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EXAMINING THE EFFECTS OF ANKYLOGLOSSIA ON SWALLOWING FUNCTION

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ABSTRACT
Oropharyngeal dysphagia (OPD) involves difficulty during one or more of the stages of swallowing, resulting in difficulty moving the bolus from the mouth to the stomach. A deficit in tongue mobility, such as that found with ankyloglossia, may affect the oropharyngeal transit time of the bolus and predispose a person to OPD. This study was conducted to examine the possible relationship between tongue tie and oropharyngeal dysphagia. Data were gathered on 8 participants (5 females, 3 males) between the ages of 12-43 years. The Lingual Frenulum Protocol (Marchesan, 2012) was used to determine tongue tie. An Iowa Oral Pressure Instrument (IOPI) measured tongue tip, tongue dorsum, and lip strength, and a combination of electromyography, and the five-finger palpation method measured laryngeal timing. Measurements were compared with normative data from Holzer (2011). Results revealed that participants with ankyloglossia had signs of oral stage dysphagia, including reduced articulator strength (tongue tip and dorsum, and lips) and reduced masseter activity. Oropharyngeal transit times were not significantly different from the norms.

Key Words: tongue tie, ankyloglossia, oropharyngeal dysphagia

INTRODUCTION
The processes of mastication and deglutition are normally executed without any conscious effort, even though over 55 pairs of muscles are involved in the processes (Seikel, Drumright, & King, 2016). Because these processes are governed largely by neural circuitry that orchestrates the precise timing of bolus movement, a deficit at any stage in the swallow can result in discoordination and life-threatening consequences should the bolus enter the airway. This study examined the effects associated with tethering of the tongue secondary to ankyloglossia. Although ankyloglossia is typically viewed as affecting swallowing function during the post-natal nursing period, the potential for the disorder to interfere with the timing and processes associated with swallowing implies that it could lead to swallowing difficulties later in life.
Oropharyngeal dysphagia (OPD) refers to a deficit in any of the stages of swallowing that results in reduction of swallowing function. Swallowing is typically viewed as including four stages: Oral preparatory, oral transit, pharyngeal, and esophageal. The oral preparatory stage involves reception and manipulation of the bolus as it is prepared for swallowing. During this stage the food bolus is repeatedly moved from the tongue to the molars and back, providing the trituration process that serves to mix the bolus with the mucoid saliva of the sublingual and submandibular glands, in preparation for the next stage. The typical oral transit stage involves elevation of the tongue tip to the alveolar ridge, forming an anterior and lateral seal that facilitates the posterior movement of the tongue as it moves the bolus toward the oropharynx. The pharyngeal stage of swallowing is initiated when the bolus reaches the faucial pillars, posterior tongue, valleculae, or variously, the posterior pharyngeal wall. Physical contact triggers a cascade of responses that can be characterized under three global functions: Protection, pressure generation, and bolus transit timing (Seikel et al., 2016).

Protection of the airway is of vital importance and is accomplished by elevation of the hyoid-larynx complex (hyolaryngeal elevation), adduction of the true vocal folds and false folds, and by lowering of the epiglottis to cover the laryngeal aditus. Hyolaryngeal elevation begins with anterior movement of the tongue base and tip during the oral transit stage. The epiglottis is mechanically linked to the tongue and hyoid, such that retraction of the tongue during the oral transit stage, coupled with the elevated larynx, causes dropping of the epiglottis to cover the laryngeal aditus. This mechanical linkage is supported by the action of the thyroepiglotticus muscle. These safeguards allow the bolus to pass over the epiglottis, split into roughly equal halves, move into the pyriform sinuses, and ultimately pass through the upper esophageal sphincter to the esophagus. The laryngeal elevation function is protective in a second way, in that it facilitates inhibition of the cricopharyngeus muscle, opening the upper esophageal sphincter. In addition to compromising pharyngeal peristalsis, failure to sufficiently elevate the larynx can result in pharyngeal residue above the level of the cricopharyngeus, leaving the individual vulnerable to aspiration of the contents should he or she become supine. Pharyngeal peristalsis, the product of sequential contraction of the pharyngeal constrictors, supports clearing residue from the pharyngeal walls, reducing collateral aspiration of residual bolus material.

