Speech Deterioration of Amyotrophic Lateral Sclerosis Before and After Diagnosis:

A Case Study of a Newscaster

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Running Title: Speech deterioration of a Newscaster with ALS

Abstract

Purpose: This case study traced speech deterioration in an individual around the time of diagnosis of amyotrophic lateral sclerosis (ALS). Our participant was diagnosed with spinal-onset, familial ALS in 2017. The speaker's occupation, a professional newscaster for 37 years, allowed a retrospective examination of her speech during news segments over 37 months around the diagnosis including pre-diagnosis.

Methods: A total of 6 time-points were selected to track auditory-perceptual and acoustic speech deterioration (2 years-, 14 months-, and 7 months-prior to diagnosis, the month of diagnosis, 7 months- and 12 months-after diagnosis). For perceptual ratings, two experts in motor speech disorders rated 17 speech dimensions on a 7-point scale. Four acoustic parameters were chosen for measurement: articulation rate, utterance duration, second formant frequency slope, and acoustic vowel space. Additionally, kinematic data were obtained from one time-point (8 months post-diagnosis) and descriptively compared to the movement of other individuals with ALS, and to healthy speakers.

Results: As expected, both perceptual and acoustic results indicated a decline in the selected speech measures as the disease progressed. More interestingly, the measures showed a consistent curvilinear appearance in which the speech parameters exhibit an improvement until immediately before and around the diagnosis, followed by sudden, drastic deterioration. Kinematic results indicated a greater degree of movement and speed compared to healthy speakers, probably due to the speaker's occupation.

Conclusions: Based on the findings, the time around diagnosis is considered a critical period with respect to speech deterioration in ALS wherein a dynamic, increasing-decreasing pattern of changes occur. This finding appears to reflect the patient's compensatory strategies as well as the speech deficits associated with bulbar involvement.

Introduction

Amyotrophic lateral sclerosis (ALS) is a degenerative neurological disease with no available cure that affects approximately 5 per 100,000 people in the United States (Mehta et al., 2018). This disease is accompanied by speech impairments in approximately 80% people with ALS, due to degeneration of upper and lower motor neurons leading to atrophy and eventual paralysis (Sitver & Kraat, 1982; Wijesekera & Leigh, 2009). The speech disorder resulting from ALS is frequently mixed flaccid-spastic dysarthria or a single form of either depending on the predominant involvement between upper and lower motor neurons (Darley, Aronson, & Brown, 1969b).

ALS progressively impairs many speech components over the course of the disease. For example, vowel distortion has been perceptually or acoustically characterized by lengthened duration, abnormal (increased or decreased) formant transition extents, reduced formant transition slopes, and compression of vowel space (Kent et al., 1989; Kent et al., 1991a; Mulligan et al., 1994; Weismer, Martin, & Kent, 1992; Weismer, Mulligan, & DePaul, 1986). Changes in vocal quality include abnormal pitch (high or low), high noise-to-harmonic ratios, tremor, and breathiness (Kent et al., 1992; Strand, Buder, Yorkston, & Ramig, 1994).

Past work has highlighted the importance of identifying the onset of ALS symptoms, especially bulbar symptoms, because bulbar onset ALS involving speech and swallowing problems is associated with quicker deterioration and a lower life expectancy than spinal onset ALS (Ball, Beukelman, Ullman, Maassen, & Pattee, 2005). Regardless of the type of onset, 85% of patients will experience bulbar symptoms at some point during the disease (Armon & Moses, 1998). In this sense, identification of dysarthria is critical in ALS because approximately 30% of patients show dysarthria as a first or predominant sign in the early stage of ALS. Further, dysarthria as an initial symptom is eight times more frequent than dysphagia in ALS (Traynor et al., 2000). Early detection of dysarthria, thus, ultimately leads to improved management of the disease including providing optimal pharmaceutical and palliative options to the patients.

Although perceptual measures have been considered the gold standard of dysarthria assessment and classification, several studies have investigated the reliability of these measures (Allison et al., 2017; Fonville et al., 2008; Zyski & Weisiger, 1987). One recent study by Allison et al. (2017) investigated speech-language pathologist (SLP)'s ability to distinguish dysarthria speech samples from healthy speech samples. They found that SLPs' judgements yielded several false-positives, attesting to the importance of supplemental objective measurements. Additionally, patients demonstrating early bulbar changes as detected by instrumentation-based measures did not report any speech or swallowing deficits, suggesting that even patients may have difficulty detecting early changes in bulbar function (Allison et al., 2017). Given the variability of perceptual analysis, instrumental measures have the potential to detect speech deterioration, even before perceptual changes are noticed.

Among dysarthria symptoms secondary to ALS, several instrumental measures have been suggested as a potential marker of bulbar involvement in ALS. Top among them is speech rate (Rong, Yunusova, Wang, & Green, 2015). A decline in speaking rate in ALS is known to precede a rapid decline in speech intelligibility (Ball, Beukelman, & Pattee, 2002; Ball et al., 2005; Niimi, 2000; Shellikeri et al., 2016). Specifically, once speaking rate "drops below 100-120 words per minute, speech intelligibility tends to decline rapidly" (Green et al., 2013).

