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Spatiotemporal movement variability in ALS: Speaking rate effects on tongue, lower lip, and jaw motor control

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Abstract

Purpose—Although it is frequently presumed that bulbar muscle degeneration in Amyotrophic Lateral Sclerosis (ALS) is associated with progressive loss of speech motor control, empirical evidence is limited. Furthermore, because speaking rate slows with disease progression and rate manipulations are used to improve intelligibility in ALS, this study sought to (i) determine between and within-group differences in articulatory motor control as a result of speaking rate changes and (ii) identify the strength of association between articulatory motor control and speech impairment severity.

Method—Ten talkers with ALS and 11 healthy controls repeated the target sentence at habitual, fast, and slow rates. The spatiotemporal variability index (STI) was calculated to determine tongue, lower lip, and jaw movement variability.

Results—During habitual speech, talkers with mild-moderate dysarthria displayed significantly lower tongue and lip movement variability whereas those with severe dysarthria showed greater variability compared to controls. Within-group rate effects were significant only for talkers with ALS. Specifically, lip and tongue movement variability significantly increased during slow speech relative to habitual and fast speech. Finally, preliminary associations between speech impairment severity and movement variability were moderate to strong in talkers with ALS.

Conclusion—Between-group differences for habitual speech and within-group effects for slow speech replicated previous findings for lower lip and jaw movements. Preliminary findings of moderate to strong associations between speech impairment severity and STI suggest that articulatory variability may vary from pathologically low (possibly indicating articulatory compensation) to pathologically high variability (possibly indicating loss of control) with dysarthria progression in ALS.

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Keywords

Speech motor control; speaking rate; movement variability; dysarthria; ALS

1. Introduction

Amyotrophic Lateral Sclerosis (ALS) is a neurodegenerative disease associated with progressive muscular weakness and loss of motor control. In bulbar ALS, motor neurons innervating head and neck muscles are affected during the early stages of the disease whereas in spinal ALS, motor neurons innervating trunk and limb muscles are initially affected. Although the decline in motor control is frequently described as a symptom of disease progression, it remains unclear if and to what extent ALS affects the motor control of orofacial movements associated with speech production.

Movement Variability as an Index of Motor Impairment Severity

The degree to which movements vary from trial to trial has been used previously to objectively quantify the severity of the motor impairment. For example, slightly elevated movement variability during walking is associated with an aging-related decline in motor control whereas considerable movement variability is thought to be disease-related (Chien et al., 2015; Owings & Grabiner, 2004). High variability of limb movements has been found in individuals with movement disorders such as Huntington's disease, Parkinson's disease, and ALS (Hausdorff et al., 1997, Hausdorff et al., 2000). Furthermore, movement variability is positively associated with motor decline in progressive movement disorders and can serve as a predictor of walking function and fall risk (Chau, Young, & Redekop, 2005; Hausdorff, 2005; Stergiou, Harbourne & Cavanaugh, 2006). Although there are fundamental differences between limb motor control and speech motor control (e.g. the use of visual feedback for limb movements versus auditory feedback for speech), several findings of studies on limb motor control have been replicated in studies on speech. For example, similar to findings of increased gait variability in older and impaired individuals, increased speech movement variability has been observed in older talkers (Wohlert & Smith, 1998) and talkers with dysarthria due to neurological conditions such as traumatic brain injury (McHenry, 2003). Similar to findings on limb motor development and learning, a decline in movement variability has been reported over the course of speech development and in response to speech motor learning (e.g., Green et al., 2002; Sadagopan & Smith, 2013; Walsh, Smith, & Weber-Fox, 2006).

With regards to ALS, however, previous findings on speech motor control have not been congruent with those documented in the limb literature. Specifically, in contrast to increased variability of walking patterns in individuals with ALS (Hausdorff et al., 2000), lip and jaw movement variability during speech was significantly lower in talkers with ALS compared to their healthy peers (Mefferd, Pattee, & Green, 2014). Although reduced variability has also been recognized as a symptom of motor pathology (Stergiou & Decker, 2011), it is intriguing that ALS would impact limb and speech movements differentially with increased movement variability in limbs but decreased variability in the bulbar system. It is possible that movement variability changes over the course of the disease with early stages being

characterized by abnormally low movement variability (as reported in Mefferd et al., 2014 for mildly affected persons with ALS) and later stages being characterized by abnormally high movement variability (as reported by Hausdorff et al., 2000 for more severely affected persons with ALS). However, speech motor control has rarely been studied across the various stages of speech deterioration and it is currently unknown if speech movement variability changes with speech impairment severity.

