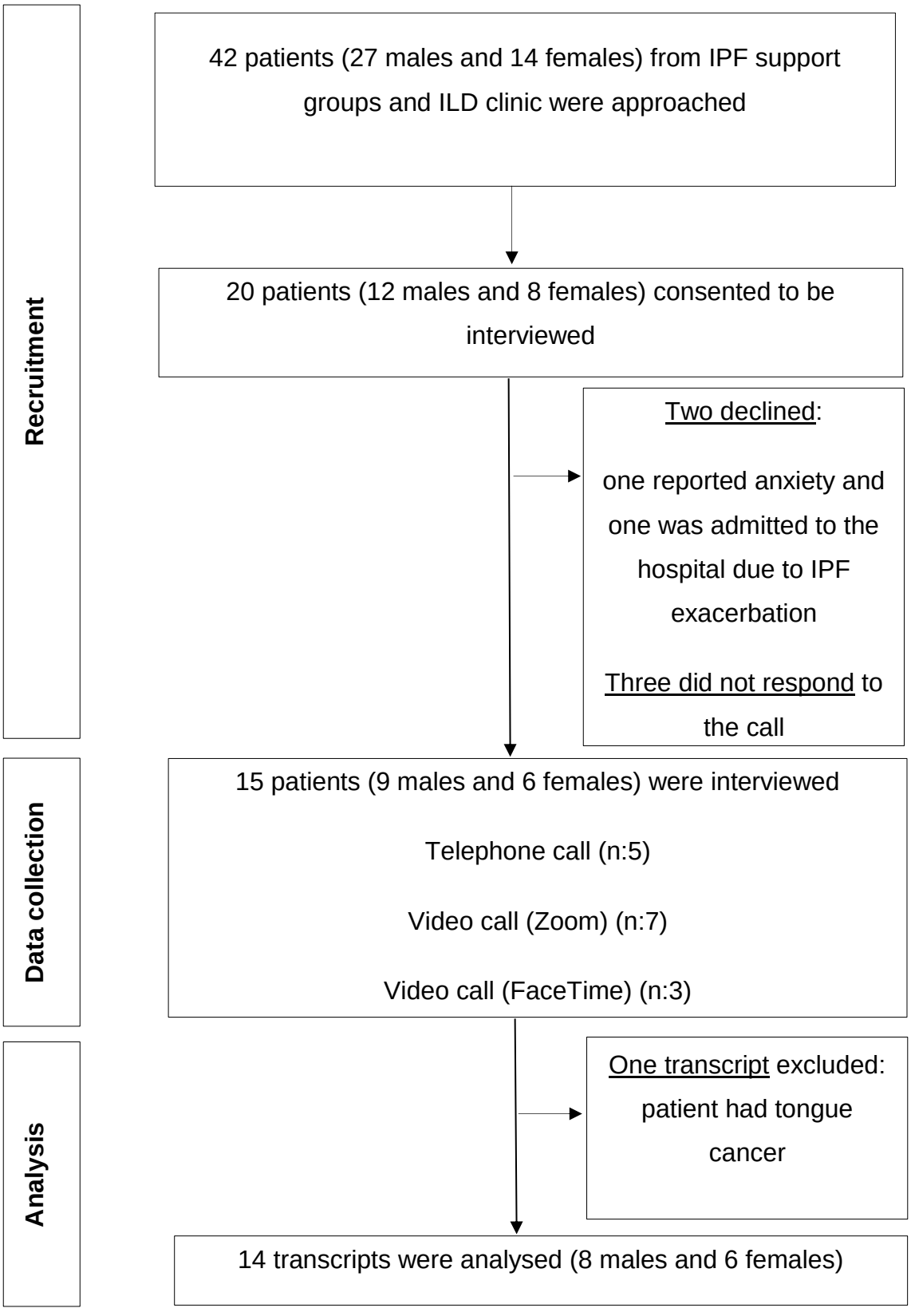

Figure 21. Consort flow diagram for the patient recruitment for the study.

6.4.1 Demographics and clinical characteristics

The patients' demographics and clinical characteristics are presented in Table 15. In total, 14 interview transcripts were analysed. The median age of patients was 71.0 years, with a range of 54 to 92 years. Eight (57%) were males. Most patients were non-smokers (10/14, 71%). Eight patients were using antifibrotic medications (8/14, 57%), and six (42.8%) patients were using Long Term Oxygen Therapy (LTOT). Most patients lived with partners (n: 10), while four patients lived alone.

Pseudonyms	Recruitment method	Interview method	Age (years)	Gender	Smoking	Using oxygen	Using antifibrotic medication	Living status
John	IPF support group	Video call (Zoom)	77	Male	Ex-smoker	No	Yes	With wife
Rob	IPF support group	Video call (Zoom)	70	Male	Ex-smoker	Yes	Yes	With wife
Sonny	IPF support group	Video call (Zoom)	64	Male	Non-smoker	No	Yes	With wife
Arthur	ILD clinic	Phone call	69	Male	Non-smoker	Yes	Yes	With wife
Henry	IPF support group	Phone call	75	Male	Non-smoker	No	Yes	With wife
Elisa	IPF support group	Video call (FaceTime)	65	Female	Non-smoker	Yes	No	With husband
Steve	IPF support group	Video call (FaceTime)	72	Male	Ex-smoker	Yes	Yes	With wife
Jackson	IPF support group	Video call (Zoom)	60	Male	Non-smoker	Yes	Yes	With wife
Ruby	IPF support group	Video call (Zoom)	73	Female	Non-smoker	Yes	No	Alone
Elizabeth	ILD clinic	Video call (Zoom)	87	Female	Non-smoker	No	No	Alone
Sarah	IPF support group	Video call (FaceTime)	92	Female	Non-smoker	No	No	Alone
Maggie	IPF support group	Video call (Zoom)	54	Female	Non-smoker	No	No	With husband
Anna	ILD clinic	Phone call	77	Female	Non-smoker	No	Yes	With husband
Russel	ILD clinic	Phone call	70	Male	Ex-smoker	No	No	Alone

Table 15. Recruitment methods and patients' characteristics

6.4.2 Themes

Three main themes were developed from the data: 1) “Eating, as such, is no longer a pleasure”; 2) “It is something that happens naturally and just try and get on with it”; and 3) “What is normal?”.

The first theme, “Eating, as such, is no longer a pleasure”, included two subthemes: 1) physical and sensory eating and drinking changes and their effects; and 2) the emotional and social impact of eating and drinking changes. This theme mainly focused on the physical and sensory changes associated with eating and drinking and their effects, and the subsequent emotional and social impact of these changes.

The second theme, “It is something that happens naturally and just try and get on with it”, included two subthemes: 1) direct strategies; and 2) indirect strategies. This theme centred on the self-determined strategies employed to manage changes to eating and drinking.

The final and third theme, “What is normal?”, included two subthemes: 1) information needs; and 2) patients’ beliefs. This theme focused on patients seeking information to better understand the changes in their eating and drinking, and the patients’ beliefs about what had changed their eating and drinking.

6.4.2.1 Theme 1: “Eating, as such, is no longer a pleasure”

This theme reports the experiences of patients with IPF that relate directly to the changes in their eating and drinking. The changes that were reported encompassed both physical and sensory aspects of the eating and drinking process and their effects and the patients’ emotional and social reactions to these changes.

Subtheme 1: physical and sensory eating and drinking changes and their effects

Patients experienced a range of problems related to their lived eating and drinking experience.

Breathlessness when eating and drinking was frequently reported by the patients. Maggie said she could not drink a cup of water without having to stop many times to catch her breath and continue drinking again, whereas before suffering from IPF that was never an issue:

“Before, I could have drunk, like, a bottle of water, taken quite a lot of it without having to stop. Now, it would be smaller mouthfuls that I would have to stop too. I couldn’t, basically: half a bottle or 500 ml of water before I continued drinking. Now, I would have just maybe a few mouthfuls and then you have to stop to get a breath and then continue drinking” (Maggie).

Rob attributed his experience with shortness of breath during meals physiologically, saying that when his stomach increased in size it reduced the amount of room for his diaphragm to move up and down and therefore reduced the amount of oxygen he could get into his lungs.

“I do have shortness of breath when I eat. If I feel like that, I put my oxygen on because I view eating in the same manner as exercising: my body needs the extra oxygen. If I eat too much, then the problem I face is that the diaphragm can’t. As the stomach expands, the diaphragm cannot expand in the normal manner so therefore it cannot get enough oxygen into my body. It makes it really difficult” (Rob).

Patients reported that it took them longer to eat and drink, due to their shortness of breath. For Maggie, this led her to plan her day in advance and adjust her routine to allow more time to finish her meal.

"It takes me longer to eat a meal because you always have to get a breath between every mouthful. Well, taking longer, like, you have all your plans worked out in your head, what you are doing next or whatever" (Maggie).

Food or drink being inhaled into the throat was mentioned by most of the patients. This had happened for Jackson a lot, especially when the food was dry, such as toast or crisps. He said:

"The other thing is when eating, the biggest thing is if I eat toast. Say I bite into some toast and the dry part of the toast is sort of inhaled into a certain part of your throat. God, I end up coughing really badly" (Jackson).

For John, he felt that something was stuck in his throat:

"There has been a sticky texture of food, and that might be, I don't know, a minute amount, and it seems as if it's lodged in my throat" (John).

This had also happened to Elisa with saliva, particularly when lying in bed.

"Drinking, I can drink and then maybe a little drop will go down the wrong way. But it does that even with saliva, quite honestly, especially when I'm in bed. And that can start me coughing. There are times when I'm eating when the food actually gets stuck, and I can't" (Elisa).

Coughing it out was reported by the patients if food or drink had gone down the wrong hole.

"Sometimes, something will, as we call [it], go down the wrong tube, like a crumb or something and then it starts [the] coughing, cough, cough, cough, although the biscuits we have, we like to nibble and break up and crunch, and a bit sometimes can whizz into the wrong tube" (John).

Almost all patients reported feeling fatigued and tired all day. Ruby, who lives alone, reported that sometimes she missed meals due to fatigue.

"I cannot cook so much, because standing can be quite exhausting, just standing. Some days I go without a meal because I'm so tired, I just stay in bed" (Ruby).

Similarly, Sarah, who lives alone, added that she lived on pre-prepared meals, *"the frozen foods"*, because she felt tired all day. Further, patients reported that they felt

tired and exhausted during and after eating, especially after their evening meal. After the evening meal, Elisa said that she always felt tired and wanted to sit on the sofa to rest. Steve also reported feeling very tired after the evening meal and normally went to bed. For Rob, eating often left him exhausted, and he saw it as similar to exercise.

Some of the other problems related to eating and drinking that patients described included mouth dryness, weight loss, and changes in taste and smell. Patients complained of persistent mouth dryness, especially at night. Most of the patients stated that they woke up a couple of times during the night to have a drink. Many kept a water bottle beside them on the bedside cabinet when they went to sleep. For example:

"I would say I drink a bit more. I drink water, perhaps once or twice in the evening, because my throat gets fairly dry, at night-time, when I'm asleep. I always carry water with me when I am going to bed" (Sonny).

Furthermore, experiencing changes in tastes while eating was reported by several patients, all of whom were on antifibrotic medications. Some patients stated that they had a metallic taste in their mouths, as described by Anna:

"Well, I couldn't taste my food properly because it has this iron-y taste in my mouth, and I couldn't taste my food. And before the fibrotic medication, I didn't have this horrible taste in my mouth" (Anna).

Other patients also experienced a change of taste. Rob mentioned that his taste had changed dramatically, and he was no longer interested in spicy food or very rich foods. He now tended to prefer plain food with very little sauce. Additionally, Arthur suffered from a loss of taste after starting antifibrotic medication.

"Well, what is the point of eating when I'm not hungry and I can't taste anything really?" (Arthur).

In addition to the changes in taste, patients reported that they had become very nauseous and felt they were less interested in food. Rob mentioned that both the sight of a plate full of food piled up and the smell of cooking food affected him. It also made him want to be sick and vomit. Steve attributed this change to the side effects of his antifibrotic medication.

"I suddenly become very nauseous and want to go to the toilet and whatever. I'd just come off nintedanib for one day, just one day. Once I've come off for one day and then it's okay, I come back to normal" (Steve).

However, Anna linked this with her overall experience with IPF:

"Well, with the IPF I didn't want to eat. I don't know" (Anna).

Several patients reported that they were no longer as interested in food as before and had lost their appetite:

"With the IPF I didn't want to eat. I don't know. I just didn't have really an appetite at all" (Anna).

"I've got a very poor appetite" (Elisa).

Due to these changes in taste, smell and appetite, portion sizes had decreased and patients tended to eat less than they used to.

"The evening meal, it's more of what a normal person would call a snack rather than a proper meal. Right, the portion of the food, that's gone down dramatically – probably down to a tea-plate size rather than, what I call, a dinner-plate size" (Rob).

Weight loss was a common concern for most of the patients.

"I've probably lost more muscle than fat, especially on my legs. I used to have quite broad thighs through my running: now they're like chicken's legs, according to my wife. That's where the majority of my muscle has gone" (Rob).

Patients thought they had lost weight because of their poor appetites and rarely feeling hungry.

"I think I've lost weight because the appetite isn't there so much, yes. Obviously, one leads to the other" (Henry).

Sarah compared her weight before IPF and now. She said in her prime she had been 9 stone, then decreased to 8 stone, and she was only 7 stone now. Other patients thought their weight loss was due to the side effects of the antifibrotic medication. Henry said:

"I weigh myself every week, as soon as I wake up on a Tuesday morning. I have lost about six pounds since starting the antifibrotic" (Henry).

Subtheme 2: the emotional and social impact of eating and drinking changes.

In this subtheme, wide ranges of negative emotions linked to eating and drinking were described by many patients. Fear of choking was the most common emotion expressed.

"Whenever this happens, you panic because you think you're going to choke. So, my reaction to that, my family laugh at me, when I get that the food seems to be stuck and I get the urges of, 'I'm going to choke', I jump to my feet" (Maggie).

Additionally, participants reported feeling worried about how their choking events might impact on others.

"I'm quite selective with food as well. I eat things that I know are not a major problem. Actually, I probably should have done, but my granddaughter comes after school, because we live next to her school, and I had had a little bit of not breathing very well when I was eating. So, I just said to her, 'If grandma is waving her hands, go and get her a drink. And if I collapse, call 999.' She's only 10, but I thought … I've worried her but I said, 'It's probably not going to happen, I just wanted you to know'" (Elisa).

The patients shared feelings of frustration and embarrassment about the increase in the duration of meals. The following quotes show how patients felt about the increase in eating time. Maggie felt frustrated about the time it took her to eat a meal.

"Whereas, before, I would have just maybe grabbed something and ran [sic]*, you can't do that now because it takes you ages to eat"* (Maggie).

Furthermore, Rob said that sometimes taking longer to eat left him not able to finish his meals:

"I don't have the time to finish them, because it takes ages and ages" (Rob).

In addition, some patients described feelings of embarrassment at taking longer to eat their meal than everyone around them. Ruby noted:

"I am definitely slower, though, because when I'm with my family, I'm always the last one, recently, to finish" (Ruby).

Eating no longer being an enjoyable or pleasurable experience was another common issue that was raised by some patients:

"It used to be. When I was fit and well, I really used to enjoy my food. Now it's something I do" (Rob).

Eating was simply seen as a task that had to be done as a means of nourishment:

"I don't think I enjoy my food as much as I used to, because of the coughing, choking, that's not easy. I don't enjoy my food. I have to eat to live" (Elisa).

A few patients outlined a general loss of interest in eating outside the home:

"I would get a bit nervous about what happens if this happens when I'm out, having a meal outside, in a restaurant or something. Because you feel like all eyes would be on you. So, yes, I suppose, because we haven't been out for a long time, I'd forgotten about that. Yes, it is a worry" (Elisa).

6.4.2.2 Theme 2: "It is something that happens naturally and just try and get on with it"

This theme covers strategies that patients developed in response to and to help cope with the changes in their eating and drinking. Adaptation strategies were divided into two categories: direct and indirect. The direct strategies were the methods of managing changes in eating and drinking that directly related to diet and eating and drinking habits. Indirect strategies involved alternative supportive methods used by patients that were not directly related to food and drink, such as using oxygen and medication, and going to sleep.

Subtheme 1: direct strategies

Patients described strategies used to manage the changes in their eating and drinking. Most reported being mindful about taking small, well-chewed bites, pacing the rate of eating and having regulated sips during the eating or drinking process. Maggie said that she chewed food much longer so as not to choke on it, and took frequent sips of a drink.

Patients also indicated that they focused on co-ordinating their breathing and swallowing to avoid breathing in while swallowing, according to Jackson:

"I've got to concentrate on my eating, to make sure that, again, I don't breathe in, for it to go – it's as if it's gone to my lung and not – it seems as if I'm coughing for a little particle to get out of my throat. It's really bad" (Jackson).

Some had developed techniques to minimise their anxiety. For example:

"I try and calm myself down, tell myself that I'm not choking, that the food will eventually go down and whenever you calm yourself down again and realise, 'Oh, the food's moving', that helps it." (Maggie).

"I'm trying not to draw my attention to it, because I think the more I realise it's happening, the more I'll think about it and make it happen" (Elisa).

A majority of patients had adapted their food preparation: for example, having softer food (steamed or mashed) and with additional sauces to reduce the likelihood of choking events and/or to reduce the effort of eating.

Patients reported that they frequently chose soft food: *"They are all mushed up"* (Sarah), and knew what food they should avoid to protect their airways: *"much softer and moist food, because I know I'm not going to choke on it, really"* (Elisa). Anna explained: *"If I do eat meat it has to be very lean and very thin"* and Steve joked that there would be no fish and chips (with laughter) as he did not like hard food, which was difficult to consume. Maggie shared: *"I have started steaming quite a lot of my food now. As I say, that helps the food stay moist, so it's easier to swallow that way"*.

Some patients reported being careful about their food choices. Fibre-rich and spicy foods were often avoided. This was due to the diarrhoea that many experienced as a side effect of the antifibrotic medication.

For example, *"I restrict my intake of fibre. If I eat too much, if I have too much fibre, which is what it is, then I can spend days on the toilet"* (Rob), *"No vegetables as such, I just can't tolerate them any more, Amal"* (Henry), and *"At one time I'd be quite happy with spicy food, now I have very bland food. I no longer enjoy spicy food"* (Sarah).

Apart from the effects of spicy food on bowel function, Maggie stated that she avoided spicy food because it caused coughing or choking.

In addition, several patients indicated that they avoided steak, because *"If I eat steak, it always seems to – even small pieces – it seems to stick in the back of my throat"*

(Maggie), *"Steak. Any meat that's hard to chew or to do anything. Any meat that is a big lump, you know what I mean?"* (Ruby), and *"It's more difficulty in swallowing"* (Elisa).

Subtheme 2: indirect strategies

Patients devised strategies not directly related to their diet and eating and drinking habits to cope with their eating and drinking changes. This was a smaller subtheme.

A few patients reported that they needed to use oxygen to help with the tiredness after their evening meal.

"I do have shortness of breath when I eat. If I feel like that, I put my oxygen on because I view eating in the same manner as exercising, my body needs the extra oxygen. If I eat too much, then the problem I face is that the diaphragm can't. As the stomach expands, the diaphragm cannot expand in the normal manner so therefore it cannot get enough oxygen into my body. It makes it really difficult" (Rob).

Others said that after eating, they needed to lie down on the sofa or go to sleep: *"After my dinner I sit down on the sofa and just recover. But I usually go to bed quite soon, I don't stay up late"* (Elisa). *"After the evening meal I normally go, I'm very tired and I normally go to bed"* (Steve). One patient mentioned that he took anxiety medication before mealtimes to calm himself down.

6.4.2.3 Theme 3: "What is normal?"

The final theme focused on patients' beliefs and information needs regarding changes in eating and drinking.

Subtheme 1: information needs

Some patients showed a desire to learn as much as possible and sought information regarding the changes in eating and drinking they experienced. For example:

"In fact, doing it with you, it wasn't the money side of it, so much, I wanted to learn more from you than probably you learnt from me" (Ruby).

Some wondered if their experience was similar to others with the same condition. Several patients stated that they had discussed the IPF changes with other patients in order to learn how they had experienced it. For example:

"I know some people do because we go to a group. We used to go to a group at Middlesbrough and there were people there who had IPF and it affected their appetite quite badly" (Henry).

"I know from other people with the disease that they go through exactly the same thing. They do have weight loss and they don't like eating, they don't like the food that they're given. I'm not an exception to the rule, if you know what I mean" (Rob). Steve added, *"I'm not alone in feeling like that. I know, from my friends who have this disease, they're in exactly the same position"* (Steve).

One patient noted that their interview made him consider swallowing services. *"After meeting you I have been thinking about swallowing service[s]. I didn't know about swallowing service[s] and nobody suggested that"* (Rob).

