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ORIGINAL ARTICLE

The Relationship Between Lingual Strength and Functional Swallowing Outcomes in Parkinson's Disease

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Abstract

The purpose of this retrospective study was to determine whether reduced lingual strength was associated with functional swallowing outcomes in individuals with Parkinson's disease (PD). Participants ($N=42$) completed evaluations of maximal lingual isometric pressure (MIP) and mean lingual swallowing pressure (MSP), and flexible endoscopic evaluations of swallowing. Regression models were used to determine the association between lingual strength and functional swallowing outcomes of airway invasion, the presence of post-swallow pharyngeal residue, and the amount of pharyngeal residue (when present). Results revealed that higher MIP ($p=0.002$, OR 0.93) and higher MSP ($p=0.001$ OR 0.88) were associated with less airway invasion of thin liquids. Both MIP and MSP were able to differentiate between those with and without dysphagia (MIP: AUC 0.7935, $p=0.001$; MSP: AUC 0.75, $p=0.026$). Neither MIP nor MSP was related to the presence of residue. However, when thin liquid oropharyngeal residue was present, both MIP ($p<0.001$, OR 0.99) and MSP ($p<0.001$; OR 0.98) were significantly associated with the amount of residue observed. Similarly, when thin liquid hypopharyngeal residue was present, MIP ($p<0.001$, OR 0.99) and MSP ($p<0.001$, OR 0.98) were associated with the amount of residue observed. These findings suggest a relationship between reduced lingual strength and worse thin liquid swallowing safety and efficiency; however, the magnitude of these effects was small. This indicates that lingual strength is one important contributing factor to functional swallowing impairments in PD and may identify those with unsafe swallowing. These findings have important clinical implications for including lingual strength in the screening, assessment, and management of dysphagia in PD.

Keywords Lingual strength · Aspiration · Residue · Parkinson's disease

Introduction

Dysphagia is highly prevalent in Parkinson's disease (PD) [1, 2] resulting in adverse pulmonary health consequences, malnutrition, dehydration, morbidity, mortality, and reduced quality of life [3–5]. Despite the known consequences of dysphagia in PD, the relative contribution of each impaired system (e.g., respiratory, oropharyngeal, laryngeal systems) on swallowing safety (i.e., airway invasion) and efficiency (i.e., pharyngeal residue) is not well understood. Lingual function is a key component of the oropharyngeal system and is of particular importance given its central role in swallowing [6, 7]. Multiple components of lingual dysfunction have been documented in PD [8–11], and include deficits in timing, speed, and direction of lingual movements [10, 11]. Lingual dysfunction has also been associated with swallowing impairments in PD. For example, lingual bradykinesia has been associated with increased oropharyngeal transit

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time and increased mealtime duration [9, 10]. Increased duration of lingual gestures (i.e., tongue "pumping") has also been associated with reduced bolus control and airway invasion [8]. While these studies have explored various components of lingual movement, no studies have specifically explored the relationship between lingual *strength* and functional swallowing outcomes, measured via instrumental evaluation (i.e., airway invasion and pharyngeal residue), in PD.

Given that reduced lingual strength has been associated with worse functional swallowing outcomes in healthy older adults [12, 13] and more generally in neurologic populations [14–17], it is likely that a similar relationship may exist in PD. Understanding the relationship between lingual strength and swallowing function in PD has important implications for improving diagnostic and treatment targets. From an evaluation standpoint, assessing lingual strength may assist in accurate dysphagia screening and contribute to a more comprehensive understanding of the underlying etiology of dysphagia, if present. From a treatment standpoint, identifying if lingual weakness is a significant contributor to dysphagia in PD may aide in the development of more targeted intervention plans for individuals with PD and dysphagia. This is of particular clinical relevance, given that lingual strength can be easily assessed using the Iowa Oral Performance Instrument (IOPI) device, lingual strength training is an existing treatment approach [18, 19], and a case study has demonstrated the possibility for lingual strengthening treatment to improve swallow function in PD [20].

Historically, there has been conflicting evidence as to whether lingual strength is reduced in individuals with PD. Despite some studies which have not found lingual strength to be reduced [21–24], a growing body of literature has identified the presence of reduced lingual strength [25–28]. In fact, a recent systematic review and meta-analysis revealed reduced anterior lingual strength in people with PD compared to healthy controls [25]. In studies that have identified reduced lingual strength in PD, it has been associated with clinical signs of dysphagia, such as lower swallowing-related quality of life, longer reported eating durations, reduced desire to eat, and diet restrictions [26, 28]. Further, recent work has sought to determine whether lingual strength differs among individuals with PD with and without self-reported dysphagia. One study identified an inverse relationship between self-reported dysphagia symptoms and maximal isometric pressure (MIP), but no relationship with mean swallowing pressure (MSP) [26]. Conversely, another study found MSP to be reduced in individuals with PD and self-reported dysphagia, as compared to those with PD and no dysphagia [29]. Importantly, dysphagia was assessed subjectively (via patient report) in both studies.

To our knowledge, there has been one study which has examined lingual strength—measured by a tongue pressure sensor sheet—during videofluoroscopy in people with PD [30]. While this study identified a relationship between increased time to lingual peak pressure duration and increased oral transit time in the dysphagic group, they found no difference in maximal lingual swallowing pressure (measured during a 5 mL bolus swallowing task) between those with and without dysphagia [30]. The authors only examined swallowing pressure and did not examine the relationship between maximal lingual strength and swallowing safety, efficiency, or severity outcomes. Additionally, flexible endoscopic evaluation of swallowing (FEES) may possess greater sensitivity for identifying functional swallowing outcomes of airway invasion and residue [31–34].

To date, lingual strength has not been correlated with functional swallowing outcomes in individuals with PD. Given that lingual strength appears to be reduced in a significant portion of individuals with PD [25], it is critical to understand what functional impact this reduction has on swallowing safety and efficiency, measured via instrumental evaluation. Therefore, the aim of this study was to determine the relationship between lingual strength—both MIP and MSP—and functional swallowing outcomes, specifically airway invasion and pharyngeal residue. We hypothesized that reduced lingual strength would be correlated with poorer functional swallowing outcomes (i.e., increased airway invasion and residue) identified via FEES. Additionally, we explored the utility of lingual strength as a screening tool to identify individuals with unsafe swallowing.

Methods

Participants

Forty-two adults with idiopathic PD participated in this study. Participants were English-speaking, community-dwelling volunteers from the greater metropolitan New York City area, living independently or with a partner or caregiver (or both). The only inclusion criterion was a diagnosis of idiopathic PD per a fellowship-trained movement disorders neurologist using UK brain bank criteria [35]. Exclusion criteria were diagnosis of significant central neurological disorders other than PD (e.g., stroke) and a history of head and/or neck cancer. Participants were not excluded based on cognitive status. Demographic information was self-reported by participants and included age, sex, and disease duration.

Study Design

This retrospective analysis was part of a larger study in which participants underwent a comprehensive evaluation of cognitive-linguistic, speech, and airway protective functioning, which included measures of lingual strength and

FEES. The evaluations were completed under supervision of a certified Speech-Language Pathologist with expertise in the evaluation of patients with PD. Evaluation appointments were scheduled when participants reported they would be in an "on" phase of their medication cycle. Written informed consent was received from all participants prior to evaluation, and all study procedures were in line with the Declaration of Helsinki. Institutional Review Board approval was received from Teachers College, Columbia University. Procedures are described below.

