Oropharyngeal Swallowing Physiology and Safety in Patients with Idiopathic Pulmonary Fibrosis: A consecutive descriptive case series

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Research Article

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Abstract

Introduction:

Dysphagia occurs in multiple respiratory pathophysiologicals, 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.

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 poorly understood, emerging data suggest an important role for aspiration [2]. Aspiration is the direct inhalation of secretions or ingested materials from the oesophagus or from the oropharynx into the airways, which can be associated with lung injury and pneumonia. To date,
investigations of IPF and aspiration have been limited to gastro-oesophageal reflux disease in which gastric content reflexes up into the oesophagus, with the potential to pass into the airway via the pharynx (microaspiration) [3]. 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 [4]. A dysregulated lung microbiome has also been implicated in high profile studies [5].

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 [6, 7]. Early evidence suggests 20% of COPD patients have a swallowing impairment, and subclinical aspiration, identified on Videofluoroscopy Swallow Studies (VFSS) [8]. Prior study found 44% of COPD patients reported swallowing difficulties symptoms on a patient-reported outcome measure [6]. 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 RVI. 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, European Respiratory Society (ERS), Japanese Respiratory Society (JRS) and Latin American Thoracic Society (LATS) clinical practice guidelines [9]. 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. The study was described in details, with full written consent taken at the outset, before study activities commenced.

**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 [10]. It has been used previously with patients with chronic respiratory disease such as COPD [6, 11, 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 20mL); 5 mL paste (custard) and solid (1/4 biscuit) mixed with radiopaque barium sulphate (E-Z-PAQUE), conducted in the lateral plane and one 10mL 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 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 [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].

Higher scores for both tools indicate poorer swallowing.

Results

We recruited 14 participants with IPF: 10M, 4F, range 51–82 years. Ten IPF patients: 7M, 3F, 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. 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 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] where the median FVC % of predicted was 72% [range: 51–92] indicating a restrictive lung pattern [18].

| 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] |
| FEV₁/FVC, %                 | 80.5 [74–93] |

Data presented as median (ranges), unless otherwise indicated. Note: BMI, body mass index; mMRC, modified medical research council dyspnoea scale; FEV1, forced expiratory volume in 1 s; FVC, forced vital capacity.

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

1. 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.

2. 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 patients’ perception of swallowing difficulty. In our study 4 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 (AECOPD) 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 [6,12]. 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, 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 [19]. 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 swallwers 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].

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 [20]. 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 [21], which has been associated with a more severe disease phenotype, higher mortality and lung function decline in people with IPF [22].

Non-sterile aspiration, related to dysphagia and unprotected by cough, therefore represents a candidate source of complex lung injury and a potential factor in life-threatening acute exacerbations. Acute exacerbations in IPF are of very particular concern as they represent the most common cause of death in IPF. 46% of deaths in IPF are preceded by an acute exacerbation and the median survival after an acute exacerbation is approximately 3 to 4 months [23].

This prospective consecutive case series is proof of concept and descriptive. Further exploration is needed to establish the association between dysphagia and IPF, and the clinical significance of such a link. Our experience indicated that such studies are possible but also underline that studies in patients with IPF 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 [24] and together with selected patient reported outcome measures, 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.

**Declarations**

All methods were performed in accordance with the approval guidelines and regulations.

**Research ethics.**

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.

**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.

**Competing interest statement.**

No competing interests or conflicts of interest.

**Consent for Publication.**

Not applicable

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John Simpson is an NIHR Research Professor.
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.

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References

1. Raghu G, Remy-Jardin M, Myers JL, et al. Diagnosis of idiopathic pulmonary fibrosis. An official ATS/ERS/JRS/ALAT clinical practice guideline. American Journal of Respiratory and Critical Care Medicine. 2018;198(5):e44-e68.

2. Brownlee I, Aseeri A, Ward C, et al. From gastric aspiration to airway inflammation. Monaldi Archives for Chest Disease. 2010;73(2).

3. Wang Z, Bonella F, Li W, et al. Gastroesophageal reflux disease in idiopathic pulmonary fibrosis: uncertainties and controversies. Respiration. 2018;96(6):571–87.

4. Raghu G, Freudenberger T, Yang S, et al. High prevalence of abnormal acid gastro-oesophageal reflux in idiopathic pulmonary fibrosis. European Respiratory Journal. 2006;27(1):136–42.

