



# Biomechanical Biomarkers of Tongue Impairment During Swallowing in Persons Diagnosed with Amyotrophic Lateral Sclerosis

Bridget J. Perry<sup>1</sup> · Kaila L. Stipancic<sup>2</sup> · Rosemary Martino<sup>3,4,5</sup> · Emily K. Plowman<sup>6,7</sup> · Jordan R. Green<sup>8,9</sup>

Received: 2 August 2019 / Accepted: 15 April 2020 / Published online: 28 April 2020  
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## Abstract

**Background** The impact of tongue dysfunction on deglutition in persons diagnosed with amyotrophic lateral sclerosis (ALS) is not well understood. This information is needed to improve our understanding of the mechanisms of swallowing impairment, for identifying risk factors of dysphagia, and for establishing impairment-specific treatments aimed at slowing the loss of swallow function.

**Objectives** The goals of this study were to determine the relation between biomechanical measures of oral tongue movements using electromagnetic articulography (EMA) and measures of swallow physiology, swallow safety and efficiency, and self-reported swallowing function.

**Methods** Participants were diagnosed with ALS by a neurologist following the El Escorial Criteria from the World Federation of Neurology. Twelve participants underwent (1) EMA to derive biomechanical measures of the tongue, (2) videofluoroscopic evaluation to measure swallow physiology, safety, and efficiency, and (3) maximal tongue strength testing using the Iowa Oral Pressure Instrument (IOPI). Participants completed self-reported functional assessments. Spearman's rank correlations assessed for associations between lingual biomechanics and swallowing physiology, swallow safety and efficiency, and self-reported bulbar function.

**Results** Results demonstrated strong associations between biomechanical and swallowing physiology, swallow safety, and self-reported measures. Notably, swallowing safety during thin liquid intake was associated with tongue speed ( $r = -0.7$ ,  $p < 0.05$ ) and range of motion ( $r = -0.71$ ,  $p < 0.05$ ), and swallowing safety during puree intake was associated with tongue strength ( $r = -0.69$ ,  $p < 0.05$ ).

**Conclusions** Our findings underscore the importance of tongue movements on swallowing physiology and safety, help improve our understanding of mechanisms of swallowing impairment, and highlight a potential clinical tool to index bulbar impairment.

**Keywords** Deglutition · Deglutition disorders · Amyotrophic lateral sclerosis · Kinematics · Electromagnetic articulography · Dysphagia · Lingual impairment

✉ Jordan R. Green  
jgreen2@mghihp.edu

<sup>1</sup> Tufts University Medical Center, 35 Kneeland Street, Boston, MA 02111, USA

<sup>2</sup> MGH Institute of Health Professions, 79/96 13th Street, Charlestown, MA 02129, USA

<sup>3</sup> Department of Speech-Language Pathology, University of Toronto, Toronto, ON, Canada

<sup>4</sup> Department of Otolaryngology-Head and Neck Surgery, Rehabilitation Sciences Institute, University of Toronto, Toronto, ON, Canada

<sup>5</sup> Krembil Research Institute, University Health Network, 160-500 University Ave., Toronto, ON M5G 1V7, Canada

<sup>6</sup> Aerodigestive Research Core, University of Florida, Gainesville, USA

<sup>7</sup> Department of Speech-Language Hearing Sciences and Neurology, University of Florida, Gainesville, FL 32610, USA

<sup>8</sup> Communication Sciences & Disorders, MGH Institute of Health Professions, 79/96 13th Street, Charlestown, MA 02129, USA

<sup>9</sup> Speech & Hearing Bioscience and Technology Program, Harvard Medical School, 79/96 13th Street, Charlestown, MA 02129, USA

## Introduction

Difficulty swallowing, caused by weakness and/or spasticity of the muscles of the oropharyngeal region is one of the most severe and debilitating symptoms of amyotrophic lateral sclerosis (ALS) [1]. Dysphagia-related complications, including malnutrition and aspiration pneumonia, increase the risk of death by almost eight fold [2]. Among oropharyngeal structures, the tongue has been reported to be disproportionately affected by the disease [3]. Although the tongue plays a critical role during swallowing [4], the mechanisms of tongue motor impairment contributing to dysphagia are not well understood. This information is needed to improve our understanding of the contribution of tongue impairment on swallowing function, to identify risk factors for dysphagia, and to establish impairment-specific treatments aimed at slowing the loss of swallow function in persons diagnosed with ALS.