Lingual Strength: Iowa Oral Performance Instrument (IOPI)

Participants were seated in an upright position and completed isometric tongue pressure tasks and saliva swallows using the IOPI (IOPI Medical LLC, Woodinville, Washington, USA). The IOPI is a hand-held portable device that uses an air-filled plastic bulb (3.5 cm long; 4.5 cm diameter, 2.8 mL internal volume) [36] to measure force generation (kPa) of the orofacial and lingual musculature.

Two measures of lingual strength were obtained: (1) maximal isometric press (MIP) and (2) mean swallowing pressure (MSP). To obtain isometric lingual pressure, the IOPI bulb was placed between the tongue blade and the alveolar ridge and participants were instructed to "press as hard as you can with your tongue against the plastic bulb." To obtain swallowing pressures, the IOPI bulb was placed between the tongue blade and the alveolar ridge and participants were instructed to "swallow your saliva like you usually would." Three trials of isometric tongue presses followed by three trials of saliva swallows were completed for each participant. Each trial of MIP and MSP was recorded during the evaluation.

Airway Invasion and Pharyngeal Residue: Flexible Endoscopic Evaluation of Swallowing (FEES)

FEES were performed by a speech-language pathologist experienced in the performance and interpretation of endoscopic evaluations, using a 3 mm diameter flexible distal chip laryngoscope (ENT-5000; Cogentix Medical, New York, USA) and video system with an integrated LED light source LCD display (Cogentix Medical, DPU-7000A). Participants were seated in an upright position and the flexible laryngoscope was passed transnasally, without the use of topical anesthetic or vasoconstrictors. The tip of the endoscope was positioned within the oropharynx in order to visualize the pharynx, larynx, and subglottis before and after swallowing, and was advanced into the laryngeal vestibule after each swallow to more closely visualize residue patterns within the larynx and subglottic space. Participants were presented with various volumes of thin liquid, including one 5 cc and one 20 cc "held and cued" bolus and up to three 10 cc and up to three 90 cc "non-held and non-cued" boluses. For held boluses, participants were instructed to "hold this in your mouth and swallow when I tell you to." For non-held boluses, participants were instructed to "swallow whenever you're ready." Liquid boluses were dyed to maximize visualization. Six drops (~0.2 cc) of blue dye (Chef-O-Van Food Coloring, Rockford, Ohio, USA), green dye (Chef-O-Van Food Coloring), or three teaspoons (~24 g) of barium powder (E-Z-PAQUE barium sulfate for suspension, 96% w/w; E-Z-EM Canada, Inc., Anjou, Canada) were added to each cup. The order of blue, green, and barium boluses were randomized across participants and all participants who received thin liquid boluses received at least one blue/green dyed liquid and one barium liquid. Solid boluses included one or two five cc teaspoons of pudding (IDDSI 4, Hunt's Food Company, California, USA), and one Sunshine Crispy Original Saltine cracker (IDSSI 7, Sunshine Biscuits, Illinois, USA). Participants were instructed to say /i/ following each bolus in order to advance the scope into the laryngeal vestibule for visualization of the larynx and subglottis. Each trial was recorded and saved for offline analysis.

Outcome Measures

Lingual Strength

The maximum MIP score across three trials was computed after the evaluation and used to represent MIP. The average MSP score across three trials was calculated after the evaluation and used to represent MSP.

Swallowing Safety and Efficiency

FEES videos were segmented into individual video clips for each bolus trial, stored digitally, and each bolus was analyzed in the order in which the video clips were recorded by a pair of blinded raters. Raters were trained in Visual Analysis of Swallowing Efficiency and Safety (VASES) [37] which was used to assess bolus clearance from the pharynx by determining the amount of pharyngeal residue remaining after each trial. VASES ratings were made for the oropharynx and hypopharynx separately, using the anatomic and temporal boundaries outlined by Curtis et al. (2021), and rated on a 100-point visual analog rating scale. The Penetration-Aspiration Scale (PAS) [38] was used to assess depth of and reaction to airway invasion.

Residue ratings were in agreement if both raters indicated residue was present (> 0%) and paired ratings differed by ≤ 10% or if both raters indicated residue was absent (0%). Residue ratings that agreed (i.e., within 10%) were taken from the first rater and used for data analysis. For residue ratings that disagreed (i.e., differed by > 10% or differed on

the presence/absence of residue), ratings were resolved by a third, blinded, expert rater. PAS ratings were only considered to agree if they matched exactly. PAS ratings that were not in agreement were resolved by the same third expert rater.

Statistical Analysis

Ordinal multilevel regression models were used to examine the relationship between lingual strength and airway invasion. Four models were run to assess the association between lingual strength and airway invasion: (1) MIP and thin liquid airway invasion, (2) MSP and thin liquid airway invasion, (3) MIP and solid bolus airway invasion, and (4) MSP and solid bolus airway invasion. For statistically significant ordinal multilevel models, receiver operating characteristic (ROC) curves were used to determine how well lingual strength differentiated between "safe" and "unsafe" swallowing. For ROC curves, the maximum PAS score across bolus trials was used to represent participants' worst swallowing function We explored two categorizations: (1) no dysphagia (PAS 1–2) vs dysphagia (PAS 3–8) and (2) non-aspirators (PAS 1–5) vs aspirators (PAS 6–8). The area under the curve (AUC) was calculated to determine the probability that lingual strength would adequately differentiate safe and unsafe swallowing. From ROC analyses, we obtained the cutoff value that maximized sensitivity and specificity. We considered an AUC of 0.7–0.8 as "adequate" and 0.8–0.9 as "excellent"[39].

Zero-inflated beta mixed effects regression models was used to examine the relationship between lingual strength and (1) the presence vs absence of pharyngeal residue and (2) the amount of pharyngeal residue when present. Separate models were used for oropharyngeal and hypopharyngeal residue—with four models for each residue location: (1) MIP and thin liquid residue, (2) MSP and thin liquid residue, (3) MIP and solid bolus residue, and (4) MSP and solid bolus residue. Alpha was set at 0.05 and Holm-Bonferroni corrections were used to account for multiple comparisons within each research question (i.e., four corrections for airway invasion models, eight corrections for oropharyngeal residue models, and eight corrections for hypopharyngeal residue models) [40]. Statistical analyses were completed using R version 4.0.1 [41].

Results

Forty-two people with PD (32 males, 10 females) met inclusion criteria. The mean age of participants was 70.12 years (SD = 8.69; Range 48–86 years) and mean disease duration was 7.93 years (SD = 5.54; Range 1–20 years). Participant demographics are detailed in Table 1. MIP was obtained from all 42 participants, mean MIP was 45.48 kPa (SD = 14.26; Range 17–73), and when compared to published normative data from healthy adults [42], MIP was reduced in 40.48% of our sample. MSP was obtained from all 42 participants, mean MSP was 18.91 kPa (SD = 8.06; Range 6.33–35), and MSP was reduced in 35.7% of our sample when compared to recent data on healthy older adults [24]. All 42 participants completed a FEES exam. Of the 42 FEES exams conducted, PAS and residue for thin liquid boluses were obtained from 41 FEES exams and PAS and residue for solid boluses were obtained from 34 FEES exams. Due to various logistical (e.g., time constraints, supply availability) or patient-specific factors (e.g., food allergy, FEES intolerance), not all participants received every bolus, as per the protocol described above. The number of participants who received each bolus size and type is presented in Appendix Table 1, the distribution of PAS scores is presented in Table 2, and residue rating scores in Fig. 1.