Despite the rapidly increasing knowledge in speech deficits in ALS using acoustic and kinematic approaches, one challenge is large variability in speech characteristics across speakers primarily resulting from varying severity and involvement of motor neurons. In this sense, the current study provides unique, valuable longitudinal speech data in which the speaker serves as her own control as this case study reports speech deterioration before and after the time of diagnosis of ALS in a female who worked as a newscaster for 37 years until June 2018. This case is rare in that longitudinal speech data in an identical form (e.g., news broadcasts anchored by the same speaker) are available on a recurring basis. More uniquely, the participant's profession allowed us the opportunity to track speech characteristics over time from *pre-diagnosis* using archived news recordings available from the broadcasting channel.

Previously speech symptoms and their progression in ALS have only been studied after the diagnosis has been confirmed. To our best knowledge, this is the first report including the pre-diagnosis speech characteristics of ALS (See Berisha et al., 2017, for a similar approach in Parkinson's disease). The current study was designed to detect and track speech changes in an individual with ALS at a relatively early stage of the disease using data from 14 months before the diagnosis to 12 months after the diagnosis. Data from 2 years before the diagnosis served as the baseline. We mostly report auditory-perceptual and acoustic data over a total of 37 months. In addition, articulatory kinematic data that were obtained 8 months after the diagnosis are reported. The overarching goal of the study was to identify speech changes around the time of diagnosis in an individual with ALS. For this purpose, the following research question was posed: Does speech systematically change around the time of diagnosis as measured by auditory-perceptual and acoustic analyses? Based on previous findings, we expect to see a gradual decrease in the target acoustic parameters over the course of the disease. In addition, we hypothesize these acoustic changes will not always be perceived by the listeners.

Methods

This study was reviewed and approved by the Institutional Review Board (IRB) of Louisiana State University. She was approached to participate in the current study after receiving voice banking services at the university's speech, language, and hearing clinic. Further, she has given written consent for her speech samples, name, age, and gender to be used in this study.

Participants

Speaker. The speaker, a Caucasian female, was 59-years-old at the time of diagnosis with familial, spinal-onset ALS in July 2017. She served as an anchor for a local news channel in Baton Rouge, Louisiana for 37 years until she retired in June 2018. During an interview with a radio station, she indicated that her first perceived symptom of ALS was stumbling while walking in September 2016.

Seven months after the diagnosis was made in July 2017, the participant visited the LSU Motor Speech Laboratory for kinematic data collection (February 2018). At that time, she reported little-to-no perceived speech problems. However, during another interview recorded in July 2018, she expressed that her speech was slow, inaccurate, and no longer fluent, which led to her decision to retire from the profession.

Listeners. Two experts in motor speech disorders with more than ten years of experience in clinical service and research participated in perceptual ratings.

Speech Samples

Time-points. Audio samples were extracted from the news station's official website, WAFB Channel 9 News, or obtained by personal contact with the news station when the samples were not publicly available. Because audio samples were extracted from news segments, all recordings were of comparable acoustic quality and did not require any pre-processing or modifications. To track speech deterioration around the time of diagnosis, the following 6 time-points were selected (Table 1).

Table 1. Time-points of speech samples extracted for the study and number of utterances and F2 slope words available for each time-point.

Time-point	Date	Relative timing from diagnosis	Number of utterances	Number of F2 slope words
1	5/21/15	26 months before diagnosis	33	8
2	5/12/16	14 months before diagnosis	38	12
3	12/28/16	7 months before diagnosis	27	9
4	7/6/17	Month of diagnosis	26	6
5	2/27/18	7 months after diagnosis	30	7
6	6/13/18	11 months after diagnosis	11	5

Time-point 1, 24 months before diagnosis, was chosen as a baseline of her speech before any signs of bulbar involvement. Taking advantage of the availability of online news segments, 24 months was chosen over 21 months which would have make the same interval of 7 months between time points.

This was also to secure a time point when the speaker had no disease-related symptoms. *Time-point 2*, 14 months before the diagnosis, was chosen based on the average of 14 months of delay between onset of symptoms and diagnosis of ALS (Makkonen et al., 2016). *Time-point 3*, 7 months before diagnosis, was chosen as the midpoint between *time-point 2* and the month of diagnosis, *time-point 4*. *Time-points 5* and *6* were selected to mirror the time intervals from pre-diagnosis. *Time-point 6* was 11 months post-diagnosis (instead of 14 months) because the participant retired at this time due to the advanced disease symptoms (June 2018). A total of 168 utterances were identified from the six time-points ranging between 11 and 38 utterances per time-point. Time-point 6 had the fewest number of utterances available from the news segment, while time-points 1 through 5 had at least 26 utterances (Table 1).

Stimuli preparations. Speech runs (i.e., chunks of the participant's speech from news segments) were first partitioned into individual utterances. An utterance was defined as a stretch of speech between pauses lasting 150 ms or longer or an audible inspiration (Tsao & Weismer, 1997). Each utterance served as the unit for acoustic analysis and perceptual ratings.

