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Early speech deterioration in amyotrophic lateral sclerosis: A case study of newscaster Donna Britt

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EARLY SPEECH DETERIORATION IN AMYOTROPHIC LATERAL SCLEROSIS:
A CASE STUDY OF NEWSCASTER DONNA BRITT

A Thesis
Submitted to the Graduate Faculty of the
Louisiana State University and
Agricultural and Mechanical College
in partial fulfillment of the
requirements for the degree of
Master of Arts

in

The Department of Communication Sciences and Disorders

by
Emily Watkins
B.A., Louisiana State University, 2016
December 2018
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Abstract

Purpose: The aim of this study is to detect and track speech changes using acoustic and perceptual measures in an individual (a local newscaster) with amyotrophic lateral sclerosis (ALS) using data from before her diagnosis, around the time of her diagnosis, and after her diagnosis.

Methods: Six time points, ranging from 37 months around her diagnosis, were analyzed from one speaker with ALS. Three acoustic parameters were measured: articulation rate, acoustic vowel space, and the slope of the second formant. Additionally, two experts with more than 10 years of experience perceptually analyzed the speech samples on 17 characteristics using a 7-point scale.

Results: The findings of this study revealed a decline in most parameters, both acoustic and perceptual, in the last time point, as expected. Additionally, a gradual increase in articulation rate, acoustic vowel space, and the number of syllables per utterance is observed before a decline after time point 4.

Discussion: The gradual increase in some parameters before a decline may reflect the speaker’s compensatory strategies. After time point 4, it is suspected the speaker could no longer compensate, and thus a decline is observed. Additionally, both perceptual and acoustic measures indicate speech changes over time; however, acoustic measures (i.e. articulation rate) appear to provide objective data to support subtle, perceptual changes between time points.
Introduction

Amyotrophic Lateral Sclerosis: Definition, Prevalence and Etiologies

Amyotrophic lateral sclerosis (ALS) is a degenerative neurological disease for which there is currently no cure. Etiology is still largely unknown, however about 5% of cases are familial or genetic, resulting from defects involving copper zinc superoxide dismutase (Rosen et al., 1993). The disease results in degeneration of both upper and lower motor neurons leading to atrophy and eventual paralysis. The disease is fatal, usually due to the weakness or paralysis of respiratory muscles (Wijesekera & Leigh, 2009). Three courses of the disease have been noted: Spinal-onset ALS is characterized by symptoms beginning in the muscles of the extremities, bulbar onset begins with involvement in the face, neck, and head and affects speech and swallowing, and a mixed onset is characterized by a somewhat simultaneous occurrence of bulbar and spinal symptoms. Bulbar onset is associated with quicker deterioration and a lower life expectancy than spinal onset ALS (Ball, Beukelman, Ullman, & Maassen, 2005). Patients may either present with bulbar-onset or spinal-onset ALS, however 85% of patients will experience bulbar symptoms at some point during the course of the disease (Armon & Moses, 1998).

According to Duffy (2013), “incidence of ALS is about 1 to 5 per 100,000 population,” and the disease tends to affect more men than women. Symptoms begin to develop, on average, between the ages of 40 and 70. While the average life expectancy is around 2-5 years post diagnosis, some people are living longer with the disease now than in the past (Duffy, 2013). Duffy (2013) proposes this could be a result of “multidisciplinary symptomatic care.”
Currently the average time from first symptoms to definitive diagnosis of ALS is about 14 months with a longer delay time for bulbar-onset ALS rather than spinal-onset ALS (Makkonen et al., 2016). The prolonged time between onset of symptoms and diagnosis inevitability results in a delay of intervention. Identification of bulbar symptoms such as dysarthria and dysphagia has potential to bridge this gap between onset of symptoms and diagnosis.

Diagnosis of bulbar involvement has potential to serve as an early indicator of the disease and a more accurate predictor of disease course and thus lead to higher quality intervention. Speech deterioration, even before perceptually noticed, indicates the onset of bulbar involvement in individuals with ALS. Specifically, speech characteristics such as a speaking rate, vowel space area, and the slope of the second formant have been reported as sensitive markers of the onset of bulbar involvement in the disease. These speech characteristics, along with intelligibility, tend to decline at a faster pace once they are affected. Specifically, once speaking rate “drops below 100-120 words per minute, speech intelligibility tends to decline rapidly” (Green et al., 2013). Early detection of bulbar involvement is imperative to provide timely intervention. Considering declination in speaking rate, vowel space area, and F2 slope can be detected acoustically before perceptually, identification of the earliest acoustic changes could lead to more timely identification of the disease and serve as a better predictor of disease course. In addition, identification of those early speech changes is essential to provide timely medical and therapeutic intervention, provide counselling for preparation of the future, maintain functional communication and improve quality of life as the disease progresses.
Aim of the Study