Table 1 Participant demographics

	Range	Mean (SD)
Age (years)	48–86	70.12 (8.69)
Disease duration (years)	1–20	7.93 (5.54)

Lingual Strength and Airway Invasion

Ordinal multilevel regression models revealed a significant association between maximal isometric pressure (MIP) and airway invasion of thin liquids ($p = 0.002$, OR 0.93, 95% CI 0.89–0.97) (Fig. 2). Specifically, a one kPa increase in MIP was associated with a 7% lower odds of worse airway invasion. Mean swallowing pressure (MSP) was also significantly associated with airway invasion of thin liquids ($p = 0.001$ OR 0.88, 95% CI 0.82–0.94), indicating that a one kPa increase in MSP was associated with a 12% lower odds of worse airway invasion. Neither MIP ($p = 1$, OR 0.99, 95% CI 0.78–1.25) nor MSP ($p = 0.934$, OR 1.01, 95% CI 0.70–1.45) were significantly associated with airway invasion of solid boluses (Table 3).

The ROC analysis demonstrated that MIP was adequately able to differentiate between those with and without dysphagia (AUC 0.7935, $p = 0.001$, 0.95% CI 0.63–0.95), with an optimal cut-point of 52.5 kPa (sensitivity = 0.77, specificity = 0.70, accuracy = 76%; Fig. 3). MIP was unable to adequately differentiate between aspirators and non-aspirators (AUC 0.67, $p = 0.101$, 95% CI 0.50–0.85), with an optimal cut-point of 53.5 kPa (sensitivity = 1.00, specificity = 0.41; accuracy = 59%). Similarly, MSP was adequately able to differentiate between those with and without dysphagia (AUC 0.75, $p = 0.026$, 95% CI 0.56–0.94), with an optimal cut-point of 25.5 kPa (sensitivity = 0.87, specificity = 0.60, accuracy = 80%; Fig. 3). MSP was not able to differentiate between aspirators and non-aspirators (AUC 0.58, $p = 0.400$,

Table 2 Distribution of PAS scores

PAS score	Frequency liquids (%)	Frequency solids (%)	Frequency Max PAS liquids (%)	Frequency Max PAS solids (%)
1	166 (59.29)	75 (92.59)	10 (24.39)	31 (91.18)
2	3 (1.07)	0 (0.00)	0 (0.00)	0 (0.00)
3	63 (22.50)	0 (0.00)	12 (29.27)	0 (0.00)
4	2 (0.71)	1 (1.23)	1 (2.44)	0 (0.00)
5	29 (10.36)	0 (0.00)	6 (14.63)	0 (0.00)
6	1 (0.36)	1 (1.23)	1 (2.44)	0 (0.00)
7	6 (2.14)	1 (1.23)	5 (12.20)	1 (2.94)
8	7 (2.50)	2 (2.47)	6 (14.63)	2 (5.88)
NA	3 (1.07)	1 (1.23)	–	–

Total number of thin liquid boluses included in the sample = 280 boluses (number of participants = 41)

Total number of solid boluses included in the sample = 81 (number of participants = 34)

NA boluses for which PAS could not be visualized

95% CI 0.39–0.78), with an optimal cut-point of 24.67 kPa (sensitivity = 0.92, specificity = 0.34; accuracy = 51%).

Lingual Strength and Residue

Oropharyngeal Residue

The presence vs. absence of liquid bolus oropharyngeal residue was not associated with MIP or MSP. However, both MIP ($p < 0.001$, OR 0.99, 95% CI 0.99–1.00) and MSP ($p < 0.001$, OR 0.98, 95% CI 0.98–0.99) were significantly associated with the *amount* of liquid bolus oropharyngeal residue, when residue was present. For every 1 kPa increase in MIP, the odds of worse oropharyngeal residue (when present) decreased by 1% and for every 1 kPa increase in MSP, the odds of worse oropharyngeal residue (when present) decreased by 2%. The presence versus absence of solid bolus oropharyngeal residue, as well as the amount of residue, when present, was not associated with MIP or MSP. See Table 4 for detailed results and Fig. 4 for predicted probabilities.

Hypopharyngeal Residue

The presence of hypopharyngeal residue was not associated with MIP or MSP. However, when thin liquid hypopharyngeal residue was present, both MIP ($p < 0.001$, OR 0.99) and MSP ($p < 0.001$, OR 0.98) were significantly associated with the amount of residue observed. For every 1 kPa increase in MIP, the odds of worse hypopharyngeal residue (when residue was present) decreased by 1% and for every 1 kPa increase in MSP, the odds of worse hypopharyngeal residue (when residue was present) decreased by 2%. MIP was also significantly associated with the amount of solid bolus hypopharyngeal residue observed ($p = 0.006$, OR 0.98), but MSP was not ($p = 1$). See Table 5 for detailed results and Fig. 5 for predicted probabilities.

Discussion

Lingual strength is frequently used in clinical swallowing evaluations to identify values outside of the normative range that may suggest the presence of a swallowing impairment. While recent data suggest that lingual strength is reduced in a subset of patients with PD [25], it is unknown what impact this has on functional swallowing. Understanding the specific contribution of lingual strength on swallowing outcomes is critical for accurate diagnosis of dysphagia and subsequent intervention. The findings of this study suggest a relationship is present between lingual strength and functional swallowing outcomes in people with PD. While this relationship is statistically significant, the relatively small magnitude of effects suggests that lingual strength is one of numerous physiologic factors that may impact functional swallowing. Lingual strength should be considered a component among the multi-factorial deficits that contribute to swallowing impairments in PD. Further, the results of the ROC analyses demonstrate that lingual strength—both MIP and MSP—have reasonable predictive ability to discriminate between those with safe and unsafe swallowing[43], highlighting the potential utility of lingual strength as a screening tool for persons with PD and suspected dysphagia.

Lingual Strength and Functional Swallowing—Airway Invasion and Residue

Maximal Isometric Press (MIP) was reduced in 40% of participants in our sample when compared to weighted normative data on healthy older adults [42]. While there has historically been discrepancy in the literature regarding