Subtheme 2: Patients' beliefs

Several patients attributed their eating and drinking problems to the ageing process rather than their IPF: *"Well, you see, when you reach my age, you've got lots of other problems as well, so you don't, but I wouldn't say it was anything to do with the IPF, I just eat a bit less now, perhaps, than I did, but it hasn't made a huge difference"* (Elizabeth), and *"That could be an age thing, but it's never, ever really thinking it's [because] of IPF"* (John).

Additionally, Henry explained his abstinence from spicy foods as a result of his age, saying, *"I won't be eating as much spicy food as I used to"* (Henry). In the opinion of Anna, as you get older, you do not need as much food. Therefore, she did not eat as much.

Some patients had not considered changes to their eating and drinking prior to their interview, as there were many other symptoms to contend with. *"Certainly, the eating and drinking and swallowing side probably is slightly worse than what it was, but I tend, maybe, not to notice that because of these other symptoms. I don't really feel I've got a problem with swallowing at the minute"* (Arthur). Furthermore, Henry added: *"Because of the other parts of the condition, like the cough and the chest discomfort, I don't tend to think much about the swallowing side of it to be honest"* (Henry).

6.5 Discussion

The aim of this chapter was to explore the lived eating and drinking experience of patients with IPF. The findings demonstrate that some patients have substantial struggles and challenges with eating and drinking, affecting them physically, emotionally and socially. The findings can be summarised under three interrelated themes: 1) “Eating, as such, is no longer a pleasure”; 2) “It is something that happens naturally and just try and get on with it”; and 3) “What is normal?” To my knowledge, this is the first study to report on IPF patients’ lived experience of swallowing changes associated with their diagnosis. Understanding patients’ experiences is beneficial in planning for patient-centred care and to develop better patient information in this area, and this information is complimentary to the earlier quantitative data in my thesis.

Patients reported breathlessness during meals. This is perhaps not surprising as dyspnoea is a commonly reported symptom of IPF across many activities of daily life (Guenther et al., 2018). Elsewhere, qualitative work has also identified breathlessness as affecting daily life activities of people with IPF, such as carrying shopping, bending at the waist and taking a shower (Swigris et al., 2005; Lyu et al., 2021). This is the first study where patients have reported this symptom also impacting on their eating and drinking.

This study focused on the patients' experience of eating and drinking, which revealed that patients frequently had to pause to catch their breath while drinking. They have also described the effects of breathlessness as not being able to get enough air in to their lungs while eating, which resulted in them taking more time to finish their meals. This could be attributed to the expansion of the stomach during digestion, which can push against the diaphragm and lungs, making it harder to breathe (Wolkove et al., 1998). Additionally, eating can also increase the demand for oxygen in the body, further exacerbating breathlessness in those patients with already compromised lung function (Ueda et al., 2002). The patients shared some techniques they applied in order to alleviate breathing discomfort, including eating more slowly, eating smaller meals and eating more frequent meals throughout the day rather than large, heavy meals. These findings are in line with previous qualitative literature in patients with COPD (Lin and Shune, 2022). On the other hand, taking longer to eat can have several consequences, such as food going cold, feeling fuller earlier and difficulty in maintaining nutritional intake. Further, these consequences can impact not only physical health but also social

life, as the pacing of eating is difficult; this may impact on the ability to maintain conversation over mealtimes. Feelings of being ashamed and frustrated when taking longer and not managing to finish meals on time when being with other people were also reported by the patients.

The interviews also shed light on other issues that contributed to the challenge that the IPF patients experienced with eating and drinking. Fatigue, a state of tiredness, was reported to have an impact on activities of daily living by the patients. This is consistent with the findings of the European IPF registry (eurIPFreg), which reported that dyspnoea, fatigue and loss of appetite were the most common clinical symptoms experienced by patients with IPF, with percentages of 90.10%, 69.20% and 67.40% respectively (Guenther et al., 2018). In this study, the patients reported experiencing fatigue and the need to rest after eating. The extra effort required for eating and drinking was likely to curtail meal times, which could potentially affect their nutritional intake. The relationship between fatigue and difficulty in swallowing has also been reported in other groups of individuals with swallowing disorders (Patterson et al., 2009).

Prior studies have examined the eating situations of patients with COPD, and the results indicate that patients often experience a decrease in oxygen saturation levels while eating or shortly afterwards, leading to the need for supplemental oxygen (Schols *et al.*, 1991; Schenkel *et al.*, 1996; Gray-Donald;Carrey and Martin, 1998; Wolkove *et al.*, 1998; Vermeeren *et al.*, 2001). Similar findings were reported by IPF patients in this study, with patients describing feeling fatigued after meals and requiring the use of oxygen and rest. In addition, IPF patients who live alone mentioned experiencing fatigue while cooking and preparing meals, resulting in their reliance on pre-made meals. A previous study on COPD patients reported that 23% of the patients experienced fatigue while preparing meals (Odencrants;Ehnfors and Grobe, 2005).

Fear of choking was the most common emotion expressed. Patients used words such as “panic” and “scary” when describing their experience of choking. This fear was typically associated with worries about being able to breathe. Choking-related fear was associated with avoidance of eating, adopted by the patients as a safety behaviour (Treasure;Cardi and Kan, 2012).

Avoidance behaviour refers to any action or behaviour taken by an individual to avoid a particular situation, task or stimulus that causes discomfort, anxiety or fear (Krypotos

et al., 2015). Avoidance behaviour can be a natural response to perceived threats or stressors, but it can also become a maladaptive coping mechanism that interferes with daily functioning and overall well-being (Krypotos et al., 2015). Avoidance behaviour in eating and drinking can manifest in various ways, including restricting food intake, avoiding certain types of food or food groups, and avoiding situations that involve eating or drinking in public or with others (Day et al., 2021).

In addition to the fear, patients in this study reported a range of emotional responses related to their experiences with eating and drinking, including embarrassment, anxiety, panic, anger, sadness and frustration. This is consistent with broader previous research on dysphagia (Eslick and Talley, 2008; Howells et al., 2021). Studies on anxiety in patients with IPF have shown a high prevalence of anxiety (31%) and its association with reduced QoL (Luppi et al., 2021). While this study did not examine emotional responses related to other daily activities, it is likely that the emotional distress reported by the IPF patients in this study contributes to the broader emotional distress observed in earlier literature (Antoniou et al., 2020).

In this study, a few patients experienced a general loss of interest in eating outside their home. However, they did not report feeling socially isolated due to their swallowing issues, which was different from other studies conducted with different populations (Ekberg et al., 2002; Patterson et al., 2009; Nund et al., 2014).

It is worth noting that the study was conducted during the COVID-19 lockdown, which meant that people were limited to socialising at home. Consequently, the study may not have fully captured the effects of eating and drinking problems on the social lives of patients with IPF, since social life was restricted during the pandemic.

Patients discussed various methods that they had tested by trial and error to enhance their overall eating and drinking experiences. Paterson and Thorne (2000) refer to this process as the “evolution of expertise”, which occurs as individuals with chronic illnesses learn to manage their symptoms over time (Thorne;Nyhlin and Paterson, 2000). These methods comprised adjusting the texture and size of their food, eating at a slower pace, chewing their food thoroughly, taking small sips of water, practising mindfulness while eating, using supplemental oxygen after the meal, resting, and going to sleep afterwards.

Previous studies in patients with COPD have reported similar results, which suggest that individuals with COPD should modify their eating habits to ease breathing difficulties (Hoit et al., 2011; Lin and Shune, 2022). Similar approaches were employed by patients with head and neck cancer, which involved consuming smaller meal portions, reducing overall food consumption and opting for softer foods (Patterson et al., 2009). It was clear that the level of eating ability varied from day to day, depending on the severity of IPF symptoms, making it challenging for the IPF patients to plan meals or social activities beforehand.

The patients often did not attribute their swallowing status to IPF. In their view, the eating and drinking changes observed were influenced by other underlying medical conditions such as dental issues, diabetes and the chronic nature of the IPF disease itself. Furthermore, several patients linked their changes in eating and drinking to old age. Previous studies in different populations have shown that individuals with swallowing difficulties tend to not seek treatment due to the perception that such problems are a natural aspect of getting older. For example, in a study conducted by Turley and Cohen (2009), only 22.6% of elderly individuals with swallowing problems sought medical help. They also showed that the most common reasons for not seeking treatment were a lack of knowledge about the treatment options available and a belief that the symptoms experienced were a natural part of the ageing process (Turley and Cohen, 2009).

6.5.1 Strengths and limitations

To my knowledge, this is the first study using qualitative research design to describe the eating and drinking experience in patients with IPF. The strength of this study includes the sample, which was diverse regarding age, gender and living status. The patients involved were from various locations in the UK. Although this study helped to shed light on patients' experiences, it has some limitations that need to be addressed. 1) The severity of IPF was not clearly defined and characterised. Its severity may potentially influence a patient's experience of eating and drinking, and the study may have not adequately accounted for the variability in the severity of IPF among the patients. However, access to the patients' medical files was not possible for the patients recruited from the pulmonary fibrosis support groups. 2) Due to the pandemic, the interviews were conducted via phone calls or videoconferencing platforms, which required participants to have access to the internet or a telephone, and it is likely that

this study was inaccessible to individuals without the required digital equipment or skills, or who might not feel comfortable participating in a remote interview due to, for example, communication problems.

6.5.2 Clinical implications

The study indicates that IPF patients face physical, social and emotional consequences due to the changes in their eating and drinking abilities, which may significantly impact their overall QoL. The findings in this study emphasise the importance of educating both patients and healthcare providers about the swallowing changes that occur in IPF. Such education could increase awareness of potential challenges in eating and drinking and might ultimately help to reduce the risk of additional health problems resulting from exacerbation of the disease. It is noteworthy that patients have been innovative in finding their own solutions to cope with these symptoms. This presents an opportunity to develop resources and materials based on their experiences. By capitalising on this patient-driven approach, valuable resources could be created to support individuals living with IPF and their specific needs related to eating and drinking.

Chapter 7. Final discussion and suggested future steps.

The aim of this research project was to explore swallowing in patients diagnosed with idiopathic pulmonary fibrosis (IPF) including perceived changes in their ability to eat and drink. To that end, this research sought to address four specific objectives: (1) describe swallowing and oropharyngeal swallowing physiology and safety using the gold standard instrumental assessment, the Videofluoroscopy Swallow Study (VFSS); (2) assess swallowing safety and performance using a simple screening Water Swallow Test (WST); (3) measure symptoms of swallowing dysfunction, laryngopharyngeal reflux and laryngeal hypersensitivity using three validated questionnaires; and (4) explore the lived eating and drinking experience using a qualitative research design.

This study employed various research designs, including both qualitative and quantitative approaches, in order to accomplish the research, aim and objectives. The core findings have been discussed in previous chapters. Hence, the purpose of this chapter is to present the overall summary of the thesis and propose potential future steps and direction.

7.1 Summary of findings

Chapter 3 of the thesis presented the initial empirical findings, aiming to describe the perception of swallowing difficulty, oropharyngeal swallowing physiology and safety among IPF patients. The analysis revealed a variety of impairments in oropharyngeal swallow physiology and safety, including instances of aspiration into an unprotected airway, observed during the VFSS. These results indicate that a subset of patients in this population may be susceptible to swallowing dysfunction, penetration and/or aspiration. Furthermore, the findings from this chapter emphasise the importance of conducting additional research to establish a clear association between dysphagia and IPF.

Chapter 4 of the thesis presented the first attempted study that aimed to screen swallowing dysfunction in patients with IPF using the WST. This study showed that the WST can be conducted online effectively in community and hospital clinics. The WST identified signs of penetration and/or aspiration, and there was evidence suggesting that swallow performance may be influenced by gender and age. Furthermore, the

study revealed that IPF patients exhibited lower swallow performance compared with age- and sex-matched individuals without dysphagia from existing published data. These findings suggest that IPF patients may potentially experience dysphagia and indicate that the WST could serve as a valuable clinical screening tool to identify IPF patients at risk of swallowing dysfunction.

Chapter 5 of the thesis presented the analysis of IPF patients' perspectives on swallowing dysfunction, laryngopharyngeal reflux and laryngeal hypersensitivity symptoms by employing specific patient-reported outcome (PRO) measures. The study findings revealed that a considerable proportion of IPF patients experience symptoms such as swallowing difficulties, laryngopharyngeal reflux and discomfort in the larynx.

In the qualitative study, some patients expressed a degree of change in eating and drinking, which affected them physically, emotionally and socially. They employed different coping strategies to manage these challenges. Additionally, the patients demonstrated a strong desire to acquire knowledge and sought information regarding the changes they were experiencing in their lived eating and drinking experience. These findings align with previous qualitative studies conducted on patients with chronic obstructive pulmonary disease (COPD). To the best of my knowledge, this study is the first of its kind to explore the lived experiences of IPF patients in relation to eating and drinking through a qualitative research approach. This understanding of patients' experiences is important for the development of patient-centred care and the improvement of patient information in this area.

7.1.1 Suggested future steps

The implications of each study are discussed in the respective chapters. In this section, I summarise and highlight suggested future steps.

The findings of these studies establish an initial knowledge base regarding swallowing dysfunction in IPF patients, an aspect of IPF pathophysiology that has not been studied. This study emphasises the importance of enhancing the detection of dysphagia, providing adequate information to the medical community and patient support groups, and exploring interventions for individuals with IPF. It is anticipated that the outcomes of these studies will contribute to enhanced healthcare provision for patients with IPF, with a specific focus on addressing swallowing issues.

- Future studies should address the need to improve both detection of dysphagia symptoms and the development of effective interventions for people with IPF.
- It would be beneficial to conduct research with larger study cohorts, including control groups, and employ longitudinal designs to achieve more conclusive results regarding swallowing in IPF.
- Investigating the oesophageal swallowing stage in conjunction with the oral and pharyngeal stages would be valuable in understanding the comprehensive swallowing process in patients with IPF. These avenues of research would contribute to the advancement of knowledge and the development of better management strategies for individuals with IPF and swallowing difficulties.

Chapter 8. Review of the impact of COVID-19 disruption

The COVID-19 pandemic had a significant impact on my previous PhD project, which focused on patients with pulmonary fibrosis. I commenced my PhD in September 2018; it involved conducting physiological measurements and using a nasoscope called the *Flexible* Endoscopic Evaluation of Swallowing (FEES), which is known to generate aerosols. I had written the full study protocol, prepared all the required documents, obtained university ethics approval for the healthy control group and applied for NHS approval for the patient group (Appendix 19: the study protocol) (Appendix 20: university ethical approval letter for healthy older volunteers).

I also started a reproducibility study for the physiological measurements and successfully recruited ten healthy volunteers, who participated in two visits each (Appendix 21: Simultaneous Assessment of Swallowing and Breathing Coordination (preliminary reproducibility study)).

Moreover, I attended various demonstration sessions where I learned to operate the PowerLab data acquisition device for the physiological measurements. As a respiratory therapist, I had no prior experience with FEES. Consequently, I underwent training provided by the speech voice swallow (SVS) associates and gained practical experience in conducting scoping sessions using FEES at Nottingham Hospital in November 2019 (Appendix 22G: FEES training certificate).

However, the COVID-19 pandemic brought about significant disruptions, leading to indefinite suspension of the ethical approval for the study, which I had prepared and submitted to the Integrated Research Application System (IRAS). In light of this unforeseen situation, my supervisory team and I adapted by conducting online meetings via Zoom. Together, we made the decision to shift the research focus towards a topic that held greater relevance and applicability within the context of the pandemic.

Throughout the pandemic, my supervisory team and Newcastle University provided immense support. Newcastle University granted me a thesis submission extension, and Imam Abdurrahman bin Faisal University provided me with a fully funded extension due to the pandemic, allowing me to successfully complete my project.

The pandemic presented us all with an unprecedented situation that impacted individuals in various ways. Despite the hardships, I recognise the valuable lessons I learned, the experiences I gained and the support I received. This challenging journey taught me the significance of resilience, flexibility and adaptability not only in academia, but also in all aspects of life.

Throughout this process, I experienced personal and professional growth. I learned to cherish the small joys in life, the moments shared with loved ones and the importance of good health. Moreover, I developed a deeper appreciation for the research process and its vital role in addressing global health challenges. It is my hope that my research contributes to advancing the understanding of pulmonary fibrosis and ultimately makes a positive difference in the lives of those affected by this debilitating disease.

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Appendices

Appendix 1: The research information sheet.

INFORMATION SHEET FOR PARTICIPANT

Research team:

Dr Ian Forrest Consultant Respiratory Physician (Chief Investigator)

Mrs. Amal Alamer (PhD student)

Prof. Chris Ward (Respiratory physiology, Academic supervisor)

Prof. Joanne M Patterson (Speech and language therapist, Academic supervisor)

Dr. Michael Drinnan (Medical physics, Academic supervisor)

Translational and Clinical Research Institute

Faculty of Medical Sciences

Newcastle University

• Introduction

My name is Amal Alamer, I am a Respiratory Therapist. I am doing my PhD at Newcastle University, United Kingdom. I would like to invite you to take part in my research. Please note that you do not have to take part in this research and you do not have to decide today. Also, feel free to talk with anyone you feel comfortable with about taking part in this research. If at any point you feel that you need more clarification, you can ask me about it. If you have any questions or concerns at a later stage, I would be happy to take the time to discuss the study with you.

• Purpose of research

The purpose is to understand the swallowing patterns of patients diagnosed with Idiopathic Pulmonary Fibrosis (which we will call pulmonary fibrosis from now on) and any perceived changes in their ability to eat and drink. It will help us to predict who may be at risk of problems when swallowing.

• Why have I been chosen?

You are being invited to take part in this research because you have pulmonary fibrosis.

- ## Type of Research

  This is a descriptive research study which will include:

- Completion of three short questionnaires about your swallowing and throat symptoms. The questionnaires will be sent to you via email or post depending on your preference.

- An interview that will be conducted via telephone or video conference (for example, Zoom, Skype or FaceTime). The interview will mainly be about your eating and drinking experience, any swallowing problems and your journey with pulmonary fibrosis. Following that, the whole interview will be transcribed verbatim in order to analyse it. The transcript will be anonymised and any other identifiable information such as names will be removed. If you feel uncomfortable answering any question, you can say no. If at any time you feel you want to stop, rest or terminate the interview, you can do so. You can also ask to destroy the interview record if you wish. The interview may take around an hour (60 minutes).

- A structured swallow test. The swallow test will be conducted to assess your swallowing function and your ability to drink. In this test, you will be asked to drink a measured amount of water. Your airway response and voice changes will be observed during and after drinking. The measured calibrated cup that will be used for administering water during the test will be sent to you via post once the date for the test is confirmed. The test will take less than30 minutes.

- ## What do I have to do if I take part?

  If you decide to take part in this research, you will have the option to choose between the four participation options:

  1. Participate in the questionnaires only.
     Or

  2. Participate in the questionnaires and interview.
     Or

  3. Participate in the questionnaires and swallow test.
     Or

  4. Participate in the whole study (questionnaires, interview and swallow test). The interview and swallow test can be completed at the same meeting or two separate meetings depending on your preference.

- **Duration**

The interview will usually take about an hour although the duration of the interview may differ slightly between participants. The swallow test usually takes 10 t0 15 minutes.

- **Do I have to take part?**

No. The decision to participate is entirely up to you. We will describe the study here, and if you have further questions then you are welcome to contact us. If you are interested, then we will ask you to sign a consent form before taking part in the study.

- **Risks**

We did not identify any risks that may be associated with you being interviewed. However, if at any point you become distressed, feel uncomfortable or tired, please let me know so we can address your concerns and/or terminate the session.

- **Benefits**

There will be no direct benefits from taking part, but the information that we get from this study will help in designing and conducting future studies.

- **Confidentiality**

If you take part in this research, all your information will be kept private and will be treated with complete confidentiality. You will be assigned a code through which you will be identified only by me and no one else will have access to this information. After the completion of the interview analysis, your name and any other identifiable information will be destroyed.

- **Sharing the results**

All the information you share with us will not be shared with anybody outside the research team and nothing will be attributed to you by name. The information gathered will be for PhD research and anonymised results may be disseminated in scientific journals and meetings so that other people who are interested in the topic may learn from the results.

- **Right to refuse or withdraw**

You do not have to take part in this research. Whether you decide to participate or not, your decision will not affect the service you receive. If at any time you feel that you want to withdraw from the research, you can do so without any concerns. You can also ask for the digital recording to be destroyed.

- ## Who to contact

If you have any questions, you can ask them now or later. If you wish to ask questions later, you may contact me by email at A.Alamer2@ncl.ac.uk or telephone on +447462649394.