The aim of this study is to detect and track speech changes in an individual with ALS using data from before her diagnosis, around the time of her diagnosis, and after her diagnosis. This speaker is a 59-year-old female local newscaster with a diagnosis of familial ALS. Her career provides the opportunity to examine speech characteristics from before her diagnosis and determine the earliest sign of bulbar involvement. This study perceptually and acoustically examines speech characteristics across six time intervals ranging from two years around her ALS diagnosis. Three acoustic parameters (articulation rate, acoustic vowel space area, and second formant frequency trajectories) were specifically chosen based on the prior research reporting their sensitivity to dysarthria speech characteristics secondary to ALS as well as to general speech intelligibility deficits in dysarthria. Tracking speech changes and determining the earliest sign of bulbar involvement hopefully inspires further research in early detection and identification of speech deterioration, and one day of the disease itself.
Literature Review

Dysarthria and Bulbar Symptoms in ALS

Dysarthria is defined as a “collective name for a group of neurologic speech disorders that reflect abnormalities in the strength, speed, range, steadiness, tone, or accuracy of movements required for the breathing, phonatory, resonantory, articulatory, or prosodic aspects of speech production” (Duffy, 2013). There are several dysarthria types depending on the location of lesion and/or affected underlying neuropathologies. For example, lesions to the cerebellum often result in ataxic type of dysarthria. In the case of ALS, 80% of patients are reported to eventually develop some form of dysarthria, frequently, flaccid, spastic, or mixed type of dysarthria between spastic and flaccid. However, symptoms may present differently and at different times in each individual patient depending on whether upper or lower motor neurons are affected initially. Regardless of type of dysarthria developed, many patients will experience a decline in speech intelligibility and will eventually require the use of augmentative alternative communication (AAC) devices a few-years post-diagnosis. Additionally, time since diagnosis is not sufficient in predicting the need for this alternative communication. Speech deterioration, specifically decline in speaking rate and intelligibility, serve as more reliable predictors for AAC need (Yorkston, Strand, Miller, Hillel, & Smith, 1993).

Approximately 30% of patients show dysarthria as a first or predominant sign in the early stage of ALS. Dysarthria as an initial symptom is 8 times more frequent than dysphagia in ALS (Traynor, Codd, Corr, Forde, Frost, & Hardiman, 2000). This highlights the reason why early detection of speech deterioration is critical for the identification of bulbar involvement in ALS, which will lead to provision for timelier medical and therapeutic intervention. While growing
research has reported early speech deterioration in ALS, very few studies have had access to speech samples from before the diagnosis. This study will perceptually and acoustically examine speech changes in ALS from pre-diagnosis to post-diagnosis, taking advantage of this speaker’s profession.

The following section reviews perceptual, acoustic, and kinematic findings of speech characteristics of individuals with ALS, which is summarized in Table 1.

Table 1. Perceptual and Acoustic Characteristics of Speech Produced by Individuals with ALS

<table>
<thead>
<tr>
<th>Abnormal speech aspects</th>
<th>Perceptual</th>
<th>Acoustic</th>
<th>Kinematic</th>
</tr>
</thead>
<tbody>
<tr>
<td>Speech Rate</td>
<td>Slowed rate (Duffy, 2013; Kent et al. 1991; Klasner &amp; Yorkston, 2005)</td>
<td>Reduced speaking rate, prolonged phonemes, and inappropriate silences</td>
<td>Reduced LL and jaw speeds (Mefferd, Green, &amp; Pattee, 2012)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>(Ball, Beukelman, Ullman, Maassen, and Pattee, 2005; Green et al., 2013; Nishio and Niimi, 2000; Shelli Keri et al. 2016)</td>
<td></td>
</tr>
<tr>
<td>Articulation</td>
<td>Imprecise consonants (Carrow, Rivera, Mauldin, Shamblin 1974; Darley, Aronson, &amp; Brown, 1969; Duffy 2013)</td>
<td>Reduced VSA (Lee, Littlejohn, &amp; Simmons, 2017; Weismer, Jeng, Laures, Kent &amp; Kent, 2001)</td>
<td>Abnormally decreased lingual and jaw variability in early ALS (Kuruvilla-Dugdale &amp; Mefferd, 2017)</td>
</tr>
</tbody>
</table>

Note. LL = lower lip; VSA = vowel space area; F2 = second formant frequency; ALS = Amyotrophic lateral sclerosis; HC = healthy control.
Perceptual Measurements

Perceptual measures have been widely used throughout the research and in the clinical setting to identify deviant speech characteristics of different types of dysarthria. It is common to use such measures as an aid for diagnosis and for measuring treatment progress. Darley, Aronson, and Brown (1969) developed a list of the most deviant speech dimensions perceived in individuals with dysarthria including those with ALS. Their results are summarized in Table 3. Salient perceptual characteristics of speech in individuals with ALS include “imprecise consonants, slowed rate, strained strangled vocal quality, hypernasality, breathiness, and low pitch” (Duffy, 2013; Klasner & Yorkston, 2005).