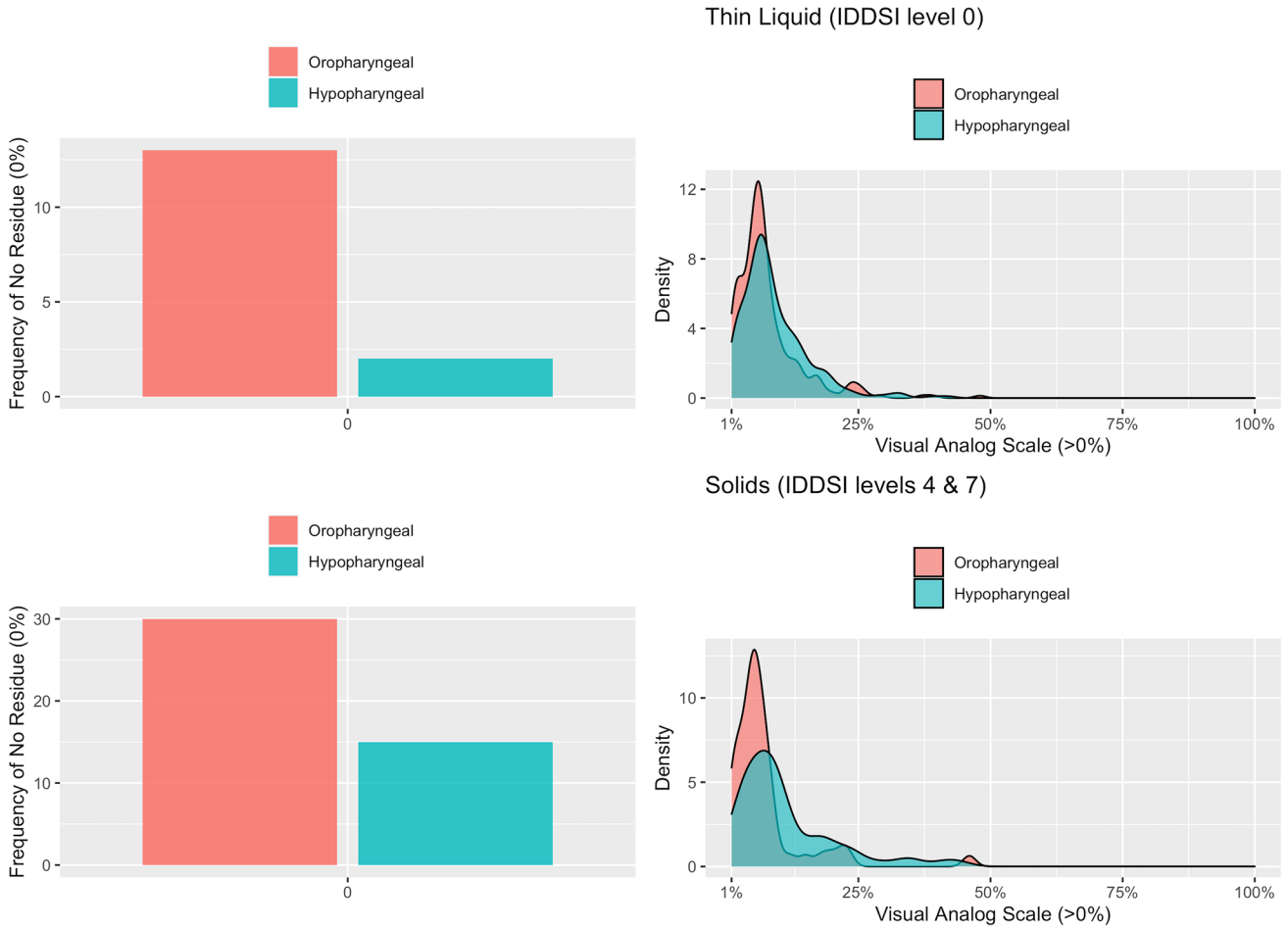

Fig. 1 Density plots of residue quantity

Table 3 Model Results: the relationship between lingual strength and airway invasion

Model	Fixed effects	Std. Error	*Z*-value	*p*-value	Effect Size	95% confidence interval	Participant random effect (SD)
Liquids[a]	MIP	0.021	−3.508	0.002*	0.93	0.89–0.97	1.45
Liquids[a]	MSP	0.037	−3.603	0.0009*	0.88	0.82–0.94	1.39
Solids[b]	MIP	0.119	−0.0901	1.00	0.99	0.78–1.25	8.42
Solids[b]	MSP	0.1864	0.0410	0.967	1.01	0.70–1.45	8.45

Effect sizes are odds ratios, calculated by exponentiating the estimates

MIP maximal isometric press, *MSP* mean swallowing pressure

All *p*-values are adjusted p-values, adjusted for four comparisons, with an "*" indicating a statistically significant finding

[a]277 bolus trials

[b]80 bolus trials

whether lingual strength is reduced in PD [22–24, 26–29], our findings are similar to recent meta-analysis results which revealed reduced lingual strength in approximately one third of individuals in a cohort of 96 persons with PD [25] and other recent studies which have reported reduced maximal lingual strength in a portion of individuals with PD [26, 28, 29]. Of note, the average MIP of persons with PD in the present study (45.48 kPa) is similar to recent reports of MIP in PD ranging from 47.97 to 49.9 kPa [25, 26, 28]), and reduced compared to the average MIP of older adults ranging

