THE PENNSYLVANIA STATE UNIVERSITY
SCHREYER HONORS COLLEGE

DEPARTMENT OF COMMUNICATION SCIENCES AND DISORDERS

The Relationship Between Spontaneous Swallowing Frequency and Clinical Measures of Swallowing in ALS

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SPRING 2023

A thesis
Submitted in partial fulfillment
of the requirements
for a baccalaureate degree in Communications Sciences and Disorders
with honors in Communications Sciences and Disorders

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ABSTRACT

This study is a preliminary investigation of spontaneous swallowing frequency (SSF) and its use as a remote swallowing evaluation tool in patients with Amyotrophic Lateral Sclerosis (ALS). Via a mobile application on patients' phones, weekly recordings of spontaneous swallowing at a home setting were sent in to be analyzed. Recordings were sent in for a total of six months. Spontaneous swallowing frequency is also compared to typical gold standard clinical evaluations used to monitor the swallowing abilities of patients. Gold standard assessments like the Modified Barium Swallow Study (MBSS) was completed at baseline and at the end of 6 months. Other clinical swallowing measures like the Eating Assessment Tool (EAT-10) and the Mann Assessment of Swallowing Ability (MASA) was given every twelve weeks. We hypothesized that spontaneous swallowing frequency will be closely associated with other measures of swallowing evaluation. A total of 5 participants were analyzed as a part of this work. Statistical analyses consisted of Spearman's correlation and independent t-tests. Spontaneous swallowing frequency was found to be lower in individuals experiencing weight loss which could be a potential link for SSF being an indicator of dysphagia severity. Over the course of six months, the study was successful in collecting data on swallowing changes in ALS individuals from a home setting.

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ACKNOWLEDGEMENTS

I would like to thank my thesis advisor, Dr. Aarthi Madhavan, for her guidance and support throughout this entire thesis process. Her extensive knowledge and passion about dysphagia made my experience rewarding and enjoyable. Without her dedication throughout, this project would not have been possible.

I am also grateful for Dr. Carol Miller's guidance over the past several years and willingness to answer questions at any time.

Lastly, thank you to the rest of my family and friends who have been there for me during this year. I could not have completed this project without your continuous love and support.

Chapter 1

Introduction

A typical healthy swallow is dependent on several nerves, reflexes, and muscles. A breakdown in the swallowing pathway can lead to difficulty swallowing. Difficulty swallowing can negatively impact the ability to obtain proper nutrition, hydration, and in certain cases lead to illness caused by aspiration. A disruption in the swallowing mechanism can be caused by a variety of reasons such as normal aging, stroke, traumatic brain injuries, and neurodegenerative diseases. One neurodegenerative disease of interest that leads to difficulty swallowing is known as amyotrophic lateral sclerosis (ALS). This thesis aims to explore evaluation methods of difficulty swallowing because of ALS.

Amyotrophic Lateral Sclerosis (ALS)

Amyotrophic lateral sclerosis is identified as a neurodegenerative disease. The disease is commonly referred to as a motor neuron disease or Lous Gherig's disease. ALS is characterized by progressive muscular paralysis due to degeneration of motor neurons in the primary motor cortex, corticospinal tracts, brainstem, and spinal cord (Onesti et al., 2017). Currently there is not a cure for patients diagnosed with ALS. Treatment for patients aims to manage symptoms, improve quality of life, and extend survival (Nakamori et al., 2016).

Patients with ALS experience a variety of symptoms that impact vital needs like eating and speech. Dysphagia, or difficulty swallowing, presents in over eighty percent of advanced stages of ALS making it an important symptom to monitor upon diagnosis (Onesti et al., 2017). Dysphagia becomes life threatening because it increases the likelihood of choking or aspirating

(Onesti et al., 2017). Aspiration into the lungs makes patients more susceptible to opportunistic infections like pneumonia (Onesti et al., 2017). Dysarthria, a result of the disease impacting speech, is described as difficulty speaking because of the inability to control the muscles used in speech which is caused by an underlying neurological disease or damage (Enderby, 2013).

In most cases, diagnoses are considered isolated, however roughly five to ten percent of diagnoses are linked to familial genes (Wijesekera et al., 2009). The best possible outcomes are seen in cases with the earliest diagnoses, however diagnosing may not occur until later in life between the ages of forty and sixty because it may be years before symptoms can appear (Hillel et al., 1989) (Wijesekera et al., 2009). Time of diagnosis can also depend on the type of onset the patient has. There are two types of onsets, either spinal onset or bulbar onset (Wijesekera et al., 2009).

Spinal Onset ALS

Spinal onset is more common, with approximately two-thirds patients with ALS diagnosed with this onset variant (Wijesekera et al., 2009). Patients with spinal onset of ALS first begin to experience developing weakness in upper or lower limbs either distally or proximally in the early stages of the disease (Wijesekera et al., 2009). Gradually over time dexterity and gait are affected as the disease progresses. The prognosis of this onset it slightly better than bulbar onset, but death due to muscular paralysis and respiratory failure is typically three to five years following diagnosis (Hillel et al., 1989).

Bulbar Onset ALS

Roughly a third of patients diagnosed with amyotrophic lateral sclerosis have bulbar onset (Onesti et al., 2017). The bulbar region of the brain describes what most would consider to be the brain stem. It is comprised of the medulla, pons, and cerebellum (Seikel et al., 2021).