Although perceptual measures have been considered the gold standard of dysarthria assessment and classification, several studies have raised a question about their reliability. Even experienced speech-language pathologists (SLPs) may find it challenging to use perceptual measures alone to determine diagnosis. One recent study by Allison et al. (2017) reported in distinguishing dysarthria speech samples from healthy speech samples, SLPs’ judgements yielded several false-positives, attesting to the importance of additional objective measurements. Additionally, when patients in the early stages of ALS reported on the function of their speech, salivation, and swallowing, most reported no deficits. This finding reveals that even the patients themselves cannot detect the earliest changes in bulbar function (Allison et al., 2017). In one case study of an adult with ALS, it was reported, unsurprisingly, that the participant’s spouse was more proficient in understanding her husband’s speech samples than unfamiliar listeners. However, after three years’ post-diagnoses, the spouse’s intelligibility rating declined and at the end of the study she understood him less than 50% of the time
(DePaul & Kent, 2000). Given the variability and subjectivity of perceptual analysis, acoustic analysis has the potential to allow for earliest possible detection of speech deterioration, even before perceptual changes are noticed.

**Acoustic Measures**

Since 1980s, acoustic measures have been frequently included in dysarthria research to provide objective, reliable data about speech characteristics. Acoustic measurements have the potential to detect subtle changes in speech that may not be perceptually detected. Among many acoustic parameters that have been applied to speakers with dysarthria, the following three variables have particularly received much attention in the studies of ALS: *speaking rate*, *acoustic vowel space*, and *second formant frequency (F2) slope*.

**Speaking Rate.** One of the earliest signs of bulbar involvement is a reduction in speaking rate, and is thus considered to be the gold standard in tracking bulbar symptoms (Shelikeri et al., 2016). The correlation between speaking rate and intelligibility has been widely studied; it has been agreed upon that a decline in speaking rate is followed by a rapid decline in speech intelligibility in individuals with ALS (Ball et al., 2005; Ball, Beukelman, & Pattee, 2002; Nishio & Niimi, 2000; Shelikeri et al., 2016). More specifically, speaking rate in ALS is known to be affected by three abnormal temporal characteristics: prolonged intervals, prolonged phonemes and inappropriate silences. These are typically not found in bulbar or pseudobulbar palsy (Darley, Aronson, & Brown, 1969).

Although longitudinal data in individuals with ALS is sparse, some researchers have been able to collect this data and attest to the decline in speaking rate. In the Nishio and Niimi (2000) longitudinal study, it was reported that speaking rate declines even before speech intelligibility
declines. This supports the idea that a decrease in speaking rate may be one of the earliest signs of bulbar involvement in individuals with ALS. As speaking rate declines, so does intelligibility. In another longitudinal case study, DePaul and Kent (2000) reported that speech intelligibility dropped significantly in the third year post-diagnosis, even for familiar listeners.

Ball, Beukelman, Ullman, Maassen, and Pattee (2005) also conducted a longitudinal study to track speaking rate in an individual with ALS via telephone. A significant decline in speaking rate was observed as the disease progressed. The first speech sample, taken in November of 2003, revealed a speaking rate of 155 words per minute. In January of 2004, the same speaker had a speaking rate of 137 words per minute. In March, his speaking rate had dropped to 102 words per minute, and in June his speaking rate was 80 words per minute and intelligibility had declined to 72%. This, although just a case study, further provides evidence that speaking rate can be used as an early detector of bulbar symptoms in ALS.

**Vowel Space Area.** A speaker’s acoustic vowel space area (VSA) is usually measured by graphing corner vowels in respect to the first and second formant frequencies (F1 and F2). The general assumption is based on the systematic relationship between formant frequencies and articulatory behaviors; F1 is inversely related to tongue height while F2 is directly related to tongue advancement (Hixon, Weismer, & Hoit, 2014). For example, a vowel such as /i/ is produced with a highly elevated and advanced tongue, which characterizes the vowel to have a low F1 and a high F2. On the other hand, a vowel such as /a/ is produced with a low and retracted tongue which characterizes the vowel to have a high F1 and a low F2, respectively. With this assumption, a speaker’s articulatory vowel space area can be inferred by the size of the acoustic vowel space area. Vowel space area can be measured as a triangular VSA by
graphing /i/, /u/ and /ɑ/, or as a quadrilateral VSA by graphing /i/, /u/, /ɑ/, and /æ/. Numerous studies have reported a strong correlation between acoustic vowel space and speech intelligibility in dysarthria. In other words, a larger vowel space area is correlated with better acoustic contrast among vowels and, therefore, enhanced speech intelligibility, while a smaller, or more centralized, vowel space area is correlated with reduced speech intelligibility in dysarthria (Kim, Kent, & Weismer, 2011; Sapir et al., 2010).

Acoustic vowel space area in individuals with dysarthria secondary to ALS has been extensively studied (Turner, Tjaden & Weismer, 1995; Weismer, Laures, Jeng, Kent & Kent, 2000). Several studies have found that individuals with ALS exhibit a more compressed vowel space area as compared to healthy adults (Lee, Littlejohn, & Simmons, 2017; Weismer, Jeng, Laures, Kent & Kent, 2001). In addition, several studies have reported a varying degree of vowel centralization among vowels such that high vowels (e.g., /i/ and /u/) are more dramatically centralized as opposed to lower vowels (e.g., /a/) (Kim, Hasegawa-Johnson & Perlman 2010; Lee, Littlejohn & Simmons 2017).