Appendix 2: Certificate of consent for the patients

The Newcastle upon Tyne Hospitals NHS
NHS Foundation Trust

An exploratory study of swallowing function and patient perceptions in IPF

Chief Investigator: Dr. Ian Forrest

Patient Identification Number for this trial:

Please initial box to Indicate agreement

1.	I confirm that I have read and understood the foregoing information, and that I have had the opportunity to ask questions about it and any questions I have asked have been answered to my satisfaction.	
2.	I understand that my participation is voluntary and that I am free to withdraw my consent at any time, without giving any reason, without my medical care or legal rights being affected.	
3.	I understand that information from my medical notes will be recorded and stored. This information will be anonymised but linked to you via a separate file kept by Dr Forrest in a locked NHS consultants' office.	
4.	I understand the data collected about me will need to be stored on paper and electronic formats for later analysis by the study team under the direction of Dr Forrest.	
5.	A copy of this completed informed consent form will be given to you. In addition the original will be securely filed.	
6.	I agree to my data being used to investigate swallowing function in IPF.	
7.	I agree that my GP can be contacted by the research team about my involvement in this study	

_____ _____ _____

Name of Patient Date Signature

_____ _____ _____

Name of Person taking consent Date Signature

Appendix 3: Oropharyngeal swallowing pathophysiology in patients with idiopathic pulmonary fibrosis: A consecutive descriptive case series. Poster presentation in the European Respiratory Society (ERS) International Congress 2020.

EUROPEAN RESPIRATORY *journal*
FLAGSHIP SCIENTIFIC JOURNAL OF ERS

Oropharyngeal swallowing pathophysiology in patients with idiopathic pulmonary fibrosis: A consecutive descriptive case series

Amal Alamer, Rhys Jones, Chris Ward, Michael Drinnan, Alexander John Simpson, Michael Griffin, Joanne Patterson, Ian Forrest
European Respiratory Journal 2020 56: 3370; **DOI:** 10.1183/13993003.congress-2020.3370

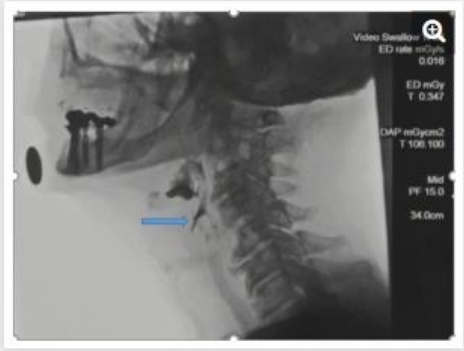
Abstract

Introduction: Research into swallowing dysfunction (dysphagia) in IPF is limited. Dysphagia is seen in other respiratory conditions such as COPD, increasing the risk of pulmonary complications secondary to aspiration.

Aim: Explore the oropharyngeal swallow of IPF patients.

Methods: Ten consecutive IPF patients from the Newcastle Interstitial Lung Disease clinic were recruited. Each had videofluoroscopy, and were given measured amounts of food and drink. Videofluoroscopies were analysed using validated scales: Penetration-Aspiration Scale (PAS); Modified Barium Swallow Impairment Profile (MBSImP).

Results: Seven males, three females, mean age 66 (52-78) were recruited. Three had airway penetration. One aspirated liquid without a cough response. Mean MBSImp for oral impairment was 4 (95%CI 4-4) and pharyngeal impairment 8.3 (95%CI 5.6–11.0), indicating mild alterations to swallowing physiology.



Method: Videofluoroscopy indicating accumulation of Barium-labelled liquid in oropharyngeal cavity with penetration (arrowed) in IPF patient.

Conclusion: To our knowledge, this is the first report on Swallowing Pathophysiology in IPF. We believe a proportion of this group may be at risk of aspiration. Further work is indicated to fully explore swallowing in this vulnerable group.

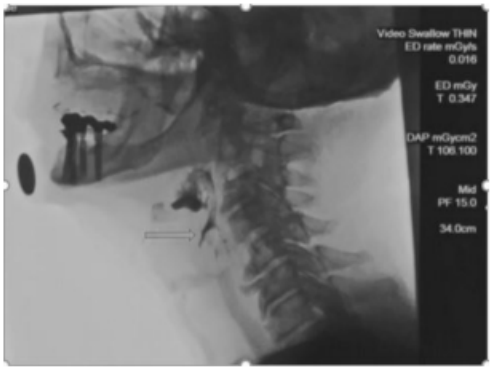
Appendix 4: Oropharyngeal swallowing pathophysiology in patients with idiopathic pulmonary fibrosis: A consecutive descriptive case series. Oral presentation at the British Thoracic Society (BTS), winter meeting, 2020.

S127 **OROPHARYNGEAL SWALLOWING PATHOPHYSIOLOGY IN PATIENTS WITH IDIOPATHIC PULMONARY FIBROSIS: A CONSECUTIVE DESCRIPTIVE CASE SERIES**

[1]A Alamer, [2]R Jones, [2]C Ward, [3]M Drinnan, [2]AJ Simpson, [3]M Griffin, [4]J Patterson, [3]I Forrest. *[1]Imam Abdulrahman Bin Faisal University, Dammam, Saudi Arabia; [2]Newcastle University, Newcastle Upon Tyne, UK; [3]Newcastle Upon Tyne NHS Trust, Newcastle Upon Tyne, UK; [4]Univerity of Liverpool, Liverpool, UK*

10.1136/thorax-2020-BTSabstracts.132

Introduction Research into swallowing dysfunction (dysphagia) in IPF is limited. Dysphagia is seen in other respiratory conditions such as COPD, increasing the risk of pulmonary complications secondary to aspiration.
Aim Explore the oropharyngeal swallow of IPF patients.
Methods Ten consecutive IPF patients from the Newcastle Interstitial Lung Disease clinic were recruited. Each had videofluoroscopy, and were given measured amounts of food and drink. Videofluoroscopies were analysed using validated scales: Penetration-Aspiration Scale (PAS); Modified Barium Swallow Impairment Profile (MBSImP).



Abstract S127 Figure 1 Videofluoroscopy indicating accumulation of Barium-labelled liquid in oropharyngeal cavity with penetration (arrowed) in IPF patient.

Results Seven males, three females, mean age 66 (52–78) were recruited. Three had airway penetration. One aspirated liquid without a cough response. Mean MBSImp for oral impairment was 4 (95%CI 4–4) and pharyngeal impairment 8.3 (95%CI 5.6–11.0), indicating mild alterations to swallowing physiology.
Conclusion To our knowledge, this is the first report on Swallowing Pathophysiology in IPF. We believe a proportion of this group may be at risk of aspiration. Further work is indicated to fully explore swallowing in this vulnerable group.

S128 **WHAT IS BEST IN THE FOLLOW UP OF UNCLASSIFIABLE PULMONARY FIBROSIS?**

[1]L Horgan, [2]L Smith, [1]T Sutherland, [1]A Boland, [1]M Darby, [1]R Bishop, [1]P Beirne. *[1]Leeds Teaching Hospitals Trust, Leeds, UK; [2]Faculty of Medicine and Health, University of Leeds, Leeds, UK*

10.1136/thorax-2020-BTSabstracts.133

Current UK guidelines suggest at least 1 year of follow-up for Unclassifiable Pulmonary Fibrosis (UPF), to identify those who develop progressive lung disease, and require therapeutic intervention. There is no reliable evidence base as to the optimum duration of follow-up. The aim of this study was to determine appropriate follow up duration, and which investigations were most informative.

A tertiary Interstitial Lung Disease (ILD) centre, UPF patients over a 8 year period (2011–2019), and a contemporaneous group of IPF patients (diagnosed at ILD MDT) were identified. Parameters collated included lung function (PFTs), oxygen saturations, symptoms, and review duration.

Diagnosis revision in the UPF group was recorded, as was its trigger (radiology, serology, symptom/PFT decline). All patient deaths were recorded, and where possible, cause of death.

Mean difference in baseline PFT values between IPF and UPF were estimated by t-tests. Linear regression models were used to estimate the mean difference adjusting for baseline values. Kaplan-Meier survival curves were calculated for IPF and UPF patients. Standard deviation and boxplots were used to describe the distribution of PFT parameters in each group.

Appendix 5: Paper: Oropharyngeal swallowing physiology and safety in patients with Idiopathic Pulmonary Fibrosis: a consecutive descriptive case series

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RESEARCH Open Access

Oropharyngeal swallowing physiology and safety in patients with Idiopathic Pulmonary Fibrosis: a consecutive descriptive case series

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Abstract

Introduction: Dysphagia occurs in multiple respiratory pathophysiologies, increasing the risk of pulmonary complications secondary to aspiration. Reflux associated aspiration and a dysregulated lung microbiome is implicated in Idiopathic Pulmonary Fibrosis (IPF), but swallowing dysfunction has not been described. We aimed to explore oropharyngeal swallowing in IPF patients, without known swallowing dysfunction.

Methods: Fourteen consecutive outpatients with a secure diagnosis of IPF were recruited and the 10-item Eating Assessment Tool (Eat 10) used to assess patient perception of swallowing difficulty. Oropharyngeal swallowing was assessed in ten patients using Videofluoroscopy Swallow Studies (VFSS). The studies were rated using validated scales: Penetration-Aspiration Scale (PAS); standardised Modified Barium Swallow Impairment Profile (*MBSImP*).

Results: EAT-10 scores indicated frank swallowing difficulty in 4/14 patients. Videofluoroscopy Studies showed that 3/10 patients had airway penetration, and one aspirated liquid without a cough response. Median *MBSImp* for oral impairment was 5, range [3–7] and pharyngeal impairment 4, range [1–14] indicating, overall mild alteration to swallowing physiology.

Conclusion: We conclude that people with IPF can show a range of swallowing dysfunction, including aspiration into an unprotected airway. To our knowledge, this is the first report on swallowing physiology and safety in IPF. We believe a proportion of this group may be at risk of aspiration. Further work is indicated to fully explore swallowing in this vulnerable group.

Keywords: Idiopathic Pulmonary Fibrosis, Videofluoroscopy Swallow Study, Oropharyngeal swallowing, The Modified Barium Swallow Impairment Profile, Aspiration, The Eating Assessment tool EAT-10

Introduction

Idiopathic pulmonary fibrosis (IPF) is a chronic life-threatening lung disease, with poor prognosis, characterised anatomically by progressive scarring of the lung and symptomatically by exertional dyspnoea, with irreversible loss of pulmonary function [1].

While the aetiology is unknown, emerging data suggest an important association with aspiration [2] linked with

†Amal Alamer, Rhys Jones and Ian A. Forrest are contributed to this work.

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gastric reflux. The term aspiration refers either to the direct inhalation of secretions or ingested materials from the oesophagus or from the oropharynx into the airways. Aspiration by either route can lead to the presence of damaging agents into the airways and the lungs, which is thought to drive the onset and/or progression of a broad range of pathological diseases that affect the airways and lungs [3]. To date, investigations of IPF and aspiration have been limited to gastro-oesophageal reflux disease in which gastric content refluxes up into the oesophagus, with the potential to pass into the airway via the pharynx (microaspiration) [4]. Concerns about the potential role of gastro oesophageal reflux-associated aspiration in IPF pathophysiology have been sufficient to prompt an influential multi-centre pilot of fundoplication [5]. A dysregulated lung microbiome has also been implicated in high profile studies [6].

The pharynx is a shared tube for breathing and swallowing. Because of this, normal swallowing is a complex process, coordinated with breathing to protect the airway. Patients with chronic respiratory diseases are at risk of oropharyngeal dysphagia and swallowing difficulties may go unreported [7, 8]. Early evidence suggests 20% of COPD patients have a swallowing impairment, and subclinical aspiration, identified on Videofluoroscopy Swallow Studies (VFSS) [9]. Prior study found 44% of COPD patients reported swallowing difficulties symptoms on a patient-reported outcome measure [7]. The parenchymal lung scarring, and hypoxaemia seen in IPF patients may act to disrupt respiratory-swallow coordination leading to dysfunction. Surprisingly we are unaware of studies investigating swallowing dysfunction in people with IPF. The purposes of this study were to explore the perception of swallowing difficulty and oropharyngeal swallowing physiology in people with IPF. This was carried out in outpatients without known or suspected swallowing problems.

Material and methods

Participants

Consecutive outpatients with a secure diagnosis of IPF according to European Respiratory Society/American Thoracic Society were recruited from the regional Interstitial Lung Disease (ILD) clinic at the Royal Victoria Infirmary, Newcastle upon Tyne between March 2014 and February 2015. Outpatients with IPF are reviewed on a regular basis at the ILD clinic at the Royal Victoria Infirmary. Recruits were drawn from within this clinic population. Written informed consent was obtained using a printed form prior to the study assessment. Inclusion criteria included competent adults (over the age of 18) with a secure diagnosis of IPF according to the American Thoracic Society (ATS), European Respiratory Society (ERS), Japanese Respiratory Society (JRS) and Latin American Thoracic Society (LATS) clinical practice guidelines [10]. Accurate diagnosis is also supported by discussions among the Newcastle Hospitals Multi-Disciplinary Team (MDT). Exclusion criteria: pregnancy; neurological disease; dementia; gastro-intestinal disease, excepting controlled reflux symptoms; head & neck pathology excepting tonsillectomy/adenoidectomy; previous thoracic surgery; stroke. The patients were screened and excluded if they had a neurological diagnosis that may be associated with dysphagia. The study was described in details, with full written consent taken at the outset, before study activities commenced. All methods were performed in accordance with the approval guidelines and regulations.

Swallowing questionnaire

Eating Assessment tool (EAT-10) is a quick, self-administered and widely used validated questionnaire, which can be used to assess dysphagia symptoms [11]. It has been used previously with patients with chronic respiratory disease such as COPD [7, 12]. It consists of 10 questions regarding swallowing difficulty. Each question is scored on a 5 point Likert scale from 0 (no problem) to 4 (severe problem). The total EAT-10 score is calculated by adding up the scores across the 10 statements (highest score = 40). A total score of 3 or more indicates swallowing difficulty [13].

Swallowing assessment

Swallowing was assessed by a Videofluoroscopy Swallow Study (VFSS). VFSS is a dynamic radiographic examination using fluoroscopy to capture and record real-time bolus flow throughout all stages of swallowing. A Speech and Language Therapist and a radiologist, performed the VFSS examinations. The examination typically includes testing different bolus volumes and constituents [14]. The participants were seated in an upright position. Test boluses were thin liquid (5 and 20 mL); 5 mL paste (custard) and solid (1/4 biscuit) mixed with radiopaque barium sulphate (E-Z-PAQUE), conducted in the lateral plane and one 10 mL liquid bolus with an anterior–posterior view. Liquids were administered first to avoid confounding the results due to remaining residue in the pharynx after ingesting solid consistencies. A penny was taped to the subject's chin during the swallowing study. The circular shape of the penny minimizes the impact of head rotation and the known diameter of the coin allows for calibration of pixels per cm and thus calculation of areas and displacement on VF.

VFSSs analysis

Studies were rated using two validated scales:

1. The standardised Modified Barium Swallow Impairment Profile *(MBSImP)*, a tool used to evaluate swal-

lowing efficiency that measures 17 physiological components of adult swallowing mechanism using ordinal scaling. Ratings of 0 to 2, 3, or 4 points *per* component, with each score representing a unique observation of either structural movement, bolus flow or both from the VFSS [14, 15]. It covers 3 functional domains of the swallow; oral (0–22), pharyngeal (0–29) and oesophageal (0–4). The Overall impression (OI) score is the worst and the most impaired score for all bolus amounts and consistencies. The Oral impairment score and Pharyngeal impairment score were calculated by summing up the OI scores [14]. Scores were interpreted according to a clinically validated classification system [16].

2. The Penetration Aspiration Scale (PAS) is a tool used to evaluate swallowing safety, ranging in value from 1 to 8, recording the presence of laryngeal penetration/sub-glottic aspiration (1 = no airway invasion, 2–5 = penetration, 6–8 = aspiration) [17]. Penetration is defined as the passage of food or fluid into the airway just above the level of the vocal cords. Whereas, aspiration is defined as the passage of food or fluid below the level of the vocal cord [18].

Higher scores for both tools indicate poorer swallowing.

Results

We recruited 14 participants with IPF: 10 males, 4 females, median: 68 years, range 51–82 years. Ten IPF patients: 7 males, 3 females, median: 63 years, range 51–77 years underwent the VFSS (Fig. 1: Consort flow diagram for study).

EAT-10: self-reported swallowing symptoms

A total of 14 participants with IPF completed the EAT-10 questionnaires. All data are presented in Fig. 2. The total median EAT-10 score was 0, range 0–25. Scores were raised in four patients, with values of 25, 15, 14 and 13. These exceeded the normal cut off < 3, indicating swallowing difficulty [13].

Swallowing physiology and safety

The clinical characteristics

The clinical characteristics for the IPF patients underwent VFSS are presented in Table 1. Overall, there were 10 IPF participants who completed the VFSS, the median age of participants was 63 years [range: 51–77]. Most participants were males (7/10, 70%) and ex-smokers (7/10, 70%). The median Body Mass Index (BMI) was 26.2 kg/m2 [range: 20.3–45.6]. The median modified Medical Research Council dyspnoea scale (mMRC) was 2 [range: 1–4]. The median FEV1/FVC% was 80.5% [range: 74–93] and the median FVC % of predicted was 72% [range: 51–92] indicating a restrictive lung pattern [19]. The median TLCO % of predicted was 60% [range: 27–78], showing impaired lung gas transfer.

The Videofluoroscopy Swallow Study (VFSS)

All data are presented in Table 2. Median *MBSImp* for oral impairment was 5 [range: 3–7] and pharyngeal impairment

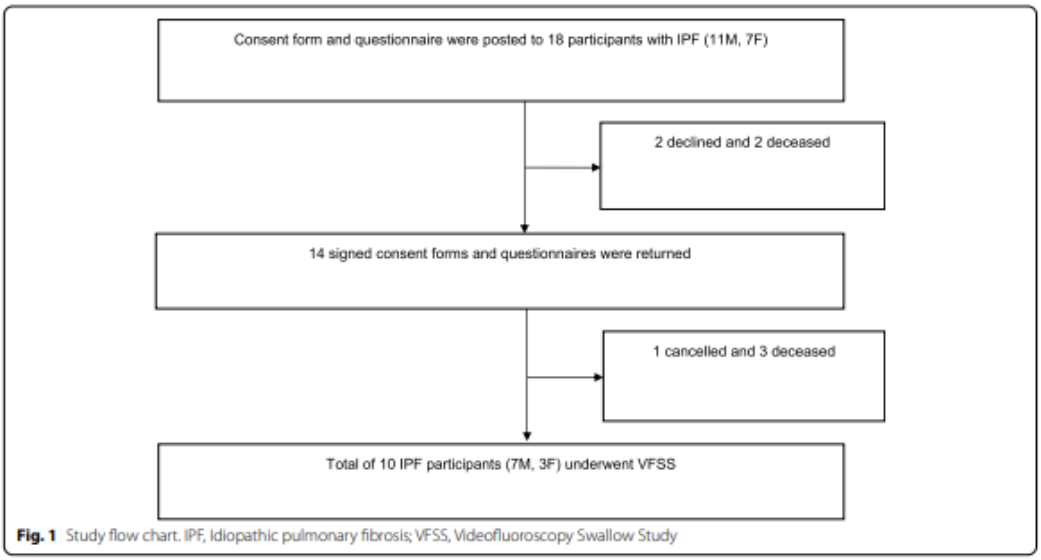

Fig. 1 Study flow chart. IPF, Idiopathic pulmonary fibrosis; VFSS, Videofluoroscopy Swallow Study

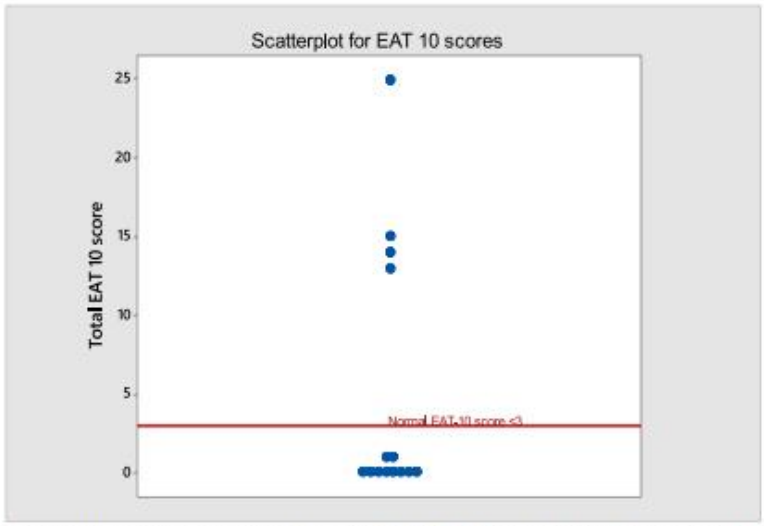

Fig. 2 Scatterplot showing: EAT-10 score (y-axis) for IPF patients. The red line shows the upper score for EAT-10. A score ≥ 3 indicates abnormal EAT-10 scores. IPF, Idiopathic pulmonary fibrosis; EAT-10; The Eating Assessment tool EAT-10

Table 1 The clinical characteristics for the IPF patients underwent Videofluoroscopy Swallow Study (VFSS)

Factor	Value
Number	10
Age,years	63 [51–77]
Sex, Male/Female	7/3
BMI, kg/m2	26.2 [20.3–45.6]
Smoking status: current smoker/ex-smoker	3/7
mMRC	2 [1–4]
FVC % predicted	72 [51–92]
FEV_1/FVC, %	80.5 [74–93]
TLCO% of predicted	60% [range:27–78]

Data presented as median (ranges), unless otherwise indicated. *BMI* Body mass index, *mMRC* Modified medical research council dyspnoea scale, *FEV1* Forced expiratory volume in 1 s, *FVC* Forced vital capacity, *TLCO* Transfer factor for carbon monoxide

4 [range 1–14] indicating, overall mild alteration to swallowing physiology. Patient 1 scored 14 in the pharyngeal impairment indicating mild/moderate impairment [16].