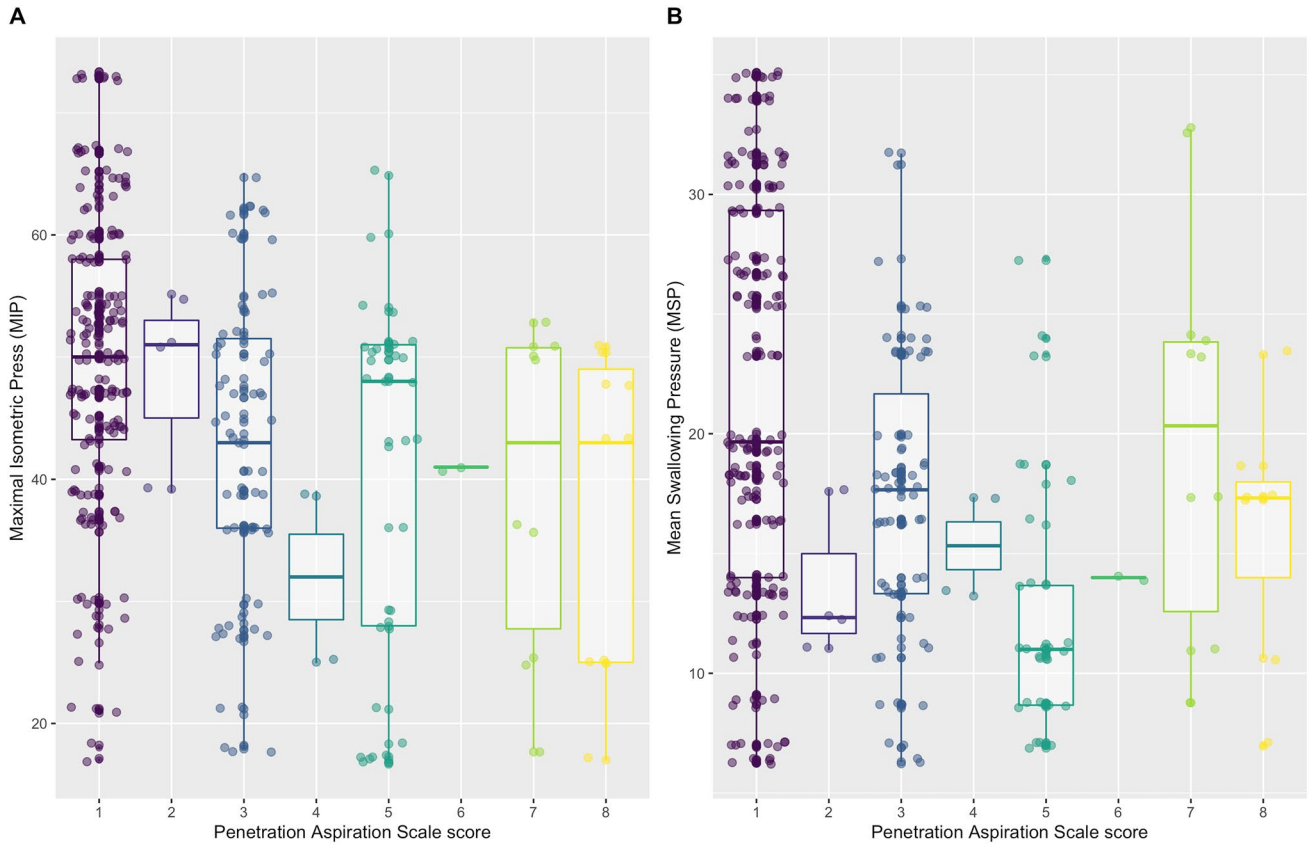

Fig. 2 The relationship between lingual strength and airway invasion

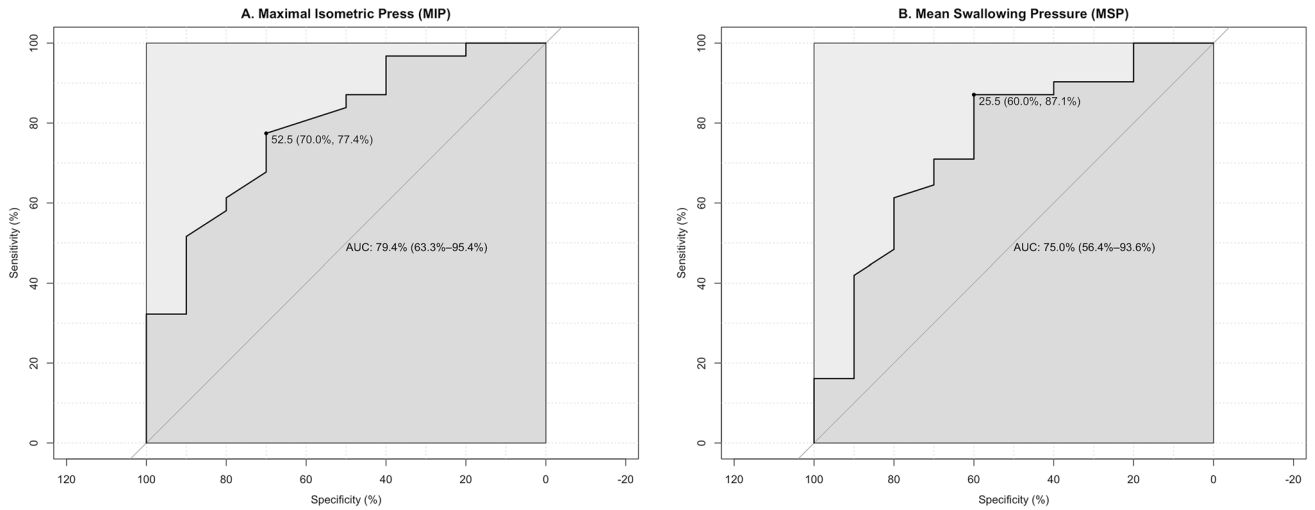

Fig. 3 Sensitivity and specificity of lingual strength to differentiate between those with and without dysphagia

from 54.5 to 57.7 kPal [25, 42]). Of note, our findings differ from a recent publication by Gandhi et al. (2022) who identified the mean MIP among 20 persons with PD to be 54.7 kPa [24]. This finding appears to be at the upper end of MIP values typically seen in PD and may be partly attributed to shorter disease duration (an average of five years since symptom onset) as compared to our sample which is characterized by an average disease duration of eight years. In sum, the reduced MIP in 40% of participants in the present study add to a growing literature to suggest that MIP

Table 4 The relationship between lingual strength and oropharyngeal residue

Model	Std. Error	*t* value	*p*-value	Effect size	95% confidence interval
MIP: Thin Liquid Residue[a]					
Beta (Intercept)	0.0995	−19.346	0.000	0.1459	0.12–0.18
Beta (MIP)	0.0021	−4.301	0.000*	0.9910	0.99–1.00
Zero-inflated (Intercept)	2.3500	−1.788	0.075	0.0150	0.00–1.53
Zero-inflated (MIP)	0. 0515	−0.311	1.000	0.9841	0.89–1.09
MSP: Thin Liquid Residue[a]					
Beta (Intercept)	0.0723	−28.342	0.000	0.1289	0.11–0.15
Beta (MSP)	0.0036	−4.322	0.000*	0.9846	0.98–0.99
Zero-inflated (Intercept)	3.0076	−2.613	0.010	0.0003	0.00–0.14
Zero-inflated (MSP)	0.1166	0.900	1.000	1.1106	0.88–1.40
MIP: Solid Bolus Residue[b]					
Beta (Intercept)	0.2517	−6.100	0.000	0.2153	0.13–0.36
Beta (MIP)	0.0055	−2.331	0.148	0.9873	0.98–1.00
Zero-inflated (Intercept)	1.0448	−0.235	0.815	0.7823	0.09–6.45
Zero-inflated (MIP)	0.0241	−1.365	0.900	0.9677	0.92–1.02
MSP: Solid Bolus Residue[b]					
Beta (Intercept)	0.1950	−11.772	0.000	0.1007	0.07–0.15
Beta (MSP)	0.0089	1.086	1.000	1.0097	0.99–1.03
Zero-inflated (Intercept)	0.7798	−1.368	0.179	0.3441	0.07–1.66
Zero-inflated (MSP)	0.0387	−0.812	1.000	0.9691	0.90–1.05

The zero-inflated portion of the model represents the odds of having residue (i.e., VAS > 0). The beta portion of the model indicates the odds of having more severe residue, when residue is present (i.e., VAS > 0)

Effect sizes are odds ratios, calculated by exponentiating the estimates. All *p*-values are adjusted *p*-values, adjusted for 8 comparisons within oropharyngeal residue models, with an "*" indicating a statistically significant finding after adjusting for multiple comparisons

[a]279 bolus trials

[b]81 bolus trials

is reduced a portion of individuals with PD. Reduced MIP may be related to sarcopenia—age-related decline in muscle strength and function [44, 45], which has been reported in up to 55% of individuals with PD [44, 46]. Sarcopenia has also been related to dysphagia [45, 47] and may contribute to the relationship between reduced lingual strength and dysphagia identified in the present study. The reduction in lingual strength in the present study is likely due to a confluence of factors, including age and disease-specific factors. Disentangling these mechanisms will require further study.

MSP was also reduced in 35.7% of participants in our sample when compared to recently published data reporting a mean MSP of 23.9 kPa in healthy older adults [24]. In fact, the mean MSP in our sample (18.91 kPa) is similar to a recent report of MSP in PD (17.3 kPa) [24]. Despite the similar MSP values, recent publications have found no difference in swallowing pressure between those with PD and healthy controls [24, 26] and no deficit in lingual swallowing pressure in persons with PD and dysphagia [30]. However, findings from our sample suggest that MSP is reduced in a portion of individuals with PD when compared to normative data, and the clinical implications of this should be considered. Given the higher prevalence of reduced MIP as compared to reduced MSP, some participants in the present study were able to recruit sufficient strength to achieve normal pressure during a saliva swallow (a functional, submaximal task), despite reduced MIP. It is possible that these participants utilized a greater percentage of their functional reserve to recruit sufficient strength necessary for the swallowing pressure task despite their reduced MIP. Functional reserve is defined as the difference between MIP and MSP [48]. Some studies have identified a similar pattern of preserved swallowing pressures (MSP) despite reduced MIP in healthy older adults [49, 50], while other studies have not found a reduction in functional reserve in healthy older adults [48, 51]. Regardless, given the importance of sufficient functional reserve during times of physiological stress [49], a reduction in functional reserve may have implications for swallowing safety. Reduced functional reserve—due to a reduction in MIP—has been suggested to increase the risk of dysphagia [50].