Kinematic data. Articulatory kinematic data were recorded one time in February 2018 (7 months post-diagnosis), using an electromagnetic articulography (EMA) system, WAVE (NDI; Waterloo, ON, Canada). A total of five sensors were glued to the articulators: two sensors to the midsagittal surface of the tongue front (TF, approximately 1 cm away from the apex) and tongue back (TB, approximately 2 cm away from TF), two sensors to the upper and lower lips (UL, LL), respectively, and one sensor to the jaw (JW). Data were biteplate-corrected using R-based customized software. Because of the single-subject and one time-point of data collection (unlike the acoustic data), the movement data were compared to the data reported by Shellikeri et al. (2016) obtained from 33 participants with ALS. For this purpose, the methods were replicated from Shellikeri et al. (2016) with respect to speech stimuli and sensor locations. Specifically, speech stimuli collected for kinematic analysis included "Buy Bobby a puppy" read at a habitual rate and loudness, five times. Additionally, to be consistent with Shellikeri et al. (2016), data from only the TF and JW sensors were used for data analysis with no jaw-decoupling procedures.

Additionally, data were segmented using the upper and lower lip aperture time history as outlined by Shellikeri et al. (2016). The movement onset for the sentence was manually identified as the minimum lip aperture immediately preceding the stop burst of *buy*. The movement offset was identified as the minimum lip aperture immediately following the CVC sequence within *puppy*.

Data Analysis

Acoustic analysis. Four acoustic parameters, articulation rate, utterance length, acoustic vowel space, and F2 slope, were selected in the study based on previous literature reporting the sensitivity of these parameters to speech deficits in dysarthria, mainly secondary to ALS and their deterioration over time. Acoustic measurements were conducted using computer software, TF 32 (Milenkovic, 2005). The definition, measurement criteria, and rationale of selection for each measure are as follows:

(1) Articulation rate (syl/sec) was computed as the number of syllables produced per second from each utterance. Many studies have identified a reduction in speech rate as one of the earliest signs of bulbar involvement in ALS. Acoustic studies have further identified prolonged phonemes and inappropriate silences in ALS as factors contributing to slow speech (Ball et al., 2005; Green et al., 2013; Niimi, 2000; Shellikeri et al., 2016).

(2) Utterance length (syl/u) was defined as the number of syllables produced per each utterance. This parameter was included based on the frequently-reported characteristic in ALS of inappropriate pauses, and its potential effect on utterance length (i.e., progressively shortening of utterance length as the disease progresses; Green, Beukelman, & Ball, 2004; Turner & Weismer, 1993; Yunusova et al., 2016).

(3) Acoustic vowel space (AVS, Hz^2) was measured as the size of the vowel quadrilateral which was constructed by first and second formant frequency values of four corner vowels (/i, æ, ɑ, u/) obtained from the temporal midpoints of each vowel (Kent & Kim, 2003). Temporal midpoints

were chosen to minimize effects of adjacent sounds (Kim, Kent & Weismer, 2011). Tokens were collected for each of the four corner vowels from each time-point. Tokens for each vowel were averaged to produce a single pair of F1 and F2 values for each corner vowel. These averages were then used to calculate the AVS for each time-point. Due to the nature of newscast speech data, phonetic contexts of each vowel and the number of vowels available were inconsistent across time-points. The number of each vowel ranged from 3 to 11 per vowel. Numerous studies have reported a reduction in AVS and its significant correlation with speech intelligibility ratings in dysarthria (Kim, Kent, & Weismer, 2011; Sapir, Ramig, Spielman, & Fox, 2010; Turner, Tjaden, & Weismer, 1995; Weismer, Laures, Jeng, Kent, & Kent, 2000).

(4) Second formant frequency (F2) slope (Hz/ms) was measured using the 20/20 rule (Kim, Weismer, Kent & Duffy, 2009; Weismer, Kent, Hodge, & Martin, 1988). First, the transition state of F2 was identified from each word using the criteria that an interval showing a spectral change in more than 20 Hz over the course of 20 ms. Then F2 slope was computed by dividing transition extent in frequency by transition duration in time from diphthongs /ɑɪ/ from the words, *crime* and *right*. These words were selected because of their relatively frequent occurrence in the participant's broadcasting segments. For example, the word *crime* was frequently used among speech samples because of a news segment, "crime stoppers" included on a daily basis. Again, due to the nature of the data, the number of tokens of the two words varied across time-points, ranging from 5 to 12 tokens per time-point (Table 1). F2 slope has been reported to be significantly correlated with speech intelligibility and thus is a relevant factor in studying bulbar function (Kent et al., 1989; Kim, Weismer, Kent, & Duffy, 2009). For example, Kent et al. (1989) reported that a single correlation among many measures (including pulmonic function data and vowel prolongation data), between speech intelligibility and the F2 slope index, was significant.