Speaking Rate Effects on Speech Movement Variability

Speaking rate is known to decline before speech intelligibility declines in talkers with ALS (Green et al., 2013). Research suggests that the slowed rate of speech may yield a compensatory role for speech production particularly, when bulbar impairment is relatively mild. For example, researchers have observed significantly reduced lip and jaw movement variability in talkers with mild dysarthria due to ALS during habitual speech compared to their healthy peers; however, when these talkers were asked to produce speech at their maximum rate, lip and jaw movement variability significantly increased whereas speech movement variability of healthy controls remained unaffected by the increase in speech rate (Mefferd et al., 2014). Interestingly, the authors also reported a significant increase in lip and jaw movement variability when talkers with ALS repeated the utterance at a speaking rate that was twice as slow as their habitual rate; a detrimental effect of slow speech that is commonly observed in healthy talkers (e.g. Kleinow et al., 2001; Mefferd et al., 2014; Smith et al., 1995). These rate-dependent findings suggest that speech motor performance observed during habitual yet slow speech rate in talkers with ALS may not provide a comprehensive account of their motor control challenges. Such insights on the effects of speech rate adjustments are also of clinical significance because speaking rate manipulations are commonly used to increase speech intelligibility and/or naturalness in individuals with mild to moderate dysarthria particularly, slowly progressing or nonprogressive dysarthrias (Yorkston et al., 2007).

Articulatory Control of the Tongue, Lower Lip, and Jaw during Speech

To date, rate-dependent changes in movement variability have only been examined in the jaw and lower lip; articulators that are known to be less affected by ALS than the tongue (DePaul & Brooks, 1988; DePaul et al., 1993). However, very little is known about tongue movement variability during typical speech and in response to speaking rate manipulations. The fact that the tongue is anatomically coupled with the jaw, however, makes it challenging to address this gap in the literature. Because of tongue-jaw coupling, changes in tongue position can, at least in part, be influenced by changes in jaw position. The relative contribution of the jaw is not trivial when examining speech movement variability because the jaw is known to produce less variable movement patterns than the lower lip (e.g., Green et al., 2002; Mefferd et al., 2014; Walsh & Smith, 2002) and also likely the tongue. If, as suggested by some kinematic studies, the jaw is more active during speech in talkers with ALS than controls (Hirose et al., 1982; Shellikeri et al., 2016; Yunusova et al., 2008), then the overall variability of tongue-jaw movements may be lowered by the stabilizing effect of the jaw in talkers with ALS than in healthy controls.

Despite methodological challenges to isolate the tongue from the jaw, important insights can be gained from observations of coupled tongue-jaw as well as lip-jaw movements during speech. Comparing such naturally occurring speech movements between talkers with ALS and their healthy peers will advance knowledge about the ability of impaired talkers to produce consistent changes in vocal tract configurations across repeated productions of the same utterance. Further, comparing the variability between coupled tongue-jaw, lower lip-jaw, as well as independent jaw movements will be an important starting point to improve our understanding about articulator-specific motor abilities in ALS.

Purpose of the Study and Research Hypotheses

The purpose of this study was to determine differences in tongue, lower lip, and jaw movement variability of talkers with ALS and their healthy peers during typical speech and in response to speaking rate manipulations. With this investigation, we aimed to replicate previous findings of speaking rate effects on lip and jaw movement variability in talkers with mild dysarthria due to ALS (Mefferd et al., 2014) and expand this line of research to establish first insights into rate effects on tongue movement variability¹. Finally, this study sought to provide preliminary data on changes in articulatory movement variability across dysarthria severity. To address this aim, the strength of association between articulatory variability and speech impairment severity indexed by a) speech intelligibility and b) speaking rate of the target sentence was examined.

Based on previous findings on speech movement variability in persons with mild dysarthria, we expected significantly reduced variability of the lower lip and jaw in talkers with mild to moderate speech decline relative to their healthy peers during typical speech (Mefferd et al., 2014). For tongue movement variability, between-group differences were difficult to predict due to the lack of research available on tongue motor control in this clinical group. With regard to speaking rate, an increase in movement variability was expected for fast speech in talkers with mild-moderate ALS but not for controls. For slow speech, however, an increase in movement variability was expected for both groups of talkers. Based on previous findings similar rate effects were expected for the jaw and lips; however, no predictions could be made for rate-dependent effects on the tongue. Finally, we expected a strong positive relation between speech movement variability and speech impairment severity (indexed by speaking rate and speech intelligibility), based on findings that show that variability of lower limb movements during walking is strongly related to the severity of the lower limb motor impairment. Because speaking rate is known to be more sensitive to speech decline than speech intelligibility, we expected the strength of relation between speech movement variability and speech intelligibility to be weaker than those of speech movement variability and speaking rate.

¹For the rest of the article we will refer to the coupled lip-jaw and tongue-jaw movements as lower lip and tongue movements even though these movements include the contributions of the jaw.