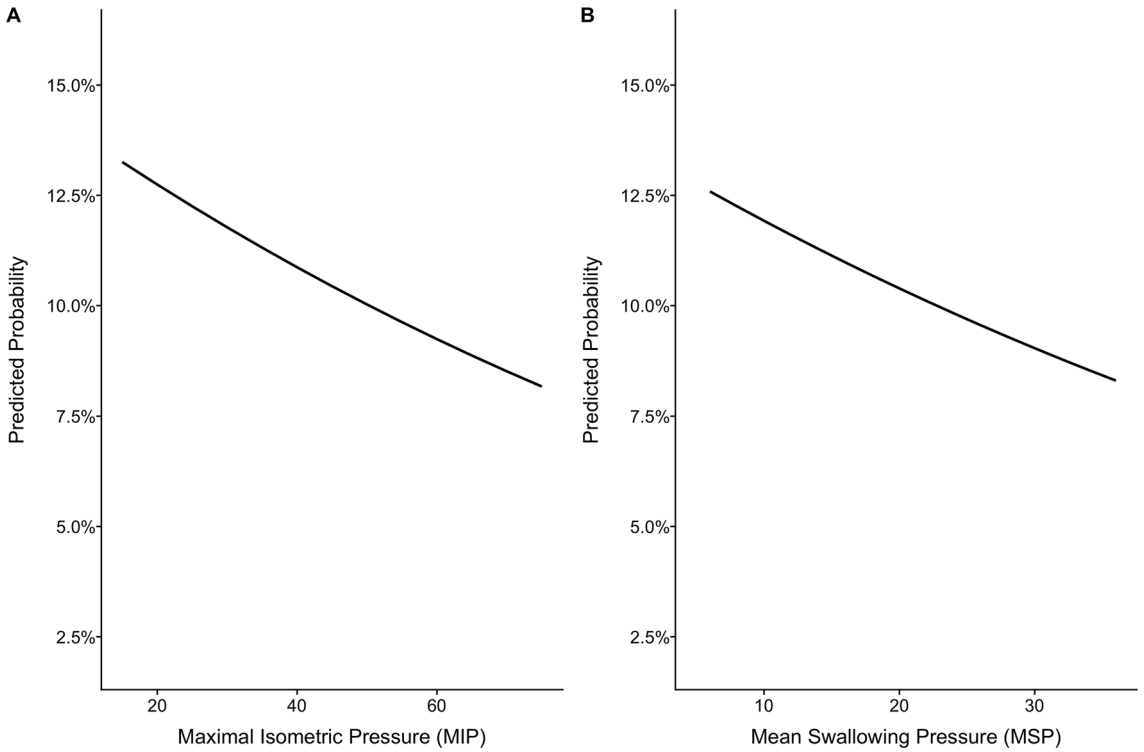

Fig. 4 Predicted probabilities of oropharyngeal residue as a function of lingual strength. *Predicted probabilities calculated for significant portions of the model only. All predicted probabilities are calculated from the beta component of the model, representing the odds of having more severe residue, when residue is present (i.e., VAS > 0). **A** predicted probability of a decrease in thin liquid residue, given an increase in MIP. **B** predicted probability of a decrease in thin liquid residue, given an increase in MSP

Indeed, in the present study, reduced MIP was significantly associated with airway invasion of thin liquids. This confirms previous reports that reduced MIP was correlated with self-reported swallowing complaints in individuals with PD [26, 28, 29]. This finding also replicates the relationship between airway invasion and lingual strength, specifically MIP, a finding which has been recently identified across a variety of other populations [14]. Using a sensitive and nuanced analysis approach was essential to adequately identifying whether a relationship exists between lingual strength and swallowing function in PD. It has been demonstrated that FEES may possess greater sensitivity for identifying and characterizing airway invasion and pharyngeal residue, as compared to videofluoroscopy [31–34]. Moreover, the use of a standardized FEES analysis approach—VASES [37]—provided a method for sensitively identifying outcomes (i.e., airway invasion and residue) of functional swallowing impairments via FEES in the present study. Given the significant relationship identified between MIP and airway invasion, MIP should be considered—among other tools—as an important component of dysphagia evaluation for individuals with PD.

Additionally, to address deficits of lingual strength, lingual strengthening paradigms focused on improving functional reserve have been suggested to preserve swallowing function, particularly in the presence of a neurodegenerative illness such as PD [49]. The efficacy of lingual strengthening to improve swallowing safety and/or efficiency has not been established in PD and has yielded mixed results in other neurologic populations [18]. Nonetheless, lingual strengthening treatment paradigms have resulted in improvements to lingual strength, with treatment gains of 9–10 kPa in some patients [52, 53] and with potential to improve swallowing safety [18]. Based on the findings of the present study, increasing someone's MIP by 10 kPa would reduce the odds of thin liquid airway invasion by 70%. Thus, despite the relatively small magnitude of effects in the present study, when considering the potential for robust treatment gains, the effect may have clinical significance. Future studies should explore the utility and efficacy of lingual strengthening treatments for individuals with PD and dysphagia.

We also identified a significant relationship between MSP and airway invasion of thin liquids in the present study. This finding confirms the relationship identified

Table 5 The relationship between lingual strength and hypopharyngeal residue

Model	Std. Error	*t* value	*p*-value	Effect size	95% Confidence interval
MIP: Thin Liquid Residue[a]					
Beta (intercept)	0.1062	17.142	0.000	0.1618	0.13–0.20
Beta (MIP)	0.0023	−6.621	0.000*	0.9850	0.98–0.99
Zero-inflated (Intercept)	1.1453	−2.947	0.004	0.0342	0.00–0.33
Zero-inflated (MIP)	0.0239	0.166	1.000	1.0040	0.96–1.05
MSP: Thin Liquid Residue[a]					
Beta (Intercept)	0.0756	28.553	0.000	0.1154	0.10–0.13
Beta (MSP)	0.0038	−4.999	0.000*	0.9812	0.97–0.99
Zero-inflated (Intercept)	0.7553	−4.925	0.000	0.0242	0.01–0.11
Zero-inflated (MSP)	0.0321	0.838	1.000	1.0273	0.96–1.09
MIP: Solid Bolus Residue[b]					
Beta (Intercept)	0.2439	−7.072	0.000	0.1782	0.11–0.29
Beta (MIP)	0.0056	−3.554	0.006*	0.9805	0.97–0.99
Zero-inflated (Intercept)	1.0037	−0.762	0.451	0.4653	0.06–3.55
Zero-inflated (MIP)	0.0218	0.071	1.000	1.0016	0.96–1.05
MSP: Solid Bolus Residue[b]					
Beta (Intercept)	0.2156	11.649	0.000	0.0811	0.05–0.13
Beta (MSP)	0.0099	−0.389	1.000	0.9961	0.98–1.02
Zero-inflated (Intercept)	0.7622	0.179	0.859	1.1463	0.24–5.37
Zero-inflated (MSP)	0.0360	−1.196	0.952	0.9579	0.89–1.03