Perceptual Analysis. For perceptual ratings, 17 speech dimensions were adapted from Darley et al. (1969a) including those identified as most deviant in ALS (see Table 2 in the Results for the list). Two measures of overall speech impression, *speech naturalness* and *speech intelligibility*, were included in the 17 speech dimensions given that the speaker mostly remained "intelligible" during the study period (<1 year post-diagnosis) despite noticeable changes in her speech especially toward time-point 6. Speech samples were played to the two listeners in a random order with respect to the time-points. The listeners rated each dimension on a seven-point scale, with 1 being the most severe and 7 being normal (Darley Aronson, & Brown, 1969b; Yorkston & Beukelman, 1978). A difference of equal to or less than two points between the two listeners was considered an agreement. If a disagreement occurred, the two listeners discussed their ratings to reach an agreement (out of 119 ratings, a disagreement only occurred once). To establish intra-rater reliability, audio from a random time-point was played a second time to be rated by the listeners. Although the audio samples were collected from televised news segments, perceptual ratings were based solely on audio.

Kinematic Analysis. As previously addressed, three articulatory parameters were obtained from the entire utterance except the utterance-final vowel /i/ according to the movement onset and offset criteria outlined by Shellikeri et al. (2016), "Buy Bobby a puppy", from two measurement points (TF and JW). The definition and measurement criteria for each measure are as follows:

(1) <u>Range of movement (mm)</u> was calculated as the Euclidean distance between the maximum and minimum values of the 3D movement.

(2) <u>Maximum speed (mm/s)</u> was calculated as the maximum absolute value of the first derivative of the 3D movement.

(3) <u>Movement duration (s)</u> was calculated as the duration between the movement onset and offset as defined by identified by the lip aperture time histories.

Statistical Analysis. Due to the nature of the data having a single value for many parameters for each time-point, most of the data was analyzed descriptively. However, when multiple data values were available (i.e., articulation rate, utterance length, and F2 Slope), a series of independent t-tests were performed between adjacent time-points to identify significant changes across time. Pearson's correlation coefficients were computed to ensure intra- and inter-measurer reliability for acoustic measurements and perceptual ratings.

Results

Our analysis focused on tracking speech changes over 6 time-points measured by 4 acoustic and 17 perceptual variables. Additionally, 3 kinematic measures were derived from a single time-point. Considering the large variability in disease presentation, the results of the current case study should be interpreted with caution and limited generalization.

Reliability

For perceptual ratings, intra-listener reliability was established by computing a Pearson's correlation coefficient for the data that were rated twice (approximately 20% of the entire data): $r(32) = .717$, $p<.05$. This was considered satisfactory. The degree of inter-listener agreement was calculated as the percentage of ratings within ± one scale value for the two expert listeners. The value, 88.2% was considered high compared to the studies with similar approaches (Kreiman et al., 1993).

For intra-measure reliability for acoustic analysis, 10% of the data was re-measured approximately two months after the original measurement. The correlation coefficient between the first and second measurement indicated high reliability; $r(56) = .912$, $p<.01$. For inter-measurer reliability, the second measurer analyzed approximately 33% of the data. The correlation coefficient between the measurers also indicated high reliability; $r(86) = .976$, $p<.01$.