Most of the literature on tongue impairment and swallowing is focused on the relation between tongue strength and swallow function [5–8]. Reduced tongue strength has been associated with a variety of oral and pharyngeal swallowing impairments [5, 7–9]; however, across studies, the effect of tongue strengthening exercises on swallowing has been highly variable [10–13]. Further, given that swallowing is a sub-maximal task, the role of strength training in dysphagia rehabilitation has been recently questioned with the suggestion that skill-based approaches (focusing on timing and coordination of specific movement patterns) in dysphagia rehabilitation may be more appropriated [14].

Research exploring the relation between movement-based measures of the tongue, including tongue range of motion and tongue speed, and swallowing function in persons with dysphagia is limited [15–17] in large part due to significant measurement challenges [18]. Using frame-by-frame digital point marking on the gold standard of swallowing assessment, videofluoroscopic assessment of swallowing (VFSS), two studies found no association between movement-based measures of the tongue and swallowing [16, 17]. However, also using frame-by-frame point marking analyses with VFSS, Kawai et al. found that for persons diagnosed with ALS, reduced movement-based measures of the posterior tongue were associated with increased swallowing impairments, including reduced oral transport and control [15].

Point tracking methods of quantifying tongue movement, such as electromagnetic articulography (EMA), have been used to quantify biomechanical features of oral tongue movements during swallowing in healthy populations [18–21] and more recently in persons diagnosed with ALS [22]. To our knowledge, however, no studies have investigated the relation between measures of swallowing

function and movement-based biomechanical measures of the oral tongue during swallowing (range of motion, speed, and coordination) using point-tracking methodologies.

The goals of this study were to first determine the association between biomechanical measures of oral tongue movements using EMA and oral tongue movements as measured by VFSS during swallowing and oral tongue strength in persons diagnosed with ALS. Secondly, we examined the association between biomechanical measures of the oral tongue during swallowing with measures of swallowing safety and efficiency on VFSS and patient perception of swallow dysfunction. We expect our findings to help improve our understanding of the impact of lingual impairment on swallow physiology and function.

## Methods

### Study Participants

Participants were recruited from the Massachusetts General Hospital (Mass General) ALS Multidisciplinary Clinic diagnosed with ALS by a neurologist following the criteria defined by the El Escorial Criteria from the World Federation of Neurology [23] who did not have a history of other neurological impairment. All participants included were recommended to undergo a VFSS for clinical management of dysphagia by a speech-language pathologist at Mass General.

### Electromagnetic Articulography

An electromagnetic tracking device (NDI Wave; Northern Digital, Inc., Shelburne, VT) was used to record swallowing movements from the lips, jaw, and tongue in a calibrated volume ( $30 \times 30 \times 30 \text{ cm}^3$ ). One 6-degree-of-freedom sensor was placed on the forehead. Five-degree-of-freedom sensors were placed on the upper lip, lower lip, center of the jaw, right corner of the jaw, and on the tongue. Tongue sensors were attached at midline at approximately 1 cm posterior to the tongue tip (T1) and at approximately 4 cm posterior to the tongue tip (T2) using a non-toxic dental glue, PeriAcryl Oral Tissue Adhesive (GluStitch Inc.). The 3D tongue and jaw data were re-expressed relative to a head-based coordinate system using the NDI system default settings.

With EMA sensors in place, participants were asked to swallow the following bolus consistencies in this order:

### 5-ml Teaspoon of Thin Liquids (1 Trial)

Participants were provided with 5-ml of thin liquid water via teaspoon and were instructed to hold the bolus in their

mouth until asked to swallow. This task only served to orient participants to the tasks.

### 5-ml Controlled Cup Sips of Thin Liquid (2 Trials)

Participants were handed a cup of thin liquid water and instructed to hold the water in their mouth until asked to swallow. If participants were unable to hold the cup secondary to arm/hand weakness, the cup was held by the clinician and cup sips were administered with clinician assistance.

### 1-tsp Puree (2 Trials)

Participants were given a teaspoon of applesauce and instructed to hold it in their mouth until asked to swallow.

### EMA Measures

A MATLAB-based program, SMASH [24], was used to analyze vertical measures of tongue movements relative to the facial plane during swallowing. A low-pass filter at 10 Hz was applied to all signals to remove high-frequency noise. Each signal was manually checked for missing data and movement artifacts. To extract tongue movement that was partially independent from that of the underlying jaw, tongue movement data were represented as the 3D Euclidian distance between the posterior tongue sensor and the jaw right sensor. Range of movement was defined as the difference between the maximum and minimum values of the vertical distance trace for the posterior tongue sensor [22, 25]. Maximum speed of movement was calculated as the maximum absolute value of the first derivative of the vertical posterior tongue distance trace [22, 25].