The second category of action, pressure generation, involves pressurizing the pharynx by elevation of the velum, retraction of the tongue in a piston-like fashion into the
oropharynx, and elevation of the larynx to open the upper esophageal sphincter. A lingual seal is essential to pressurization, providing separation of the oral and pharyngeal spaces. The relatively higher pressure of the pharynx moves the bolus into the lower-pressure esophagus.

The third category of action, oropharyngeal timing, is the complex and highly orchestrated product of an intact nervous system. Bolus manipulation and transit timing are governed by central pattern generators (CPGs) that capitalize upon the presence of reflexes, such as the sucking reflex, chewing reflex, and palatal reflex (e.g., Jean, 1984). The chewing center of the posterior brainstem is responsible for the mastication responses resulting in the rotary chew necessary for grinding the bolus on the molars. The respiratory center of the brainstem has CPGs responsible for rhythmic breathing as well as respiration in response to gas chemical changes in the blood.

There are two CPGs for swallowing, both housed within the dorsal medulla. The dorsal swallowing group (DSG), located near the nucleus ambiguous, generates the patterns associated with swallowing, while the ventral swallowing group (VSG) is responsible for the muscle activation. The DSG entrains the reflexive responses associated with the pharyngeal swallow (i.e., adduction of the vocal folds, elevation of the velum, elevation of the larynx) and organizes them into a sequential response that moves the bolus seamlessly to the waiting esophagus. The VSG is responsible for activation of these muscles in the highly orchestrated fashion that accomplishes the task. The result is oral transit of the bolus requiring up to one second and pharyngeal transit that is accomplished in less than a second (see Dodds, Stewart, & Logemann, 1990 and Seikel et al., 2016). The DSG and VSG ensure that the sequence is accomplished with appropriate timing, assuming normal physical and neuroanatomical structures. Alterations in the physical parameters of swallowing change the timing equation, such that, for example, increasing bolus viscosity or volume alters timing of both initiation and transit of the bolus (e.g., Kendall, Leonard & McKenzie, 2001). Similarly, as an individual ages, sensory and motor systems associated with swallowing change, modifying the swallow timing (e.g., Selley, Flack, Ellis & Brooks, 1989). At issue in the present study is whether the tethering of the tongue found in ankyloglossia is sufficient to interrupt the timing inherent in the system.

It is important to distinguish between signs of a swallowing problem, and the actual deficit in the swallow. A sign is the physical manifestation that indicates that there is a deficit, whereas the deficit is the abnormality in a structure, if any, that affects the function of the structure. Some common signs of
Oropharyngeal dysphagia are pocketing food in the lateral or anterior sulci, increased oropharyngeal transit time of the bolus, and nasal regurgitation. For these signs, the respective deficits may be weak buccal musculature, weakened tongue muscles, and slowed velar elevation (Seikel et al., 2016).

This study examined the relationship between a structural variation in humans, ankyloglossia, and swallow function. Ankyloglossia is a genetic oral condition characterized by an abnormally short lingual frenulum (Messner, Lalakea, Aby, Macmahon, & Bair, 2000; Yoon et al., 2017). The wide variety of characteristics, signs, range of severity, and effects of ankyloglossia contribute to the difficulty in diagnosing it and in determining prevalence (Ballard, Auer, & Khoury, 2002; Bai and Vaz, 2014; Buryk, Bloom, & Shope, 2011; Hogan, Westcott, and Griffith, 2005; Kotlow, 1999; Lalakea & Messner, 2003; Williams & Waldron, 1985; Yoon et al., 2017). Degrees of impact from ankyloglossia have been reported on feeding, speech, social issues, and sleep (Buryk et al., 2011; Huang, Quo, Berkowski, & Guilleminault, 2015; Messner & Lalakea, 2000; Yoon et al., 2017).