**F2 Slope.** The second formant frequency (F2) trajectories during the production of diphthongs have been used as an index of tongue movement. The slope of the second formant frequency (F2 slope) has been reported as significantly correlated with speech intelligibility and thus is a relevant factor in studying bulbar function (Kent et al., 1989; Kim, Weismer, Kent & Duffy, 2009).

Individuals with dysarthria secondary to ALS are likely to produce a shallow F2 slope. This is unsurprising as the trend for these individuals is to decline in speaking rate and
intelligibility (Kent et al. 1989; Weismer, Martin, Kent & Kent 1992; Yunusova et al., 2012). This is especially so when intelligibility is less than 70% (Duffy, 2013).

**Kinematic Measures.** Research on speech kinematics in individuals with ALS is limited although rapidly growing. Kinematic measures are collected using 3D motion capture systems such as electromagnetic articulography or ultrasound systems. The focus of kinematic measures of individuals with ALS has been the speed and distance travelled of the tongue, lower lip, and jaw.

**Articulator Rates and Variability.** In the kinematic data, it has been reported that individuals with mild to severe ALS exhibit slower movements of the lower lip and jaw (Mefferd, Green, & Pattee, 2012). Additionally, it has been reported that in the early stages of bulbar involvement, individuals with ALS tend to have abnormally low variability in their tongue and jaw movements. It has been proposed that this may be a compensatory strategy to control articulators. However, as the dysarthria becomes more severe, articulatory variability increases as the individuals lose the ability to use the compensatory strategy (Kuruvilla-Dugdale & Mefferd, 2017). Yunusova, Weismer, Westbery and Lindstrom (2008) used kinematic data to determine that although speakers with ALS do not necessarily move their articulators less than healthy controls, they take much longer to move their articulators the given distance. Interestingly, Yunusova, Green, Lindstrom, Ball, Pattee and Zinman (2009) found that in two of their three participants with ALS, jaw speed increased immediately before a decline in speech intelligibility. Why this is so remains unclear, it is possible that an increase in jaw movements was used as a compensatory strategy for decreased tongue movement.
ALS Disease Progression

Most research on the speech characteristics and deterioration in ALS patients is done using a cross-sectional design due to the lack of pre-diagnostic data. However, the nature of this speaker’s career lends to the rare opportunity to study speech samples pre-diagnosis. Thus far, only one study has collected pre-diagnosis data in an individual with dysarthria secondary to Parkinson’s disease. Berisha et al. (2017) collected speech samples from the famous boxer Muhammad Ali from before his diagnosis of parkinsonism and tracked his speech deterioration over time. The authors concluded that Ali exhibited a significant decline in speaking rate and decreased vowel space area over time.

The career of this study’s speaker provides a similar opportunity to study speech data including pre-diagnosis speech samples. This study is the first analyzing pre-diagnosis speech samples produced by patients with ALS. The overarching goal of this study is to track speech deterioration in this individual with ALS, using both pre- and post-diagnosis speech data, as a way to determine the earliest signs of bulbar involvement. Specifically, the following questions were posed:

1. **(Perceptual analysis) Do listeners (experts in the field of motor speech disorders) reliably identify the declination of speech among speakers with ALS?**

2. **(Acoustic analysis) How does ALS disease progression affect the following acoustic measures: articulation rate, vowel space area, and F2 slope.**

Based on the previous findings, findings of this study are expected to include a reduction in the three acoustic parameters, articulation rate, vowel space area, and F2 slope and to identify
gradual decreases in those parameters over the progress of the disease. In addition, it is hypothesized that such acoustic changes are not always perceived by listeners.
Methods

Participants

Speaker. The speaker was a 59-year-old Caucasian female with a diagnosis of familial ALS. She was officially diagnosed in June of 2017 and her symptoms began as spinal-onset. She was a local newscaster in Baton Rouge, Louisiana. She retired one year after diagnosis, in June 2018. She has given written consent to use her speech samples, name, age and gender in this study.

Listeners. Two experts in the field of motor speech disorders with more than 10 years of experience participated in the study to assess speech abnormality using a 7-point scale, with 1 corresponding to most deviant and 7 corresponding to most normal. Considering that the speaker is a local public figure, it was impossible to hide the identity of the speaker from the listeners. The speech samples ranged from 4 to 8 breath groups with the exception of one sample, which consisted of one long breath group containing 16 syllables. The samples from each time point were played via desktop computer in a quiet room in random order. In addition, the listeners were blind to the time point of each sample.