These areas of the brain are responsible for motor movement, coordination, balance, and involuntary life functions like breathing (Seikel et al., 2021). Due to the onset of the disease in the brain stem, decline tends to be more rapid, and support is generally needed sooner. Over time progressive paralysis leads to respiratory failure and death usually within two to three years (Wijesekera et al., 2009). This type of onset is often responsible for causing a myriad of related symptoms associated with swallowing, speech, and respiration (Hillel et al., 1989). Patients with bulbar onset often initially present with signs of dysarthria and dysphagia for solids or liquids (Wijesekera et al., 2009). Weakness in lower limbs can often present at around the same time (Onesti et al., 2017).

The Typical Swallow

A healthy, typical swallow is a complex movement involving a variety of structures. In short, a series of sensorimotor signals are received by the brain and sent to several muscles cohesively, to trigger a safe and efficient swallow. Information about the material being swallowing like the viscosity, volume, temperature, and texture is also sent back to the brain. Sensory information regarding any material in the airway or remaining residue, if present, allows for a motor response of coughing or a second swallow, as needed.

Nervous System Involvement

In a healthy swallow the electrical signal begins in the central nervous system and passes onto peripheral nervous system to trigger muscles used in the swallow (Seikel et al., 2021). The signal is controlled by the medulla oblongata, a part of the central nervous system (Jean, 2001). There are two groups of neurons located in the medulla oblongata involved in the neurological signal that is sent to other structures needed in the swallow movement. One group is located within the dorsal part of the medulla also known as the backside of the medulla. This part contains the generator neurons involved in triggering, shaping, and timing the sequence of the swallowing pattern (Jean, 2001). The second group is located more ventrally or in the frontal portion of the medulla. This region helps distribute the swallowing drive to the various pools of motor neurons that innervate individual muscles involved in swallowing (Jean, 2001). Once the signal is sent and distributed, the motor movement is coordinated by 5 cranial nerves and 3 peripheral nerves (Shaw et al., 2013). The process of swallowing is typically broken down into 4 phases that are closely linked to one and other.

Oral preparatory phase

The first stage of swallowing is known as the oral preparatory stage. This phase begins when the food is visualized, and we get sensory information from the smell and perhaps prior knowledge or anticipation of taste. This is the stage where mastication or chewing takes place and the food is prepared for swallowing (Seikel et al., 2021). During this process saliva in the mouth, triggered starting with that initial visualization of the food, helps to break down and liquify food. This helps to form more of a cohesive paste to swallow, referred to as bolus (Palmer et al., 1992). The lips also help to seal and contain the food as it is chewed (Seikel et al., 2021). Additionally, the soft palate also known as the velum closes to seal off nasal cavity to prevent nasal aspiration (Palmer et al., 1992). Once the correct consistency of bolus is reached then the next stage of swallowing begins.

Oral Transport Phase

This phase of swallowing involves moving the bolus through the oral cavity to the oropharynx or back of the mouth. The most important anatomical structure for this phase is the tongue. Movement and collection of chewed food into one cohesive bolus is achieved by the tongue pushing food to the back of the mouth (Palmer et al., 1992).

Pharyngeal Phase

The pharyngeal phase is marked by the involuntary reflex controlled by the amount of food pushed to the back of the oral cavity from the last stage (Palmer et al., 1992) Triggering the reflex to send the bolus down the esophagus depends on size and viscosity (Shaw et al., 2013). Bolus is propelled down the pharynx via contraction of pharyngeal constrictors (Seikel et al., 2021). Prior to propelling food down the esophagus, respiration ceases, vocal folds close, and the epiglottis seals the trachea to protect the airway (Seikel et al., 2021). This stage of swallowing is the shortest of the stages, only lasting about a second (Shaw et al., 2013).

Esophageal Phase

Once the bolus enters the esophagus via the upper esophageal sphincter, the final stage of swallowing begins. (Shaw et al., 2013). Relaxation of the cricopharyngeal muscle facilitates opening of the upper esophageal sphincter allowing for the bolus to pass through (Palmer et al., 1992). Involuntary peristaltic motion carries the bolus through the esophagus to the lower esophageal sphincter into the stomach for digestion (Seikel et al., 2021).

Clinical and Instrumental Measures of Swallowing

Swallowing is a critically important complex skill that is required for life. The evaluation of swallowing physiology for safety and efficiency is achieved either via a clinical evaluation or instrumental. Clinical assessment tools are typically administered at the bedside and aim to capture dysphagia signs and symptoms (Martino et al., 2013). Instrumental assessment tools measure dysphagia physiology via technology (Martino et al., 2013). The combination of these measurement methods allows for a holistic evaluation of swallowing.

Common Clinical Measures of Swallowing

One of the most common clinical assessments administered to patients is known as the MASA or Mann Assessment of Swallowing Ability. This examination uses twenty-four different areas to gauge a person's swallowing ability (Mann, 2002). Some examples of areas measured include respiratory rate, tongue movement, saliva, tongue coordination, and pharyngeal response (Mann, 2002). These areas are then converted into points and scored on a scale of two hundred (Mann, 2002). The MASA is used across various diseases and effective for predicting the incidence of aspiration and determining the severity of dysphagia (Kwon et al., 2019). This evaluation is often used as a basis or comparison for other assessments in development in the clinical field.