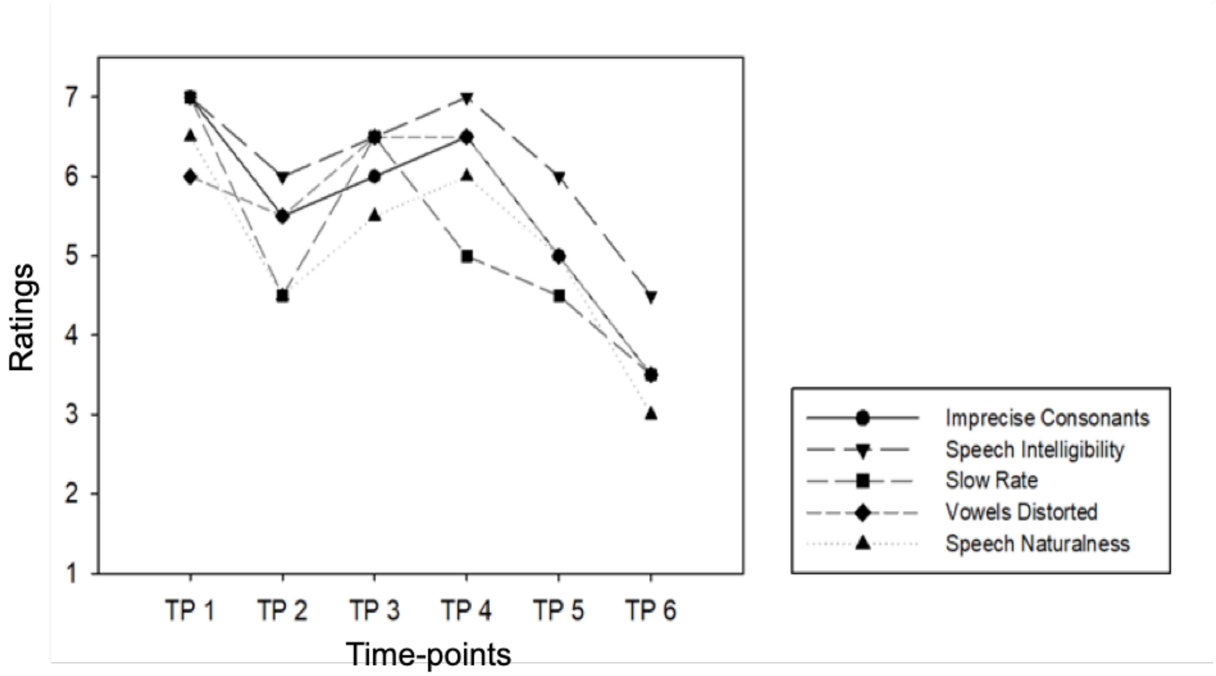
Perceptual Results

Table 2 indicates the mean of perceptual ratings for the 17 dimensions across 6 time-points. With no exceptions, the lowest ratings occurred at time-point 6. Also, most ratings showed a peak around time-points 4 and 5 and then a significant decline toward time-point 6. Figure 1 demonstrates the change of five selected dimensions.

Table 2. Mean values of perceptual ratings for 17 speech dimensions across 6 time-points (TP) on a 7 point scale.

Speech Dimension	**TP1**	**TP2**	**TP 3**	**TP 4**	**TP 5**	**TP 6**
Imprecise Consonants	7	5.5	6	6.5	5	3.5
Hypernasality	5.5	5.5	6	5.5	5.5	5.5
Harsh Voice	7	5.5	6	6	6	6
Slow Rate	7	4.5	6.5	5	4.5	3.5
Monopitch	7	5.5	6	7	7	5
Phrases Short	7	5	5	7	6.5	5
Vowel Distortion	6	5.5	6.5	6.5	5	3.5
Low Pitch	7	5	5.5	7	6	4
Monoloudness	7	5.5	6	7	6.5	4.5
Equal and Excess Stress	7	5	5.5	7	5.5	4
Speech Naturalness	6.5	4.5	5.5	6	5	3
Reduced Stress	7	5	6	7	6	4.5
Strained-Strangled Voice	7	5.5	6	6	6	5
Breathy Voice	7	5.5	5.5	7	6	5
Audible Inspirations	6	5.5	6	6	5	4.5
Inappropriate Silences	6.5	4.5	5.5	6.5	6.5	5
Speech Intelligibility	7	6	6.5	7	6	4.5

Figure 1. Perceptual ratings of five selected speech dimensions across the six time-points.



Acoustic Results

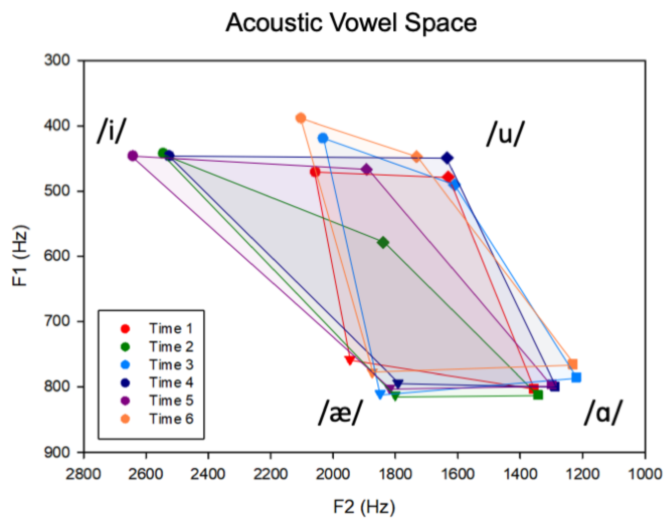
Table 3 summarizes descriptive results of the 4 acoustic measures across 6 time-points. Figure 2 demonstrates the changes in the acoustic parameters across 6 time-points.

Articulation rate (syl/s). T-test results indicated a significant difference between time-points 4 ($M = 5.40$, $SD = 0.84$) and 5 ($M = 4.75$, $SD = 0.72$); $t(52) = 3.068$, $p = 0.034$. Articulation rate continued to decrease from time-point 5 ($M = 4.75$, $SD = 0.72$) to time-point 6 ($M = 4.28$, $SD = 0.52$). However, the difference failed to attain statistical significance; $t(38) = 2.012$, $p = 0.051$.

Utterance length (syl/u). T-test results revealed no significant difference between paired time-points.

Acoustic vowel space (AVS; Hz^2). Because of the single AVS value for each time-point, a t-test was not performed. Figure 2 demonstrates the change in acoustic vowel space across six time-points, which shows a boomerang pattern of expanding and shifting to the left (time-points 4 and 5) before contracting and returning to an AVS comparable to time-point 1 (see time-points 1 and 6).

Figure 2. Acoustic vowel space across six time-points.