2. Methods

2.1 Participants

The experimental group comprised of 10 people with definite ALS (7 males; 3 females) who displayed mild to severe dysarthria at the time of assessment and presented with either bulbar- or spinal-onset ALS (see Table 1). Participants in the ALS group reported no comorbid neurological diagnoses such as Parkinson's disease, stroke, or traumatic brain injuries. Prior to diagnosis, individuals in the ALS group had no history of speech, language, and hearing problems. Eleven age and gender matched healthy individuals (7 males, 4 females) who had no history of neurological, speech, language, hearing, and cognitive problems comprised the control group. In addition to self-reports, we also administered the Montreal Cognitive Assessment (Nasreddine et al., 2005) on all the controls (mean score = 25.09; SD = 1.81) and 6 of 10 participants with ALS (mean score = 23.5; SD = 4.09). For 2 of the 10 participants with ALS we administered the Mini Mental Status Examination (Folstein & Folstein, 2012) and the two participants passed with 30/30 points. Cognitive screenings were included because frontotemporal dementia is known to occur in a subgroup of persons with ALS (Strong et al., 2003). Therefore, it was important to ensure that none of the participants had a significant cognitive impairment. For the remaining two participants with ALS a formal cognitive test score was not available but the patients and family members denied a cognitive impairment. Hearing was determined to be within normal limits through a hearing screening or self-reports of normal hearing when an audiometer was not available. All participants followed conversations and task instructions at normal loudness levels without any signs of hearing problems. None of the participants reported wearing hearing aids.

Table 1 displays the intelligibility scores and speaking rates of participants with ALS and the means of these measures for the control group. Both measures were calculated from sentences recorded as part of the Sentence Intelligibility Test (SIT; Yorkston, Beukelman, Hakel, & Dorsey, 2007). More detailed information about how these measures were obtained is available under section 2.5.3. It can be seen that the last two talkers (ALS 9 and ALS 10) were more severely impaired in their speech compared to the rest of the talkers in the ALS group.

2.2 Experimental Tasks

Participants were asked to produce 12 consecutive repetitions of the sentence '*Say that I owe you a yo-yo today*' at habitual, slow, and fast speaking rates. For the habitual rate, subjects were asked to repeat the sentence at the rate and loudness that they use for everyday conversations. For the slow condition, participants were asked to reduce their speaking rate to half their habitual rate whereas for the fast condition they were asked to go as fast as possible. No specific instructions were provided for loudness but feedback about loudness was provided if, for example, talkers spoke very softly during the slow condition or very loudly during the fast condition. In such instances, subjects were reminded to maintain habitual loudness levels despite the change in rate. Task order was mixed across participants with regards to fast and slow speech. Typical speech was always elicited first. None of the study participants had difficulty following instructions for the various rate conditions and

participants were able to adjust speaking rate consistently across all the 10 repetitions of each condition. The target sentence was developed for a different study to examine speech kinematics and acoustics simultaneously in an utterance loaded with vowels in order to track formants continuously. Although the sentence was not specifically designed for the current project, the utterance was deemed suitable for the purpose of this study because it can elicit movements that are of similar complexity (peaks and troughs) across all articulators of interest (tongue tip, tongue dorsum, lower lip, and jaw).

2.3 Kinematic Data Acquisition

Tongue movement data was acquired by the first author using the Wave Speech Research System (NDI Inc., Canada) for all the healthy controls and 7 of 10 subjects with ALS. For the remaining three subjects with ALS movement data was collected by the second author using the AG501 (Carstens Medizinelektronik, GmbH). The dynamic tracking accuracy for both articulographs is comparable and is less than 0.5 mm (Berry, 2011; Stella et al., 2013). Eight sensors were affixed to the orofacial region including three on the tongue, one on the midline of the lower lip at the vermillion border, and two on the mandible just below the canines on the right and left side. The three tongue sensors were affixed using non-toxic glue (Glustitch Inc.) to the tongue tip (1 cm from the tip), tongue blade (2.5 cm from the tip), and tongue dorsum (4 cm from the tip). In some patients with ALS, only two sensors (T1 and T3) were attached to the surface of the tongue to reduce the time for experimental setup. Because T2 data was unavailable for some of the subjects, only data from the tongue-tip (T1) and dorsum (T3) were included in the analysis. Head sensor(s) served as reference(s) to create a local coordinate system to express movement from each of the orofacial sensors in the x, y, and z axis. For sessions using the Wave system, one 6DOF (degrees of freedom) head sensor was taped to the forehead using double-sided clear tape so that the orientation of the sensor could be monitored during the session (see Figure 1a, b, & c). For sessions using the AG501, three head sensors were attached to a pair of plastic goggles, which were then worn by the participant during the data collection. The sampling rate for the movement data was 400 Hz (Wave System) and 250 Hz (AG501). A microphone placed about 20 cm from the mouth for each participant allowed for simultaneous capture of audio and tongue movement data during sentence productions. The sampling rate for the audio data was 22,050 Hz (Wave System) and 44,100 Hz (AG501).

2.4 Data Analysis Procedures

The recorded lower lip and tongue movements also included contributions of the jaw due to their anatomical coupling. In previous kinematic studies, lower lip movements have been decoupled from jaw movements by linear subtraction (e.g. Grigos & Patel, 2007; Mefferd et al., 2014). However, decoupling tongue and jaw movements is a much more complex undertaking. One reason is the jaw's pitching rotation, which can result in a greater relative contribution of the jaw on anterior tongue movements than on posterior tongue movements (Westbury, 2002). Furthermore, tongue segments (tongue tip, tongue blade, tongue dorsum) differ in the way they are anatomically coupled with the jaw; for example, the tip of the tongue moves more independently from the jaw than the tongue blade and tongue dorsum. Thus, linear subtraction of jaw movements from tongue movements is not possible because it can introduce large errors (Westbury, 2002). One way to experimentally decouple tongue

and jaw is to ask talkers to produce speech while fixating the jaw with a bite block. However, bite block speech is not typical speech and will not allow insights into typical speech motor performance because the tongue has to adapt to an unnatural jaw position. Therefore, the recorded lower lip and tongue movement trajectories used for the purpose of this study were composite movements that included the contributions of the lower lip and jaw as well as tongue and jaw.