The Eating Assessment Tool (EAT-10) is another common clinical measure of swallowing using a self-administered, symptom-specific survey to evaluate for dysphagia (Belafsky et al., 2008). The survey consists of ten statements that a patient rates on a scale of 0-4, with 0 indicating no problem to 4 indicating a severe problem (Belafsky et al., 2008). A similar scale system is used to measure swallowing using the Functional Oral Intake Scale (FOIS). The FOIS is a reliable tool for the daily level assessment of the functional oral intake of food and liquids in patients using varying consistencies of liquids and foods (Crary et al., 2005). Following the evaluation, the patient is assigned a specific level on the FOIS scale indicating the oral intake abilities of the patient.

Common Instrumental Swallowing Assessment

A Modified Barium Swallow Study (MBSS) is a routine instrumental evaluation that uses radiographic examination of swallowing process for different consistencies (Sandidge et al., 2009). X-ray imaging focuses on the pharynx and esophagus to show how anatomical structures are moving during the swallow (Sandidge et al., 2009). Barium mixed into the different viscosities helps to illuminate the upper GI tract for a better view of anatomical features and the physiology of swallowing different boluses. Clinicians can make observations regarding the shape and size of the pharynx and esophagus during the swallow. Observations are made regarding the safety and efficiency of swallowing based on airway entry, residue in various areas, symmetry of movement etc. Using instrumental data, clinicians can diagnose the severity of the patient's dysphagia. Identification of the location in which the swallowing mechanism is impaired is essential in choosing appropriate therapy methods.

The Penetration Aspiration Scale and the Pooling score commonly referred to as a P-score is an example of a standardized and objective scoring while completing a MBSS. This test grades the severity of swallowing dysfunction according to observation of aspiration events using a scale (Rosenbek et al., 1996). The MBSImp scale (Modified Barium Swallow Impairment Scale) is another tool that measures 17 components of the swallow in each swallowing domain. Each of the 17 components is given an overall impression score across varying tested bolus consistencies, rated on a Likert scale (Martin-Harris et al., 2008).

Novel Telemonitoring Assessment Methods

As modern medicine advances, researchers look to improve efficiency of diagnostics tests while minimizing the invasiveness of the examination. Reducing the time it takes to complete routine evaluations becomes another area for improvement for those diagnosed with progressive diseases. For those diagnosed with a progressive disease, like ALS, the ongoing monitoring of symptoms through these diagnostic evaluations becomes a heavy burden for patients and loved ones. The combination of travel and long appointments can be very time consuming (Stephens et al., 2016). Thus, searching for techniques to monitor progression that reduce the burden placed on patients is an important goal.

Spontaneous Swallowing Frequency

Recently, there has been research regarding the use of spontaneous swallowing frequency (SSF) as a method to monitor the progression of dysphagia as a result of ALS. SSF has been used as a diagnostic tool in several studies with patients experiencing dysphagia secondary to other diagnoses. Spontaneous swallowing frequency involves collecting a ten-minute sound sample of a patient spontaneously swallowing. Microphones are attached anteriorly on the neck to record each swallow and the number of swallows in the ten-minute period are counted and recorded. Based on the number of swallows, an inference about the patient’s condition could be made. SSF offers a non-invasive and less time-consuming technique to monitor patients. It also reduces the inconvenience of travel as it can be done remotely and then sent via mobile app to be analyzed. Data collection is also easy as it only involves the attachment of a microphone on the front of the neck.

Patients with head and neck cancer and stroke survivors are two populations in which the application of SSF as a diagnostic evaluation tool has been studied (Hillel et al., 1989). In a published study, SSF was used to screen for dysphagia in patients who were recently admitted into the hospital following a stroke. SSF was compared to gold standard clinical measures like the MASA and MBSS that were also performed on the patients. Results showed a strong correlation between the standard clinical evaluations and SSF as well as a high accuracy of dysphagia identification in acute stroke patients (Carnaby et al., 2019). Similarly, in a study published in 2021, SSF was found to provide a useful method to identify and monitor dysphagia in patients with head and neck cancer following cancer treatment because of a strong correlation found between SSF and standard clinical swallowing assessments (Carnaby et al., 2022). Applying SSF to other populations experiencing dysphagia like ALS patients still needs to be explored to evaluate its validity but offers a possible solution to the burdens ALS patients face regarding supportive care.

To understand the value of SSF, a score for a typical healthy adult must be defined. Previous studies have found that the spontaneous swallowing frequency for a typical healthy adult to be between 0.3-6.7 swallows per minute (Bulmer et al., 2021). A normative result for spontaneous swallowing frequency is indicative of a clear airway. Having a clear airway reduces the likelihood of secondary illnesses developing as a result of aspiration from impaired swallowing. A reduced spontaneous swallow frequency indicates dysphagia, as validated against gold standard tools. (Bulmer et al. 2021, Carnaby et al., 2019, Carnaby et al., 2022).