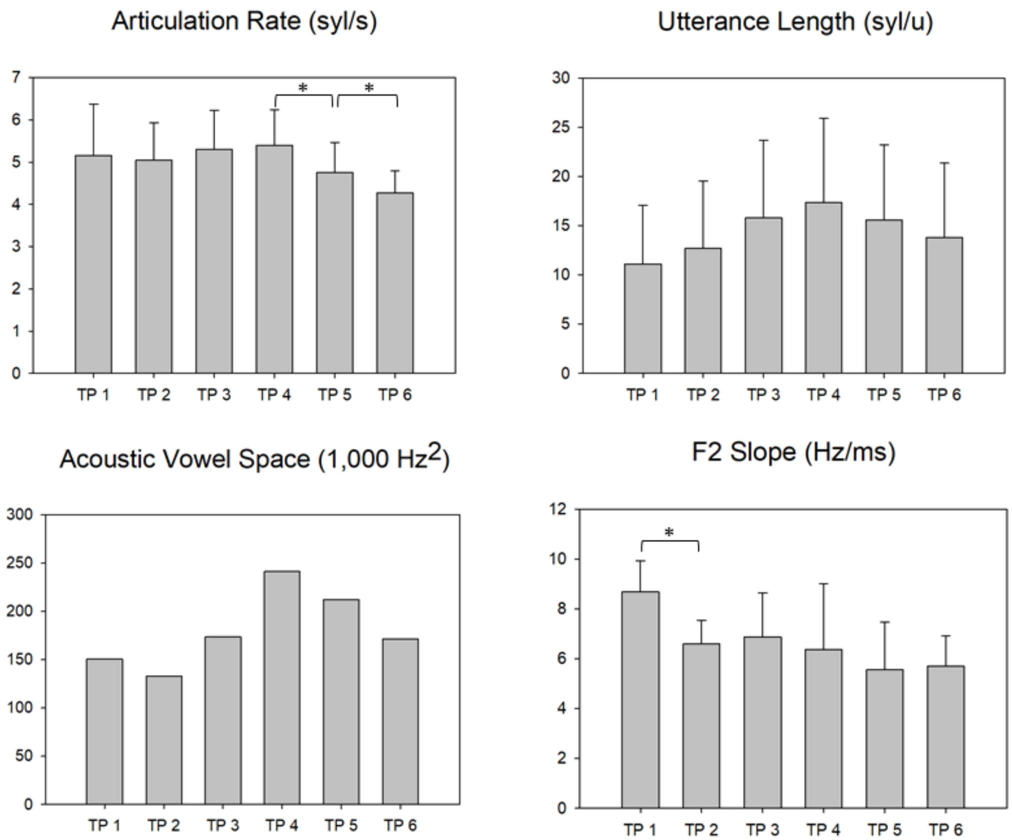
F2 slope (Hz/ms). T-test results detected a significant difference only between time-points 1 (M = 8.69, SD = 1.15), and time-point 2 (M = 6.58, SD = 0.91); $t(18) = 4.316$, $p < 0.001$.

Table 3. Mean, standard deviation) of acoustic results across 6 time-points. Standard deviation is not available for acoustic vowel space because of the single data point for each time-point measurements.

Acoustic Measures	TP 1			TP 2			TP 3			TP 4			TP 5			TP 6		
	M	**SD**	**CV**	**M**	**SD**	**CV**	**M**	**SD**	**CV**	**M**	**SD**	**CV**	**M**	**SD**	**CV**	**M**	**SD**	**CV**
Articulation Rate (syl/s)	5.16	1.2	0.24	5.05	0.9	0.17	5.31	0.9	0.17	5.4	0.8	0.16	4.75	0.7	0.15	4.28	0.5	0.12
Utterance Length (syl/u)	11.1	6	0.54	12.7	6.9	0.54	15.8	7.8	0.50	17.4	8.5	0.49	15.6	7.6	0.49	13.8	7.2	0.52
F2 Slope (Hz/ms)	8.69	1.2	0.13	6.58	0.9	0.14	6.87	1.7	0.24	6.37	2.4	0.38	5.56	1.8	0.32	5.7	1.1	0.19
Acoustic Vowel Space (Hz^2)	150447.42			133149.46			173626.43			241257.19			212265.71			171196.66		

TP: time-points, M: mean, SD: standard deviation, CV: coefficient of variation

Figure 3. Bar graphs displaying the means of tokens for each of the 6 time-points (TP). Standard deviation error bars are not available for acoustic vowel space because of the single data point for each time-point. Asterisks indicate significant differences between paired adjacent time-points at the .05 level.



Kinematic Results

Table 4 summarizes the mean and standard deviation for kinematic measures. For comparison purposes, Table 4 also includes results reported by Shellikeri et al. (2016) for mild, moderate, and severe ALS, as well as healthy speakers. Descriptively, data from the current study is overall comparable to the data of Shellikeri et al. (2016) with our speaker consistently demonstrating a greater speed and range of motion for tongue and jaw movement.

Table 4. Mean (standard deviation) of kinematic results collected from 7 months post-diagnosis. Data for mild, moderate, and severe ALS, and healthy speakers are reported in Shellikeri et al. (2016), replicated with permission from the publisher.