The zero-inflated portion of the model represents the odds of having residue (i.e., VAS > 0). The beta portion of the model indicates the odds of having more severe residue, when residue is present (i.e., VAS > 0)

Effect sizes are odds ratios, calculated by exponentiating the estimates. All *p*-values are adjusted *p*-values, adjusted for 8 comparisons within hypopharyngeal residue models, with an "*" indicating a statistically significant finding after adjusting for multiple comparisons

[a]277 bolus trials

[b]81 bolus trials

between swallowing pressure and self-reported dysphagia in persons with PD [29] but differs from a study by Fukuoka et al. (2019) which did not identify a relationship between swallowing pressure and dysphagia in persons with PD[30]. There were numerous differences in methodology between these studies. Fukuoka et al. (2019) used a sensor sheet inserted into the oral cavity (instead of the IOPI), measured barium bolus swallows (instead of saliva swallows), used videofluoroscopy (instead of FEES), and lastly, patients were grouped into dysphagic vs non-dysphagic groups based on airway invasion or residue amount (instead of using continuous data). These classifications may have reduced the ability to identify the presence of a relationship. Our finding also differs from a recent analysis of a diverse sample of over 400 patients, in which no relationship was found between MSP and PAS scores; but a significant relationship was identified between MIP and PAS scores [14]. While Curtis et al.'s (2021) sample included 51 participants with neurologic diagnoses, it is not specified how many participants had PD. Therefore, these findings may not be indicative of persons with PD. Further, this study also utilized videofluoroscopy and the present study's utilization of FFES may have allowed for increased sensitivity in detecting airway invasion and residue [33–36]. In summary, the present study adds to the literature to suggest that, while MSP may only be impaired in a small sub-set of people with PD, when it is impaired, may suggest the presence of a functional swallowing impairment.

Moreover, ROC analysis suggests that both MIP and MSP may be used to differentiate between safe (PAS 1–2) and unsafe (PAS 3–8) swallowing in persons with PD. The MIP cut-off score that optimized sensitivity and specificity was 52.5 kPa. Seventy-seven percent of participants who penetrated or aspirated demonstrated a MIP equal to or less than 52.5 kPa (sensitivity—the proportion of positive cases identified correctly). Seventy percent of participants who did

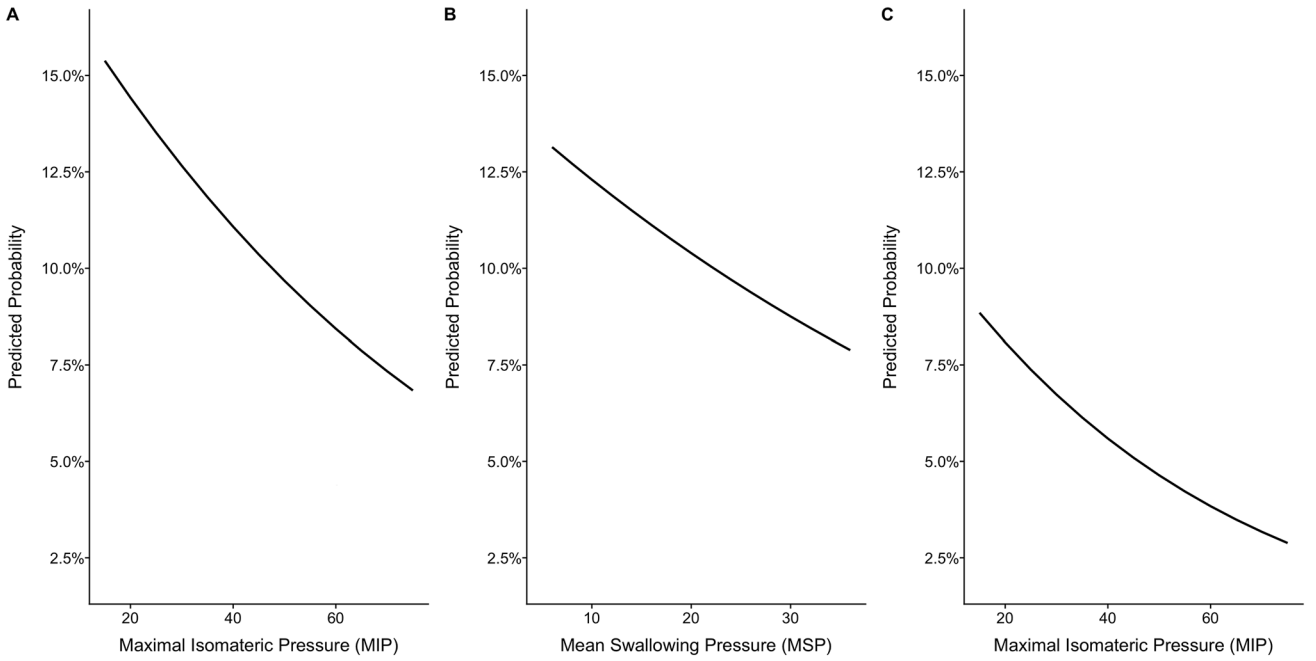

Fig. 5 Predicted probabilities of hypopharyngeal residue as a function of lingual strength. *Predicted probabilities are calculated for significant portions of the model only. All predicted probabilities are calculated from the beta component of the model, representing the odds of having more severe residue, when residue is present (i.e., VAS > 0). **A** predicted probability of a decrease in thin liquid residue, given an increase in MIP. **B** predicted probability of a decrease in thin liquid residue, given an increase in MSP. **C** predicted probability of a decrease in solid bolus residue given an increase in MIP

not penetrate or aspirate demonstrated a MIP above 52.5 kPa (specificity—the proportion of negative cases identified correctly). The AUC value of 0.79 suggests that a MIP of less than 52.5 kPa would accurately predict dysphagia 79% of the time [43]. The MSP cut-off score that best discriminated between safe and unsafe swallowing was 25.5 kPa. Eighty-seven percent of participants who penetrated or aspirated demonstrated an MSP equal to or less than 25.5 kPa (sensitivity—the proportion of positive cases identified correctly). Sixty percent of participants who did not penetrate or aspirate demonstrated an MSP above 25.5 kPa (specificity—the proportion of negative cases identified correctly). The AUC value of 0.75 suggests that an MSP less than 22.5 kPa accurately predicts dysphagia 75% of the time [43]. These findings suggest the utility of obtaining MIP and/or MSP, and using the above-described optimal cut-offs, to screen for dysphagia in persons with PD. This analysis should be replicated in larger sample sizes to ensure it maintains its predictive value.