Oropharyngeal dysphagia indicates difficulty during one or more of the stages of swallowing that causes difficulty preparing the bolus for swallowing or moving the bolus from the mouth to the stomach (Logemann, 1998). There are many causes for oropharyngeal dysphagia, including structural changes that affect the oropharyngeal swallow. Ferres-Amat, Pastor-Vera, Ferres-Amat, Mareque-Bueno, Prats-Armengol, and Ferres-Padro (2016) state that ankyloglossia causes atypical deglutition due to insufficient palatal support to produce a mature adult swallow. As discussed, the mature swallow requires adequate oral seal and posterior movement of the tongue, processes that are restricted in the infant and developing child with hypertrophic lingual frenulum.

The tongue is the propelling structure that pushes and squeezes the bolus to the faucial pillars during the oral stage of the swallow, and a deficit in its mobility may affect the oropharyngeal transit time of the bolus. Francis, Krishnaswami, and McPheeters (2015) state, “Not all patients identified with ankyloglossia may have difficulties breastfeeding and/or need surgery. However, no data exists to differentiate how patients may fare later in life” (p. 1463). There is suggestion of a relationship between ankyloglossia and swallowing. Olive (2016) conducted a study of 8 participants with ankyloglossia comparing them to norms for the measurements of oropharyngeal transit time, masseter contraction, laryngeal timing, and measure of force based on IOPI of tongue tip, dorsum, and lips. Results reported all participants having a noticeable delay in oropharyngeal transit time.
for all bolus consistencies, a noticeable
difference in masseter contraction, a marked
delay in laryngeal timing, and reduced tongue
tip, dorsum, or lip strength (Olive, 2016). As
this was the first study linking ankyloglossia
and oropharyngeal dysphagia more studies in
this area are needed. The question of the
present study is: Do individuals with tongue-tie
differ from the norms of individuals without
tongue-tie in measures associated with
oropharyngeal dysphagia?

METHODOLOGY
The purpose of this study was to increase the
evidence regarding whether or not individuals
with ankyloglossia vary from the norm in
measures of oral pharyngeal dysphagia. The
study (IRB-FY2017053) was approved by the
Human Subjects Committee, Office for
Research at Idaho State University.

Participants
Participants included 5 females (mean age 25
yrs.) and 3 males (mean age 13.3 years),
between 12 and 43 years of age.

Instruments and Materials
The following parameters were recorded:
presence and degree of ankyloglossia, tongue
dorum and tip strength, lip strength, degree of
masseter activity, and oropharyngeal transit
time.

The Lingual Frenulum Protocol (LFP)
(Marchesan, 2012) was used to confirm the
presence of ankyloglossia in all participants, as
well as to obtain case history. All subjects
completed a demographic survey to obtain
information on participants’ birth date, gender,
history of serious medical conditions or
disorders, and preference of food types.
Results lead to the exclusion of participants
with a history of cleft palate or cleft lip,
intellectual or motor limitations, neurogenic
structural impairments to the head or neck,
neurogenic disorders, or traumatic brain injury
with coma. Participants with a history of a
concussion resulting in a period of
unconsciousness lasting no more than 5
minutes, and who had no reduction in motor or
cognitive function, were not excluded from the
study.

Clinical observation and professional judgment
were used to measure subjective variables,
which consisted of presence of open or closed
mouth posture at rest, presence of tongue
protrusion during swallow, rated cohesion of
bolus, and residue on the tongue after the
swallow. Superior tongue and lip strength were
measured using the Iowa Oral Performance
Instrument (IOPI) (Breakthrough model 1.5).
Degrees of masseter contraction and oral
pharyngeal transit time were measured using a
two channel Infiniti EMG (Thought Technology)
by means of surface electrodes. Food and
liquids that were administered to the
participants included Snack Pack chocolate pudding, water, and Triscuit crackers. A syringe calibrated for volume, measured in cubic centimeters, was used to measure pudding amounts and water.

**Procedures**

A counterbalanced order of presentation of measurement was used by developing three protocols (available upon request). Participants were assigned to one of the three groups (refer to Table 1). The counterbalanced order helped to control for measurement presentation effect such as fatigue or familiarity. The *Lingual Frenulum Protocol* was administered first for all three groups as it was critical for diagnosing an individual with tongue tie. Participants were evaluated in the Idaho State University Speech, Language, and Hearing Clinic or in their homes. All participants were tested in a quiet environment, free of distractions. Participants were seated in an upright, comfortable position, and were allowed to drink water after completion of each bolus trial.