Swallow physiology: the MBS impairment profile The *MBSImP* scores suggested none-to-mild swallowing impairment during oral and pharyngeal stages [16].

Lip closure, bolus preparation and transport, soft palate elevation were uniformly normal. However, all participants had evidence of oral residue, six had a reduction in tongue control and eight participants had a late initiation of pharyngeal swallow.

For the pharyngeal stage of swallowing five participants had evidence of incomplete laryngeal closure and one participant had a bilateral bulging of both pharyngeal walls. Eight participants had a reduction in tongue base retraction. Nine participants had evidence of post-swallow pharyngeal residue.

Swallow safety: the penetration/aspiration scale On PAS, 3/10 patients (Patients 2, 4 and 8) had airway penetration. Only patient 1 aspirated liquid without a cough response; this patient had reduced laryngeal elevation and incomplete laryngeal vestibular closure, resulting in the residue laying below the true vocal chords without a response to eject the aspirated liquid (Fig. 3).

Discussion

To our knowledge, dysphagia has not been described in people with IPF. The purpose of this study was to describe oropharyngeal swallowing physiology and safety in unselected patients with a secure, mixed-disciplinary, diagnosis of IPF, without previous evidence for swallowing difficulty.

The widely used patient- reported symptoms of swallowing impairment, EAT-10 tool was used to assess

Table 2 Videofluoroscopy Swallow Study (VFSS) and the Eating Assessment tool -10 results

	Oral Domain							Pharyngeal Domain											
Participant	Lip Clousre	Tongue Control	Bolus Preparation	Bolus Transport	Oral Residue	Initation of Pharyngeal Swallow	oral impairment	Soft Palate Elevation	Laryngeal Elevation	Anterior Hyoid Excursion	Epiglottic Movement	Laryngeal Vestibular Closure	Pharyngeal Stripping Wave	Pharyngeal Contraction	Pharyngoesophageal Segment Openin	Tongue Base Retraction	Pharyngeal Residue	pharyngeal impairment	Penetration Aspiration scale	EAT-10
1	0	1	0	0	0	3	4	0	1	1	1	2	1	3	1	2	2	14	8	25
2	0	2	0	0	2	3	7	0	1	1	1	2	1	0	1	2	2	11	2	0
3	0	1	0	0	2	3	6	0	1	0	0	1	0	0	0	2	2	6	1	0
4	0	2	0	0	2	3	7	0	1	0	1	0	0		1	0	2	5	2	1
5	0	0	0	0	2	3	5	0	0	0	0	0	1	0	1	0	2	4	1	0
6	0	2	0	0	2	3	7	0	0	0	0	0	0	0	0	0	2	2	1	0
7	0	2	0	0	0	3	5	0	1	0	0	0	0	0	0	0	2	3	1	14
8	0	0	0	0	0	3	3	0	1	0	0	1	0	0	0	0	2	4	2	0
9	0		0	0	2	2	4	0	0	1	0	0	1		0	0	0	2	1	0
10	0	2	0	0	0	2	4	0	0	0	0	1	0		0	0	0	1	1	13

Results for all patients relating to the 17 physiological components showing the Overall impression (OI) score, oral and pharyngeal impairment scores, PAS and EAT-10. EAT-10, Eating Assessment tool; Green cells indicate no dysfunction; yellow cells mild dysfunction; orange cells moderate dysfunction; red cells indicate severe dysfunction and white blank cells denote missing data

patients' perception of swallowing difficulty. In our study the median EAT-10 score was 0, range 0 -25. Four out of 14 patients (29%), had markedly raised total EAT-10 scores, with values of 25, 15, 14 and 13.

We are unaware of previous EAT-10 data in people with IPF. A recent study in 30 patients with Acute Exacerbations of COPD showed that 67% of patients had a raised EAT-10 score, compared with 23% of patients with cardiac disease [12].

In our study, subjective dysphagia symptoms reported by the EAT-10 tool were not consistently related to the nature and severity of the oropharyngeal swallow impairment observed during VFSS. Patient 1 who demonstrated aspiration on videofluorocopy had the highest EAT-10 score of 25, but patients who scored 13 and 14 in the EAT-10 tool had relatively normal swallow physiology detected during VFSS (Table: 2). Our exploratory findings in a limited number of patients are consistent with previous studies in COPD which have previously shown a weak association of EAT-10 with objective measurements of dysphagia [7, 12]. Further studies are therefore indicated in people with IPF, within which it is important to identify patients who may have difficulty swallowing regardless of whether aspiration is present. The EAT-10 tool helps to extend understanding about broad aspects of swallowing,

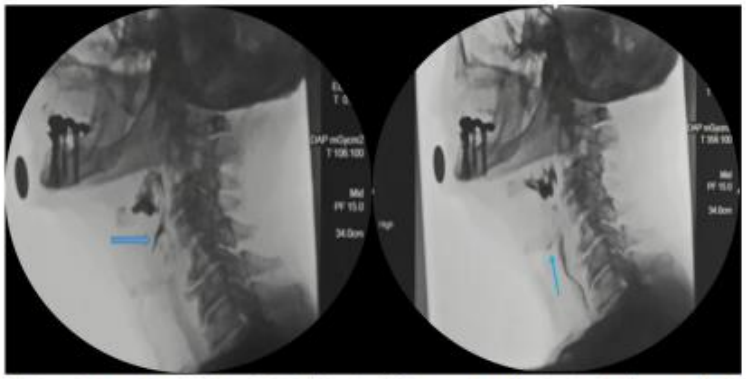

Fig. 3 Oblique sagittal views during swallow of 20 mL thin liquid showing (left) penetration of barium into the larynx and (right) tracheal aspiration with contrast medium below the region of penetration

which includes patient centred social and emotional information, not captured by objective instrumental tests.

Videofluoroscopy studies demonstrated a range of physiology in the ten patients studied. The swallow from laryngeal elevation onwards was consistently and highly disrupted in patient 1 and 2; these two patients had an abnormal physiology according to the *MBSImP* classification system [16]. However, patient 2 had no airway invasion, despite objectively the worst physiology of all, suggesting that a high score on *MBSImP* may not correspond to an unsafe swallow. Higher scores on some components of the *MBSImP* may be regarded as 'normal'; for example, the bolus may enter the pharynx before swallow is initiated even in healthy individuals and some features may be part of a healthy ageing profile [20]. On PAS patients 2, 4 and 8 had airway penetration, we also noted that Patient 4 and 8 had relatively normal physiology by *MBSImP*. In the scarce literature, normal older swallowers sometimes have scores of 2 and 3 in PAS and our findings may therefore represent the combined effects of both normal aging and IPF pathophysiology and require further study [17]. There is no available evidence in the literature of which we are aware comparing the prevalence of swallowing impairments in IPF patients compared to the general population of similar age, with no known neurological disorders.

Parenchymal lung scarring and hypoxaemia may disrupt the complex coordination of normal swallowing and breathing function and in principle dysphagia may contribute to a complex dysregulated aerodigestive homeostasis in people with IPF. The true incidence of dysphagia in IPF is unknown but oral dysbiosis has been linked with a range of lung diseases including, pneumonia, COPD, and lung cancer [21]. The oral cavity has been shown to be a source of diverse bacteria and it is of interest that this can include non-gastric reservoirs of *Helicobacter pylori* [22], which has been associated with a more severe disease phenotype, higher mortality and lung function decline in people with IPF [23].

As fibrotic changes progress and lung function declines in IPF, the affected lung may be expected to become more susceptible to external challenges such as aspiration. Non-sterile aspiration, related to dysphagia and unprotected by cough, therefore represents a candidate source of complex lung injury and microbiome dysregulation. Aspiration is noted to be a trigger of threatening acute exacerbations [24]. Acute exacerbations in IPF are of high concern as they represent the most common cause of death in IPF. Just under a half of deaths in IPF are preceded by an acute exacerbation and the median survival after an acute exacerbation is approximately 3 to 4 months [25].

This prospective consecutive case series is descriptive in a limited number of patients. Further exploration is needed to establish the association between dysphagia and IPF, and the clinical significance of such a link. It would be of interest for further studies to assess if the prevalence of dysphagia in IPF is above that expected in a population of a similar age and to compare our IPF findings to other patient groups with comorbidities that could increase the risk of swallowing problems/aspiration (e.g. frailty). Our experience indicated that such studies are possible in people with IPF but also underline that these are challenging. Of 18 patients approached in our study five died, and three deaths occurred in fourteen consented patients, before videofluoroscopy could be performed.

In other settings, simple bedside tests of dysfunction are clinically informative in swallowing pathophysiology [26] and together with selected patient reported outcome measures, [27] such approaches may be useful in frail patients. Safe approaches to augmented personalised therapy in selected patients, including speech and language intervention, could be rapidly implemented given the established model of mixed disciplinary care in IPF, if dysphagia is confirmed in further studies.

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Authors' contributions
Mrs Amal Alamer: analysed the study and wrote all drafts of the manuscript. Mr Rhys Jones: designed and recruited for the study and contributed to analysis and writing. Dr. Michael Drinnan: analysed and co-wrote the study. Prof. A. John Simpson: study design and patient recruitment. Prof. Mike Griffin: study design and protocol development. Prof. Joanne M Patterson: study design, protocol development and writing the study. Mr Abdullah Althuwaybi study discussion and literature review. Prof. Chris Ward designed, analysed and wrote the study. Dr. Ian Forrest initiated, designed, recruited, analysed and was clinical lead for the study. The author(s) read and approved the final manuscript.

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John Simpson is an NIHR Research Professor.

Availability of data and materials
The anonymised patient-level data used for this study cannot be shared for reasons of information governance. Data may be available to affiliated researchers given the North West- Preston Research Ethics committee, REC reference 14/NW/1056 ethical approval, and are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
All methods were performed in accordance with the approval guidelines and regulations.
The study was approved by the Health Research Authority (HRA), North West-Preston Research Ethics committee, REC reference 14/NW/1056 and carried out in accordance with approval guidelines. All participants provided written informed consent.

Consent for publication
Not applicable.

Competing interests
No competing interests or conflicts of interest.

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Publisher's Note

Appendix 6: Approval letter

NHS
Health Research Authority
National Research Ethics Service

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01 July 2014

Mr Rhys Jones
Oesophagogastric Unit, Ward 36,
Royal Victoria Infirmary,
Newcastle-upon-Tyne
NE14LP

Dear Mr Jones

Study title:	**Videofluoroscopic assessment of oropharyngeal swallow competence in idiopathic pulmonary fibrosis.**
REC reference:	**14/NW/1056**
IRAS project ID:	**130314**

The Research Ethics Committee reviewed the above application at the meeting held on 27 June 2014. Thank you for attending by telephone to discuss the application.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details, unless you expressly withhold permission to do so. Publication will be no earlier than three months from the date of this favourable opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to withhold permission to publish, please contact the REC Manager Mrs Carol Ebenezer, nrescommittee.northwest-preston@nhs.net.

Ethical opinion

The members of the Committee present gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below. .

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Appendix 7: The Eating Assessment Tool-10.

Subject Initial: ..... Date:...../...../.....

Eating Assessment Tool (EAT-10)

To what extent are the following scenarios problematic for you?

(Please put a tick in the score box as applicable to your symptom)

Circle the appropriate response	**0 = No problem**				**4 = Severe problem**
1. My swallowing problem has caused me to lose weight.	0	1	2	3	4
2. My swallowing problem interferes with my ability to go out for meals.	0	1	2	3	4
3. Swallowing liquids takes extra effort.	0	1	2	3	4
4. Swallowing solids takes extra effort.	0	1	2	3	4
5. Swallowing pills takes extra effort.	0	1	2	3	4
6. Swallowing is painful.	0	1	2	3	4
7. The pleasure of eating is affected by my swallowing.	0	1	2	3	4
8. When I swallow food sticks in my throat.	0	1	2	3	4
9. I cough when I eat.	0	1	2	3	4
10. Swallowing is stressful.	0	1	2	3	4
				Total EAT-10:	

Thank you for completing the questionnaire.

Appendix 8: Swallowing safety and performance in patients with idiopathic pulmonary fibrosis: evidence from the water swallow test. Poster presentation at the British Thoracic Society (BTS), winter meeting, 2022.

'Scar Wars' – The pot pourri of ILD

PDF

P25 Swallowing safety and performance in patients with idiopathic pulmonary fibrosis: Evidence from the water swallow test

A Alamer [1, 2], J Patterson [3], M Drinnan [4], I Forrest [5], C Ward [2]

Abstract

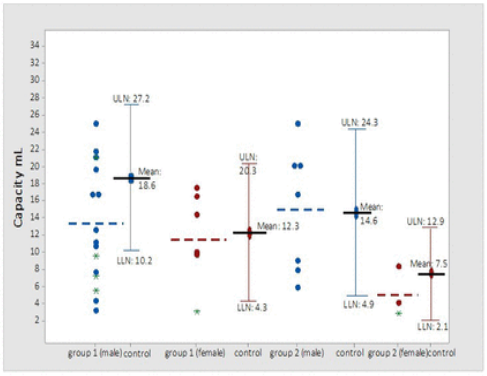
Introduction Awareness of swallowing abnormalities in respiratory conditions such as (COPD) is growing. However, research into swallowing dysfunction in IPF is limited.

Aim Explore the swallowing safety and performance of the Water Swallow Test (WST) in patients with IPF.

Methods Thirty-four IPF patients were recruited from pulmonary fibrosis support groups around the UK or from the Newcastle Interstitial Lung Disease (ILD) clinic between January and October 2021. The WST was conducted via teleconference call or face-to-face in the ILD clinic. Patients took three volumes of water: 5 mL single-sip, 10 mL single-sip and 100 mL consecutive sips. Signs of penetration or aspiration (airway response and/or wet voice after swallow) indicated a WST fail. Three swallowing performance parameters were calculated: swallow volume (mL per swallow), capacity (mL per second) and speed (time per swallow). Swallowing performance parameters were reported by age, gender and% predicted FVC. The cohort was compared with published healthy controls (Hughes and Wiles, 1996). Patients' HR, RR and SpO2 pre and post WST were obtained.

Results Thirty-three IPF patients (23 M, 10 F) completed the WST, median age 72 (52–92) years. Ten patients (10/33, 30%) were on Long Term Oxygen Therapy (LTOT). IPF patients had poorer swallow performance than the healthy people (figure 1) and six patients (18%, 4 M, 2 F) failed the WST.

Post-test SpO2 was higher (p=0.004). Females have lower swallow volume than males (p=0.006, mean rank difference 8.2 mL). However there was no difference for swallow capacity and speed.



Appendix 9: Approval letter

NHS
Health Research Authority

Skipton House
80 London Road
London SE1 6LH

Email: hra.approval@nhs.net

Dr Ian Forrest
Consultant Respiratory Physician
Newcastle upon Tyne Hospitals NHS Foundation Trust
Department of Respiratory Medicine
Royal Victoria Infirmary Newcastle upon Tyne
NE1 4LP

04 April 2018

Dear Dr Forrest

Letter of HRA Approval

Study title:	**The feasibility of respiratory muscle training as part of an interstitial lung disease pulmonary rehabilitation programme**
IRAS project ID:	**211628**
Protocol number:	**n/a**
REC reference:	**18/NE/0037**
Sponsor	**Newcastle upon Tyne Hospitals NHS Foundation Trust**

I am pleased to confirm that **HRA Approval** has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further from the HRA.

How should I continue to work with participating NHS organisations in England?
You should now provide a copy of this letter to all participating NHS organisations in England, as well as any documentation that has been updated as a result of the assessment.

This is a single site study sponsored by the site. The sponsor R&D office will confirm to you when the study can start following issue of HRA Approval.

It is important that you involve both the research management function (e.g. R&D office) supporting each organisation and the local research team (where there is one) in setting up your study. Contact details of the research management function for each organisation can be accessed here.

How should I work with participating NHS/HSC organisations in Northern Ireland, Scotland and Wales?
HRA Approval does not apply to NHS/HSC organisations within the devolved administrations of Northern Ireland, Scotland and Wales.

If you indicated in your IRAS form that you do have participating organisations in one or more devolved administration, the HRA has sent the final document set and the study wide governance

Section 4: Review bodies for the amendment

Please note: This section is for **information only**. Details in this section will complete automatically based on the options selected in Sections 1 and 2.

	Review bodies																	
	UK wide:						England and Wales:				Scotland:				Northern Ireland:			
	REC	Competent Authority MHRA - Medicines	Competent Authority MHRA - Devices	ARSAC	Radiation Assurance	UKSW Governance	REC (MCA)	CAG	HMPPS	HRA and HCRW Approval	REC (AWMA)	PBPP	SPS (RAEC)	National coordinating function	HSC REC	HSC Data Guardians	Prisons	National coordinating function	Category:
Change 1:						(Y)				(Y)									A
Change 2:						(Y)				(Y)									C
Change 3:						(Y)				(Y)									C
Overall reviews for the amendment:																			
Full review:						N				N									
Notification only:						Y				Y									
Overall amendment type:	Non-substantial, no study-wide review required																		
Overall Category:	A																		

Appendix 10: Water swallow test evaluation form.

Participant ID:	Number of swallows	Time	Criteria for test failure or termination				
			In ability to drink the whole amount of water	Coughing or chocking during and after the swallow	Presence of wet Voice before the test	Presence of wet voice after the test or is not able to clear throat sufficiently	Comments
5 mL							
5 mL							
10 mL							
100 mL							

Appendix 11: Patient reported assessment of swallowing and throat symptoms and reflux in patients with idiopathic pulmonary fibrosis, oral presentation at the European Respiratory society congress, Milan, 2023.

Patient reported assessment of swallowing and throat symptoms and reflux in patients with idiopathic pulmonary fibrosis

Amal Alamer, Chris Ward, Ian Forrest, Michael Drinnan, Joanne M Patterson

European Respiratory Journal 2023 62: OA1431; **DOI:** 10.1183/13993003.congress-2023.OA1431

Abstract

Introduction: Aim: to describe IPF patients' perceptions of swallowing dysfunction, laryngopharyngeal reflux and laryngeal hypersensitivity symptoms.

Methods: A cross sectional observational study. Recruitment was undertaken through UK pulmonary fibrosis support groups and the Newcastle Interstitial Lung Diseases clinic, between (January 2021- November 2021). Three validated questionnaires were used (1) The Eating Assessment Tool-10 (EAT-10), (normal <3), (2) The Reflux Symptoms Index (RSI), (normal ≤13) and (3) The Newcastle Laryngeal Hypersensitivity (LHQ) Questionnaire.

Results: Forty IPF patients were recruited (26 M: 14 F, age median 71, range 52-92 years) 30% used oxygen. EAT-10 mean score was 5.9 (±6.5), range 0 to 20, (23/40) 57% of patients had swallowing difficulty evident by a total EAT-10 score >3. The most common symptoms reported were coughing on eating and food sticking in the throat. The RSI mean score was 15.6 (±8.3), range from 2 to 31, (24/40) 60% had an RSI score >13, suggesting the presence of extra-oesophageal reflux in some patients. The Newcastle LHQ scores indicated that 25 patients reported alterations to laryngeal sensation, and had a total LHQ scores that were below the lower level of 95% CI of published normal LHQ score (17.8).

Conclusion: In this study, a majority of IPF patients reported swallowing difficulty, reflux and laryngeal sensation symptoms. Further work is indicated to explore these symptoms fully in this vulnerable group.

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Appendix 12: The Reflux Symptom Index (RSI).

Subject Initial: ..... Date:...../...../.....