### Tongue Strength

Maximum tongue strength was measured in kilopascals (kPa) using the Iowa Oral Performance Instrument [26]. An IOPI pressure bulb was attached to a tongue blade using double-sided tape. The tongue blade was inserted into the oral cavity and placed against the hard palate. A bite block was inserted between the back teeth and participants were instructed to clench their teeth against the bite block prior to pressing their tongue against the pressure bulb in order to stabilize the jaw and avoid jaw contribution to the tongue movement. Participants were then asked to push their tongue against the pressure bulb as hard as they could for 3 s. This task was repeated 3 times. The maximum value derived from the set of 3 trials was used for data analysis.

### Videofluoroscopic Evaluation of Swallowing (VFSS)

VFSS was conducted by a licensed speech-language pathologist. VFSS was conducted with a pulse rate of 30 fps and recorded at a frame rate of 30 fps using a TIMS DICOM system (TIMS Medical, Chelmsford, MA). Participants were asked to swallow one 5-ml teaspoon of Varibar thin liquid barium, one 5-ml controlled cup sip of Varibar thin liquid barium, one teaspoon of Varibar Pudding, and varying quantities and consistencies of Varibar barium needed by the clinician in order to gather clinically important information about the swallow for her purposes. During controlled swallow trials, participants were instructed to hold the bolus in their mouth until asked to swallow.

### VFSS Measures

Two unblinded licensed speech-language pathologists registered in the Modified Barium Swallow Impairment Protocol (MBSImP™) [27] reviewed each VFSS from digital recordings. Scores from the initial thin liquid teaspoon swallow were used to orient participants to the task and were not included in the present study. The following measures were derived for each swallow on the VFSS in accordance with the MBSImP™ protocol for comparison with physiologic measures because they represented measures of lingual function: tongue control during bolus hold (MBSImP™), bolus transport/lingual motion (MBSImP™), initiation of pharyngeal swallow (MBSImP™), tongue base retraction (MBSImP™), oral residue (MBSImP™), and pharyngeal residue (MBSImP™).

In order to assess the relation between biomechanical measures of swallowing and overall oral and pharyngeal swallow function, an Oral Total Sum Score (MBSImP™) and a Pharyngeal Total Sum Score (MBSImP™) were calculated. The Oral Total Sum Score (MBSImP™) was calculated by summing the worst scores from each consistency on tongue control during bolus hold (MBSImP™), bolus transport/lingual motion (MBSImP™), initiation of pharyngeal swallow (MBSImP™), and oral residue (MBSImP™) and could range from 0 indicating no impairment to 15 indicating the most severe impairment. Because a solid bolus was not tested across all participants, MBSImP™ component 3 (bolus preparation/mastication) was not included in the oral total sum score; MBSImP™ component 1 (lip closure) was not included in the oral total sum score since several VFSS's did not capture the anterior portion of the mouth. The Pharyngeal Total Sum Score (MBSImP™) was calculated by summing the worst scores from each consistency on soft palate elevation (MBSImP™), laryngeal elevation (MBSImP™), anterior hyoid excursion (MBSImP™), epiglottic movement (MBSImP™), laryngeal vestibular closure

(MBSImP™), pharyngeal stripping wave (MBSImP™), pharyngoesophageal segment opening (MBSImP™), tongue base retraction (MBSImP™), and pharyngeal residue (MBSImP™). Scores could range from 0 indicating no impairment to 26 indicating the most severe impairment. Because a swallow in the AP view was not administered in all the studies, MBSImP™ component 13 (pharyngeal contraction) was not included in the scoring of the pharyngeal total sum scores. Penetration/Aspiration Scale (PAS) scores were also calculated [28]. PAS scores range from 1 to 8 with 1 indicating no penetration/aspiration and 8 indicating aspiration with no effort to clear from the larynx. A PAS score was derived for each swallow captured on the VFSS. The worst PAS score for each consistency was entered into the analysis.

### Patient Reported Outcome Measures

Several self-reported bulbar function scale scores were collected and compared with biomechanical measures of the tongue. Both total scores and bulbar subsection scores were derived using the ALS Functional Rating Scale-Revised (ALSFRS-R) [29]. ALSFRS-R total scores range from 0 to 48 with 48 indicating no impairment and 0 being most impaired; ALSFRS-R bulbar subscales scores range from 0 to 12 with 12 indicating no impairment and 0 being most impaired. Self-reported symptom-directed dysphagia impairment scores were calculated using the Eating Assessment Tool (EAT-10) [30]. EAT-10 scores range from 0 to 40 with 0–3 indicating normal swallowing and 40 indicating severe impairment. Functional oral intake of food and liquid in persons with dysphagia were captured using the Functional Oral Intake Score (FOIS) [31]. FOIS scores range from 1 to 7 with 1 being most severe and 7 indicating no impairment.