Interestingly, neither MIP nor MSP significantly influenced airway invasion of solid boluses. Given the tongue's role in mastication and bolus propulsion, this finding was unexpected. Previous studies have identified that thicker consistencies require greater tongue-palate pressures [13, 54] and increased tongue pumping has been observed in patients with PD when swallowing thicker (i.e., pudding) boluses [55]. It has been reported that solid bolus airway invasion occurs less frequently than thin liquid airway invasion and is not a prevalent finding in PD [55–57]. In fact, in the present study, of participants who received solid bolus trials, 91% did not exhibit any deficits in swallowing safety (i.e., worst PAS score of 1). This finding may also be due to fewer solid bolus trials included in swallowing assessments. All three of the participants who exhibited aspiration on solid boluses also exhibited pharyngeal residue and aspiration on thin liquids. Therefore, we hypothesize that these participants likely had worse dysphagia overall, due to multifactorial deficits, and thus, lingual strength was not a significant predictor variable. Future research involving more solid bolus trials across PD severity levels is needed to elicit a wider distribution of PAS scores and determine the role of lingual strength in solid bolus consumption in PD.

Lingual strength was not related to the presence/absence of oro- or hypo-pharyngeal residue. This may be partly

explained by the high prevalence of small amounts of residue identified in this study and supports previous studies in PD that have identified trace to small quantities of vallecular and piriform sinus residue in most swallows in persons with PD [58]. Further, trace to small quantities of residue are not expected to contribute to swallowing safety impairments [59]. However, lingual strength—both MIP and MSP—was significantly related to the *amount of residue* observed, when thin liquid residue was present. Greater quantity of thin liquid residue in both the oropharynx and hypopharynx was associated with lower MIP and lower MSP, supporting the importance of lingual strength for bolus propulsion through the pharynx [7]. However, the magnitude of this effect was extremely small—with the odds of worse pharyngeal residue decreasing by 1% for every 1 kPa increase in MIP and decreasing by 2% for every 1 kPa increase in MSP. Given the multifactorial nature of dysphagia in PD, it is likely that functional swallowing impairments result from deficits across multiple systems (i.e., respiratory, laryngeal) and that a confluence of swallowing parameters are driving the presence of pharyngeal residue and airway invasion in this population. Thus, future research evaluating a variety of screening and evaluation tools that assess multiple components of airway protective function is needed to further understand how lingual strength interacts with other impaired systems and contributes to functional swallowing impairments.

Limitations

Due to the retrospective design of this study, there was variability in the number of boluses consumed by each participant in our sample and not all participants trialed each bolus volume and consistency. It is possible that variability in number of boluses resulted in different opportunities for airway invasion to occur for participants during the FEES. Therefore, future prospective well-controlled studies are needed to replicate the findings of the present study. Further, our sample was characterized by overall mild dysphagia, with only 29% of participants who received thin liquid boluses exhibiting a maximum PAS score of six, seven, or eight (i.e., aspiration) on thin liquids and only 8.8% of participants who received solid boluses aspirating on solids. The relatively small amount of residue identified further highlights the mild nature of dysphagia across the sample. It is also important to note that residue was measured after the final clearing swallow and the number of swallows required to clear the bolus was not included as an outcome. Therefore, the functional deficits in patients for whom multiple swallows were required to clear pharyngeal residue may not have been captured. Additionally, assessments of lingual strength occurred in the context of a standardized protocol where MIP was performed before MSP. It is important to note that completing MIP (a maximal task) prior to MSP (a submaximal task) may alter the swallow and could potentially inflate MSP values. Future prospective studies may consider counterbalancing these tasks.

Conclusion

This study revealed that reduced lingual strength was associated with worse penetration, aspiration, and pharyngeal residue in PD. This has important implications for assessment and management of dysphagia, suggesting that when lingual strength is reduced, it may represent the presence of a functional swallowing impairment. Further, findings suggest that the relationship between lingual strength and thin liquid airway invasion may be identified via maximal isometric lingual pressure (MIP) and/or mean swallowing pressure (MSP), in individuals with PD. Both MIP and MSP also demonstrated adequate discriminant ability to differentiate those with safe (PAS 1–2) vs unsafe (PAS 3–8) swallowing. These data provide support for utilizing lingual strength—MIP and/or MSP—as a component of a screening or assessment to identify possible risk of a functional swallowing impairment (i.e., thin liquid airway invasion) in individuals with PD. However, the magnitude of effects across models were generally small. Together, these findings suggest that while lingual strength plays a significant role, functional swallowing impairments of both safety and efficiency in individuals with PD are likely influenced by a multitude of contributing deficits across the swallow mechanism. To this end, future work should explore other components of swallowing function which may be key drivers of swallowing safety and efficiency. This will inform what unique combination of screening and/or evaluation tools best predict functional swallowing outcomes in PD and support the development of targeted treatment paradigms. Future work may also explore the utility of lingual strengthening as a treatment approach for improving swallowing safety and efficiency in individuals with PD.

Appendix

See Table 6

Table 6 The number of boluses administered to each participant

ID	Thin liquid boluses								Solid boluses			
	5 ml		20 ml		10 ml		90 ml		Pudding		Cracker[a]	
1	1		1		3		3		0		1	
2	1		1		3		3		2		0	
3	1		1		3		3		2		1	
4	1		1		3		3		2		1	
5	1		0		3		3		–		–	
6	1		1		3		2		1		1	
7	1		1		3		3		2		1	
8	0		0		2		2		–		–	
9	0		0		1		1		2		1	
10	0		0		3		3		–		–	
11	1		1		3		1		2		0	
12	1		1		3		3		2		1	
13	1		1		3		3		–		–	
14	1		0		3		0		1		0	
15	1		1		2		2		–		–	
16	1		1		3		3		–		–	
17	–		–		–		–		1		2[1]	
18	1		1		2		1		0		1	
19	1		1		3		3		2		1	
20	1		1		3		0		2		1	
21	1		1		3		3		2		1	
22	1		1		3		2		2		1	
23	1		1		2		1		2		0	
24	1		0		0		1		1		1	
25	1		1		3		0		2		1	
26	1		1		3		3		0		1	
27	1		1		3		3		2		1	
28	1		1		3		1		2		0	
29	1		1		3		3		2		1	
30	1		0		3		3		2		1	
31	1		1		3		3		1		0	
32	1		1		2		1		1		1	
33	1		0		3		3		2		0	
34	1		1		3		3		0		1	
35	1		1		3		3		2		1	
36	1		1		3		3		2		1	
37	1		1		3		3		2		1	
38	1		1		3		2		–		–	
39	1		1		3		3		–		–	
40	1		1		3		3		2		1	
41	1		1		3		3		2		1	
42	1		1		3		3		1		1	
Total *n*	5 ml 1 trial	*n* = 38 *n* = 38	20 ml 1 trial	*n* = 33 *n* = 33	10 ml 3 trials 2 trials 1 trial	*n* = 40 *n* = 34 *n* = 5 *n* = 1	90 ml 3 trials 2 trials 1 trial	*n* = 38 *n* = 26 *n* = 5 *n* = 7	Pudding 2 trials 1 trial	*n* = 30 *n* = 23 *n* = 7	Cracker 2 trials 1 trial	*n* = 27 *n* = 1 *n* = 27

Total number of thin liquid boluses per participant, mean (SD) [range]
6.8 (1.68) [2–8]
Total number in protocol: 8

Total number of solid boluses per participant, mean (SD) [range]
2.35 (0.77) [1–3]
Total number in protocol: 3

[a]One saltine cracker was administered and PAS and residue ratings were made following consumption of the full cracker. Participant 17 received 1 saltine cracker, however, first consumed one third of the cracker, following which PAS and residue ratings were made (bolus 1), and subsequently took an additional bite of the cracker (bolus 2)

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