Research Question

The purpose of this research is a preliminary exploration of spontaneous swallowing frequency and its use as a swallowing evaluation tool in patients diagnosed with ALS. SSF will also be compared to typical gold standard clinical evaluations used to monitor the swallowing abilities of the patients. Currently, little research has been performed exploring the use of SSF in ALS. By determining the relationship between SSF and other clinical measures of swallowing in ALS, we can form a preliminary understanding of its utilization as a standalone assessment measure, that is quick, non-invasive, and easy to complete. We hypothesize that SSF will be closely associated with other measures of swallowing evaluation. Baseline knowledge and expectation can be achieved and applied in further research.

This research is part of a larger grant exploring the use of telemonitoring of speech and swallowing in patients with ALS. The grant exclusively includes patients with ALS and applies spontaneous swallowing frequency as a form of telemonitoring for dysphagia.

Chapter 2

Methods

A cohort of 5 individuals from the larger grant were analyzed for this study. The participants enrolled in the study had to demonstrate a high probability of exemplifying measurable bulbar changes over the length of study. The length of study for each individual was 24 weeks from beginning to end. During these 24 weeks, standard clinical swallowing assessment was administered during clinic visits to collect baseline and comparative data at specific weeks. The Modified Barium Swallow Study (MBSS) and the Eating Assessment Tool (EAT-10) were administered at the beginning to collect baseline information (week 0) and at the conclusion of the study (week 24). The Mann Assessment of Swallowing Ability (MASA) was given during weeks 0 (beginning), 12 (midway), and 24 (conclusion). In addition to clinical measures, SSF was collected from each patient on a weekly basis.

Collection of SSF

Spontaneous swallowing frequency is defined as the number of spontaneous swallows that occur during a specific period of time. In the case of this study, the specified period of time was ten minutes. SSF is then reported as swallows per minute (SPM). As the sample was collected, participants watched a short video on mute or read a passage while performing natural, spontaneous swallows. Participants did not eat or drink anything during the collection period because it would interfere with data results.

Materials Needed for Collection

To accurately record a ten-minute swallowing sample, the participant used their smartphone, Lavalier microphone covers, attachable Lavalier microphones, and alcohol wipes. Previously, SSF was collected using a separate audio recording device, such as the Olympus DM-720. The steps explained below involve data collection using the recorder, however, they are closely matched to the steps the participants employed in the study using their own smartphones. The Lavalier microphone covers consisted of a felt covering placed over the microphone to dampen inferring noise like wind or rustling. The Lavalier microphone covers also came with an adhesive tape to attach the microphone to the body. Alcohol wipes were used to gently wipe the area of attachment for the microphone ensuring the success of microphone adhesion to the skin. The Lavalier microphones used plugged into the recording device and recorded the audio of each swallow collected during the collection period.

Figure 1 Olympus DM-720 Audio Recorder

Figure 2 Lavalier Microphone Covers

Figure 3 Voice Technologies VT506 Omni Lavalier Microphone

Procedure for Assessment

The spontaneous swallowing recording was recorded on a weekly basis by the participant and then sent in via a mobile application. Participants were shown how to apply the device correctly during their in-person visits and as needed via video telemonitoring during the recording period. To begin the application process one Lavalier felt microphone cover was applied to the microphone. The brown paper of the Lavalier cover was removed to reveal the adhesive backing of the undercover. The adhesive backing was then placed on to the microphone with the microphone circle side down.

Figure 4 Microphone in Circle Side Down Position

Once the adhesive backing was added, a singular felt microphone cover was placed on top of the microphone and sticky undercover. Following the Lavalier microphone cover setup, the participant then used an alcohol pad to wipe down the area of application on the neck. The microphone was placed on either side of the neck based on the participant's preference. Next, the microphone was applied flat against the skin in the proper placement area that was just cleaned. The microphone should be lateral to the larynx, about three quarters up the neck. In a tactile sense, this location should feel "fleshy" and a gap can be felt.

Figure 5 Lateral Placement of Microphone on Neck

Following mounting the microphone to the neck, the microphone was plugged into the Olympus DM-720 audio recorder in the microphone port. The audio recorder was powered on using the power button on the side of the device. To begin recording the ten-minute swallowing sample the participant pressed the button with the red circle labeled "rec". After ten minutes the participant pressed the stop button with the black square. The sound file was then uploaded into the mobile application provided to the participant and sent for analysis. After completion of the recording, the microphone coverings were removed and discarded. The same microphone and set up is used for each weekly recording.

Procedure for Data Analysis

The weekly sound files were analyzed using a sound editing software called WavePad. Weekly files were downloaded onto a desktop and then uploaded to WavePad for listening. Bose wireless noise cancelling headphones were used to listen to each recording. WavePad allows for the sound file to be analyzed each minute by the time markers at the bottom of the screen. Visualization of the sound waves during the recording was also provided.