### Scoring Reliability

Because MBSImP™ and PAS scores are subjective and clinician derived, inter-rater reliability for MBSImP™ and PAS scores was calculated using a consensus exact agreement method. Each MBSImP™ component score and each PAS score was compared between the 2 raters for each swallow. In instances where scores did not exactly agree, the same two clinicians reviewed the swallow on VFSS, and differing scores were discussed. If a score could not be agreed upon, a final score was adjudicated by a third rater.

### Data Analysis

Descriptive statistics were used to summarize biomechanical measures of the tongue, measures of tongue physiology during swallowing, and our swallowing outcome measures. Spearman rank correlations were derived between

biomechanical measures of tongue movement (speed, range, and strength), measures of tongue physiology during swallowing (tongue control, bolus transport, initiation of pharyngeal swallow, tongue base retraction, oral impairment, and pharyngeal impairment), swallow safety and efficiency measures (PAS, oral residue, pharyngeal residue), and patient-reported outcome measures (EAT-10, FOIS, ALSFRS-R). To determine the discriminant ability of biomechanical measures of tongue function to detect impairments in swallowing safety, receiver operating characteristic (ROC) curves were created between statistically significant associations of biomechanical measures of tongue function and PAS scores. Area under the curve (AUC), sensitivity, specificity, and optimal cut points were calculated between penetration/aspiration of water and tongue speed and tongue range of motion and between penetration/aspiration of puree and tongue strength. These analyses were conducted with two different binary categories. The first explored the ability to detect penetrators/aspirators ( $PAS \geq 3$ ) vs. non-penetrators/aspirators ( $PAS < 3$ ). The second explored the ability to detect aspirators ( $PAS \geq 6$ ) vs. non-aspirators ( $PAS < 6$ ). Finally, Spearman's Rho correlation coefficients were derived between biomechanical measures of the tongue during swallowing and self-reported measures of bulbar function (ALSFRS-R, EAT-10, FOIS). Statistical tests were two-sided with an alpha of 0.05. Statistical analyses were conducted in RStudio version 1.2.1335 [32].

## Results

### Participant Characteristics

A total of 12 participants diagnosed with ALS between the ages of 50–74, with a mean age of 61 years, were included in this study. Fifty percent of participants were female ( $n = 6$ ). Seven participants had onset of their initial symptoms in the spinal regions and five had initial symptom onset in the bulbar region. All participants had bulbar symptoms at the time of assessment. As calculated using the Speech Intelligibility Test [33], the mean speaking rate was 107 words per minute ( $SD \pm 73.5$ ) and mean speech intelligibility was 73.1% ( $SD \pm 38.6$ ). The mean time from symptom onset to assessment was 386 days ( $SD \pm 350$ ). Mean ALSFRS-R was 34.8 ( $SD \pm 5.6$ ) and mean ALSFRS-R Bulbar subscale score was 7.6 ( $SD \pm 2.4$ ). All participants underwent an assessment of swallowing function via VFSS followed by a data collection session where oral tongue movements during swallowing were assessed using EMA. The average time between VFSS and EMA was 12 days ( $SD \pm 13$  days) (Table 1). Median scores and ranges of biomechanical measures of tongue movement, measures of tongue physiology

**Table 1** Measures of tongue movement, tongue strength, tongue physiology, and patient-reported outcomes

Measure	Consistency	Median (95% CI)
Range of motion (mm)	Thin	11.6 (8.6, 14.7)
	Puree	12 (8.6, 13.6)
Speed (mm/s)	Thin	40.6 (28.2, 53.8)
	Puree	38.4 (28.1, 46.8)
Strength (kPa)	NA	16 (7, 24)
Tongue control during bolus hold	Thin	1.5 (0, 2)
	Puree	1 (0, 2)
Bolus transport/lingual motion	Thin	1 (0, 3)
	Puree	3 (0, 3)
Swallow initiation	Thin	1 (0, 3)
	Puree	0 (0, 0.5)
Tongue base retraction	Thin	1.5 (1, 2)
	Puree	1.5 (1, 2.5)
Oral residue	Thin	1.5 (1, 2)
	Puree	2 (1.5, 2)
Pharyngeal residue	Thin	1.5 (1, 2)
	Puree	1.5 (1, 2)
Oral total score	Thin	5.5 (3, 8.5)
	Puree	6 (2, 7.5)
Pharyngeal total score	Thin	6 (3.5, 9)
	Puree	5 (3, 8.5)
Penetration/aspiration scale score	Thin	3 (2.5, 6)
	Puree	4 (3, 8)
Eating assessment tool-10	NA	15.4 (4.5, 26.7)
Functional oral intake scale	NA	5 (5, 6)
ALS functional rating scale-bulbar	NA	8 (7, 9.5)
ALS functional rating scale-total	NA	36 (32, 39)