The Reflux Symptom Index (RSI)

Within the past month, how did the following affect you? (Please circle the appropriate response)	**0= No problem**					**5: Severe problem**
Hoarseness or a problem with your voice?	0	1	2	3	4	5
Clearing your throat?	0	1	2	3	4	5
Excess throat mucus or postnasal drip?	0	1	2	3	4	5
Difficulty swallowing food, liquid or pills?	0	1	2	3	4	5
Coughing after you ate or lie down?	0	1	2	3	4	5
Breathing difficulties or choking episodes?	0	1	2	3	4	5
Troublesome or annoying cough?	0	1	2	3	4	5
Sensations of something sticking in your throat or a lump in your throat?	0	1	2	3	4	5
Heartburn, chest pain, indigestion, or stomach acid coming up?	0	1	2	3	4	5
Total						

Thank you for completing the questionnaire.

Appendix 13: The Newcastle Laryngeal Hypersensitivity Questionnaire.

Subject Initial: ..... Date:...../...../.....

The Newcastle Laryngeal Hypersensitivity Questionnaire (LHQ)

Please circle the best answer that best describes you currently. Be sure to only select one response:

Please circle the best answer	All of time (1)	Most of the time (2)	A good bit of the time (3)	Some of the time (4)	A little of the time (5)	Hardly any of the time (6)	None of the time (7)
There is abnormal sensation in my throat:	1	2	3	4	5	6	7
I feel phlegm and mucous in my throat:	1	2	3	4	5	6	7
I have pain in my throat:	1	2	3	4	5	6	7
I have a sensation of something stuck in my throat:	1	2	3	4	5	6	7
My throat is blocked:	1	2	3	4	5	6	7
My throat feels tight:	1	2	3	4	5	6	7
There is an irritation in my throat:	1	2	3	4	5	6	7
I have a sensation of something pushing on my chest:	1	2	3	4	5	6	7
I have a sensation of something pressing on my throat:	1	2	3	4	5	6	7
There is a feeling of constriction as though needing to inhale a large amount of air:	1	2	3	4	5	6	7
Food catches when I eat or drink:	1	2	3	4	5	6	7

Appendix 14: The eating and drinking experience in patients with IPF: a qualitative study, poster presentation at the European Respiratory society congress, Milan, 2023.

The eating and drinking experience in patients with IPF: a qualitative study

Amal Alamer, Chris Ward, Ian Forrest, Michael Drinnan, Joanne M Patterson

European Respiratory Journal 2023 62: PA3452; DOI: 10.1183/13993003.congress-2023.PA3452

Abstract

Introduction: The purpose of this study is to explore IPF patients' eating and drinking experiences, and the impact of any changes subsequent to their diagnosis and any coping mechanisms.

Methods: This qualitative study used purposive sampling to recruit IPF patients from IPF support groups and clinics. Semi-structured, in-depth interviews explored patients' experiences of their eating and drinking, using an evidence-informed topic guide. Interviews were recorded via video or telephone call, transcribed, and data coded and analysed using a reflexive thematic analysis.

Results: Fifteen IPF patients (9 M, 6 F), median age 71, range (54-92) years were interviewed via telephone call (n: 5), video call (n: 10). Three main themes were identified, along with several sub-themes. These were 1) 'Eating, as such, is no longer a pleasure'. This theme focused on the physical and sensory changes in eating and drinking, and the subsequent emotional and social impact. 2) 'It is something that happens naturally and just try and get on with it'. This theme centred on the self-employed strategies used to manage changes to eating and drinking. 3) 'What is normal and the new normal'. This theme focuses on patients' information seeking to understand eating and drinking changes and what are their expectations about these changes.

Conclusion: To our knowledge, this is the first study to report on IPF patients' lived experience of swallowing changes due to their diagnosis Findings demonstrate that some patients have substantial struggles and challenges with eating and drinking, affecting them physically, emotionally and socially. There is a need to provide better patient information for this area.

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Appendix 15: Paper, Eating and drinking experience in patients with idiopathic pulmonary fibrosis: a qualitative study.

Open access Original research

BMJ Open

Eating and drinking experience in patients with idiopathic pulmonary fibrosis: a qualitative study

Amal Ahmad Alamer [1], Christopher Ward [2], Ian Forrest,[3] Michael Drinnan,[4] Joanne Patterson[5]

To cite: Alamer AA, Ward C, Forrest I, *et al*. Eating and drinking experience in patients with idiopathic pulmonary fibrosis: a qualitative study. *BMJ Open* 2024;**14**:e078608. doi:10.1136/bmjopen-2023-078608

► Prepublication history and additional supplemental material for this paper are available online. To view these files, please visit the journal online (https://doi.org/10.1136/bmjopen-2023-078608).

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ABSTRACT

Objective To explore eating and drinking experiences of patients with idiopathic pulmonary fibrosis (IPF), the impact of any changes associated with their diagnosis and any coping mechanisms developed by patients.

Setting Pulmonary fibrosis support groups around the UK and the regional Interstitial Lung Diseases Clinic, Newcastle upon Tyne.

Participants 15 patients with IPF (9 men, 6 women), median age 71 years, range (54–92) years, were interviewed. Inclusion criteria included competent adults (over the age of 18 years) with a secure diagnosis of IPF as defined by international consensus guidelines. Patients were required to have sufficient English language competence to consent and participate in an interview. Exclusion criteria were a history of other lung diseases, a history of pre-existing swallowing problem of other causes that may be associated with dysphagia and individuals with significant communication or other memory difficulties that render them unable to participate in an interview.

Design A qualitative study based on semistructured interviews used purpose sampling conducted between February 2021 and November 2021. Interviews were conducted via video videoconferencing call platform or telephone call, transcribed and data coded and analysed using a reflexive thematic analysis.

Results Three main themes were identified, along with several subthemes, which were: (1) Eating, as such, is no longer a pleasure. This theme mainly focused on the physical and sensory changes associated with eating and drinking and their effects and the subsequent emotional and social impact of these changes; (2) It is something that happens naturally and just try and get on with it. This theme centred on the self-determined strategies employed to manage changes to eating and drinking; and (3) What is normal. This theme focused on patients seeking information to better understand the changes in their eating and drinking and the patients' beliefs about what has changed their eating and drinking.

Conclusions To our knowledge, this is the first study to report on IPF patients' lived experience of eating and drinking changes associated with their diagnosis. Findings demonstrate that some patients have substantial struggles and challenges with eating and drinking, affecting them physically, emotionally and socially. There is a need to provide better patient information for this area and further study.

STRENGTH AND LIMITATIONS OF THIS STUDY

⇒ The sample was diverse regarding age, gender and living status. The patients involved were from various locations in the UK.
⇒ The severity of idiopathic pulmonary fibrosis could not be formally recorded due to the recruitment method via pulmonary support groups, however the researcher was able to gain an understanding of the disease's severity through patients' perceived symptoms.
⇒ Due to the pandemic, the interviews were conducted remotely, potentially excluding individuals without the required equipment or skills.

INTRODUCTION

Idiopathic pulmonary fibrosis (IPF) is a chronic and progressive interstitial lung disease characterised by unknown causes. It is primarily defined by the progressive scarring and fibrosis of lung tissue, leading to irreversible loss of pulmonary function.[1] Typical symptoms of IPF include chronic exertional dyspnoea and a persistent dry cough.[2] Risk factors for IPF include older age, male sex, genetic susceptibility, lung microbiome dysregulation and a history of cigarette smoking.[3]

IPF is the most common type of fibrotic interstitial lung disease. In the UK, its estimated incidence is between 5.3 and 7.3 cases per 100 000 individuals per year, while in the USA, rates range from 31.1 to 93.7 cases per 100 000.[4] The mortality rates associated with IPF have been increasing over time. In the UK, the rates rose from 4.6 cases per 100 000 individuals per year in 2001 to 9.4 cases per 100 000 individuals per year in 2011, while in the USA, a similar increasing trend was observed between 2000 and 2010, with rates escalating from 4.4 to 5.3 cases per 100 000 individuals per year.[5] IPF is more frequently diagnosed in the sixth or seventh decade of life.[6] The median survival for patients with IPF is approximately 3–5 years if left untreated.[7]

Several qualitative research studies have been published on patients' experiences with IPF.[8–12] These studies have identified issues with patients obtaining a correct and timely diagnosis; accessing suitable information about their condition; and substantial physical and psychosocial impacts on themselves and carers, affecting their overall Quality of Life (QOL).[10 13] We have also published preliminary work which noted that around a third of patients with IPF reported difficulties with eating and drinking using a patient-reported measure.[14] Further qualitative work is warranted to understand this problem in more depth.

Qualitative studies on eating and drinking issues have rarely been published in patients with chronic respiratory disease, with limited work in Chronic Obstructive Pulmonary Disease (COPD).[15 16] Coughing and shortness of breath were the most common symptoms experienced by patients with COPD during eating and drinking resulting in anxiety, fear and embarrassment.[16] As a result, they developed coping strategies including modifying the way they ate, for example, eating soft food, chewing well and taking small, regulated sips.[16] These findings supported survey findings (n=133) which identified 75% of patients had breathing discomfort when eating or drinking and similar strategies were employed.[17]

Furthermore, there has been limited research on nutritional issues of patients with IPF. Known problems include sustained weight and muscle loss, loss of appetite and vitamin D deficiency, negatively affecting IPF prognosis.[18] A December 2023 action on pulmonary fibrosis report commissioned to form the fundamentals of a new national integrated care pathway identified the need for research and 'support with diet'. The extent of the unmet need identified by the pulmonary fibrosis community was reflected in the title of the report 'I wish it was cancer: Experience of Pulmonary Fibrosis in the UK'.[19]

This study aims to explore eating and drinking experiences of patients with IPF, the impact of any changes associated with their diagnosis and any coping mechanisms they deploy. Eating and drinking is multidimensional, including physical, emotional and social aspects. In light of this complexity, a qualitative approach was chosen to facilitate a deep and diverse understanding of their lived experiences.

METHODS

Interviews were recorded using a password-encrypted audio recording device (Sony ICD-TX650, slim digital PCM/MP3 stereo voice recorder) or videoconferencing call platform (Zoom).

The interview transcripts were coded for confidentiality and anonymity before analysis and kept in secure, locked storage which was only accessible to the research team. In the transcripts, any details specific to the patients' demographics were removed, for example, country of origin, name of the doctor treating them and other names mentioned during the conversation. Pseudonyms were used instead of the real names of the patients for the purpose of presenting these quotations. In accordance with the guidelines of the National Institute for Health and Care Research, participants were compensated for their time with vouchers, delivered either via e-gift cards or mailed gift cards.

Research design

This is a descriptive qualitative study to gain a deeper understanding of IPF eating and drinking experiences from the patients' perspective, using semistructured interviews. The research objectives were to (1) understand the perceived eating and drinking changes experienced by patients with IPF, (2) understand the impact of eating and drinking changes on patients with IPF and (3) explore any coping strategies for and adjustments for these changes.

Patient and public involvement

This qualitative study focused on the opinions of patients and their families. There was no direct involvement of patients in the design of the study.

Participants and recruitments

Patients were recruited from six pulmonary fibrosis support groups around the UK and the regional Interstitial Lung Diseases (ILD) Clinic, Royal Victoria Infirmary hospital, Newcastle upon Tyne, from February 2021 to November 2021.

Inclusion criteria included competent adults (over the age of 18 years) with a secure diagnosis of IPF as defined by international consensus guidelines.[20] Patients were required to have sufficient English language competence to consent and participate in an interview. Exclusion criteria included a history of other lung diseases; a history of pre-existing swallowing problem of other causes such as head and neck pathology excepting tonsillectomy/adenoidectomy, previous thoracic surgery, stroke and neurological diagnosis that may be associated with dysphagia; and individuals with significant communication difficulties, cognitive impairment or memory difficulties that render them unable to participate in an interview.

The recruitment process was designed to encompass all eligible patients from both clinic and support groups. Specifically, during clinic recruitment, the researcher approached all eligible patients for study participation. For support group recruitment, leaders within the support groups facilitated the distribution of study details and encouraged patients to directly reach out to the researcher. Subsequently, all individuals meeting the eligibility criteria were approached.

Patients were recruited based on purposive sampling method using maximum variation sampling approach, to ensure heterogeneity in the sample as well as to elicit diverse views and experiences. In the first few interviews, the sample was based on age (≤65 years vs >65 years) and gender (male or female), because eating and drinking

can vary by age and gender. After a few interviews, the selection criteria were discussed by the research team. Living status (alone or with someone) appeared to have an influence on eating and drinking experiences; therefore, it was included as a selection criterion for maximum variation.

The number of conducted was informed interviews by the concept of 'information power'. Information power is an alternative approach to 'data saturation', which indicates that the more information the sample holds, relevant for the actual study, the lower number of participants is needed.[21–24] Indeed, researchers have shown that indices of saturation in qualitative interview-based studies are often inadequately described, and authors focus on the number of participants in order to convince readers (or themselves) that a sufficiently large sample was recruited to achieve their study aim. Instead, many qualitative researchers have shifted to describing quality findings as sufficient, which covers both analytical sufficiency and data sufficiency.[21 25] Given the limitations of the data saturation concept, information power is considered a more appropriate measure of evaluation sufficiency.[25] The use of information power to determine whether qualitative findings are sufficient involves assessing five items: (1) their study aim, (2) sample specificity, (3) use of established theory, (4) quality of dialogue and (5) analysis strategy, together with an iterative analysis of the results and discussion with research team, which were sufficient to inform the decision on information power and when to stop recruitment.[21 25]

Data collection

The semistructured interviews were conducted by a PhD respiratory researcher (AAA), who had no role in the patients' care. The interviews took place either via telephone or virtually through video conferencing call platforms (Zoom or FaceTime), depending on the participants' preference due to COVID-19 restrictive measures. The mean duration of the interviews was 40 min.

An interview topic guide (online supplemental material) was devised and piloted in two online interviews to elicit detailed responses from each patient. The interview topic guide followed existing guidelines for the design of qualitative interviews.[26 27] It was developed following a comprehensive review of relevant medical literature.[10 12 28 29] Input from healthcare practitioners with expertise in IPF and discussion with the research team were also included. The interviewer asked open-ended questions about each area of interest in the guide, and in addition, a range of sub-questions and prompts were developed to explore the issues in more depth. When necessary, questions were rephrased to aid a participant's understanding. Interviewing is an iterative process in nature; data collection and analysis are conducted simultaneously, as in all qualitative research. An ongoing and iterative analysis of the transcripts was conducted. After each interview, the research team reviewed the recording and reflected on the interviewer's responses and the patient's responses. This was done to inform the direction of the next interviews to ensure sufficient depth is reached.

Analysis

The data analysis was conducted using inductive thematic analysis 'analyst-driven' approach described by Clarke and Braun,[30 31] who suggested the six phases analysis methods which were followed in this study.[30 31] Briefly, these included initial reading and familiarisation of the transcripts, coding and identification of candidate themes, followed by checking that themes reflected the entire dataset. Finally, themes were named and the story behind them was considered before writing up the analysis. Using thematic analysis, the researchers were able to draw themes across the interview transcripts for patients with IPF, on a semantic and explicit level. The six phases process of analysis of the transcripts was iterative and reflective with a substantial overlap between the six phases with an attitude of inquiry and interpretation. Common codes and themes were discussed and approved by the research team to ensure an accurate reflection of the data and to help guide the process and validity of the analysis. The analysis was performed using the qualitative data analysis software package NVivo V.R1.6 (QSR International). The 15-point checklist criteria for good thematic analysis was followed to ensure the quality of the thematic analysis.[31]

RESULTS

42 patients were invited to participate. 22 patients did not respond to the invitation, and 15 patients with IPF (9 men and 6 women) were interviewed. Of those who consented to be interviewed, 10 were recruited from the IPF support groups and 5 from the ILD Clinic. Patients were interviewed via telephone calls (n: 5), Zoom video calls (n: 7) and FaceTime video calls (n: 3). One interview transcript was excluded from the analysis as during the interview, the patient reported a history of tongue cancer, making him ineligible for the study (figure 1).

Demographics and clinical characteristics

The patients' demographics and clinical characteristics are presented in table 1. In total, 14 interview transcripts were analysed. The median age of patients was 71.0 years (range 54–92 years). Eight (57%) patients were men. Most patients were non-smokers (10/14, 71%). Eight patients were using antifibrotic medications (8/14, 57%), and six (42.8%) patients were using long-term oxygen therapy. Most patients lived with partners (n: 10), and 4 patients lived alone.

Themes

Three main themes were developed from the data: (1) Eating, as such, is no longer a pleasure; (2) It is something that happens naturally and just try and get on with it; and (3) What is normal? (table 2).

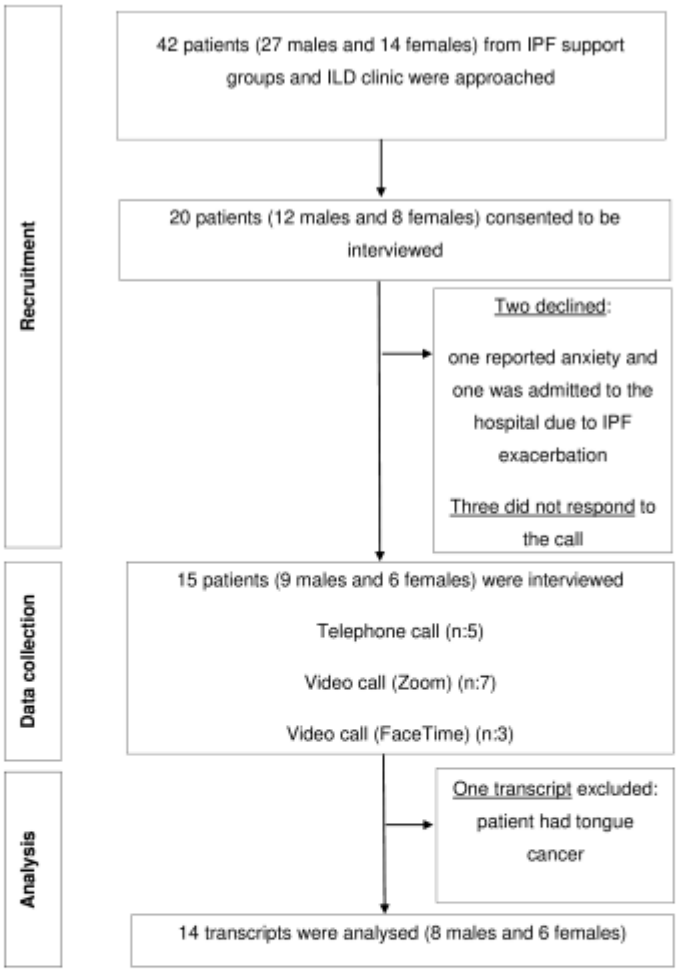

Figure 1 Consolidated Standards of Reporting Trials flow diagram for the patient recruitment for the study. ILD, Interstitial Lung Diseases; IPF, idiopathic pulmonary fibrosis.

Theme 1: Eating, as such, is no longer a pleasure

This theme reports the experiences of patients with IPF who relate directly to the changes in their eating and drinking. The changes that were reported encompassed both physical and sensory aspects of the eating and drinking process and their effects and the patients' emotional and social reactions to these changes.

Subtheme 1: physical and sensory eating and drinking changes and their effects

Patients experienced a range of problems related to their eating and drinking lived experience. Breathlessness when eating and drinking was frequently reported by the patients, forcing adjustments in the pace and frequency of swallowing. One IPF patient said she cannot drink a cup of water, without having to stop many times to catch her breath and continue drinking again, whereas before the IPF that was never an issue:

> Before, I could have drunk, like, a bottle of water, taken quite a lot of it without having to stop. Now, it would be smaller mouthfuls that I would have to stop too. I couldn't, basically: half a bottle or 500ml of water before I continued drinking. Now, I would have just maybe a few mouthfuls and then you have to stop to get a breath and then continue drinking.

Table 1 The patients' demographics and clinical characteristics

Factor	Level	Value
N		14
Age in years, median (minimum–maximum)		71 (52–92)
Gender	Female	6 (42.8%)
	Male	8 (57.2%)
Recruitment method	IPF support group	10 (71.4%)
	ILD Clinic	4 (28.6%)
Interview method	Video call (Zoom)	7 (50%)
	Video call (FaceTime)	3 (21.4%)
	Phone call	4 (28.6%)
Smoking	Non-smoker	10 (71.4%)
	Ex-smoker	4 (28.6%)
Using oxygen	Yes	6 (42.8%)
	No	8 (57.2%)
Using antifibrotic medication	Yes	8 (57.2%),
	No	6 (42.8%)
Living status	With someone	10 (71.4%)
	Alone	4 (28.6%)

ILD, Interstitial Lung Diseases; IPF, idiopathic pulmonary fibrosis.

Another IPF patient attributed this to physiological changes, linking stomach expansion to restricted diaphragmatic movement, impeding oxygen intake during meals:

> I do have shortness of breath when I eat. If I feel like that, I put my oxygen on because I view eating in the same manner as exercising: my body needs the extra oxygen. If I eat too much, then the problem I face is that the diaphragm can't. As the stomach expands, the diaphragm cannot expand in the normal manner so therefore it cannot get enough oxygen into my body. It makes it really difficult.