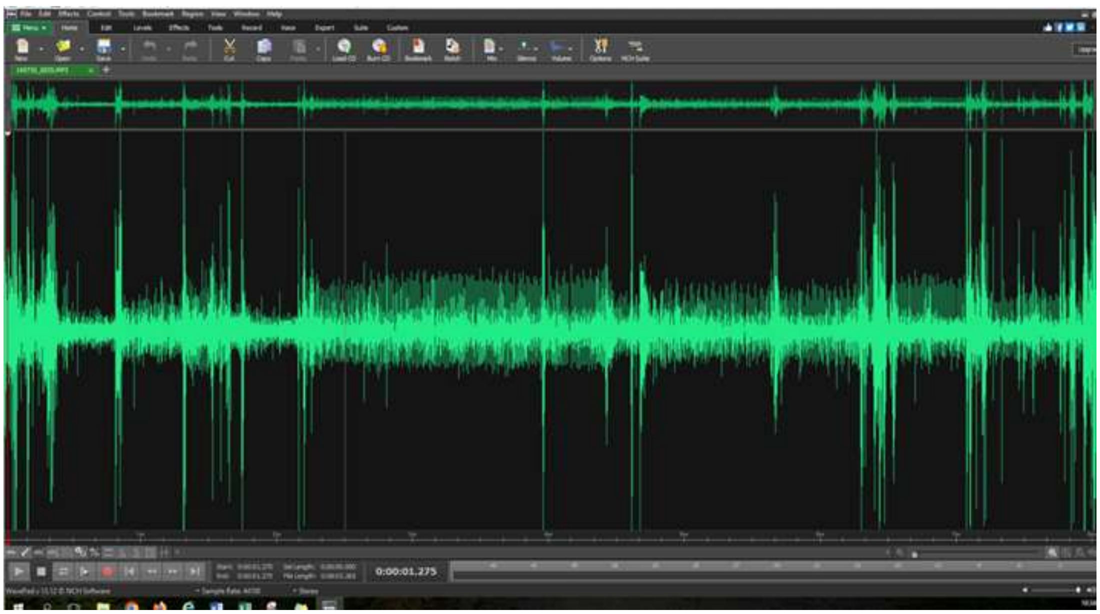

Figure 6 WavePad Sound Editing Software

As the recording played, the number of swallows that occurred during each minute is recorded into an Excel spreadsheet. Each recording was labeled with the date and participant number. After the number of swallows per minute was counted for the entire ten-minute recording, the total number of swallows was computed and recorded. The total number of swallows was then divided by ten to calculate the spontaneous swallowing frequency of the sample. Figure 7 shows how data was logged in an Excel spreadsheet and Figure 8 shows the overall calculation for spontaneous swallowing frequency.

Figure 7 Example Excel Spreadsheet Data Log

To calculate Swallows Per Minute (SPM):

1. Calculate the total number of swallows from the recording duration.
    a. Minute 1 total + Minute 2 total + Minute 3 total + Minute 4 total + Minute 5 total + Minute 6 total + Minute 7 total + Minute 8 total + Minute 9 total + Minute 10 total = *total number of swallows*.
2. Divide the total number of swallows by the total recording duration time (in this case, the recording duration time should be 10 minutes).
    a. $\frac{total\ number\ of\ swallows}{10\ minutes}$ = swallows per minute (SPM)

Figure 8 Calculation for Spontaneous Swallowing Frequency

Statistical Analysis

We used Spearman's correlation and independent sample t-tests to analyze the data. Running these statistical analyses allowed to identify any significant correlations between variables in the study to draw discussion from.

Spearman's Correlation

The data collected in the study is a form of quantitative data known as continuous data. Continuous data when graphed, forms a distribution of values along a continuum (Dettori, 2018). Some key examples of continuous variables that were used to find correlations included weight, age, SSF values, and scores from clinical assessments. The type of data collected, nominal, categorical, continuous, was one factor that led to choosing Spearman's correlation because it was better suited to continuous data. The second factor that made Spearman's a better type of statistical correlation analysis was that the data was not normally distributed. Most of the participants in the study are all in later stages in life which does not allow for an even distribution of age. Spearman's correlation is more appropriate to evaluate non-normally distributed data to give a better assessment of correlations between variables.

As correlations were calculated, they were grouped into different degrees of significance based on the value of r (Spearman's correlation). There were three degrees of correlation defined as high (±0.50- ± 1.0), moderate (±0.30- ±0.49), and low (and below) (Dettori, 2018).

T-Test

In addition to evaluating correlations in the data between continuous variables, comparing means between the variables also allowed for determining the difference between two groups. A t-test is frequently used to evaluate a hypothesis regarding means of two or more continuous variables. By comparing the means of two independent groups, it can determine whether the statistical evidence associated with the population means is significantly different. In other words, the larger the difference between the two means the more unlikely it would be observed just by chance (Whitley, 2002). Specifically, the independent samples t-test was used because there was no relationship between the participants in each sample tested. This meant that when dividing participants into two groups to compare means, subjects in one could not also be in another group, no subjects in either group can influence subjects in the other group, and no group can influence the other group tested.

When using the independent samples t-test, standard deviation and standard error of the mean were two measures used to evaluate significance of results. Standard deviation represents the variability of the data about the mean. Standard error of mean estimates how closely the sample means will represent the true population mean. Both statistical tools were used to evaluate results. A p-value will determine the significance of the difference of means. P-values greater than 0.05 are not significant findings and p-values lower are significant.