For each speaking rate condition, a single file containing tongue, lower lip, and jaw movement and audio data of all 12 sentence repetitions was loaded in SMASH which is a MATLAB-based software tool that allows us to view and parse kinematic data (Green, Wang, & Wilson, 2013). A 10 Hz LP filter was applied to all movement data. The vertical displacement of the tongue tip sensor was used to determine the onset and offset points to segment each repetition of the sentence. More specifically, for the sentence '*Say that I owe you a yo-yo today*', displacement of the tongue tip sensor in the y-dimension was used to parse out '*I owe you a yo-yo*' from the carrier phrase by using peak displacement of the final/t/in *that* (onset marker) and peak displacement of the initial/t/in *today* (offset marker; see Figure 2a). Once the individual sentences were segmented from the train of consecutive repetitions for each condition, the parsed files were loaded in SMASH to compute spatiotemporal variability of articulatory movement patterns for each rate condition (see Figure 2b). Because the spatiotemporal variability index (STI) estimates the consistency of tongue movement patterns over repeated productions, inconsistent utterances characterized by prolonged pauses, false starts, and/or interruptions of vocal quality were excluded from the STI analysis. A minimum of 9 repetitions was included for the STI analysis for all subjects.

2.5 Measurements

2.5.1 Spatiotemporal Variability Index (STI)—STI analysis was conducted on the articulatory movements in the vertical dimension because this is the primary axis for vocal tract constrictions during speech. The STI was calculated as previously described (e.g., McHenry, 2003; Mefferd et al., 2014; Smith et al., 1995).

Specifically, articulatory movement trajectories for the 12 repetitions of each word were time and amplitude normalized (see Figure 3). For time normalization, a cubic spline procedure was used to adjust each movement trajectory onto a constant axis length of 1000 points. Amplitude was normalized by subtracting the mean of the displacement signal and dividing by its standard deviation (SD). Finally, the SD of the 12 normalized articulatory movement trajectories was calculated at fixed 2% intervals in relative time. The 50 SD were added to get the movement variability for each condition. A high STI score indicates high movement variability whereas a low STI score indicates less movement variability.

2.5.2 Sentence Durations—Sentence durations (time between onset and offset of movement in seconds) were used to evaluate speaking rate modulations.

2.5.3 Speech Intelligibility and Rate—The SIT (Yorkston et al., 2007) was used to calculate sentence intelligibility and speaking rate for each individual. Once the samples were collected, three listeners (student assistants) naïve to the study were asked to listen to

each sentence and transcribe the sentences exactly as they heard them. Intelligibility was calculated by counting the number of correctly identified words and dividing by the total number of words possible, multiplied by 100. Speaking rate was calculated in words per minute by dividing the total number of words attempted by the total time taken to produce the sentences (converted from seconds to minutes). Intelligibility scores and speaking rate measures were averaged for each participant based on the scores provided by the three student assistants. Inter-rater reliability was determined for intelligibility and speaking rate data from the mild-moderate speakers using Spearman's rank order correlation. There was a strong agreement between raters for sentence intelligibility ($r_s = .82$, $p = .05$) and speaking rate ($r_s = .95$, $p = .01$). For the severe talkers, inter-rater reliability of speaking rate and intelligibility was determined by calculating the absolute mean difference across the three analysts. Absolute mean difference was 2.42 for sentence intelligibility ($SD = 1.72$) and 4.72 for speaking rate ($SD = 1.90$).

3. Statistical Analysis

The data of the two severely impaired talkers were excluded from the statistical analysis of speaking rate effects on movement variability because their values stood out as outliers relative to the rest of the ALS group means. Therefore, only data from talkers with mild to moderate dysarthria ($n = 8$) and age- and sex-matched controls were used to examine speaking rate effects on speech movement variability. However, within this severity range the examination of rate effects is particularly relevant because a) speaking rate adjustments are commonly used to maximize intelligibility and/or naturalness in these talkers whereas more severely impaired talkers are trained on communication devices and b) findings of this study can be compared to previous findings on talkers with mild to moderate dysarthria to achieve convergence and expand upon previously established knowledge.

Although eight talkers with mild to moderate dysarthria due to ALS and eleven controls were enrolled, the sample size within each group varied slightly due to sensor tracking errors. Due to the relatively small number of participants within each group, non-parametric statistics were completed. Between-group effects were tested using Mann Whitney U tests, within-group effects were evaluated using Friedman's test for each group, and post-hoc simple comparisons were conducted using Wilcoxon Signed Rank Tests.