The prolonged duration of meals due to shortness of breath became a common experience, impacting daily routines:

> It takes me longer to eat a meal because you always have to get a breath between every mouthful. Well, taking longer, like, you have all your plans worked out in your head, what you are doing next or whatever .

Patients conveyed challenges with food inhalation, especially with dry items like toast, leading to coughing and discomfort:

> The other thing is when eating, the biggest thing is if I eat toast. Say I bite into some toast and the dry part of the toast is sort of inhaled into a certain part of your throat. God, I end up coughing really badly .
>
> There has been a sticky texture of food, and that might be, I don't know, a minute amount, and it seems as if it's lodged in my throat.
>
> Sometimes, something will, as we call, go down the wrong tube, like a crumb or something and then it starts coughing, cough, cough, cough, although the biscuits we have, we like to nibble and break up and crunch, and a bit sometimes can whizz into the wrong tube.

This has also happened with saliva, particularly when lying in bed:

Table 2 Themes, subthemes and codes

Eating, as such, is no longer a pleasure		It is something that happens naturally and just try and get on with it		What is normal?	
Physical and sensory eating and drinking changes and their effects	Emotional and social impact of eating and drinking changes.	Direct strategies	Indirect strategies	Information needs	Patients' beliefs
Breathlessness	Fear	Portion size	Oxygen	Desire to learn	Being old
Fatigue	worry	Regulated sips	Rest	Comparing experience with other patients with IPF	Other underlying health issues
Taking longer to eat and drink	Frustration	Being slower	Medications	Swallowing services	
Weight loss	Embarrassment	Chew more	Sleep		
Poor appetite	Loss of enjoyment	Softer food	Clam		
Mouth dryness	Social life	Avoid some foods	Distraction		
Food or drink goes to the wrong pipe			Fear reduction		
Nauseous					
Smaller portion size					
Changes in taste and smell					

> Drinking, I can drink and then maybe a little drop will go down the wrong way. But it does that even with saliva, quite honestly, especially when I'm in bed. And that can start me coughing. There are times when I'm eating when the food actually gets stuck, and I can't.

Fatigue was reported by all patients, living alone, even missed meals due to exhaustion: "I cannot cook so much, because standing can be quite exhausting, just standing. Some days I go without a meal because I'm so tired, I just stay in bed". Similarly,another patient, who lives alone, added that she lived on pre-prepared meals, 'the frozen foods', because she felt tired all day.

Further, patients reported they felt tired and exhausted during and after eating, especially after their evening meal. After the evening meal, one patient said that she always felt tired and wanted to sit on the sofa to rest. Another patient also reported feeling very tired after the evening meal and normally went to bed. For another patient, eating often left him exhausted, and he saw it as similar to exercise.

Patients noted persistent mouth dryness, night-time thirst and changes in taste and smell.

> I would say I drink a bit more. I drink water, perhaps once or twice in the evening, because my throat gets fairly dry, at nighttime, when I'm asleep. I always carry water with me when I am going to bed.

The metallic taste reported by patients seemingly linked to antifibrotic medication use, with associated altered food experiences:

> Well, I couldn't taste my food properly because it has this irony taste in my mouth, and I couldn't taste my food. And before the fibrotic medication, I didn't have this horrible taste in my mouth .

The patients conveyed a profound shift in their relationship with food, marked by a significant loss of appetite and diminished interest in eating. One patient questioned the purpose of consuming food when both hunger and the ability to taste were compromised. Others echoed this sentiment, expressing a general lack of appetite: "With the IPF I didn't want to eat. I don't know. I just didn't have really an appetite at all" and "I've got a very poor appetite". Another patient mentioned that both the sight of a plate full of food piled up and the smell of cooking food affected him. It also made him want to be sick and vomit. Another attributed this change to antifibrotic medication side effects: "I suddenly become very nauseous and want to go to the toilet and whatever. I'd just come off nintedanib for one day, just one day. Once I've come off for one day and then it's okay, I come back to normal". In reflecting on the broader experience of IPF, one patient expressed a diminished desire to eat: "Well, with the IPF I didn't want to eat. I don't know".

This change in appetite and taste preferences manifested in altered eating behaviours, with patients describing evening meals reduced to what one patient termed a 'snack' rather than a full dinner, reflected in significantly smaller portion sizes.

> The evening meal, it's more of what a normal person would call a snack rather than a proper meal. Right, the portion of the food, that's gone down dramatically – probably down to a tea-plate size rather than, what I call, a dinner-plate size.

Weight loss was a common concern for most of the patients: "I've probably lost more muscle than fat, especially on my legs. I used to have quite broad thighs through my running: now they're like chicken's legs, according to my wife. That's where the majority of my muscle has gone" . Patients attributed weight loss to poor appetite and infrequent feelings of hunger. One patient linked his weight loss to antifibrotic medication, as indicated by a 6 lb reduction since starting the treatment. Another patient compared her weight before IPF and now. She said in her prime she had nine stones, which later decreased to eight stones and now she had only seven stones.

Subtheme 2: the emotional and social impact of eating and drinking changes

In this subtheme, wide ranges of negative emotions linked to eating and drinking were described by many patients. Fear of choking was the most common emotion expressed: "Whenever this happens, you panic because you think you're going to choke. So, my reaction to that, my family laugh at me, when I get that the food seems to be stuck and I get the urges of, 'I'm going to choke', I jump to my feet".

Additionally, patients reported feeling worried about how their choking events might affect others:

> I'm quite selective with food as well. I eat things that I know are not a major problem. Actually, I probably shouldn't have done, but my granddaughter comes after school, because we live next to her school, and I had had a little bit of not breathing very well when I was eating. So, I just said to her, 'If grandma is waving her hands, go and get her a drink'. And if I collapse, call 999. She's only 10, but I thought… I've worried her but I said, 'It's probably not going to happen, I just wanted you to know'.

Frustration, embarrassment and a sense of diminished enjoyment turned meals into challenging experiences. The following quotes show how patients felt about the increase in eating time. One patient felt frustrated about the time it took her to eat a meal: "Whereas, before, I would have just maybe grabbed something and ran, you can't do that now because it takes you ages to eat". Furthermore, another patient said that sometimes taking longer to eat left him not able to finish his meals: "I don't have the time to finish them, because it takes ages and ages". Additionally, some patients articulated feelings of embarrassment and shame as they took longer to eat compared to those around them. One patient acknowledged, "I

am definitely slower, though, because when I'm with my family, I'm always the last one, recently, to finish".

The loss of pleasure in eating, expressed by several patients, mirrored the emotional toll: "It used to be. When I was fit and well, I really used to enjoy my food. Now it's something I do".

Eating was simply seen as a task that had to be done as a means of nourishment. Another patient expressed "I don't think I enjoy my food as much as I used to, because of the coughing, choking, that's not easy. I don't enjoy my food. I have to eat to live" .

A few patients outlined a general loss of interest in eating outside home, eating out, "I would get a bit nervous about what happens if this happens when I'm out, having a meal outside, in a restaurant or something. Because you feel like all eyes would be on you. So, yes, I suppose, because we haven't been out for a long time, I'd forgotten about that. Yes, it is a worry" .

Theme 2: It is something that happens naturally and just try and get on with it

This theme covers strategies that patients developed in response to and to help cope with the changes in their eating and drinking. Adaptation strategies were divided into two categories, direct and indirect. The direct strategies were the methods of managing changes in eating and drinking that directly related to diet and eating and drinking habits. Indirect strategies involved the alternative supportive methods used by patients that were not directly related to food and drink, such as using oxygen, medication and going to sleep.

Subtheme 1: the direct strategies

Patients described strategies used to manage the changes in their eating and drinking. Most reported being mindful about taking small, well-chewed bites, pacing rate of eating and having regulated sips during the eating or drinking process. One patient mentioned that she chewed food much longer so as not to choke on it and took frequent sips of a drink.

Co-ordinating breathing and swallowingbecame a conscious effort for some patients:

> I've got to concentrate on my eating, to make sure that, again, I don't breathe in, for it to go– It's as if it's gone to my lung and not– It seems as if I'm coughing for a little particle to get out of my throat. It's really bad.

A majority of patients had adapted their food preparation, for example, having softer food (steamed or mashed) and with additional sauces to reduce the likelihood of choking events and/or to reduce the effort of eating.

Patients reported that they frequently chose soft food: "They are all mushed up", and knew what food they should avoid to protect their airways: "much softer and moist food, because I know I'm not going to choke on it, really" . One patient explained: "If I do eat meat it has to be very lean and very thin" and Steve joked that there would be "no fish and chips" (with laughter) as he did not like hard food which was difficult to consume. Another patient shared: "I have started steaming quite a lot of my food now. As I say, that helps the food stay moist, so it's easier to swallow that way".

Some patients reported being careful about their food choices. Fibre-rich and spicy foods were often avoided. This was due to the diarrhoea which many experienced as a side effect of the antifibrotic medication. For example, "I restrict my intake of fibre. If I eat too much, if I have too much fibre, which is what it is, then I can spend days on the toilet" ; "No vegetables as such, I just can't tolerate them any more, Amal"; and "At one time I'd be quite happy with spicy food, now I have very bland food. I no longer enjoy spicy food".

Besides the effects of spicy food on bowel function, one patient stated that she avoided spicy food because it caused coughing or choking.

In addition, several patients indicated that they avoided steak, because "If I eat steak, it always seems to – even small pieces – it seems to stick in the back of my throat" ; "Steak. any meat that's hard to chew or to do anything. any meat that is a big lump, you know what I mean?"; and "It's more difficulty in swallowing".

Subtheme 2: the indirect strategies

Patients devised strategies not directly related to their diet and eating and drinking habits, to cope with their eating and drinking changes. This was a smaller subtheme.

A few patients reported that they needed to use oxygen to combat post-meal fatigue and the need for rest or sleep after eating:

> I do have shortness of breath when I eat. If I feel like that, I put my oxygen on because I view eating in the same manner as exercising, my body needs the extra oxygen. If I eat too much, then the problem I face is that the diaphragm can't. As the stomach expands, the diaphragm cannot expand in the normal manner so therefore it cannot get enough oxygen into my body. It makes it really difficult .

> After my dinner I sit down on the sofa and just recover. But I usually go to bed quite soon, I don't stay up late.

> After the evening meal I normally go, I'm very tired and I normally go to bed.

Patients developed self-soothing techniques like calming, reassurance and distraction to manage anxiety during meals: "I'm trying not to draw my attention to it, because I think the more I realise it's happening, the more I'll think about it and make it happen" . In another approach, a patient mentioned using anxiety medication before meals to reduce fear and induce calmness.

Theme 3: what is normal?

The final theme focused on patients' beliefs and information needs regarding changes in eating and drinking.

Subtheme 1: information needs

Patients expressed a desire for information to understand and contextualise their experiences. One patient statement, "It wasn't the money side of it, so much, I wanted to learn more from you than probably you learnt from me", illustrates a deeper desire for knowledge beyond tangible considerations.

Seeking insights from others with IPF, served as a way to gauge the normalcy of their challenges. Reflection on group sessions, one patient said "We used to go to a group, and there were people there who had IPF, and it affected their appetite quite badly". Another patient emphasised this by stating, "I know from other people with the disease that they go through exactly the same thing. I'm not an exception to the rule".

Additionally one patient added, "I'm not alone in feeling like that. I know, from my friends who have this disease, they're in exactly the same position". Apart from the practical side of things, one patient expressed how comforting it is to know that others are going through similar challenges: "It was comforting to know that my struggles were shared by a community going through the same journey".

One patient noted that their interview made him consider swallowing services: "After meeting you I have been thinking about swallowing service[s]. I didn't know about swallowing service[s] and nobody suggested that".

Subtheme 2: patients' beliefs

Beliefs about the ageing process as a contributor to eating changes were expressed by some patients, deflecting attribution solely to IPF. One patient acknowledged the multifaceted nature of ageing, stating, "When you reach my age, you've got lots of other problems as well". Her perspective suggests a recognition that various aspects of health can influence eating habits. Similarly, another patient cautiously considered the role of age in his changes, noting, "That could be an age thing, but it's never, ever really thinking because it's because of IPF".

Another patient shared a reflective insight into her experience, highlighting the complex balance between adapting her eating habits and the natural ageing process: "Age brings its own set of challenges. I find myself adjusting my eating habits, not just because of IPF but also as part of the natural ageing process. It's a delicate balance between the two".

One patient's decision to avoid spicy foods was explained through the lens of ageing, offering a unique angle to dietary modifications. Another patient contributed to this narrative by suggesting that as individuals get older, they may find they need less food, influencing their overall eating patterns.

Furthermore, some patients revealed that considerations about changes in eating and drinking were not at the forefront of their minds before the interview. One patient noted, "Certainly, the eating and drinking and swallowing side probably is slightly worse than what it was, but I tend, maybe, not to notice that because of these other symptoms". This observation suggests that it is hard for patients to decide which health issues are most important, and it is challenging for them to notice changes in their eating habits because they have many health concerns.

Another patient expanded on this idea, expressing, "Because of the other parts of the condition, like the cough and the chest discomfort, I don't tend to think much about the swallowing side of it to be honest". Another patient further emphasised the intricate nature of symptoms, describing swallowing as "a smaller piece of a much larger puzzle". This metaphor highlights the challenge patients' face in isolating and understanding the impact of specific symptoms within the broader context of their health.

In summary, the themes are interconnected in a way that reflects the progression from the raw experiences of patients (theme 1) to their coping mechanisms (theme 2) and the context and beliefs that influence these experiences (theme 3). For example, the physical challenges outlined in theme 1, such as breathlessness and fatigue, prompt patients to develop coping mechanisms like pacing their eating or using oxygen, which are discussed in theme 2. Fears of choking, frustration, embarrassment and loss of pleasure in eating discussed in theme 1 contributes to the development of both direct and indirect coping strategies. Examples include, patients may choose softer foods, co-ordinating breathing when swallowing or seeking calmness during meals. Patients' beliefs, as discussed in theme 3, serve as a context for their experiences. Beliefs about the ageing process or the multifaceted nature of health challenges shape how patients perceive and interpret their eating and drinking changes, as highlighted in theme 1.

DISCUSSION

To our knowledge, there is no published research exploring the eating and drinking experiences of patients with IPF using qualitative research methods. The aim of this study was to explore the eating and drinking lived experience for patients with IPF. This provides rare data that directly addresses an unmet need for research and 'support with diet' identified by the UK-based charity Action for Pulmonary Fibrosis in a December 2023 report 'I wish it was cancer: Experience of Pulmonary Fibrosis in the UK'.[19] Our novel findings indicate that some patients have substantial struggles and challenges with eating and drinking, affecting them physically, emotionally and socially. Our findings can be summarised under three inter-related themes: (1) Eating, as such, is no longer a pleasure; (2) It is something that happens naturally and just try and get on with it; and (3) What is normal?

Patients reported breathlessness during meals. This is perhaps not surprising as dyspnoea is a commonly reported symptom of IPF across many activities of daily life.[32] Elsewhere, qualitative work has also identified breathlessness as affecting daily life activities of people

with IPF, such as carrying shopping, bending at the waist and taking a shower.[15 33] This is the first study where patients have reported this symptom also affecting their eating and drinking.

This study focused on the patients' experience of eating and drinking, which revealed that patients frequently had to pause to catch their breath while drinking. They have also described the effects of breathlessness as not being able to get enough air in to their lungs while eating, which resulted in them taking more time to finish their meals. This could be attributed to the expansion of the stomach during digestion, which can push against the diaphragm and lungs, making it harder to breathe.[34] Additionally, eating can also increase the demand for oxygen in the body, further exacerbating breathlessness in those patients with already compromised lung function.[35] The patients shared some techniques they applied in order to alleviate breathing discomfort, including being slower, eating smaller meals and eating more frequent meals throughout the day rather than large, heavy meals. These findings are in line with previous qualitative literature in patients with COPD.[16] On the other hand, taking longer to eat can result in various consequences, including food going cold, challenges in maintaining nutritional intake and an increased risk of overeating due to extended exposure to food. Further, these consequences can affect not only the physical health but also social lives, as the pacing of eating is difficult, this may affect the ability to maintain conversation over mealtimes. Feelings of being ashamed and frustrated when taking longer and not managing to finish meal on time when being with other people were also reported by the patients.

The interviews also shed light on other issues that contribute to the challenge the IPF patients' experience with eating and drinking. Fatigue, a state of tiredness, was reported to have an impact on activities of daily living by the patients. This is consistent with the findings of the European IPF registry (eurIPFreg, 2018), which reported that dyspnoea, fatigue and loss of appetite were the most common clinical symptoms experienced by patients with IPF with a percentage of 90%, 69% and 67%, respectively.[32] In this study, the patients reported experiencing fatigue and the need to rest after eating. The extra effort required for eating and drinking was likely to curtail mealtimes, which could potentially affect nutritional intake. The relationship between fatigue and the difficulty of swallowing has also been reported in other groups of individuals with swallowing disorders.[36]

Prior studies have examined the eating situations of patients with COPD, and the results indicate that patients often experience a decrease in oxygen saturation levels while eating or shortly afterward, leading to the need for supplemental oxygen.[34 37–40] Similar findings were reported by patients with IPF in this study, with patients describing feeling fatigued after meals and requiring the use of oxygen and rest. In addition, patients with IPF who live alone mentioned experiencing fatigue while cooking and preparing meals, resulting in their reliance on premade meals. The previous study on patients with COPD reported that 23% of the patients experienced fatigue while preparing meals.[15]

Fear of choking was the most common emotion expressed. Patients used words such as 'panic' and 'scary' when describing their experience of choking. This fear was typically associated with worries about being able to breathe. Chocking-related fear was associated with avoidance of eating, adopted by the patients as a safety behaviour.[41]

Avoidance behaviour refers to any action or behaviour taken by an individual to avoid a particular situation, task or stimulus that causes discomfort, anxiety or fear.[42] Avoidance behaviour can not only be a natural response to perceived threats or stressors but it can also become a maladaptive coping mechanism that interferes with daily functioning and overall well-being.[42] Avoidance behaviour in eating and drinking can manifest in various ways, including restricting food intake, avoiding certain types of food or food groups and avoiding situations that involve eating or drinking in public or with others.[43]

In addition to the fear, patients in this study reported a range of emotional responses related to their experiences with eating and drinking, including embarrassment, anxiety, panic, anger, sadness and frustration. This is consistent with broader previous research on dysphagia.[44 45] Studies on anxiety in patients with IPF have shown a high prevalence of anxiety (31%) and its association with reduced quality of life.[46] While this study did not examine emotional responses related to other daily activities, it is likely that the emotional distress reported by the patients with IPF in this study contributes to the broader emotional distress observed in previous literature.[47]

In this study, a few patients experienced a general loss of interest in eating outside their home. However, they did not feel socially isolated due to their eating and drinking issues, which was different from other studies conducted with different populations.[36 48 49] It is worth noting that the study was conducted during the COVID-19 lockdown, which meant that people were limited to socialising at home. Consequently, the study may not have fully captured the effects of eating and drinking problems on the social lives of patients with IPF since social life was restricted during the pandemic.

Patients discussed various methods that they had tested by trial and error, to enhance their overall eating and drinking experiences. Thorne *et al* refer to this process as the 'evolution of expertise,' which occurs as individuals with chronic illnesses learn to manage their symptoms over time.[50] These methods comprised adjusting the texture and size of their food, eating at a slower pace, chewing their food thoroughly, taking small sips of water, practising mindfulness while eating, using supplemental oxygen after the meal, resting and going to sleep afterwards.

Previous studies in patients with COPD have reported similar results which suggest that individuals with COPD modify their eating habits to ease breathing difficulties.[16 17]

Similar approaches were employed by patients with head and neck cancer, which involved consuming smaller meal portions, reducing overall food consumption and opting for softer foods.[36] It was clear that the level of eating ability varied from day to day depending on the severity of IPF symptoms, making it challenging for the patients with IPF to plan meals or social activities beforehand.

The patients often did not attribute their eating and drinking status to IPF. In their view, the eating and drinking changes observed were influenced by other underlying medical conditions such as dental issues, diabetes and the chronic nature of the IPF disease itself. In their view, the eating and drinking changes observed were influenced by other underlying medical conditions such as dental issues, diabetes and the chronic nature of the IPF disease itself. Furthermore, several patients linked their changes in eating and drinking to old age. IPF is considered an age-related disease, as highlighted by the BTS national registry, where the average age of presentation was reported as 74 years.[51] This age range aligns with the median age of 71 years observed in this study. In regard to swallowing performance, previous researches have shown age-related changes in various aspects, such as the time it takes for a swallow response, prolonged transition time between the oral and pharyngeal stages of swallowing, extended hyolaryngeal excursion, reduced pharyngeal pressure generation and loss of muscle mass, which affect swallowing.[52–57] Previous studies in different populations have shown that individuals with swallowing difficulties tend to not seek treatment due to the perception that such problems are a natural aspect of getting older. For example, in a study conducted by Turley and Cohen, only 23% of elderly individuals with swallowing problems sought medical help. They also showed that the most common reasons for not seeking treatment were a lack of knowledge about the treatment options available and a belief that the symptoms experienced were a natural part of the ageing process.[58]

Future directions and conclusion

We have previously provided quantitative data showing that dysregulated swallowing physiology can occur in patients with IPF, which included fluoroscopic evidence of aspiration.[14] Non-sterile aspiration represents a candidate source of complex lung injury and microbiome dysregulation and the present study provides novel complimentary information that provides qualitative data regarding eating and drinking in patients with IPF.