Kinematic Measures	Our Participant 178 WPM		Data Reported by Shellikeri et al. (2016)							
			Mild > 160 WPM		Moderate 120 – 160 WPM		Severe < 120 WPM		Healthy M = 221.4 WPM	
Tongue + Jaw										
Movement Range (mm)	18.14	(± 1.30)	16.22	(± 3.91)	12.75	(± 3.34)	12.56	(± 4.44)	14.18	(± 3.25)
Maximum Movement Speed (mm/s)	185.60	(± 32.36)	151.31	(± 36.03)	114.81	(± 25.1)	68.29	(± 25.55)	121.79	(± 29.92)
Jaw										
Movement Range (mm)	10.66	(± 1.98)	8.55	(± 2.4)	6.73	(± 2.68)	8.28	(± 3.99)	7.14	(± 3.28)
Max Movement Speed (mm/s)	209.91	(± 41.99)	97.08	(± 30.59)	67.27	(± 25.54)	60.83	(± 27.77)	86.07	(± 40.13)
Movement Duration (s)	0.88	(± 0.09)	1.09	(± 0.35)	1.14	(± 0.44)	2.05	(± 0.68)	1.18	(± 0.25)

Discussion

Dysarthria research has been largely conducted using cross-sectional group comparisons with healthy controls post-diagnosis. Due to the rapid progress of the disease and short life expectancy, longitudinal data are sparse particularly for ALS. Another complicating factor of ALS is the large variability of speech symptoms among speakers with ALS due to many factors, including the primary involvement of upper or lower motor neurons. Therefore, longitudinal examinations of case studies of ALS in which the speaker serves as his/her own control may provide valuable information on the course of disease progression especially regarding speech (Kent et al., 1991b).

The focus of the current case study was tracking speech changes in the early stage of ALS, especially around the time of diagnosis. More interestingly, this study provided the first insights of speech deterioration in ALS using speech samples from 26 months before the speaker's diagnosis of ALS. Despite the relatively short post-diagnosis time, the 11 months interval was considered critical and sensitive to disease progression based on the findings of significant changes in acoustic measures during this period across the six time-points (24-months pre- to 11-months post-diagnosis) as well as the speaker's self-report. The participant expressed drastic deterioration in speech between time-points 5 and

6, leading her to discontinue broadcasting in June 2018. She noted feeling a decrease in her speech rate and described the "crisp delivery" required for presenting news segments began to suffer (interview with *The Advocate*, July, 2018).

Speech Deterioration in the Early Stage of ALS

As expected, most perceptual and acoustic parameters indicated the most severe deterioration for time-point 6. Among the 17 speech dimensions, *speech naturalness, slow rate, imprecise consonants, and vowel distortions* were perceived to be the most severe at time-point 6. Speech intelligibility was consistently rated higher than speech naturalness across six time-points although the two parameters were highly-correlated ($r = .98$, $p < .01$). This is consistent with prior work reporting that two functional outcome measures, speech naturalness and intelligibility, may provide different insight into speech deficits in dysarthria, as speech intelligibility is primarily affected by articulatory deficits while speech naturalness is by prosodic disturbances (Anand & Stepp, 2015; De Bodt, Hernández-Díaz Huici, & Van De Heyning, 2002; Plowman-Prine et al., 2009). Although the two measures covary with 81% variance accounted for, speech naturalness was constantly rated lower than speech intelligibility by 1-1.5 point across 6 time-points, which indicates good sensitivity to speech deficits in ALS while speech still remains intelligible.

Acoustic deterioration was apparent in compression of acoustic vowel space and reduction in speech rate, consistent with previous research comparing speakers with ALS with healthy controls (Lee, Littlejohn, & Simmons, 2017; Weismer, Jeng, Laures, Kent, & Kent, 2001).

Further, most parameters, including acoustic and perceptual, showed a consistent pattern across the time-points. That is, the parameters reached the highest point around time-points 3 or 4 followed by a significant decline toward time-points 5 and 6 (Figures 1 and 3). We argue that this curvilinear appearance possibly reflects that the speaker was using compensatory strategies even prior to the diagnosis of ALS or self-perception of significant changes in speech. The latter is based on the speaker's description that no significant changes or challenges were noted until time-point 5 (conversation occurred in February 2018). The use of compensatory strategies would account for the increase in speech parameters until time-points 3 and 4, followed by an abrupt decline in these variables probably due to the loss of the compensation ability. This may be consistent with ALS data in which comparisons are made across different severity groups. For example, the findings from Shellikeri et al. (2016) indicate greater movement for the mild ALS group compared to not only the more severe ALS groups (moderate, severe) but also the healthy group for all kinematic parameters (Table 4). In other words, the mildly-involved ALS group showed a greater range and speed for tongue and jaw movement than healthy controls. Other acoustic and kinematic studies have also suggested jaw movements increase before decreasing in individuals with ALS, possibly secondary to the use of compensatory strategies in response to decreasing speed in tongue movement (Weismer et al., 1992; Yunusova et al., 2010). In addition, it has been reported that ALS patients with normal speech intelligibility (i.e., no prominent dysarthria symptoms yet) show even higher speech intelligibility scores than healthy controls, which could also be explained by compensatory strategies of ALS speakers such as hyperarticulation (Green et al., 2013).