Chapter 3

Results

Demographics of Participants

The cohort of five individuals from the larger grant consisted of two males and three females. One individual withdrew from the study halfway through the data collection period. All participants were older adults, between the ages of 66-80 years old. In addition to similar age, all individuals involved in the study were 3 months since the onset of bulbar symptoms in their ALS progression. Each participant was given the ALSFRS-R, a clinical assessment test given to determine the severity of their ALS diagnosis. The minimum score on the assessment is 0 meaning mild and the maximum score depicting the most severe cases is 48 (Cedarbaum et al., 1999). The scores from the 5 individuals ranged from 30-42.

Baseline and 6-Month Correlation Results

Table 1 Spearman's Correlation for Baseline Data

			Age	ALSFRS	SSF_Baseline	MASA_Baseline	EAT10_Baseline	MBSImp_Baseline
Spearman's rho	Age	Correlation Coefficient	1.000	-.500	-.053	-.500	.300	.300
		Sig. (2-tailed)	.	.391	.933	.391	.624	.624
		N	5	5	5	5	5	5
	ALSFRS	Correlation Coefficient	-.500	1.000	.369	.500	-.100	-.100
		Sig. (2-tailed)	.391	.	.541	.391	.873	.873
		N	5	5	5	5	5	5
	SSF_Baseline	Correlation Coefficient	-.053	.369	1.000	.158	-.158	-.158
		Sig. (2-tailed)	.933	.541	.	.800	.800	.800
		N	5	5	5	5	5	5
	MASA_Baseline	Correlation Coefficient	-.500	.500	.158	1.000	-.900*	-.900*
		Sig. (2-tailed)	.391	.391	.800	.	.037	.037
		N	5	5	5	5	5	5
	EAT10_Baseline	Correlation Coefficient	.300	-.100	-.158	-.900*	1.000	1.000**
		Sig. (2-tailed)	.624	.873	.800	.037	.	.
		N	5	5	5	5	5	5
	MBSImp_Baseline	Correlation Coefficient	.300	-.100	-.158	-.900*	1.000**	1.000
		Sig. (2-tailed)	.624	.873	.800	.037	.	.
		N	5	5	5	5	5	5

*. Correlation is significant at the 0.05 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

There were three moderate positive correlations in the baseline data collected from the individuals. The correlation between age of participant and their EAT-10 score was 0.30. The correlation coefficient for age and MBSImp score was also 0.30. The correlation between spontaneous swallowing frequency and baseline ALSFRS-R score was 0.369. MASA had a -0.50 correlation with age and a -0.9 correlation with both EAT-10 and MBSImp.

Table 2 6-Month Spearman's Correlation Results

Correlations

			Age	ALSFRS	SSF_Final	MASA_Final	EAT10_Final
Spearman's rho	Age	Correlation Coefficient	1.000	-.632	.400	-.500	.200
		Sig. (2-tailed)	.	.368	.600	.667	.800
		N	4	4	4	3	4
	ALSFRS	Correlation Coefficient	-.632	1.000	.105	-.866	-.632
		Sig. (2-tailed)	.368	.	.895	.333	.368
		N	4	4	4	3	4
	SSF_Final	Correlation Coefficient	.400	.105	1.000	-.500	-.800
		Sig. (2-tailed)	.600	.895	.	.667	.200
		N	4	4	4	3	4
	MASA_Final	Correlation Coefficient	-.500	-.866	-.500	1.000	.500
		Sig. (2-tailed)	.667	.333	.667	.	.667
		N	3	3	3	3	3
	EAT10_Final	Correlation Coefficient	.200	-.632	-.800	.500	1.000
		Sig. (2-tailed)	.800	.368	.200	.667	.
		N	4	4	4	3	4

The MBSImp Score variable was not analyzed in the 6-month correlation data because of the participant that withdrew and did not complete testing. There was a positive moderate correlation between age and spontaneous swallowing frequency (0.40). MASA scores were negatively correlated with age (-0.50) and ALSFRS-R score (-0.866). Spontaneous swallowing frequency was negatively correlated with EAT-10 score (-0.80).

Baseline and 6-Month T-Test Results

Participants were divided into two groups: those who experienced weight loss and those who did not. Their response to the first question of the EAT-10 assessment was used to classify the 4 individuals into their weight loss group. Any score reported higher than a 0 to this question indicated weight loss possibly due to dysphagia Weight loss was used in the t-test because it is a surrogate marker for nutrition and can be a sign for dysphagia.

Table 3 Baseline T-Test Grouping Results

Group Statistics

	1=weightLoss; 0=NoWeightLoss	N	Mean	Std. Deviation	Std. Error Mean
MASA_Baseline	0	3	165.00	15.524	8.963
	1	2	144.50	21.920	15.500
SSF_Baseline	0	3	.33	.153	.088
	1	2	.25	.071	.050
Age	0	3	70.33	3.512	2.028
	1	2	73.00	9.899	7.000
ALSFRS	0	3	34.00	4.583	2.646
	1	2	37.00	7.071	5.000
EAT10_Baseline	0	3	5.00	4.359	2.517
	1	2	26.00	11.314	8.000
MBSImp_Baseline	0	3	6.33	3.055	1.764
	1	2	12.00	1.414	1.000

On average, participants experiencing weight loss had a lower MASA score (M =144.5) than those not experiencing weight loss (M=165). Similarly, participants experiencing weight loss had a worse MBSImp score (M= 12) than those not experiencing weight loss (M= 6.33) with a relatively low standard error (weight loss: 1.0, no weight loss: 1.764). Individuals experiencing weight loss were also found to have higher EAT-10 scores (M = 26) than those not losing weight (M = 5).