**Table 1. Presentation Order of Measurement Tasks.**

<table>
<thead>
<tr>
<th>Group A</th>
<th>Group B</th>
<th>Group C</th>
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</thead>
<tbody>
<tr>
<td>Lingual Frenulum Protocol</td>
<td>Lingual Frenulum Protocol</td>
<td>Lingual Frenulum Protocol</td>
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<tr>
<td>IOPI force</td>
<td>EMG masseter contraction</td>
<td>EMG laryngeal timing</td>
</tr>
<tr>
<td>EMG masseter contraction</td>
<td>EMG laryngeal timing</td>
<td>IOPI force</td>
</tr>
<tr>
<td>EMG laryngeal timing</td>
<td>IOPI force</td>
<td>EMG masseter contraction</td>
</tr>
</tbody>
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**Tongue, Lip, and Masseter Strength.**

IOPI measurements for tongue tip strength, tongue dorsum strength, and lip strength were each done three times, recording the force exerted in micropascals. Masseter activity was recorded using EMG surface electrodes placed superficially on the belly of the masseter. Electrodes were first placed to obtain a masseter baseline and to measure masseter contraction. To identify the masseter belly, participants were asked to clench their back teeth while the researcher palpated the masseter. The EMG electrodes were placed bilaterally on the masseter belly in a vertical plane, with the right and left masseters assigned Channel A and Channel B, respectively. The ground electrodes were placed on the participants’ clavicle. A masseter activity baseline was recorded while participants clenched their molars, as an indication of maximum masseter function. This provided a comparison for masseter activity during swallow trials. Participants were asked to bite down with their back teeth as hard as
possible and then to relax, repeated for a total of three trials. Subsequently, groups were presented each bolus in three trials (½ teaspoon of pudding, 1 ½ teaspoon of pudding, 10 cc of water, and a Triscuit cracker). Each subject was to hold the bolus until told to swallow. The researcher palpated the lateral neck and submental region during the swallow, using the five-finger method (Logemann, 1998). At the initiation and termination of the swallow, the researcher pushed the spacebar of the laptop computer, which placed a mark on the EMG recording. The Triscuit cracker was an exception to this; in this case, participants chewed until they were prepared to swallow, and then swallowed with their own timing. This was determined to be minimally disruptive to the swallow timing itself. EMG was recorded for all boluses. Again, the researcher marked timing initiation and termination of the swallow using the spacebar. Each stimulus was presented three times.

**EMG and Behavioral Laryngeal Timing**

Instrumental and behavioral measurements of oropharyngeal transit timing were used with the goal of identifying the initiation and termination of the swallow. Initiation was defined as movement of the tongue and was instrumentally measured by recording the EMG of the submental region. Termination was defined as the depression of the larynx following swallow. As the myogenic response of depression is variable, this was also measured behaviorally using the 5-finger palpation method.

Channel A of the EMG was placed on the submental region, approximating the mylohyoid muscle. The first electrode was placed approximately two centimeters posterior from the chin point, and the second electrode placed two centimeters posterior to the first. Channel B had one electrode placed to the left of the thyroid notch with the second electrode placed two centimeters posterior to the first. Participants were presented with ½ teaspoon of pudding and instructed to clean the spoon and then to swallow when ready, repeated three times. The five-finger palpation method of Logemann (1998) was used as a behavioral measurement of laryngeal timing. The researcher also depressed the spacebar of the laptop computer at initiation of the swallow and at depression of the larynx, placing a marker on the EMG recording.

The researcher pulled down the subject's lower lip during a swallow to observe lingual function. The participant was presented with 1 ½ teaspoons of pudding and instructed to clean the entire spoon and swallow when ready. The researcher observed any tongue protrusion during the bolus preparation, the swallow, and for completion of the task. Presence or absence of tongue protrusion was indicated on the protocol. The remaining boluses were also