during swallowing, measures of swallowing safety and efficiency, and patient-reported outcome measures are detailed in Table 1.

### Tongue Physiology and Biomechanical Measures of the Oral Tongue

With cup sips of water, strong negative correlations were found between tongue speed and tongue control during bolus hold, bolus transport/lingual motion, swallow initiation, and oral impairment score (Table 2). With teaspoons of puree, strong negative correlations were found between tongue speed and bolus transport/lingual motion (Table 2).

Strong negative correlations were found between tongue range of motion and bolus transport/lingual motion, swallow initiation, bolus transport/lingual motion, and tongue base retraction with cup sips of thin liquids (Table 2). With teaspoons of puree, strong negative correlations were found with bolus transport/lingual motion and oral impairment score (Table 2).

Tongue strength was negatively correlated with bolus transport/lingual motion, swallow initiation, and oral impairment score with cup sips of water (Table 2). With teaspoons of puree, tongue strength was negatively correlated with bolus transport/lingual motion and oral impairment score (Table 2).

### Swallowing Safety, Efficiency, and Biomechanical Measures of the Oral Tongue

Tongue speed was negatively correlated with swallowing safety (PAS scores) during cup sips of water (Table 3).

**Table 2** Correlations between biomechanical measures of the tongue (speed, range of motion, strength) and MBSImP™ © measures of tongue physiology

	Speed		Range of motion		Strength	
	Water	Applesauce	Water	Applesauce	Water	Applesauce
Tongue control during bolus hold	<b>-0.77</b>	-0.45	-0.37	-0.42	-0.66	-0.64
Bolus transport/lingual motion	<b>-0.88</b>	<b>-0.73</b>	<b>-0.69</b>	-0.46	<b>-0.69</b>	<b>-0.82</b>
Swallow initiation	<b>-0.71</b>	-0.52	<b>-0.81</b>	-0.1	<b>-0.74</b>	-0.52
Oral impairment score	<b>-0.93</b>	-0.42	<b>-0.81</b>	-0.33	<b>-0.82</b>	<b>-0.74</b>
Tongue base retraction	-0.35	-0.45	<b>-0.69</b>	-0.21	-0.26	-0.56

The bold values denote statistical significance at  $p < 0.05$

**Table 3** Correlations between biomechanical measures of the tongue and measures of swallowing safety and efficiency

	Speed		Range of motion		Strength	
	Water	Applesauce	Water	Applesauce	Water	Applesauce
PAS score	<b>-0.71</b>	-0.5	<b>-0.7</b>	-0.11	-0.61	<b>-0.69</b>
Oral residue	-0.52	-0.47	-0.52	-0.13	-0.35	-0.3
Pharyngeal residue	-0.45	-0.2	<b>-0.78</b>	-0.04	-0.43	-0.4

The bold values denote statistical significance at  $p < 0.05$

Tongue range of motion was negatively correlated with swallowing safety and pharyngeal residue with cup sips of water (Table 3). Tongue strength was negatively associated with swallowing safety with teaspoons of puree (Table 3).

### Self-reported Scales of Bulbar Function and Biomechanical Measures of the Oral Tongue

On self-reported scales of bulbar function, tongue speed was positively correlated with FOIS scores, ALSFRS-Bulbar scores, ALSFRS-Total scores, and negatively correlated with EAT-10 scores during cups sips of thin liquids (Table 4).