Data Collection and Analysis

Speech Samples. Since the speaker was a local newscaster, the audio of speech samples was extracted from her news station’s official website, WAFB channel 9 news, or personal contact with the news station. Speech samples were acoustically and perceptually analyzed from six time intervals which are summarized in Table 2.
Table 2. Time Points of Speech Samples

<table>
<thead>
<tr>
<th>Time Point</th>
<th>Date</th>
<th>Interval</th>
</tr>
</thead>
<tbody>
<tr>
<td>Time Point 1</td>
<td>5/21/15</td>
<td>24 months before diagnosis</td>
</tr>
<tr>
<td>Time Point 2</td>
<td>5/12/15</td>
<td>14 months before diagnosis</td>
</tr>
<tr>
<td>Time Point 3</td>
<td>12/28/16</td>
<td>7 months before her diagnosis</td>
</tr>
<tr>
<td>Time Point 4</td>
<td>7/6/17</td>
<td>Month of diagnosis</td>
</tr>
<tr>
<td>Time Point 5</td>
<td>2/27/18</td>
<td>7 months after her diagnosis</td>
</tr>
<tr>
<td>Time Point 6</td>
<td>6/13/18</td>
<td>12 months after her diagnosis</td>
</tr>
</tbody>
</table>

It was decided that 24 months before her diagnosis would provide a baseline of her speech before any signs of bulbar involvement. Since Makkonen (2014) reported the average delay time between onset of symptoms and diagnosis is around 14 months, this time interval was chosen. Additionally, a sample from around the time of her diagnosis was chosen, and several months after her diagnosis to track speech deterioration. The data were analyzed at the LSU Motor Speech Lab.

**Perceptual Ratings.** The 17 perceptual parameters were selected from the list of most deviant speech dimensions in ALS suggested by Darley, Aronson and Brown (1969). Table 3 below lists these dimensions in order from most deviant to less deviant with the following exceptions: It was decided to exclude *intervals prolonged* and *phonemes prolonged* from Darley, Aronson and Brown (1969)’s original list and replace them with *speech naturalness* and *speech intelligibility*. This change was made due to the assumption that the original characteristics were redundant of *slow rate* and that speech naturalness and intelligibility were important to this particular study. Additionally, the dimension, *nasal emissions*, was omitted from the original list due to its difficulty to identify perceptually using only audio.
Table 3. Selected Deviant Speech Dimensions in Dysarthria Secondary to Amyotrophic Lateral Sclerosis from DAB (1969)

<table>
<thead>
<tr>
<th>Number</th>
<th>Dimension</th>
<th>Number</th>
<th>Dimension</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Imprecise Consonants</td>
<td>10</td>
<td>Excess and Equal Stress</td>
</tr>
<tr>
<td>2</td>
<td>Hypernasality</td>
<td>11</td>
<td>Speech Naturalness</td>
</tr>
<tr>
<td>3</td>
<td>Harsh Voice</td>
<td>12</td>
<td>Reduced Stress</td>
</tr>
<tr>
<td>4</td>
<td>Slow Rate</td>
<td>13</td>
<td>Strained-Strangled Voice</td>
</tr>
<tr>
<td>5</td>
<td>Monopitch</td>
<td>14</td>
<td>Breathy Voice</td>
</tr>
<tr>
<td>6</td>
<td>Phrases Short</td>
<td>15</td>
<td>Audible Inspiration</td>
</tr>
<tr>
<td>7</td>
<td>Vowels Distorted</td>
<td>16</td>
<td>Inappropriate Silences</td>
</tr>
<tr>
<td>8</td>
<td>Low Pitch</td>
<td>17</td>
<td>Speech Intelligibility</td>
</tr>
<tr>
<td>9</td>
<td>Monoloudness</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Data Analysis

Acoustic analysis and perceptual analysis measures are described below.

**Acoustic Analysis**

The following three acoustic variables were measured using TF 32 (Milenkovic, 2005).

**Articulation Rate.** Articulation rate in the number of syllables per second was analyzed for all time points. Pauses that exceeded 150 milliseconds and audible inspirations were excluded when calculating the number of syllables per second (Kim & Choi, 2017). Articulation rate was computed by dividing the total number of syllables in each breath group by the utterance duration. The number of syllables per utterance was also reported considering the potential decrease over the course of the disease due to her respiratory and phonation function.
declination. The number of utterances analyzed per time point ranged from 26 to 38 for the first five time points. However, for the last time point, she produced significantly less utterances and thus only 11 utterances were available for analysis.

**Vowel Space Area.** Vowel space area for all time points was measured by taking an average of F1 and F2 values for each available token of the corner vowels from each time point: (/i/, /u/, /ɑ/ and /æ/). The temporal mid-point of the vowel was obtained from a spectrogram display and used to determine F1 and F2 values. Due to the nature of the speech samples, there were not an equal amount of tokens among every time point, however at least two tokens were obtained for each corner vowel from each sample.

**F2 Slope.** F2 slope was measured using the 20/20 rule (Kim, Weismer, Kent & Duffy, 2009; Weismer, Kent, Hodge, & Martin, 1988). That is, a spectral change in more than 20 Hz over the course of 20 milliseconds was defined as the transition state of F2. Once the transition was determined, F2 slope was computed by dividing transition extent by transition duration. To analyze F2 slope, the diphthong /aɪ/ was used from the word “crime.” This word comes from the speaker’s segment, “crime stoppers” and thus is a fairly consistent use of the sound among samples. One exception was for the last time point, the speaker did not use this word, and thus the word “right” was analyzed. Although these news segments are not ideal typical or conversational speech, the samples all come from this setting in order to maintain consistency among the speaking style.