Our preliminary analysis of the association between speech movement variability and overall speech decline included the two talkers with severe dysarthria in addition to the talkers with mild and moderate dysarthria ($n = 10$). The strength of relation between movement variability and speech severity was evaluated by submitting STI values of typical speech and measures that index speech decline (i.e., speech intelligibility and speaking rate) to bivariate correlational analyses for each articulator.

4. Results

4.1 Task Performance

Figure 4 displays each group's mean target utterance durations for each speaking rate. Findings revealed a significant effect of speaking rate on target utterance durations for the

ALS group [$\chi^2(2) = 14.250, p < .001$] and the control group [$\chi^2(2) = 22.000, p < .001$]. Within each group, pairwise comparisons of speaking rate conditions were all significant at a p -level of $p = .017$ in the ALS group and $p = .003$ in the control group. Further, the between-group effect was significant for typical ($U = 9.000, p = .003$) and fast speech ($U = 6.500, p = .002$); no significant between-group difference was found for slow speech.

4.2 Task Effects on Movement Variability

4.2.1 Tongue movement variability—For tongue tip movement, no significant within or between-group effects were found for speaking rate in talkers with ALS or controls.

However, as can be seen in Panel A of Figure 5, STI values of talkers with ALS tended to be lower than those of controls, particularly during typical speech. For the tongue dorsum, the effect of speaking rate was approaching significance for talkers with ALS [$\chi^2(2) = 5.429, p = .066$] but not for controls. As Panel B of Figure 5 indicates STI values of controls did not significantly vary with speaking rate, whereas for talkers with ALS slow speech tended to yield higher STI values than fast and typical speech. With regards to between-group differences, talkers with ALS had significantly lower tongue dorsum STI values than controls during typical speech ($U = 5.000, p = .002$) and fast speech ($U = 13.00, p = .033$), but not during slow speech.

4.2.2 Lower lip movement variability—For lower lip movements, a significant within-group speaking rate effect was found for talkers with ALS [$\chi^2(2) = 7.000, p = .030$], but not for controls. As can be seen in Panel C of Figure 5, fast and typical speech yielded significantly lower STI scores compared to slow speech in the ALS group ($p = .025$). By contrast, STI scores did not vary with rate in the control group. A significant between-group effect was observed for typical speech ($U = 15.000, p = .027$). Further, as can be seen in Panel C of Figure 5, talkers with ALS tended to have lower STI scores of lower lip movements than controls during fast speech.

4.2.3 Jaw movement variability—Finally, for jaw movements, no significant within-group speaking rate effects were found for talkers with ALS or controls. However, as can be seen in Panel D of Figure 5, STI scores tended to be higher during slow speech than typical and fast speech within the ALS group. Between-group differences were significant for typical speech ($U = 19.00, p < .041$). Further, jaw STI values also tended to be lower in talkers with ALS compared to controls during fast speech ($U = 22.000, p = .075$).

4.3 Associations between Impairment Severity and Movement Variability

Panels A–D of Figure 6 displays the associations between the sentence intelligibility scores indexing speech severity and the STI scores of each articulator. STI scores were available for the two talkers with severe dysarthria for the tongue tip, tongue dorsum, and lower lip, but the jaw STI score was available only from one talker with severe dysarthria. The relations between speech impairment severity and speech movement variability was evaluated by submitting the STI score for typical speech and each talker's corresponding sentence intelligibility score (an index of speech severity) to a curve estimation analysis. Sentence intelligibility scores were log-transformed to reduce the skewness of the sample.

Linear fits and exponential fits yielded virtually identical results; therefore, we selected to report results for linear fits. Significant relations were found for sentence intelligibility and STI scores of the tongue tip ($R^2 = .47$, $p = .034$), tongue dorsum ($R^2 = .49$, $p = .037$), as well as the lower lip ($R^2 = .58$, $p = .01$). However, no significant associations were found for sentence intelligibility and jaw STI scores.

Associations between speaking rate and STI scores could be best described by non-linear, exponential fits. As can be seen in Figure 7, nonlinear relations were strongest for speaking rate and STI scores of the tongue tip ($R^2 = .82$, $p < .001$) and tongue dorsum ($R^2 = .82$, $p = .001$), followed by relations between speaking rate and STI of the lower lip ($R^2 = .56$, $p = .013$), and the jaw ($R^2 = .47$, $p = .04$).

5. Discussion

The primary aim of this investigation was to determine between and within-group differences in speech movement variability for typical speech and in response to speaking rate adjustments. A secondary aim was to determine if speech movement variability was associated with speech impairment severity as indexed by intelligibility, rate, and utterance duration. Our main findings were (i) during habitual speech talkers with mild to moderate dysarthria had lower tongue and lip movement variability compared to the control group, (ii) for slow speech, the ALS group had significantly greater tongue and lip movement variability; however, no significant rate effects were observed for the controls, and (iii) speech severity indexed by speaking rate and intelligibility showed significant associations with tongue and lip movement variability during habitual speech for the ALS group. However, the findings on the associations between speech impairment severity measures and STI values should be considered preliminary because only two severely impaired talkers were included in this analysis and had a heavy influence on the strength of association.