**Table 4** Correlations between biomechanical measures of the tongue and self-reported scales of bulbar function

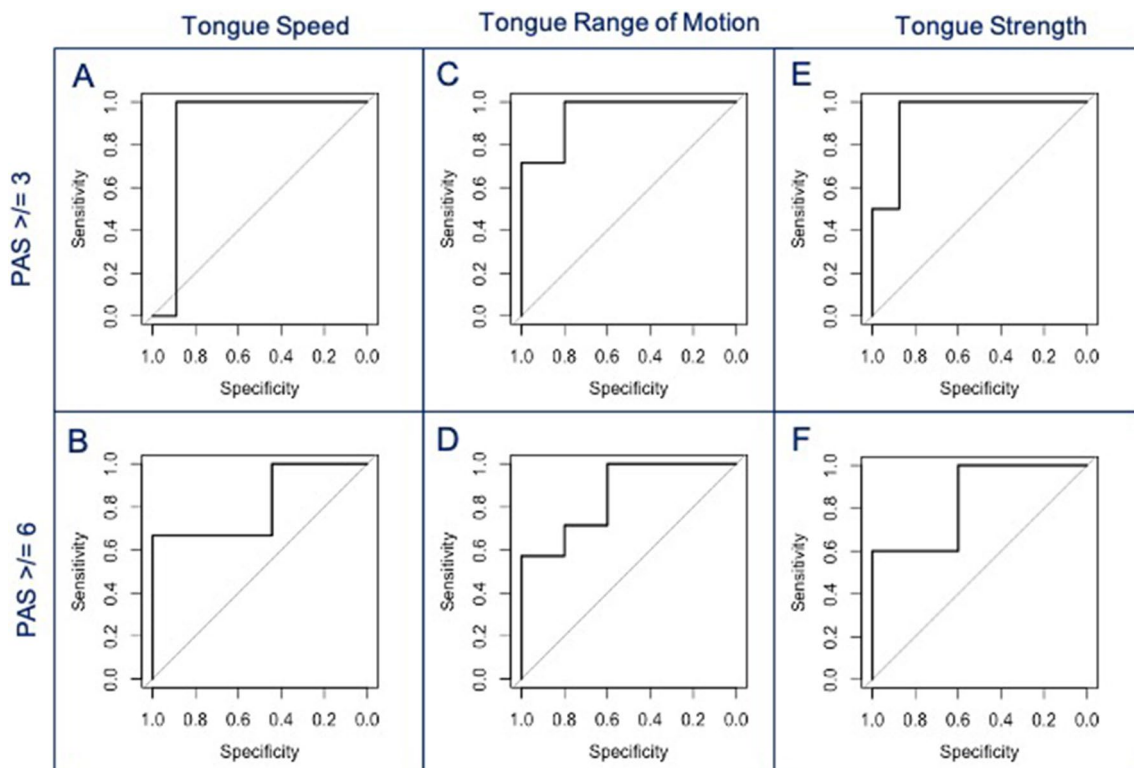
	Speed		Range of motion		Strength
	Water	Applesauce	Water	Applesauce	
FOIS	<b>0.87</b>	<b>0.7</b>	<b>0.81</b>	<b>0.66</b>	<b>0.87</b>
ALSFRS-bulbar	<b>0.68</b>	<b>0.72</b>	0.48	<b>0.78</b>	0.65
ALSFRS-total	<b>0.78</b>	0.41	<b>0.67</b>	0.4	0.65
EAT-b	<b>-0.8</b>	-0.5	-0.57	-0.43	<b>-0.67</b>

The bold values denote statistical significance at  $p < 0.05$

With teaspoons of puree, tongue speed was positively associated with ALSFRS-Bulbar scores and FOIS scores (Table 4). Tongue range of motion was positively correlated with FOIS score during cups sips of thin liquids and with ALSFRS-Bulbar scores with teaspoons of puree (Table 4). Tongue strength was positively correlated with FOIS scores, and negatively correlated with EAT-10 scores (Table 4).

### Discriminant Ability of Biomechanical Measures of Tongue Impairment for Detecting Penetration/Aspiration

Tongue speed during water swallows detected ALS penetrators/aspirators [AUC 0.89] at a cut point of 43.8 mm/s with sensitivity of 100% (95% CI 1, 1) and a specificity of 88% (95% CI 0.67, 1) (Fig. 1a). Tongue speed during water swallows detected ALS aspirators [AUC 0.942] at a cut point of 35.7 mm/s with sensitivity of 100% (95% CI 0.71, 1) and a specificity of 80% (95% CI 0.6, 1) (Fig. 1b). Tongue range of motion during water swallows detected ALS penetrators/aspirators [AUC 0.81] at a cut point of 16.4 mm with sensitivity of 66% (95% CI 0, 1) and a specificity of 100% (95% CI 1, 1) (Fig. 1c). Tongue range of motion during water swallows detected ALS aspirators [AUC 0.86] at a cut point of 8.5 mm with sensitivity of 100% (95% CI 0.43, 1)



**Fig. 1** Receiver operator curves (ROC) for the ability of biomechanical assessment measures to identify ALS patients who **a** penetrate or aspirate ( $PAS \geq 3$ ) and **b** aspirate ( $PAS \geq 6$ )

s and a specificity of 60% (95% CI 0.6, 1) (Fig. 1d). Tongue strength detected ALS penetrators/aspirators [AUC 0.84] at a cut point of 21.5 kPa with sensitivity of 100% (95% CI 1, 1) and a specificity of 87.5% (95% CI 0.625, 1) (Fig. 1e). Tongue strength detected ALS aspirators [AUC 0.84] at a cut point of 10 kPa with sensitivity of 100% (95% CI 0.2, 1) and a specificity of 60% (95% CI 1, 1) (Fig. 1f).