**Perceptual Analysis**

The listeners used a seven-point scale to rate the aforementioned 17 different speech characteristics. The samples were played in random order. A difference of equal to or less than
two points between the two listeners was considered as an agreement. If a disagreement occurred, the two listeners discussed to reach an agreement (out of 119 ratings, this difference only occurred once).

**Statistical Analysis**

Because of the nature of the data that have a single value for most parameters and in each time point, the results mostly remain descriptive. However, t-tests were used to calculate differences between paired time points when multiple data values were available (articulation rate, utterance length). For example, t-tests were conducted between the first and second time point, the second and third point, the third and fourth time point, and so on.
Results

The results are reported separately for perceptual ratings and acoustic variables.

Perceptual Ratings

The mean rating values from the two listeners for each of the 17 perceptual characteristics are reported in Table 4. Perceptual ratings greatly decreased in the last time point for most characteristics, with significant decreases noted in imprecise consonants, vowels distorted, slow rate, and speech naturalness.

Table 4. Perceptual Ratings Results across Time Points

<table>
<thead>
<tr>
<th>Speech Dimensions</th>
<th>Mean Rating per Each Time Point</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Time Point 1</td>
</tr>
<tr>
<td>Imprecise Consonants</td>
<td>7</td>
</tr>
<tr>
<td>Hypernasality</td>
<td>5.5</td>
</tr>
<tr>
<td>Harsh Voice</td>
<td>7</td>
</tr>
<tr>
<td>Slow Rate</td>
<td>7</td>
</tr>
<tr>
<td>Monopitch</td>
<td>7</td>
</tr>
<tr>
<td>Phrases Short</td>
<td>7</td>
</tr>
<tr>
<td>Vowels Distorted</td>
<td>6</td>
</tr>
<tr>
<td>Low Pitch</td>
<td>7</td>
</tr>
<tr>
<td>Monoloudness</td>
<td>7</td>
</tr>
<tr>
<td>Equal and Excess Stress</td>
<td>7</td>
</tr>
<tr>
<td>Speech Naturalness</td>
<td>6.5</td>
</tr>
<tr>
<td>Reduced Stress</td>
<td>7</td>
</tr>
<tr>
<td>Strained-Strangled Voice</td>
<td>7</td>
</tr>
<tr>
<td>Breathy Voice</td>
<td>7</td>
</tr>
<tr>
<td>Audible Inspirations</td>
<td>6</td>
</tr>
<tr>
<td>Inappropriate Silences</td>
<td>6.5</td>
</tr>
<tr>
<td>Speech Intelligibility</td>
<td>7</td>
</tr>
</tbody>
</table>
**Acoustic Analysis Results**

**Articulation Rate.** Table 5 summarizes the speaker’s mean articulation rate (number of syllables per second), which is graphed in Figure 1. The articulation rate was measured from breath groups containing 2 to 35 syllables. This speaker’s mean articulation rate for time point 1 is 5.16 syllables per second. This provides a baseline for her articulation rate, as this time point is two years prior to her diagnosis. Time points 3 and 4 show a trend of an increase in articulation rate, although this increase was not found to be statistically significant. However, statistical significance was found between time points 4 and 5 ($p = .003$). Furthermore, when comparing the last two time points, the difference is approaching significance ($p = .051$). In addition, the number of syllables per utterance is reported in Figure 2. Although no statistical significance was found for this measure, a similar pattern was found in which the number of syllables steadily increases until time point 4 and then dramatically decreases for the last two time points.

![Figure 1. Articulation Rate (syl/sec) across all Time Points](image)
Table 5. Articulation Rate (syl/sec): Mean and Standard Deviation

<table>
<thead>
<tr>
<th>Time Point</th>
<th>Time Point 1</th>
<th>Time Point 2</th>
<th>Time Point 3</th>
<th>Time Point 4</th>
<th>Time Point 5</th>
<th>Time Point 6</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean</td>
<td>5.16</td>
<td>5.05</td>
<td>5.31</td>
<td>5.40</td>
<td>4.75</td>
<td>4.28</td>
</tr>
<tr>
<td>Standard Deviation</td>
<td>1.22</td>
<td>0.88</td>
<td>0.92</td>
<td>0.84</td>
<td>0.72</td>
<td>0.52</td>
</tr>
</tbody>
</table>

Figure 2. Number of Syllables per Utterance

**Vowel Space Area.** Table 6 summarizes this speaker’s acoustic vowel space area and mean F1 and F2 values across the time points which is graphed in Figure 3. As seen in Figure 3, this speaker’s vowel space area shows a pattern of increasing between time points 1 through 4, then decreasing for the last time point. Specifically, the vowels /u/ and /ɔ/ are affected. The vowel /ɔ/ in the last speech sample has a lower F1 value and a higher F2 value than in other samples. The vowel /u/ shows a trend of increasing in F1 and F2 between time points 1 and 2 and then again between time points 4 and 5.
Figure 3. Vowel Space Area across all Time Points