Reduced Spatiotemporal Variability during Habitual Speech in Mild to Moderate Dysarthria

As expected, we found decreased tongue and lip movement variability during habitual speech in talkers with ALS compared to typical controls. A similar decrease in variability has also been observed previously in talkers with mild dysarthria due to ALS who produced highly consistent lip and jaw movements during typical speech (Mefferd et al., 2014). Historically, relatively low movement variability has been associated with a healthy motor system whereas high spatiotemporal variability during habitual speech has been associated with disordered or aging speech motor systems (e.g., Kleinow et al., 2001; Smith et al., 1995; Wohlert & Smith, 1998). More recent discussions, however, suggest that there is an optimal state of variability associated with typical motor performance, for example during speech or locomotion, and that a deviation from this state due to increased articulator stiffness or a rigid type of articulation can lead to decreased movement variability (Dromey, Boyce, & Channell, 2014; Stergiou & Decker, 2011). In fact, studies on stroke patients suggest that fixation of specific body segments may be an adaptive strategy to achieve motor control (Bernstein, 1967; Vereijken, van Emmerik, Whiting, & Newell, 1992). Although this compensatory strategy is implemented to improve motor control, it has been shown to limit mobility in stroke patients (Cirstea & Levin, 2000). Prior speech kinematic research in

talkers with ALS indirectly supports this notion that increased rigidity results in reduced movement. Kuruvilla and colleagues demonstrated that individuals with moderately impaired speech who are likely to use compensatory strategies to cope with rapid speech decline, showed the greatest reduction in independent movements of tongue segments (i.e., tongue tip, tongue base, tongue dorsum) even in comparison to talkers with severe dysarthria (Kuruvilla, Green, Yunusova, & Hanford, 2012).

Although low spatiotemporal variability was observed in talkers with relatively mild speech impairment, the preliminary findings of the two talkers with severe dysarthria who were included in the regression analysis suggest that articulatory movements become highly variable when speech is severely affected as can be seen in the trendline analysis of speech severity measures and STI values in which the two severely impaired talkers are included (Figures 6 & 7). The increased speech movement variability seen in more severely affected talkers with ALS is congruent with previous findings on limb movement variability in more severely affected persons with ALS (Hausdorff et al., 2000). This change from pathologically low to high variability with disease progression may be explained by the inability to make compensatory adjustments; for example rigid articulatory behaviors or increased jaw contributions to speech production. Such strategies have been discussed as potential facilitators of motor control during the early stages of speech decline (Green et al., 2013; Kuruvilla et al., 2012). However, their implementation may become increasingly difficult with disease progression. Further, a disease-related increase in neuromuscular noise, muscle weakness, and atrophy which are thought to negatively impact movement variability may further explain the significant increase in variability at later disease stages (Schmidt et al., 1979; Wohlert & Smith, 1998).

In ALS, the hypoglossal (lingual) motor neurons are considered to be disproportionately affected compared to the trigeminal and facial motor neurons that innervate the jaw and lips, respectively (DePaul & Brooks, 1988; DePaul et al., 1993). Because the tongue and lower lip were not decoupled from the jaw in this study, it is possible that the performance of the jaw dominated the performance that was observed for the tongue and the lips. Therefore, the reduced tongue and lip variability observed in talkers with ALS during typical speech may result from greater relative contribution of the jaw that may facilitate lower tongue and lip movement variability relative to healthy talkers. Increased reliance on the jaw has been reported for talkers with ALS previously; for example, in terms of greater jaw movement speeds in talkers with a perceivable speech impairment compared to talkers with no perceivable speech impairment (Green et al., 2013).

Speaking Rate Effects on Movement Variability

In both groups, our hypothesis about speaking rate effects was partially supported. That is, in talkers with ALS, slow speech elicited the predicted effect on tongue and lip movement variability whereas fast speech did not. The elevated movement variability for slow speech seen in our talkers with ALS has been documented in prior speech motor control studies on both healthy and disordered talkers (Kleinow et al., 2001; Mefferd et al., 2014; Smith et al., 1995; Wohlert & Smith, 1998). For the slow speech condition in the current study, participants were asked to reduce their rate to half their typical rate. In addition, they were

specifically instructed to prolong each word so that articulation time rather than pause time was significantly prolonged for the slow speech condition. Thus, the cognitive load to monitor speaking rate manipulation was likely greater during slow speech than fast speech. According to Dromey (2000), the increased attentional demands required to constantly monitor articulatory movements during atypical conditions such as slow speech may overburden the finite pool of resources allocated to speech production. The reallocation of resources to meet attentional demands could have a detrimental effect on speech performance in talkers with neurological conditions like ALS (Bunton & Keintz, 2008; Dromey & Bates, 2005; Dromey & Benson, 2003; Dromey & Shim, 2008; Radovanovic et al., 2014). As observed in this study, slow speech can negatively affect an impaired talker's ability to produce consistent articulatory movements and may not be suited as a treatment option to improve speech intelligibility.