The study indicates that patients with IPF face physical, social and emotional consequences due to the changes in their eating and drinking abilities, which can significantly impact their overall quality of life. The findings in this study emphasise the importance of educating both patients and healthcare providers about the eating and drinking changes that may occur in IPF disease. Such education could increase awareness of potential challenges in eating and drinking and might ultimately help to reduce the risk of additional health problems resulting from exacerbation of the disease. It is noteworthy that patients have been innovative in finding their own solutions to cope. This presents an opportunity to develop resources and materials based on their experiences. By capitalising on this patient-driven approach, valuable resources can be created to support individuals living with IPF and their specific needs related to eating and drinking and further research is required.

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Appendix 16: Interview topic guide initial interviews.

Interview guide

1. **Introduction**

'Thank you for agreeing to take part in this interview / swallow assessment. Your participation is very much appreciated. As you know, I will be recording the interview. This is so a member of the project team can type out what we both say later for analysis. Any identifying information about you (including your name) will be removed from the typed version of the interview transcription. All the information will be stored securely. You may see me writing while you are talking this is only to help me for further discussion. Our discussion will last approximately 1 hour'

2. Demographics

'First I'd like some basic information about yourself. Can you tell me your age? Can you remember when you were diagnosed with IPF? Do you use oxygen? Do you work? (if so ask type of work) are you a current or ex-smoker?'

Now we will move on to the main part of the interview. Please let me know at any point if you would like to stop or take a break. I will be covering three main areas, focusing on your eating, drinking and diet. I'll ask a range of questions to help direct our discussion in each area. Is it OK to start the interview?

3. **Main interview**

Can we start off by you describing how IPF affects your life right now?

4. **Eating and drinking experience**

Lets now think about your eating and drinking

Topic	Questions
1. Physical changes to eating	**Have you noticed any changes to the way you eat and drink since you were diagnosed with IPF?** **Probe: any coughing?** **Things going down the wrong way?** **Taking longer over meals** **Needing a drink with a meal** **Lack of saliva** **Shortness of breath** **Chest tightness** **Feeling tired after eating**
2. Changes to diet	**Have you experienced any changes to the type of diet you have?** **Probes: softer foods (if so who / how are these prepared)** **Portion size** **Lighter meals** **Avoiding certain foods (if so, which ones?)**

3. Changes to enjoyment	**Has the amount you enjoy eating and drinking altered?** **Probes: prefer to eat alone (eat with family / friends?)** **Avoid going out to pubs / cafes / restaurants** **Change in appetite?** **Change to ability to taste food?** **Worry about eating / drinking?**
4. Changes to lifestyle **** only ask if interviewee has noticed changes in Q 1-3**	**How have these changes impacted on your day to day life?** **Probes: altered social life?** **Altered daily routines?** **Less pleasure?** **Low mood / worry?** **Influence on the ability to work? financial security**
5. Adaptations **** only ask if interviewee has noticed changes in Q 1-3**	**Have you found ways to adapt to these changes?** **Probes: using oxygen** **Changes to way of preparing food** **Changes to the way of eating**
6. Advice and support **** only ask if interviewee has noticed changes in Q 1-3**	**What sort of advice or support have you received for these changes?**

5. Finally, to wrap up the interview at the end you mentioned ……is there anything you feel we have not covered in the interview about your experience of eating and drinking since your diagnosis that you would like to add now?

That concludes all my questions. Thank you for taking the time to be interviewed about your eating and drinking

Appendix 17: Interview topic guide final interviews.

Interview guide

6. **Introduction**

'Thank you for agreeing to take part in this interview / swallow assessment. Your participation is very much appreciated. As you know, I will be recording the interview. This is so a member of the project team can type out what we both say later for analysis. Any identifying information about you (including your name) will be removed from the typed version of the interview transcription. All the information will be stored securely. You may see me writing while you are talking this is only to help me for further discussion. Our discussion will last approximately 1 hour'

7. Demographics

'First I'd like some basic information about yourself. Can you tell me your age? Can you remember when you were diagnosed with IPF? Do you use oxygen? Do you work? (if so ask type of work) are you a current or ex-smoker?'

Now we will move on to the main part of the interview. Please let me know at any point if you would like to stop or take a break. I will be covering three main areas, focusing on your eating, drinking and diet. I'll ask a range of questions to help direct our discussion in each area. Is it OK to start the interview?

8. **Main interview**

Can we start off by you describing how IPF affects your life right now?

9. **Eating and drinking experience**

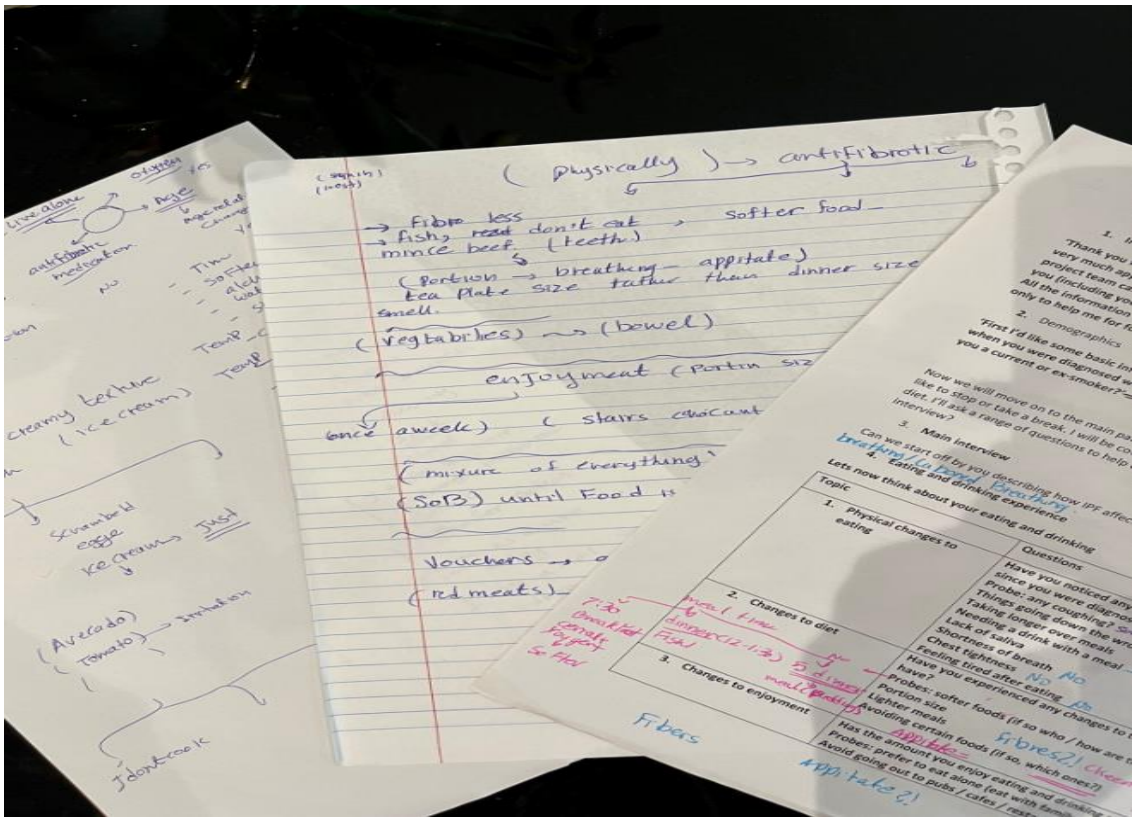
Lets now think about your eating and drinking

Topic	**Questions**
7. Physical changes to eating	
8. Changes to diet	
9. Changes to enjoyment	
10. Changes to lifestyle **** only ask if interviewee has noticed changes in Q 1-3**	
11. Adaptations **** only ask if interviewee has noticed changes in Q 1-3**	
12. Advice and support **** only ask if interviewee has noticed changes in Q 1-3**	

10. Finally, to wrap up the interview at the end you mentioned ……is there anything you feel we have not covered in the interview about your experience of eating and drinking since your diagnosis that you would like to add now?

That concludes all my questions. Thank you for taking the time to be interviewed about your eating and drinking.

Appendix 18: Example of note taking during interview.



Appendix 19: An exploratory study of swallowing function and patients' perceptions in IPF. Study protocol.

An exploratory study of swallowing function and patients' perceptions in IPF

Amal Alamer, MSc, BSRC

Primary Investigator:

Dr. Ian Forrest

Supervisors:

Prof. Chris Ward

Prof. Joanne M Patterson

Dr. Michael Drinnan

Translational and clinical research Institute

Medical School

Newcastle University

Background

The pharynx is a shared tube for breathing and swallowing. Because of this normal swallowing is a surprisingly complex process, coordinated with breathing in order to protect the airway. Breathing has to be temporarily suspended during swallowing to avoid food, liquid and saliva being aspirated down into airway. The integrity of the swallowing system, the respiratory system and the links between the two are critical for maintaining safe swallowing (Martin-Harris et al., 2005). However, swallowing and swallowing ventilation coordination can be altered and compromised in patients with chronic lung diseases, As a result, patients are more likely to suffer a rapid deterioration of their lung function and be admitted more frequently to hospital. The problem of swallowing difficulties (dysphagia) followed by aspiration in chronic lung diseases such as: (Chronic Obstructive Pulmonary Disease) has begun to be recognised in the literature (Ghannouchi *et al.*, 2016b). However, to date, this research has not considered the association between dysphagia and other, less common chronic lung diseases, such as idiopathic pulmonary fibrosis (IPF). Idiopathic pulmonary fibrosis (IPF) is a disease where the lungs scar for unknown reasons and there is a high mortality of 50% 5 years after the first diagnosis (Raghu *et al.*, 2016). New ways of helping patients are therefore urgently required.

The aim of this research is to perform a small, exploratory study to understand the swallowing patterns of patients diagnosed with IPF and associated perceptions of ability to eat and drink. This will indicate whether such studies are possible in people with IPF. This could lead to larger studies to identify the role of dysphagia in Idiopathic pulmonary Fibrosis patients. This initial exploratory study therefore represents a first step in an important gap in knowledge which may have translational significance in a disease where new treatments are very much needed.

Aim:

To determine whether it is possible to evaluate possible swallowing-breathing dysfunction and the potential existence of dysphagia in IPF patients.

Objectives:

Development of practical methods to simultaneously inform swallowing and breathing coordination.

Explore swallowing function and perceptions in IPF.

Explore swallowing-breathing dysfunction and the potential existence of dysphagia in IPF patients.

Inclusion/Exclusion Criteria

Inclusion criteria:

60 years old or more.

Working diagnosis of IPF made by a multi-disciplinary team.

Ability to complete the questionnaires and follow instructions.

Exclusion Criteria

History of head and neck surgery.

Current diagnosis or history of dysphagia.

History of stroke.

Inability to complete the questionnaires.

Patients with neurological impairment or neurological disease.

Any contraindication for FEES examination including a history of vasovagal attack.

History of other lung diseases.

Inability to follow instructions, for example: learning difficulty.

Methods

The aim is to perform a small exploratory study in Ten IPF patients. The participants will be recruited from the regional Interstitial Lung Disease (ILD) Clinic. Participants will be required to visit the Clinical Research Facility (CRF) department at Royal Victoria Infirmary Hospital (RVI) once only. The visit will last for 60 minutes. The given meal will consist of a cheese and tomato sandwich, milk and a biscuit. Special monitors, and a nasopharyngeal scope or camera, to study swallow will be used. All procedures, equipment, consumables and the patients recruitment will be carried out by the study team under the clinical supervision of Dr. Ian Forrest (ILD lead consultant) and Prof. Joanne M Patterson (academic speech and language therapist).

Protocol

Questionnaires

At the beginning of the visit, participants will be asked to complete three questionnaires regarding swallowing, reflux and throat symptoms. Participants will complete the questionnaires by themselves, and if they need any guidance, a researcher will be available to help.

The questionnaires are:

Reflux Symptoms Index (RSI).

The Newcastle Laryngeal Hypersensitivity Questionnaire (LHQ).

The Eating Assessment Tool-10 (EAT-10).

Physiological measurements

Objective measurements of breathing and swallowing coordination will be obtained by using the respiratory and swallowing monitors listed below. These monitors will be connected via an integrated data acquisition device called Powerlab.

Nasal pressure Cannula

A Nasal pressure cannula connected to a pressure transducer will be used to monitor respiratory airflow. It is a simple, non-invasive and sensitive method to detect respiratory events.

Non-invasive Respiratory pneumobelt

The respiratory belt will be wrapped around the participant's chest, it is a non-invasive monitor designed to measure changes in chest diameter resulting from breathing. These measurements can indicate inhalation and expiration. The belt comes with an adjustable strap that make it comfortable to wear, rugged and reliable.

Submental Electromyography sEMG

The submental electromyography electrode will be taped beneath the chin over the geniohyiod-mylohyoid-anterior digastric muscle complex. Because these muscles are laryngeal elevator, their activity provides information concerning the onset and duration of oropharyngeal swallowing.

Throat Microphone

The throat microphone will be tapped at the middle of the neck. Swallow sound will be recorded from the throat microphone to mark the moment of swallow.

Flexible Endoscopic Evaluation of Swallowing (FEES)

FEES (*Flexible* endoscopic evaluation of swallowing) uses a thin nasopharyngeal scope for studying swallowing. It is inserted into the nose and investigates the pharyngeal stage of swallowing. It is able to detect abnormal aspects such as aspiration of fluids or food into the airways and identifying bolus residue in the pharynx and pharyngeal-laryngeal area. The scope is thin, flexible, easy to use, well tolerated and less costly than other techniques and does not involve radiation exposure.

The FEES will be conducted by a speech and language therapist with the required level of competency as set out by the Royal College of SALT guidelines. The FEES will be recorded and anonymised at the time of the examination. The patient's study

number will be used for the purposes of blinded post-rating patient identification and analysis.

Pre-examination

Patients will be seated in an upright position in a chair with back support. The tip of the scope will be inserted in each nostril to assess which side provided the wider access. The nasendoscope will be coated in a water based lubricant to ease its passage through the inferior meatus of the nasal passage. The tip of the scope will be deflected downward into the oropharynx, below the tip of the uvula. A full view of the laryngopharynx will be maintained as best as possible throughout the examination.

Examination

The participant will be given a meal consisting of a cheese and tomato sandwich, milk and a biscuit. The patient will be given the instruction to swallow as they would do normally. The participant will keep their head in a neutral position throughout the test procedure. Compensatory swallowing manoeuvres or postures should not be performed during the research examination.

Study procedures:

Screening

All participants will be screened for any contraindications prior to the beginning of the study. The volunteers will be briefed about the study protocol and will sign a written informed consent.

Study day

The participants will arrive at the Clinical Research Facility department (CRF) at Royal Victoria Infirmary Hospital (RVI). The visit will last for 60 minutes. Participants will be asked to fill in Reflux Symptoms Index questionnaire, The Newcastle Laryngeal Hypersensitivity questionnaire and the EAT-10 questionnaire, first. Then a nasal cannula will be placed on the participant's nose with the prongs seated comfortably inside the nostrils and the tubes placed over the ears. A non-invasive pneumobelt will be wrapped around the chest. Additionally, Submental EMG will be tapped beneath

the chin and throat microphone will be tapped at the middle of the neck. These monitors will be connected via an integrated data acquisition device called power lab. Simultaneously, Fiberoptic endoscopic evaluation of swallowing will be performed while the participant is having a meal that consists of a cheese and tomato sandwich, milk and biscuit. All participants will be given a 10 minutes adaptation period before they eat the standard meal.

Adverse event (AE)/ serious adverse event (SAE):

Any unwanted medical condition will be reported to the primary investigator. Including death, life-threatening condition, and requirement of hospitalization.

End of study

Study will be ended once the sample size has been achieved and the review bodies will be notified within 90 days.

Appendix 20: Faculty of medical sciences: ethical approval for the study titled "Assessment of normal swallowing-breathing mechanism in healthy older volunteers.

08 July 2019

Amal Alamer
Institute of Cellular Medicine

Faculty of Medical Sciences
Newcastle University
Medical School
Framlington Place
Newcastle upon Tyne
NE2 4HH

FACULTY OF MEDICAL SCIENCES: ETHICS COMMITTEE

Dear Amal

Title: Assessment of normal swallowing-breathing mechanism in healthy older volunteers
Application No: 1692/12464/2019
Start date to end date: 15/08/2019 to 15/08/2020

On behalf of the Faculty of Medical Sciences Ethics Committee, I am writing to confirm that the ethical aspects of your proposal have been considered and your study has been given ethical approval.

The approval is limited to this project: **1692/12464/2019**. If you wish for a further approval to extend this project, please submit a re-application to the FMS Ethics Committee and this will be considered.

During the course of your research project you may find it necessary to revise your protocol. Substantial changes in methodology, or changes that impact on the interface between the researcher and the participants must be considered by the FMS Ethics Committee, prior to implementation.*

At the close of your research project, please report any adverse events that have occurred and the actions that were taken to the FMS Ethics Committee.*

Best wishes,

Yours sincerely

M Holbrough

Marjorie Holbrough
On behalf of Faculty Ethics Committee

cc.
Professor Daniel Nettle, Chair of FMS Ethics Committee
Mrs Kay Howes, Research Manager

*Please refer to the latest guidance available on the internal Newcastle web-site.

Appendix 21: Simultaneous Assessment of Swallowing and Breathing Coordination (preliminary reproducibility study)

Methods

Aim

The main aim of this study was to measure the level of agreement using repeated measures of respiratory/swallow variables with healthy volunteers.

Study objectives

To determine the effect of bolus volume on swallow apnoea duration.

To examine the respiratory swallow phase pattern.

Participants

This was a cross-sectional study of a convenience sample of healthy adults. Participants were aged 18 and over with no disease that might cause dysphagia. They were recruited from Newcastle University in the period February–March 2020. The sample comprised ten participants, seven of whom attended two visits.

Protocol

Breathing- swallowing -coordination measurements

Objective measurements of breathing and swallowing coordination were obtained using the respiratory and swallowing monitors listed below. These monitors were connected via an integrated PowerLab data acquisition device (see Figure 1).

Nasal pressure cannula

A nasal pressure cannula, connected to a pressure transducer, was used to monitor respiratory airflow. This is a simple, non-invasive and sensitive method used to detect respiratory events.

Non-invasive respiratory pneumobelt

A respiratory pneumobelt, which is wrapped around the participant's chest, is a non-invasive monitor designed to measure changes in chest diameter resulting from breathing. These measurements can indicate inhalation and expiration. The belt comes with an adjustable strap that makes it comfortable to wear, and it is robust and reliable.

Submental electromyography

A submental electromyography (sEMG) electrode was taped beneath the chin over the geniohyoid-mylohyoid-anterior digastric muscle complex. Because these muscles are laryngeal elevators, their activity provides information concerning the onset and duration of oropharyngeal swallowing.

Throat microphone

A throat microphone was taped to the middle of the neck. The sound of swallowing was recorded by the throat microphone to mark the moment of swallowing.

Breathing–swallowing coordination assessment

The participants were instructed to drink measured amounts of water using a calibrated cup (5 ml, 5 ml, 10 ml, 10 ml, 20 ml, 20 ml). An additional 5 ml was given at the beginning of the study for training purposes (see Figure 1).