As previously reviewed, speech rate, among many speech parameters, has received much attention in studies of ALS because of its sensitivity to bulbar involvement. Research has reported that speakers with ALS demonstrate declines in speech rate prior to reduction in speech intelligibility (Ball et al., 2002; Ball et al., 2005; Niimi, 2000; Shellikeri et al., 2016). Green et al. (2013) also found that ALS patients with normal speech intelligibility demonstrate even higher speech intelligibility than healthy controls but significantly lower speech rate (measured in words per minutes). That is, some ALS patients slow their speech rate in order to maintain a high degree of speech intelligibility. The current data supports these observations. Unlike most perceptual dimensions which showed the greatest value at the time-point 4, perceptions of speech rate peaked at time-point 3 followed by clear decline (Figure 3). It is noteworthy that the acoustic measure of speech rate, articulation rate, shows a declining pattern from time-point 4. Although the difference is subtle and not statistically significant (time-point 3: 5.31 syl/s, time-point 4: 5.40 syl/s, Table 3), one possible explanation for the gap between perceptual ratings and acoustic measures is the role of pauses. The current study did not include pause measures separately and our speech rate measure, articulation rate, excluded pauses longer than 150 ms from speech runs of news segments. However, previous studies have ascribed reduced speech rate in ALS to increased pause duration (e.g., Green et al., 2013).

Furthermore, it was our interest to see if there exists a certain pattern in acoustic vowel space (AVS). As previously noted, Figure 2 demonstrates a boomerang pattern of expanding and shifting to the left (time-points 4 and 5) before contracting and returning to an AVS comparable to time-point 1 (see time-points 1 and 6). The variability associated with the AVS appears to be mainly driven by the high vowels /i/ and /u/, in contrast to the relatively stable low vowels /æ/ and /a/. This finding is consistent with reports of high vowels being most dramatically affected by a loss of lingual function in ALS. Lee et al. (2016) found that speakers with varying severities of dysarthria secondary to ALS demonstrated reduced acoustic vowel space area predominantly due to the reduction of vowels /i/ and /u/. In contrast, the low vowels /æ/ and /a/ remain relatively consistent across all time-points likely due to the effective use of jaw compensation (DePaul, Abbs, Caligiuri, Gracco, & Brooks, 1988).

At time-point 5, the speaker was observed declining in several acoustic (articulation rate, AVS) and perceptual measures (speech intelligibility, speech naturalness, imprecise consonants). Despite the perceptual and acoustic speech deficits at this time point (i.e., month of the kinematic data collection), the kinematic results revealed that the participant's range and speed of articulatory movement were greater and quicker than individuals with varying severities of ALS and healthy speakers, as reported by Shellikeri et al. (2016). We argue that this finding is not surprising considering the speaker's occupation as a newscaster. It is likely that the speaker's experience with professional broadcasting is responsible for the observed large and fast articulatory movement. In addition, large and fast articulatory movement alone does not account for adequate intelligible speech even when intelligibility impairment is mild. Other factors such as articulatory precision, apart from large and fast articulatory movement, are likely more responsible for the observed reduction in perceptual and acoustic measures.

While the clinical implications from the current study are largely limited due to the single case-study design, the current findings contribute to a growing body of literature reporting an increase before a rapid declination in several acoustic and perceptual measures. As previously stated, this pattern is likely compensatory. Although the disease course is highly variable among patients with ALS, the current study may contribute to the general expectations and treatment plan for patients with spinal-onset ALS. For example, tracking patients' bulbar involvement using the methods in the current study may inform the timing of services (e.g., voice banking, augmentative and alternative communication training).

Conclusions and Limitations

Our speaker's occupation as a local newscaster allowed us the opportunity to examine speech samples before and after her diagnosis. Owing to this opportunity, the study adds data on speech changes before and after the time of diagnosis of ALS. Most consistently and interestingly, a pattern of apparently increasing articulatory precision was revealed in most perceptual and acoustic measures over the 37 months of study period.

Despite the advantage of this rare case study including that the speaker served as her own control, several limitations must be noted that result from the retrospective, single subject design of the study. Firstly, the speech samples were collected from past news segments. As such, some parameters, such as F2 slope, inevitably included a limited, unbalanced number of tokens available for analysis across time-points. Secondly, the linguistic, semantic, and phonetic contexts for many of the tokens used for analysis could not be controlled across time-points. Thirdly, the speaker's profession, a 37-year career of professional newscaster, should be considered as an important factor in data interpretation. For example, a unique articulatory speech profile characterized by larger and faster movements compared to other speakers (both healthy speakers and speakers with ALS) may reflect both her professional articulation and exaggerated compensatory strategies. Therefore, the measures reported within the current study should be interpreted with caution as the generalizability of our findings is relatively limited. Lastly, kinematic data were collected only from one time-point which limited our ability to make inferences about disease progression regarding articulatory movement. Future studies should utilize the retrospective single case study design, when possible, to investigate speech declination across disease progression in ALS.

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