5. Spagnolo P, Molyneaux PL, Bernardinello N, et al. The role of the lung’s microbiome in the pathogenesis and progression of idiopathic pulmonary fibrosis. International Journal of Molecular Sciences. 2019;20(22):5618.

6. Garand KL, Strange C, Paoletti L, et al. Oropharyngeal swallow physiology and swallowing-related quality of life in underweight patients with concomitant advanced chronic obstructive pulmonary disease. International Journal of Chronic Obstructive Pulmonary Disease. 2018;13:2663.

7. Lin T-F, Shune S. Chronic Obstructive Pulmonary Disease and Dysphagia: A Synergistic Review. Geriatrics. 2020;5(3):45.

8. B. Mokhlesi JAL, A.W. Rademaker, C.A. <oropharyngeal dysfunction in COPD patient.pdf>. Chest 2002;121(2):361–9.

9. Raghu G, Collard HR, Egan JJ, et al. An official ATS/ERS/JRS/ALAT statement: idiopathic pulmonary fibrosis: evidence-based guidelines for diagnosis and management. American Journal of Respiratory and Critical Care Medicine. 2011;183(6):788–824.
10. Belafsky PC, Mouadeb DA, Rees CJ, et al. Validity and reliability of the Eating Assessment Tool (EAT-10). Annals of Otology, Rhinology & Laryngology. 2008;117(12):919–24.

11. Regan J, Lawson S, De Aguiar V. The Eating Assessment Tool-10 predicts aspiration in adults with stable chronic obstructive pulmonary disease. Dysphagia. 2017;32(5):714–20.

12. Lindh MG, Janson C, Johansson MB, et al. Swallowing dysfunction in patients hospitalised due to a COPD exacerbation. ERJ Open Research. 2021;7(2).

13. Cheney DM, Siddiqui MT, Litts JK, et al. The ability of the 10-item eating assessment tool (EAT-10) to predict aspiration risk in persons with dysphagia. Annals of Otology, Rhinology & Laryngology. 2015;124(5):351–4.

14. Martin-Harris B, Brodsky MB, Michel Y, et al. MBS measurement tool for swallow impairment—MBSImp: establishing a standard. Dysphagia. 2008;23(4):392–405.

15. Martin-Harris B, Humphries K, Garand KL. The modified barium swallow impairment profile (MBSImp™©)—innovation, dissemination and implementation. Perspectives of the ASHA Special Interest Groups. 2017;2(13):129–38.

16. Beall J, Hill EG, Armeson K, et al. Classification of physiologic swallowing impairment severity: a latent class analysis of modified barium swallow impairment profile scores. American Journal of Speech-Language Pathology. 2020;29(2S):1001–11.

17. Robbins J, Coyle J, Rosenbek J, et al. Differentiation of normal and abnormal airway protection during swallowing using the penetration–aspiration scale. Dysphagia. 1999;14(4):228–32.

18. Aaron SD, Dales RE, Cardinal P. How accurate is spirometry at predicting restrictive pulmonary impairment? Chest. 1999;115(3):869–73.

19. Dua KS, Ren J, Bardan E, et al. Coordination of deglutitive glottal function and pharyngeal bolus transit during normal eating. Gastroenterology. 1997;112(1):73–83.

20. Pathak JL, Yan Y, Zhang Q, et al. The role of oral microbiome in respiratory health and diseases. Respiratory Medicine. 2021;185:106475.

21. Kashyap D, Baral B, Verma TP, et al. Oral rinses in growth inhibition and treatment of pylori infection. BMC Microbiology. 2020;20(1):1–18.

22. Bennett D, Bargagli E, Refini RM, et al. Helicobacter pylori seroprevalence in patients with idiopathic pulmonary fibrosis. European Respiratory Journal. 2014;43(2):635–8.

23. Collard HR, Ryerson CJ, Corte TJ, et al. Acute exacerbation of idiopathic pulmonary fibrosis. An international working group report. American Journal of Respiratory and Critical Care Medicine. 2016;194(3):265–75.

24. Patterson JM, McColl E, Carding PN, et al. Swallowing performance in patients with head and neck cancer: a simple clinical test. Oral Oncology. 2009;45(10):904–7.

Tables

Table 2 is available in the Supplementary Files section.
Figures

Figure 1

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

Figure 2

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

Figure 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.

Supplementary Files

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- Table2.jpg