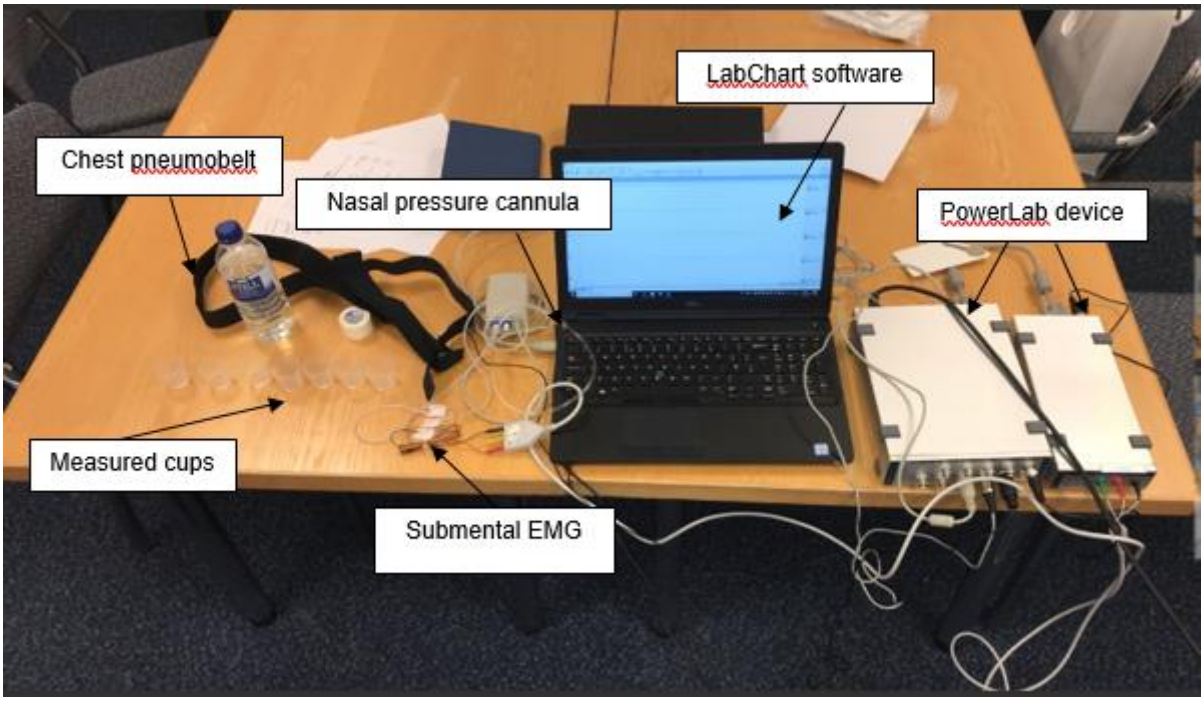


Figure 1: Respiratory -swallowing coordination measurement instruments.

Outcome measures

- Swallow apnoea duration (SAD) was measured in seconds from the onset of respiratory apnoea to the offset of respiratory apnoea.
- The respiratory/swallow phase was identified by locating the onset and offset of swallow apnoea and determining if inhalation or exhalation was present immediately before swallow apnoea onset and immediately after swallow apnoea offset.

Analysis

Continuous variables were presented using means and medians. For interrater reliability, data were analysed using average measurement, absolute agreement, 2 way mixed effect model. Interclass correlation coefficients (ICC) and 95% Confidence interval (CI) were calculated. All statistical analyses and graphic presentations in this study were performed using Statistical Package for the Social Sciences (SPSS, IBM SPSS Statistics 26) and Microsoft Excel 2016 (Microsoft Corporation, Washington, DC, USA).

Results

Establishing the level of agreement between two measurements

The scatter plots in Figure 2 present good agreement in SAD between the first and second trials (n:7), as the points lie close to the line of equality. The ICC= 0.943, and the 95% CI o= 0.52– .99. Based on the ICC result, the test–retest reliability is moderate to excellent.

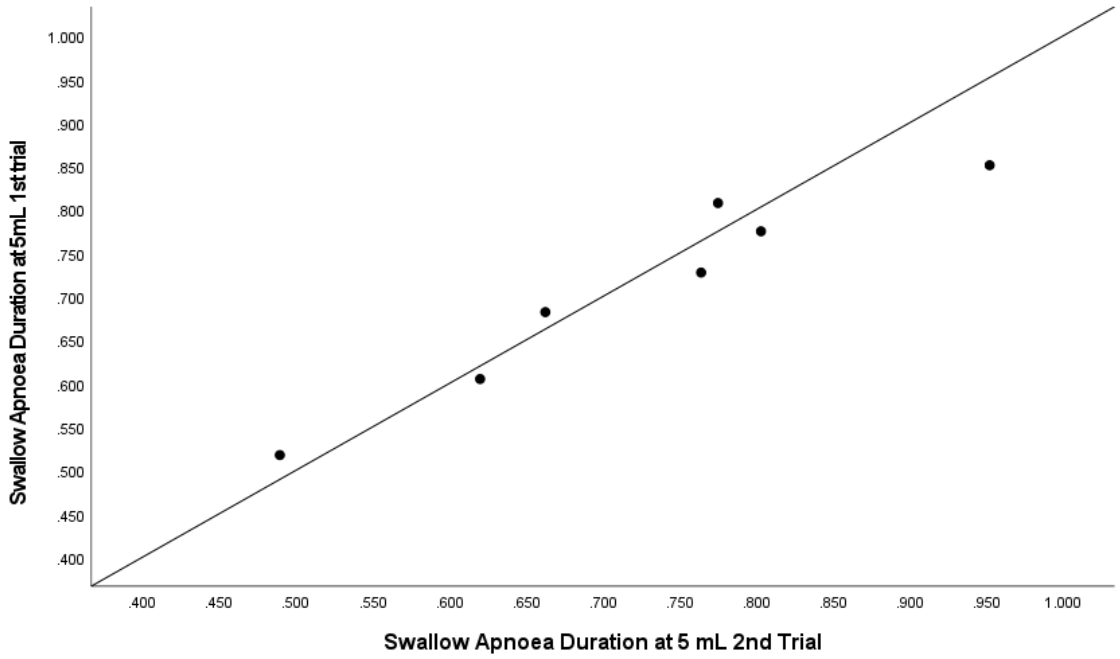


Figure 2. Scatter plots for swallow apnoea duration at 5 ml in the 1st and 2nd trials (n:7). Black dots represent the swallow apnoea duration for both first and second trials, the black solid line represent the line of equality between first and second trial.

Effect of bolus volume on SAD

The mean for SAD across the three different bolus volumes, 5 ml, 10 ml and 20 ml were measured. When the bolus volume increases, SAD also increases (see Figure 3).

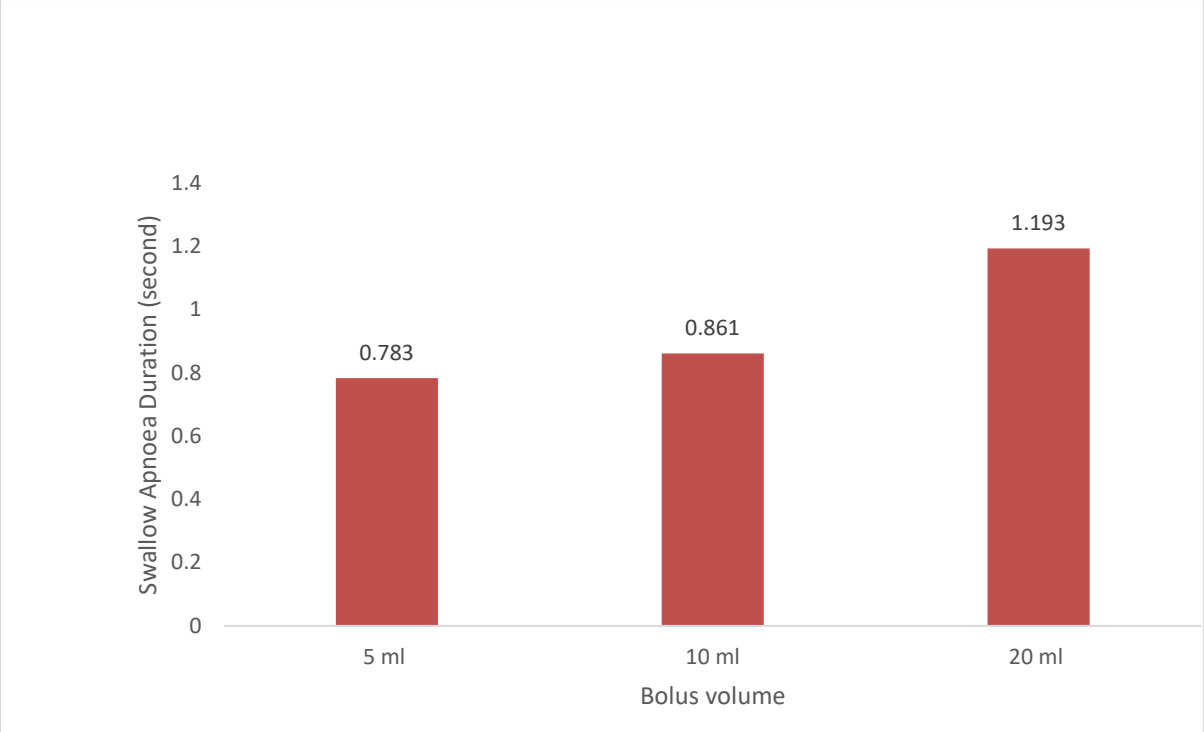


Figure 3. Mean for swallow apnoea duration across the three different bolus volumes (5ml, 10 ml and 20 ml).

Respiratory swallow phase pattern

This study observed four different respiratory phase patterns. In Figure 4, swallowing happens during exhalation, and respiration resumes with exhalation (A: Ex/Ex pattern). In Figure 5, swallowing occurs during inhalation, and respiration resumes with inhalation (B: IN/IN pattern). In Figure 6, swallowing occurs during inhalation, and respiration resumes with exhalation (C: IN/EX pattern).

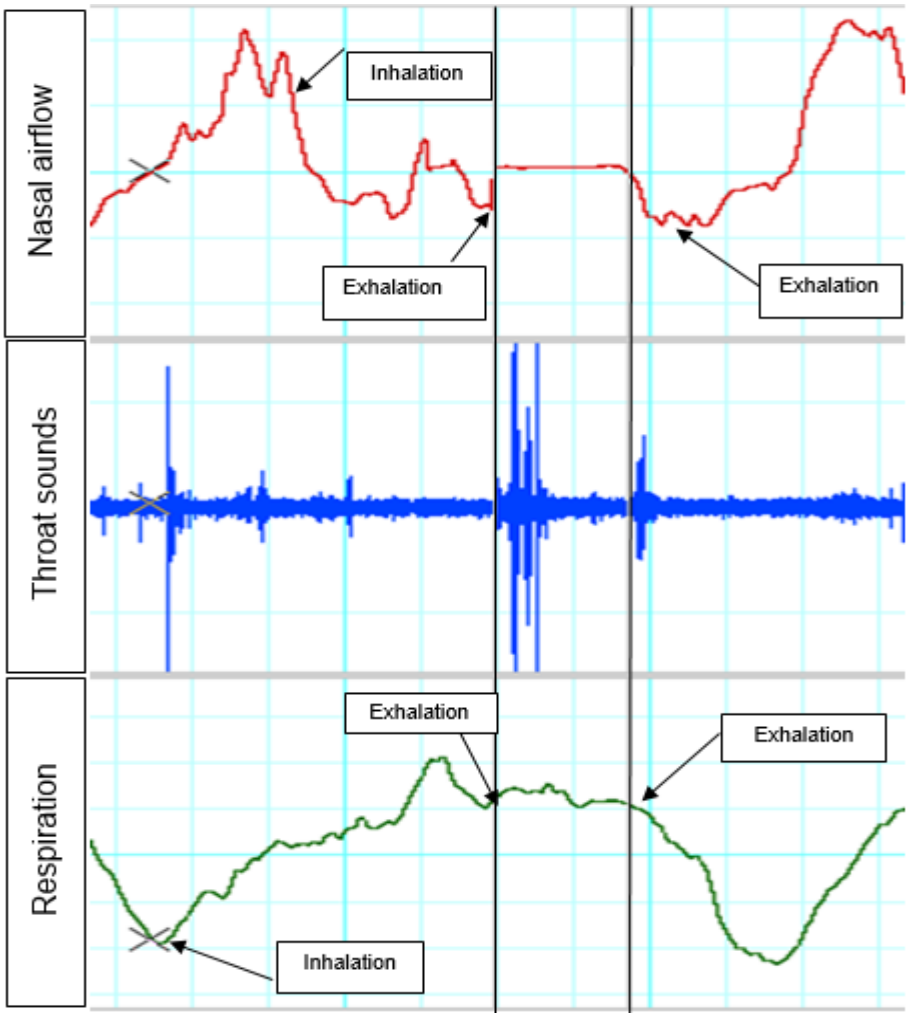


Figure 4. Exhalation/ Exhalation respiratory swallow phase pattern

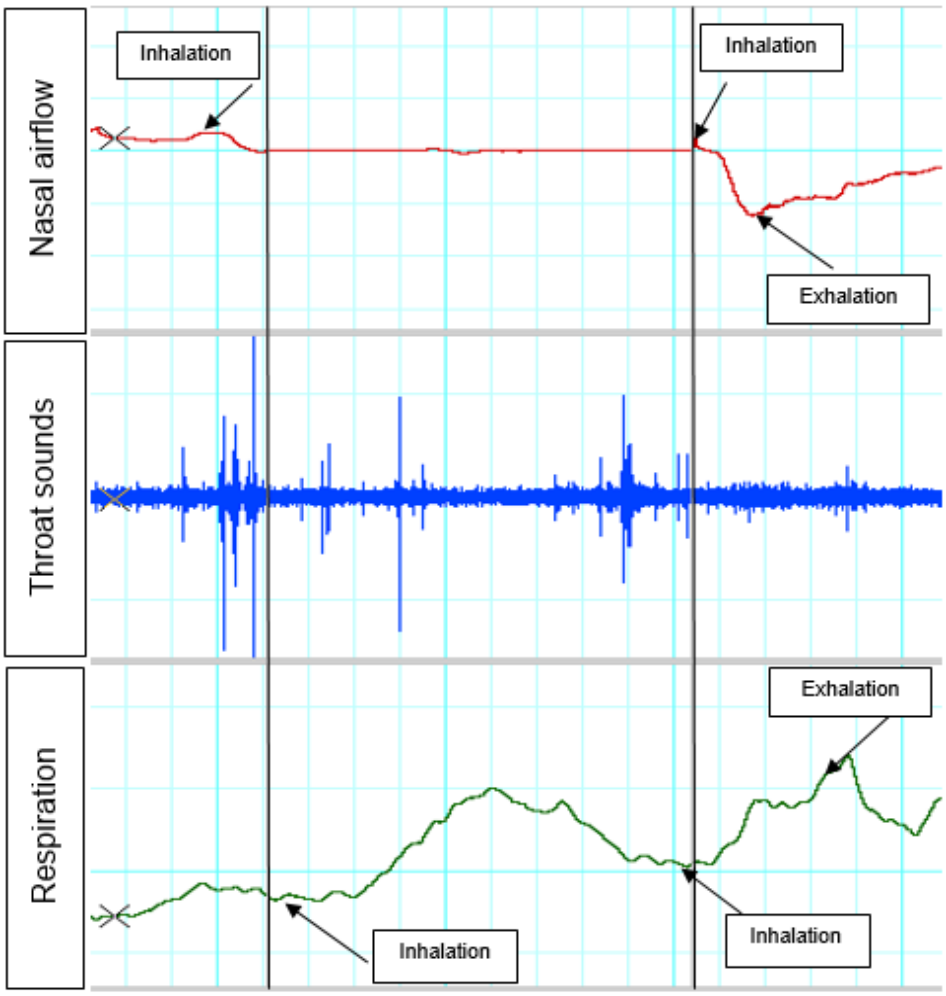


Figure 5. Inhalation/Inhalation respiratory swallow phase pattern.

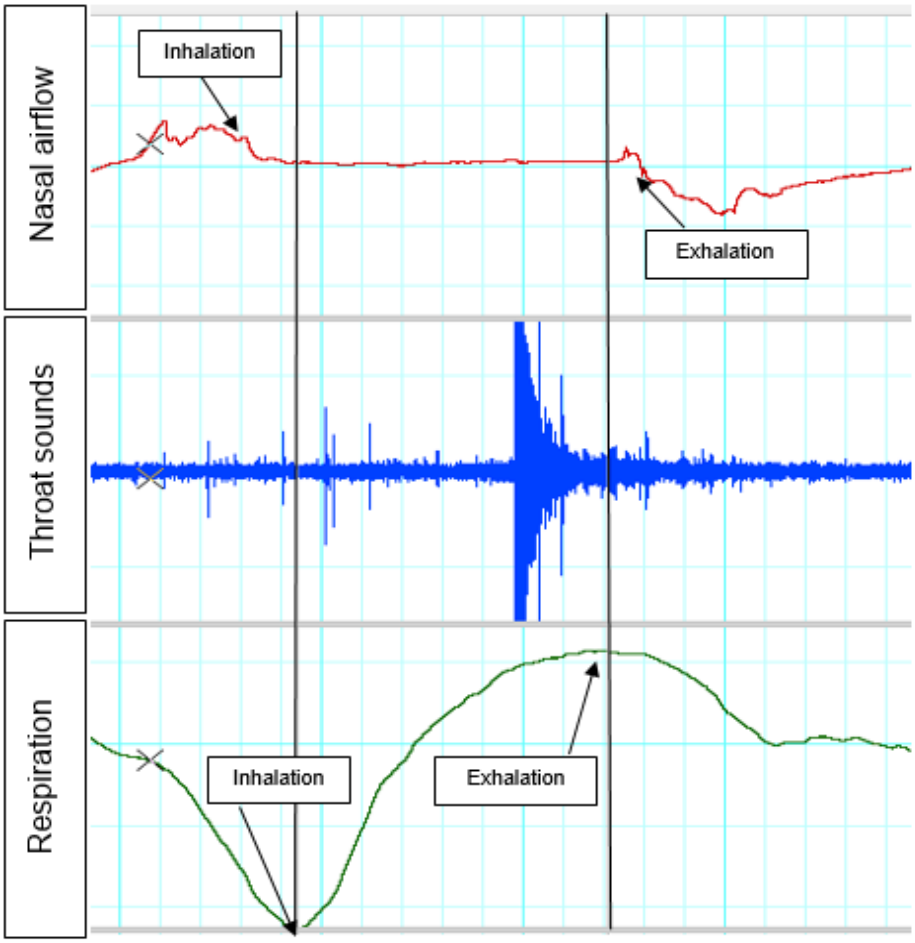


Figure 6. Inhalation/ Exhalation respiratory swallow phase pattern.

The respiratory phase surrounding swallow apnoea in each participant during the 1st and 2nd trials was measured. The Ex/Ex respiratory pattern is the most frequently reported across participants in both trials (see Table 1).

	Bolus volume (1st trial)						Bolus volume (2nd trial)					
Participant	5 mL	5 mL2	10 mL	10 mL2	20 mL	20 mL2	5 mL	5 mL	10 mL	10 mL	20 mL	20 mL
1	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	In/Ex
2	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex
3	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	In/Ex	In/Ex	In/Ex	Ex/Ex	Ex/Ex	In/In
4	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	In/Ex	In/Ex	Ex/Ex	Ex/Ex	In/Ex	In/Ex	In/Ex	In/Ex
5	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/In	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex
6	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex
7	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	In/In	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex	Ex/Ex

Table 1. Respiratory phase surrounding swallow apnoea (SA) across the three different bolus volumes during 1st and 2nd trials.
Ex/Ex: Exhalation/Exhalation; Ex/In: Exhalation/Inhalation; In/Ex: Inhalation/Exhalation; In/In: Inhalation/Inhalation.

Impact of COVID-19 on this study

The main aim of this study was to measure the level of agreement between two different time points. This study demonstrated different respiratory phases during swallowing and helped provide an understanding of whether the newly developed respiratory/swallow measurement technique would be able to attain the same results when repeating the measurements under identical circumstances. The main intention was to use these baseline data for the purpose of ensuring repeatability in a study involving healthy older volunteers (control group) and patients with Idiopathic Pulmonary Fibrosis (IPF) (disease group). However, due to the COVID-19 pandemic, neither the control nor the IPF group studies were conducted.

Appendix 22: FEES training program certificate

SVS
speech - voice - swallow
associates

This is to certify that

Amal Al Amer

attended

FEES TRAINING PROGRAMME

Foundation Level: 7th-8th November 2019

Clinical Skills Education Centre, Nottingham University Hospitals NHS Trust

Training provided by SVS Training Ltd

Signed:

H Fiona Robinson MSc PGCMedEd FRCSLT
SVS Training Ltd

Suzanne Slade MA(RM) BSc MRCSLT
SVS Training Ltd

Date: 8th November 2019

Appendix 23: Good for Clinical Practice (GCP) certificate.

NIHR | National Institute for Health Research

Certificate of Completion

Amal Alamer

has completed

Introduction to Good Clinical Practice (GCP)

A practical guide to ethical
and scientific quality standards in clinical research

on

10th December 2019

Including EU Directives, Medicines for Human Use (Clinical Trials) Regulations & the Department of Health UK Policy Framework for Health & Social Care Research, as applied to the conduct of Clinical Trials & other studies conducted in the NHS

Modules completed:

Introduction to Research and the GCP standards
Preparing to deliver your study
Identifying and recruiting participants: eligibility and informed consent
Ongoing study delivery and data collection
Safety Reporting
Study closure
Course summary and knowledge check

This course is worth 6 CPD credits

CPD
The CPD Certification Service

Delivering research to make patients, and the NHS, better