Table 6. Vowel Space Area and Mean F1 and F2 Values for Corner Vowels

<table>
<thead>
<tr>
<th></th>
<th>Time Point 1</th>
<th>Time Point 2</th>
<th>Time Point 3</th>
<th>Time Point 4</th>
<th>Time Point 5</th>
<th>Time Point 6</th>
</tr>
</thead>
<tbody>
<tr>
<td>Vowel Space Area (Hz^2)</td>
<td>129089.37</td>
<td>210350.60</td>
<td>277614.28</td>
<td>254770.68</td>
<td>223823.20</td>
<td>130589.42</td>
</tr>
<tr>
<td>Mean F1 /i/ (Hz)</td>
<td>416.00</td>
<td>384.44</td>
<td>366.88</td>
<td>445.30</td>
<td>454.33</td>
<td>332.86</td>
</tr>
<tr>
<td>Mean F2 /i/ (Hz)</td>
<td>1763.86</td>
<td>2283.77</td>
<td>1779.00</td>
<td>2543.60</td>
<td>2583.33</td>
<td>1803.43</td>
</tr>
<tr>
<td>Mean F1 /u/ (Hz)</td>
<td>343.75</td>
<td>400.50</td>
<td>309.58</td>
<td>449.00</td>
<td>473.75</td>
<td>321.00</td>
</tr>
<tr>
<td>Mean F2 /u/ (Hz)</td>
<td>1018.18</td>
<td>1392.25</td>
<td>1227.00</td>
<td>1634.33</td>
<td>1887.00</td>
<td>1022.00</td>
</tr>
<tr>
<td>Mean F1 /a/ (Hz)</td>
<td>642.00</td>
<td>721.88</td>
<td>687.13</td>
<td>797.70</td>
<td>799.22</td>
<td>570.00</td>
</tr>
<tr>
<td>Mean F2 /a/ (Hz)</td>
<td>1085.20</td>
<td>1194.00</td>
<td>1068.00</td>
<td>1296.10</td>
<td>1182.90</td>
<td>1484.66</td>
</tr>
<tr>
<td>Mean F1 /æ/ (Hz)</td>
<td>578.58</td>
<td>722.11</td>
<td>810.33</td>
<td>826.00</td>
<td>810.29</td>
<td>680.50</td>
</tr>
<tr>
<td>Mean F2 /æ/ (Hz)</td>
<td>1455.37</td>
<td>1600.33</td>
<td>1849.00</td>
<td>1745.66</td>
<td>1818.43</td>
<td>1640.13</td>
</tr>
</tbody>
</table>
**F2 Slope.** As seen in Table 7, the slope of the speaker’s second formant began around approximately 8.72 Hz/sec, decreased to 5.88 Hz/sec, rose again to 8.75 Hz, and finally dropped off again during time points 4 and 5. Two instances of /aɪ/ were analyzed from each sample for time points 1, 2, and 4 and the average is reported below. For time points 3, 5, and 6 only one instance per sample of the diphthong /aɪ/ was analyzed due to limited data. With the exception of the last time point, the diphthong was measured from the target word “crime.” In the last time point, the speaker did not use this word and thus the word “right” was analyzed.

Table 7. F2 Slope across Time Points

<table>
<thead>
<tr>
<th>Time Point</th>
<th>F2 Slope Token 1 (Hz/ms)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Time Point 1</td>
<td>8.72</td>
</tr>
<tr>
<td>Time Point 2</td>
<td>5.88</td>
</tr>
<tr>
<td>Time Point 3</td>
<td>8.75</td>
</tr>
<tr>
<td>Time Point 4</td>
<td>5.94</td>
</tr>
<tr>
<td>Time Point 5</td>
<td>6.77</td>
</tr>
<tr>
<td>Time Point 5</td>
<td>10.56</td>
</tr>
</tbody>
</table>

Reliability

**Acoustic Analysis.** 10% of the data was re-measured approximately 2 months after the original measurement in order to ensure intra-measurer reliability. Pearson’s correlation coefficient between the first and second measurement indicated high reliability; \( r (56) = .912 \). In order to ensure inter-measurer reliability, another measurer analyzed approximately 33% of
the data. Pearson’s correlation coefficient between the two measurers indicated high reliability; 
\[ r(86) = .976. \]

**Perceptual Analysis.** Intra-measurer reliability was established by asking the listeners to rate one the speech samples twice during the listening task. A Pearson’s correlation coefficient for intra-measurer reliability was found to be \[ r (32) = .717, \] indicating high reliability.
Discussion

This study sought to answer the following questions: (1) Are the abnormal patterns of speech in the early stage of ALS perceptually identified over the progress of the disease? And (2) how does ALS disease progression affect the following acoustic measures: articulation rate, vowel space area, and F2 slope? For this study, both perceptual and acoustic results reflected the speaker’s most significant deterioration in the last time point, as expected. However, interestingly, acoustic measures showed a gradual, positive change over the time points until time point 4 where a significant decline occurs to 3 out of 4 acoustic parameters, articulation rate, the number of syllables per utterance and vowel space area.