### Inter-rater Reliability

Inter-rater reliability for MBSIm<sup>PTM</sup>© and PAS scores was calculated using a consensus exact agreement method. While there were some initial differences in MBSIm<sup>PTM</sup>© and PAS scores between reviewers, after discussion between the two reviewers, inter-rater reliability was 100%.

### Discussion

The current findings underscore the importance of tongue movement on swallowing safety and help improve our understanding of the underlying mechanisms of swallowing impairment in persons with ALS. Tongue speed and range of motion were associated with swallowing safety during swallows of thin liquids. Maximum tongue strength was associated with swallowing safety during swallows of puree. These findings serve as a first step towards identifying tongue motor impairments as predictors of aspiration in this at risk patient population.

#### Reductions in Biomechanical Measures of the Tongue are Associated with Impairments in Swallowing

Our findings suggest strong associations between radiographically confirmed oral swallowing impairments and measures of tongue speed, range of motion and strength. Although the presence of oral swallowing impairments in persons with ALS are well documented [15, 34–36], the mechanisms underlying these impairments are not well understood. In our study, correlations between biomechanical measures of the tongue and thin liquid swallows were particularly strong, suggesting that thin liquids placed greater demands on the tongue in persons diagnosed with ALS than did puree consistencies.

To our knowledge, the relation between tongue movement speed and oral swallow function has not been explored in persons with swallowing impairments. During speech, reduced tongue speed in persons diagnosed with ALS has been associated with functional speech impairments including dysarthria and reduced speech intelligibility [37, 38]. Similarly, our study found associations between tongue speed and tongue control, bolus transport, and swallow

initiation, suggesting a potential causal relation between tongue speed and swallowing function.

Reduced range of motion of the tongue was associated with impairments in bolus transport/lingual motion. These findings are corroborated by a study by Kawai et al. that describes swallowing deficits in holding and transporting the bolus through the oral cavity due to reduced contact between the anterior tongue and the hard palate, and the posterior tongue and the soft palate in this patient population [15]. In our study, we found that during thin liquid swallows, reduced posterior tongue range of motion was also associated with reduced tongue base retraction, which could affect pharyngeal swallowing function. This finding extends those of Kawai et al. [15] who suggested that impairments in tongue range of motion are associated with impairments in both the oral and pharyngeal stages of swallowing.

We also observed that reductions in maximum tongue strength were associated with impairments in the transport of thin liquids and purees, and with the initiation of thin liquid swallows. These findings are consistent with previous reports demonstrating the effects of tongue weakness on oral swallowing impairments such as poor bolus formation, reduced bolus containment in the oral cavity, and reduced bolus transport [5, 7]. Our observation of the relation between tongue strength and swallow initiation adds to these findings and raises the possibility that posterior tongue weakness contributes to a delayed in the initiation of the pharyngeal swallow [39].

The association between biomechanical measures of the oral tongue and the initiation of pharyngeal swallowing was also robust. This finding is clinically meaningful because, when occurring with other physiologic impairments of swallowing, delays in swallow initiation may increase the risk for penetration/aspiration before the swallow [39, 40]. While the timing of swallowing onset has been found to be highly variable [41], reductions in tongue speed, tongue range of motion, and tongue strength were all associated with delays in swallow initiation.

Finally, strong associations were noted between all biomechanical measures of the tongue and MBSIm<sup>PTM</sup>© Oral Total Sum scores derived from VFSS. These findings, collectively, provide preliminary evidence suggesting that impairments in tongue speed, tongue range of motion, and tongue strength all contribute to oral swallowing dysfunction.

#### Reduced Biomechanical Measures of the Tongue are Associated with Reduced Swallowing Safety

Our study noted strong associations between swallowing safety with tongue speed and tongue range of motion with cup sips of thin liquids and with tongue strength during puree swallows. Similarly, a systematic review by Steele

and Chicero reported an association between reduced tongue strength and reduced swallowing safety [42]. Additionally, reduced tongue driving force combined with reduced pharyngeal contraction amplitudes has been associated with reduced swallowing safety in persons with ALS [43]. These findings highlight the potential of continued research directed at finding clinically accessible methods of bulbar assessment that are highly sensitive to bulbar impairment.