Table 4 Baseline T-Test Results

Independent Samples Test

		Levene's Test for Equality of Variances		t-test for Equality of Means							
						Significance		Mean Difference	Std. Error Difference	95% Confidence Interval of the Difference	
		F	Sig.	t	df	One-Sided p	Two-Sided p			Lower	Upper
MASA_Baseline	Equal variances assumed	.598	.496	1.254	3	.149	.299	20.500	16.351	-31.537	72.537
	Equal variances not assumed			1.145	1.686	.194	.389	20.500	17.905	-72.043	113.043
SSF_Baseline	Equal variances assumed	1.396	.323	.696	3	.268	.537	.083	.120	-.298	.465
	Equal variances not assumed			.822	2.894	.237	.473	.083	.101	-.246	.413
Age	Equal variances assumed	11.084	.045	-.457	3	.339	.679	-2.667	5.837	-21.244	15.910
	Equal variances not assumed			-.366	1.171	.385	.769	-2.667	7.288	-68.732	63.398
ALSFRS	Equal variances assumed	1.154	.361	-.593	3	.297	.595	-3.000	5.055	-19.088	13.088
	Equal variances not assumed			-.530	1.577	.330	.661	-3.000	5.657	-34.813	28.813
EAT10_Baseline	Equal variances assumed	16.800	.026	-3.093	3	.027	.054	-21.000	6.791	-42.610	.610
	Equal variances not assumed			-2.504	1.202	.103	.207	-21.000	8.386	-93.327	51.327
MBSImp_Baseline	Equal variances assumed	1.396	.323	-2.365	3	.049	.099	-5.667	2.396	-13.292	1.958
	Equal variances not assumed			-2.795	2.894	.036	.071	-5.667	2.028	-12.255	.921

The difference in means for MBSImp baseline score had a one-sided p-value of 0.049. No other significant p-values were found in any of the variables tested due to the small cohort studied.

Table 5 6-Month Grouping T-Test Results

Group Statistics

	1=weightLoss; 0=NoWeightLoss	N	Mean	Std. Deviation	Std. Error Mean
Age	0	2	72.00	2.828	2.000
	1	2	73.00	9.899	7.000
ALSFRS	0	2	35.00	1.414	1.000
	1	2	32.00	5.657	4.000
SSF_Final	0	2	1.75	1.061	.750
	1	2	.20	.283	.200
MASA_Final	0	2	144.50	6.364	4.500
	1	1	145.00	.	.
EAT10_Final	0	2	12.00	7.071	5.000
	1	2	27.50	10.607	7.500

One of the four remaining individuals in the weight loss did not have a final MASA score recorded and so standard deviation and standard error mean could not be calculated. On average, those experiencing weight loss had a lower SSF (M = 0.20) than those not experiencing weight loss (M = 1.75). Similarly, those having weight loss had a higher EAT-10 score (M = 27.5) than those not having weight loss as a symptom (M = 12).

Table 6 6-Month T-Test Results

Independent Samples Test

		Levene's Test for Equality of Variances		t-test for Equality of Means							
						Significance				95% Confidence Interval of the Difference	
		F	Sig.	t	df	One-Sided p	Two-Sided p	Mean Difference	Std. Error Difference	Lower	Upper
Age	Equal variances assumed	.	.	-.137	2	.452	.903	-1.000	7.280	-32.324	30.324
	Equal variances not assumed			-.137	1.162	.455	.911	-1.000	7.280	-67.941	65.941
ALSFRS	Equal variances assumed	.	.	.728	2	.271	.543	3.000	4.123	-14.740	20.740
	Equal variances not assumed			.728	1.125	.294	.588	3.000	4.123	-37.487	43.487
SSF_Final	Equal variances assumed	.	.	1.997	2	.092	.184	1.550	.776	-1.790	4.890
	Equal variances not assumed			1.997	1.142	.135	.271	1.550	.776	-5.845	8.945
MASA_Final	Equal variances assumed	.	.	-.064	1	.480	.959	-.500	7.794	-99.535	98.535
	Equal variances not assumed			.	.	.	.	-.500	.	.	.
EAT10_Final	Equal variances assumed	.	.	-1.720	2	.114	.228	-15.500	9.014	-54.284	23.284
	Equal variances not assumed			-1.720	1.742	.123	.246	-15.500	9.014	-60.335	29.335

Due to having a small cohort, no significant p-values are found in any of the variables tested at 6-months.

Chapter 4

Discussion

Previous studies of spontaneous swallowing frequency in other predisposed dysphagic populations, people with acute stroke and head and neck cancer patients, have demonstrated swallowing decline trends. In the small group of individuals with ALS observed from a larger grant, similar swallowing decline trends were seen among various clinical measures.

Baseline data showed positive correlations between age and EAT-10 score (0.30), age and MBSImp score (0.30), SSF and ALSFRS-R scores (0.369). Negative correlations were between MASA score and age (-0.50), MASA and EAT-10 (-0.90), and MASA and MBSImp (-0.90). 6-month correlation data showed positive correlations between age and SSF (0.40). Negative correlations were seen between MASA score and age (-0.50), ALSFRS-R score and MASA score (-0.866), and SSF and EAT-10 score (-0.80).