Speech Deterioration in the Early Stages of ALS

As expected, in general, perceptual and acoustic parameters indicated the most severe speech deterioration for time point 6. That is, consistent with previous research, vowel space area was found to be more centralized and articulation rate was found to be the slowest at the last time point. For example, the more compressed vowel space area found in this study is consistent with previous research of vowel space area in individuals with ALS (Lee, Littlejohn, & Simmons, 2017; Weismer, Jeng, Laures, Kent & Kent, 2001). This reduction in vowel space area indicates that the speaker is restricted in her tongue movements needed to produce articulate speech. Specifically, the vowel /u/ exhibits a higher F1 and F2 value during time points 4 and 5 as opposed to the first three time points. This indicates that the speaker is producing this vowel with a lower and more protruded tongue position, possibly due to decreased ability to reach the exaggerated position required for this vowel. This is consistent with previous findings that high vowels are particularly affected in this population (Kim, Hasegawa-Johnson & Perlman
2010; Lee, Littlejohn & Simmons 2017). Also, the vowel /ɑ/ drastically changes in the last time point. It is hypothesized this is secondary to an inability to effectively lower the jaw needed to produce this vowel. More research should be conducted on vowel space area and kinematics of individuals with ALS.

For the perceptual analysis, most speech dimensions were rated as more deviant for the last time point. As shown in Table 4, an obvious difference can be seen between the first and last time point. The first time point contained all ratings of 6 or 7, whereas the last had most ratings between 3 and 5. Speech intelligibility did not significantly differ among time points, which is not surprising considering this speaker’s tendency to over-articulate secondary to her career. However, *imprecise consonants, vowels distorted, speech naturalness*, and *slow rate* declined the most.

**Perceptual Ratings vs. Acoustic Measurements**

Although changes were eventually detected perceptually, especially toward the last time point where speech disturbance was most apparent, acoustic measures not only detected changes before they were noticed perceptually but also showed better sensitivity to changes in speech functions. For example, the mean value given for *slow rate* during the perceptual analysis was 5 for time point 4 and 4.5 for time point 5; however, a statistically significant change was noticed for articulation rate between these time points when measured acoustically. It should be noted that time point 4 is the month in which she was diagnosed; however, she was not reporting speech deficits at this time. Consistent with the hypothesis, acoustic measures appear to provide sensitive information regarding speech deterioration than perceptual measures.
Additionally, it should be noted that this speaker produced significantly less breath groups in the last sample as compared to the other samples. This is most likely due to her slow rate and the increased effort needed for speech. She retired shortly after the last time point.

In addition, most acoustic measures tended to increase until time point 4, then began to significantly decrease, while perceptual ratings did not show this pattern. It is possible that the speaker was using compensatory strategies even before changes in speech were detected perceptually. The use of compensatory strategies would account for the increase in vowel space area, articulation rate, and number of syllables per utterance up until time point 4. After this time point, it is suspected the speaker lost the ability to compensate and thus declination in these variables is observed. This explanation would be consistent with limited data from previous research. For example, acoustic and kinematic findings suggest jaw movements increase before decreasing in individuals with ALS, possibly secondary to the use of compensatory strategies (Weismer, Martin, Kent, & Kent, 1992; Yunusova, Green, Lindstrom, Ball, Pattee & Zinman, 2009).

**Summary, Limitations, and Future Research**

Overall, articulation rate, the number of syllables per utterance, and vowel space area show a pattern of increasing until around the time of diagnosis, then decreasing. A decline in articulation rate was detected acoustically before changes in F2 slope, or vowel space area. Additionally, acoustic measures tend to be more sensitive to speech deterioration than perceptual measures.

The current study is not without limitations. First, being a case study, speech samples and data collected were limited. Additionally, data could not be controlled such as which
phrases were said and how many tokens of corner vowels could be analyzed. Future research should use pre-diagnosis data when studying individuals with ALS, possibly collecting speech samples from home videos and analyzing a larger sample size.

**Clinical Implications**

The current study highlights the importance of early identification of speech deterioration in individuals with ALS using acoustic and perceptual data, because at least some measures reflect changes in her speech pattern (compensation or deterioration) even before her diagnosis or self-complaints of speech problems. Additionally, it should be noted that acoustic measures (i.e. articulation rate) detected speech deterioration before experts detected this deterioration perceptually. This indicates the value acoustic measures hold in a clinic setting. While certain characteristics are difficult to detect acoustically (i.e. nasality), clinicians should supplement perceptual analysis with acoustic analysis to determine deviations that may not be detected otherwise.
References


VITA

Emily Watkins, born in Lafayette, Louisiana, received her bachelor’s degree from Louisiana State University in Communication Sciences and Disorders in December of 2016. She then began the master’s program at Louisiana State University for Communication Sciences and Disorders in January of 2017 and plans to graduate on December 14, 2018. Her interests include working with individuals with traumatic brain injuries, neurodegenerative diseases, and other cognitive-communicative disorders. Following graduation, Emily plans to work as a Speech-Language Pathologist in a hospital or rehabilitation setting.