Biomechanical measures of tongue function were not found to be strongly associated with measures of swallowing efficiency. This unexpected finding may be, in part, due to the relatively restricted range of residue scores across our cohort, which was primarily between 1 and 3.

### Biomechanical Measures of the Tongue are Associated with Self-reported Measures of Bulbar Function

During cup sips of thin liquids, we observed a strong relation between tongue speed and scores on validated self-reported scales of bulbar function including the EAT-10, FOIS, and ALSFRS-B. While prior studies have shown each of these measures to be sensitive to dysphagia severity [31, 44, 45], to our knowledge, this is the first study to examine the association between objective measures of tongue motor impairment with self-reported swallowing function scales. Both measures of tongue range of motion and tongue strength demonstrated strong correlations with the FOIS and ALSFRS-R and the EAT-10 and FOIS, respectively. Because the FOIS scale is a measure of functional oral intake, strong associations between all three biomechanical measures of the tongue and FOIS scores reflect the critical role of lingual transport for oral intake.

### Clinical Implications, Limitations, and Future Work

Although these preliminary data were collected in a small sample size, they serve to motivate future studies in this area. Physiologic predictors of penetration/aspiration among persons with ALS are currently not well defined [42]; although preliminary, the current findings suggest that tongue motor impairment maybe a major risk factor for aspiration in this patient population. These findings motivate the need for larger cohort studies to further establish the predictive value of biomechanical measures of the tongue on swallowing safety.

Of the three biomechanical measures explored in this study, tongue speed, particularly during thin liquid swallows, appeared to be most closely related to impairments in tongue function, swallow safety, and self-reported bulbar function scales. This finding is most likely because thin liquids move quickly and are more difficult to control. At present, except for maximum tongue strength, the

clinical assessment of tongue function is typically limited to observations of tongue range of motion. Future work exploring objective, sensitive, and clinically accessible ways of measuring tongue speed may provide added value to oral motor assessment and contribute to establishing a widely accepted evidence-based bulbar assessment for persons diagnosed with ALS.

Finally, oral swallowing impairments, swallowing safety, and self-reported bulbar function measures demonstrated stronger associations with biomechanical measures of the tongue during cup sips of thin liquids than with teaspoons of puree. Similar to findings from Hazelwood et al. [46], our findings highlight the importance of conducting comprehensive swallowing evaluations using a variety of bolus consistencies to elucidate all physiologic impairments of swallowing in persons with dysphagia.

Although the current findings advance our understanding of the contribution of tongue impairment to deficits in functional swallowing, the study is limited by its small sample size, making it difficult to extrapolate results to the larger ALS population. Additionally, due to the limited number of participants, we were unable to extrapolate associations between biomechanical measures of the tongue and the timing of aspiration. Understanding how measures of speed, range of motion, and strength are associated with the timing of aspiration (before, during, or after the swallow) is critical to understanding the impact of tongue dysfunction on impairments in swallowing safety.

Predicting the decline of swallow function in persons diagnosed with ALS is a significant challenge, making clinical management of dysphagia in this patient population difficult. To our knowledge, specific guidelines for managing bulbar dysfunction do not exist [47]. Future work is needed to translate instrumentally based biomechanical methodologies into objective, valid, and clinically accessible assessments of tongue function that predict dysphagia in persons diagnosed with ALS.

**Funding** This work is funded by the National Center for Advancing Translational Sciences Grant TL1 TR002546, the National Institute on Deafness and Other Communication Disorders Grant F31 DC015941-01A1, the National Institute on Deafness and Other Communication Disorders Grant R01 DC0135470, the National Institute on Deafness and Other Communication Disorders Grant K24DC016312, and the National Institute on Deafness and Other Communication Disorders Grant R21 DC016664.

### Compliance with Ethical Standards

**Conflict of interest** The authors declare that they have no conflicts of interest.

**Ethical Approval** All procedures performed in studies involving human participants were in accordance with the ethical standards of the insti-



tutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

**Informed Consent** Informed consent was obtained from all individual participants included in the study.

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**Bridget J. Perry** PhD, CCC-SLP

**Kaila L. Stipancic** MA, CCC-SLP

**Rosemary Martino** PhD

**Emily K. Plowman** PhD

**Jordan R. Green** PhD