Findings for the fast rate effect on articulatory control were in contrast to findings that describe fast speech as an unstable mode of speech associated with higher STI scores in persons with ALS (Mefferd et al., 2014). However, fast speech effects have not been robust in healthy controls in previous studies. For example, STI scores of lower lip movements were elevated during fast speech in one study (McHenry, 2003), reduced in another (Mefferd et al., 2014), and even comparable to habitual speech in a third study (Kleinow et al., 2001). Based on previous and current findings of fast speech effects in talkers with ALS, it is likely that an accelerated rate does not yield robust effects on speech movement variability in this clinical group either. One reason for these variable effects on speech motor control may be the different articulatory strategies speakers may use to achieve fast speech. For example, it has been shown that some talkers prefer to increase articulatory speed to achieve faster speech whereas other talkers prefer to reduce their articulatory amplitude (e.g. Kuehn & Moll, 1976). More research is needed to further investigate how talkers with ALS modulate articulatory movements to meet rate demands and the effects of various speaker strategies on speech movement variability. Most importantly, however, findings on fast speech effects question the idea that the habitually slow speech evident in talkers with ALS is a compensatory mechanism for motor control. If the habitually slow speech rate is indeed a compensatory mechanism adopted to improve speech motor control, precision, and intelligibility then movement variability should increase from the typical to fast condition because the strategy to stabilize the motor system is not available to the speaker anymore.

In controls, no significant speaking rate effects were observed for any articulator in the current study. This finding was surprising given the relatively robust effects of slow speech on movement variability in the literature on controls (e.g., Kleinow et al., 2001; Smith et al., 1995; Wohlert & Smith, 1998). However, it should be noted that in the present study movement variability was already relatively high during typical speech for controls. That is, in comparisons to other studies (e.g. Kleinow et al., 2001; Mefferd et al., 2014; Wohlert & Smith, 1998) that report mean STI scores below 20 for typical speech, the mean STI score in the present study were approximately 25 across all articulators. It is therefore possible that the effect of slow speech was dampened by relatively high variability during typical speech in controls. Relatively high movement variability during typical speech may be explained by the nature of the speech utterance which predominantly contains vowel and semivowel sounds but lacks stop consonants (e.g. in comparison to "Buy Bobby a puppy"). It is

possible that the production of consonants throughout an utterance may facilitate stability by providing relatively concrete articulatory targets (bilabial closure). It is important to note that movement variability during typical speech was also greater for talkers with ALS in the current study compared to the variability scores reported in the previous study (Mefferd et al., 2014). Therefore, between-group differences that were observed in this study were not driven by unusual high movement variability in the controls. Future studies should expand upon findings of the current study by including utterances that specifically target each articulator (bilabial for lip movements, alveolar sounds for tongue tip movements, and palatal and velar sounds for tongue blade movements).

Movement Variability as an Index of Speech Impairment Severity

Variability of gait dynamics has been shown to serve as a predictor of limb function impairment such as walking ability and fall risk (Chau, Young, & Redekop, 2005; Hausdorff, 2005; Stergiou, Harbourne & Cavanaugh, 2006). In speech, several measures are used to indicate impairment severity, most commonly speech intelligibility and speaking rate. In order to gain preliminary insights on the relations between STI scores and clinical speech measures that index speech impairment severity we examined the associations between STI and intelligibility and rate during habitual speech. Associations between intelligibility and STI scores are difficult to interpret because the two severe talkers dominate the nature and strength of the regression analysis. However, a visual inspection of the data dispersion (see Figure 6) suggests that ceiling and floor effects of perceptually-based severity ratings result in rather weak associations between the speech intelligibility as a speech severity index and speech movement variability in this clinical group. By contrast, the two severe talkers have less dramatic influence on associations between speaking rate and speech movement variability (particularly the tongue tip and tongue dorsum STI; see Figure 7) suggesting that with speaking rate decline, speech movement variability increases. Due to the small number of participants however, this finding should be considered preliminary and further studies are warranted to replicate these findings. We also examined the associations between STI and articulation rate (overall speech time minus pause times larger than 200ms; Green, Beukelman, & Ball, 2004) to determine if the correlations between speech rate and STI were influenced by extensive pause times in the severely impaired talkers. However, correlations of articulatory rate and STI were almost identical to those of speaking rate and STI values.

Speaking Rate Adjustments

As expected, both the healthy controls and talkers with mild to moderate dysarthria were able to manipulate their typical speaking rates to produce the target sentence with significantly longer as well as shorter durations for the slow and fast conditions, respectively. During fast speech, both groups produced the target sentence 79% faster than their typical rate. Even though both groups increased duration by the same percentage for fast speech, the ALS group was significantly slower than the healthy controls. For the slow condition, the controls slowed their speech by 360% while the individuals with dysarthria slowed down by 240%. Despite these differences, both groups had comparable mean durations for slow speech. Similar findings were reported by Kleinow et al. (2001) who showed an 80% increase in speaking rate for fast speech and a 190% slowing for slow speech by their

Parkinson's group. McHenry (2003) reported an 82% increase in speaking rate and a 216% slowing of rate compared to typical rate in talkers with mild dysarthria secondary to TBI. This disproportionate adjustment in duration for slow compared to fast speech suggests that both ALS and typical talkers are operating close to the upper limits of their physiological capabilities at typical rates (Jaeger et al., 2000; McHenry 2003). Alternatively, it also suggests that there may be more constraints imposed on the speech motor system by fast compared to slow speech which may limit the extent to which duration can be manipulated in the fast condition.