Baseline t-tests using weight loss as a grouping variable shows individuals experiencing weight loss had worse clinical scores in MBSImp, EAT-10, and MASA compared to individuals not experiencing weight loss. The difference in MBSImp score amongst those experiencing weight loss and those was a significant difference with a p-value of 0.049. 6-month t-test using the same grouping variable showed participants experiencing weight loss had lower SSF and a higher EAT-10 score than participants not experiencing weight loss. Due to this study being a preliminary study, no significant p-values were found with the 6-month data.

Clinical Measures Trends

Overall scores of clinical measures of swallowing were seen declining over time indicating an increase in difficulty swallowing. This trend is expected to be seen as participant's ALS diagnosis progresses and severity of symptoms persists.

MASA Trends

As participants continued in the study, MASA scores were seen to decrease over time showing an increase in severity of swallowing impairment. Negative correlations with age show that as age increases MASA scores decrease showing decline in swallowing. Previous studies have found that as adults age, reductions in muscle mass and strength reduce critical muscle movements like chewing and swallowing (Cichero, 2018). Individuals diagnosed with a degenerative neuromuscular disease like ALS, will experience this at an accelerated rate as they age, (Onesti et al., 2017) resulting in the decline in MASA scores. Similar negative correlation trends with the EAT-10 assessment and MBSImp assessment further verify swallowing decline.

As swallowing declines due to the progression of the disease, weight loss can be a symptom. According to the t-tests, individuals experiencing weight loss were found to have lower scores than those not experiencing weight loss. Previous studies have associated weight loss with dysphagia secondary to the ALS diagnosis (Hillel et al., 1989, Onesti et al., 2017).

EAT-10 Trends

EAT-10 scores were seen to generally increase over time amongst participants, which shows an increase in severity of symptoms relating to swallowing. Individual's scores were positively correlated with age, which is expected. When diagnosed with a progressive neurodegenerative disease like ALS, patients experience a decline in swallowing abilities as the disease progresses over time (Onesti et al., 2017).

EAT-10 scores were also consistently found to be worse in those experiencing weight loss than those not experiencing weight loss. This was found in both baseline and 6-month t-test data. This is expected, because as swallowing declines the individual will have increased difficulty with various consistencies of food and liquid (Onesti et al., 2017, Sura et al., 2012). Difficulty with consistencies of food and liquid could lead to potential weight loss if not managed appropriately.

ALSFRS-R Trends

ALSFRS-R scores varied over the progression of the study. In some cases, individuals had higher scores at the end of the 6-month period showing increased impairment of abilities due to the continuance of the disease. ALSFRS-R was seen positively correlated with MASA scores. This means that as ALSFRS-R scores increase MASA scores were also found to increase. While this is not what was expected to be seen over the course of the study, it was recorded that three of the individuals had ALSFRS-R scores improve at the end of the 6-month period. It should be noted that the ALSFRS-R is a clinical evaluation that can give varying scores depending on the observations of the clinician administering the assessment.

MBSImp Trends

MBSImp was shown to be negatively correlated with the MASA assessment. This is expected given lower scores on the MASA indicate poor swallowing and higher scores on the MBSImp also indicate poor swallowing. MBSImp was also seen to have a significant (0.049) difference of means when testing weight loss as a grouping variable. Those experiencing weight loss were seen to have a higher MBSImp than those not experiencing weight loss. This is consistent that if a higher level of difficulty swallowing is experienced, diet and food intake can be affected resulting in weight loss (Sura et al, 2012).

Spontaneous Swallowing Frequency Trends

SSF was found to be positively correlated with age, ALSFRS-R score, and EAT-10 score. This does not support the hypothesis that SSF will be closely associated with other gold standard clinical measures of swallowing. Since this is a preliminary study part of a larger grant, the cohort was relatively small affecting the accuracy of representing the true population we are trying to measure. As an example of not being able to accurately represent the true population, there was one individual that was an outlier in terms of SSF, but due to the small number of individuals being analyzed specific statistical analyses for outliers could not be applied.

6 month t-test data did support that individuals experiencing weight loss had a lower SSF than those not experiencing weight loss. This is an expected trend as ALS negatively impacts swallowing abilities which can affect overall intake. Furthermore, this additional potential support for SSF being related to the severity of dysphagia (Carnaby et al., 2019).

Conclusions

Preliminary conclusions from the present study are based on a relatively small cohort of bulbar onset ALS individuals. While previous studies recorded similar trends observed by data in this study, smaller cohorts limit generalizing findings because they may not be fully representative of the larger true population. Nonetheless, the small cohort of individuals exhibited changes in swallowing over the study period, a primary goal of the study. Reliability of data is another consideration in these preliminary conclusions. Most spontaneous swallowing frequency recordings were performed at home by the individuals themselves or other non-medical personnel (caregivers) limiting the quality and length of recordings sent in. However, the success of collecting data to monitor swallowing changes from a home setting to limit travel was achieved. Thus, this small cohort shows that home telemonitoring may be a viable option for frequent monitoring of swallowing in patients with ALS. If the relationship between SSF and gold standard measures of swallowing are determined to be validated, patient and caregiver burden can be significantly reduced.

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