Limitations

It is important to point out that despite the relatively small sample size statistical significance was reached for the between-group comparisons of the tongue dorsum STI, lower lip STI, and jaw STI at habitual speech. Yet, the between-group comparison for the tongue tip STI measure for the habitual and fast speech conditions as well as the lower lip STI measure for the fast speech condition remained non-significant. Given the small sample size, statistical power was too low for these between-group differences to reach statistical significance (e.g., only 26% power for comparisons of tongue tip STI at habitual rate). To mitigate the low power and reach statistical significance, a sample size of at least 37 talkers with ALS would be required (if critical p -value is set to 0.05). Tongue kinematic data of a sample this size is very difficult to obtain given that the disease is relatively rare and that individuals with ALS progress relatively fast once speech production deteriorates. It should also be mentioned that critical p -value were not adjusted for multiple between-group comparisons. Given the fact that the significant between-group effects reported in the present study were congruent with those of a previous study on lip and jaw movement variability, the risk of falsely accepting our research hypotheses to be true is relatively low. Nevertheless, the importance of replicating findings from the present study cannot be emphasized enough as the statistical power of studies on tongue kinematics in ALS is typically low.

6. Conclusions

During typical speech, talkers with mild-moderate dysarthria had less variable tongue and lip movement patterns in comparison to healthy controls. Even though the underlying mechanisms of reduced variability are poorly understood, there are several possibilities for pathologically low articulatory variability such as (i) rigid articulation resulting from compensatory adjustments to stabilize the speech motor system, (ii) increased spasticity or stiffness as a result of upper motor neuron involvement, and/or (iii) greater jaw involvement as a compensatory response to reduced tongue movement. With regard to speaking rate adjustments, slow but not fast speech resulted in a significant increase of movement variability in talkers with ALS. Our results suggest that movement variability shows promise as an index of motor impairment severity and is expected to increase with dysarthria severity in talkers with ALS. Further support for this stems from the strong associations between STI scores and speaking rate, which is known to be a sensitive indicator of bulbar deterioration.

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Highlights

- Reduced movement variability seen in ALS compared to controls for habitual speech
- Predicted increase in movement variability for slow speech was observed in ALS group
- Fast speech did not elicit predicted increase in movement variability for ALS group
- Moderate to strong associations for habitual speaking rate and movement variability

Figure 1(a). Setup of the Wave Speech Research System (NDI Inc.)

Figure 1(b). Placement of jaw right (JR), jaw left (JL), and lower lip (LL) sensors

Figure 1(c). Placement of tongue sensors T1 (tip), T2 (mid), and T3 (back)

Figure 1.
Setup of the Wave Speech Research System

Figure 2.

Figure 2(a). Tongue onset and offset markers used to parse the target sentence from the carrier phrase. 2(b) Vertical movement pattern of the tongue dorsum for a single repetition (above) and 10 repetitions of the target sentence (below).

Figure 3. An example of amplitude and time-normalized vertical displacement trajectories of the lower lip (LLy), tongue-tip (T1y), tongue-back (T3y), and jaw (JLy) for ‘*I owe you a yo-yo*’ taken from a control participant.

Figure 4. Mean target utterance durations ($\pm$ *SD*) of talkers with mild to moderate dysarthria due to ALS and controls across all three speaking rates. Brackets indicate significant between-group comparisons. All pairwise comparisons were significant within both groups. Note: * $p < .05$.

Figure 5.

Mean STI scores ($\pm$ SD) of talkers with mild to moderate dysarthria due to ALS and controls across all three speaking rates. Solid brackets indicate significant between-group comparisons. The dashed bracket indicates a between-group comparison with approaching significance ($p = .075$). For within-group pairwise comparisons please see text. Note: ** $p = .002$, * $p < .05$.

Figure 6. Associations between Sentence Intelligibility and STI scores for talkers with ALS and controls. Trendlines indicate linear fits.

Figure 7. Associations between speaking rate (based on Sentence Intelligibility Test) and STI scores for talkers with ALS and controls. The trendlines indicate exponential fits.

Table 1

Demographics, sentence intelligibility, and speaking rate of participants with ALS

Group	Age (years. months)	Sentence Intelligibility (%)	Speaking Rate (WPM)
	Mean (SD)	Mean (SD)	Mean (SD)
Controls	58.24 (10.73)	98.48 (2.17)	197.60 (30.90)
ALS	61.16 (10.59)	82.42 (17.51)	136.68 (54.97)
ALS 1	54.06	98.48	230.27
ALS 2	52.05	97.27	150.52
ALS 3	61.06	95.91	116.28
ALS 4	73.09	86.97	116.63
ALS 5	65.10	96.33	164.39
ALS 6	64.06	80.30	121.88
ALS 7	68.04	79.71	137.98
ALS 8	52.00	71.09	141.68
ALS 9	78.08	55.45	79.57
ALS10	44.07	48.79	33.42