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Differential effects of velopharyngeal dysfunction on speech intelligibility during early and late stages of Amyotrophic Lateral Sclerosis

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Abstract

The detrimental effects of velopharyngeal dysfunction (VPD) on speech intelligibility in persons with progressive motor speech disorders are poorly understood. In this study, we longitudinally investigated the velopharyngeal and articulatory performance of 142 individuals with varying severities of amyotrophic lateral sclerosis (ALS). Our goal was to determine the mechanisms that underlie the effects of VPD on speech intelligibility during early and late stages of ALS progression. We found that during the early stages of the disease, the effect of VPD on intelligibility was partially mitigated by an increase in articulatory (e.g., lower lip and jaw) movement speed. This apparent articulatory compensation eventually became unavailable during the late stages of disease progression, which led to rapid declines of speech intelligibility. The transition across the early and late stages was characterized by the slowing of the composite movement of lower lip and jaw below 138 mm/s, which indicated the onset of precipitous speech decline and thus, may provide important timing information for helping clinicians to plan interventions.

Index Terms: velopharyngeal dysfunction, articulatory compensation, speech intelligibility, amyotrophic lateral sclerosis

1. Introduction

Velopharyngeal dysfunction (VPD) is a speech disorder caused by the failure of proper closure of the velopharyngeal port, which can result from anatomic defects such as cleft palate or neuromuscular disorders that affect the vagus nerve and/or the relevant muscles. Prior research on VPD has established its global impact on speech acoustics (e.g., reduced oral acoustic energy, altered spectral structures, increased nasal resonance, etc.), which combined have an impact on the acoustic distinctiveness of phonemes [1, 2] and in turn, reduce the overall speech intelligibility [3, 4]. In particular, studies of dysarthria - a motor speech disorder that typically affects multiple speech subsystems (i.e., articulation, resonance, phonation, and respiration) - identified VPD as one of the most detrimental factors that contribute to the overall decline of speech intelligibility, and the reduction of phonetic contrasts underlying speech intelligibility loss [3, 4, 5]. Despite the detrimental effect of VPD on speech intelligibility, it is largely understudied compared to the extant literature on articulatory impairments in dysarthria. To provide a more comprehensive understanding of the physiologic mechanism underlying dysarthric speech impairment, we investigated the impact of VPD on speech intelligibility in persons with amyotrophic lateral sclerosis (ALS) - a motor neuron disease that affects all of the four speech subsystems and is characterized by progressive decline of speech intelligibility.

Because of the progressive nature of ALS, understanding the pattern of disease progression is especially important for improving diagnosis and prognosis. Prior studies on the progression of subsystem impairments in ALS found that VPD has much less of an impact on speech intelligibility during the early stages of the disease than during the late stages [3, 6]. The differential effects of VPD on the early and late stages of speech intelligibility decline have two contrasting explanations: (1) velopharyngeal function only starts to decline when muscle weakness progresses to a critical level; (2) VPD in the early stages of progression is mitigated by behavioral compensations that become unavailable during the late stages. The potential for compensation is supported by previous studies on VPD resulting from cleft palate showing that the acoustic and perceptual effects of VPD on speech could be minimized using oropharyngeal articulation strategies [7, 8]. Although these articulatory strategies have not been assessed in persons with dysarthria, they led to a possible explanation for the differential effects of VPD on early- versus late-stage of speech decline in ALS.

The primary aim of this study was to determine the mechanisms that underlie the differential effects of velopharyngeal dysfunction on speech intelligibility in persons with ALS. To address this aim, we hypothesized that (1) articulatory compensations are made to partially mitigate the effect of VPD on speech intelligibility during the early stages of ALS; and (2) oropharyngeal articulatory compensation becomes unavailable during the late stages of ALS due to the decline of articulatory function, resulting in rapid declines of speech intelligibility. The findings of this study might (1) provide crucial information about the timing of critical speech changes that can assist disease monitoring and clinical speech assessment, and (2) help us understand why some individuals are capable of maintaining intelligible speech despite velopharyngeal impairments.

2. Methods

2.1. Participants

We recruited 142 participants with ALS (90 male + 52 female) aged from 40 to 88 years old ($M = 59$, $SD = 10$) and 75 age-matched healthy participants. The participants with ALS varied in both overall and bulbar (i.e., speech and swallowing) disease severity, as measured by the total score and bulbar subscore on the ALS Functional Rating Scale - Revised (ALSFRS-R), respectively. The ALSFRS-R score was obtained from 12 survey questions that assessed the degree of functional impairment with the score of each question ranging from 4 - least impaired to 0 - most impaired. The ALSFRS-R scores of the participants with ALS ranged from 15 to 48 ($M = 37$, $SD = 7$). The bulbar subscore, estimated based on the first three questions of the

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scale, assessed the bulbar function with a maximum score of 12 ranged between 5 and 12 ($M = 10$, $SD = 2$).

2.2. Data collection

All participants with ALS were studied longitudinally throughout multiple visits; the duration between the first and last visits ranged from 42 to 2100 days ($M = 485$ days, $SD = 417$ days). The healthy participants only visited once. All participants performed two different tasks during each visit. The first task was the Sentence Intelligibility Test (SIT), in which the participants read a list of 10 randomly generated sentences of varying length (5-15 words) [9]. Based on the SIT recordings, speech intelligibility (percentage of words correctly transcribed by a naive listener out of the total words produced) and speaking rate (words produced per minute [WPM]) were obtained to serve as indicators of the overall speech performance.

The second task assessed articulatory and velopharyngeal performance. In this task, the participants were asked to read the sentence "Buy Bobby a puppy" at their normal speaking rate and loudness. This sentence was used because (1) it contains a number of pressure consonants (e.g., /p/, /b/) that are known to be sensitive to VPD [10]; and (2) its articulation mainly involves lip and jaw movements, which are less affected by ALS than the tongue [11], resulting in a relatively large capacity for making adjustments to compensate for VPD.

To assess articulatory performance, one of the following two systems was used to record articulatory movements while the participants repeated the sentence for 10 times. One of the systems was a high resolution 3D optical motion capture system (Motion Analysis Corp.), which recorded the positions of a set of reflective markers attached to the participants' forehead and the vermilion border of the lower lip (LL) at a sampling rate of 120 Hz. Alternatively, a WAVE Articulography (NDI) was used to record the positions of a set of electromagnetic sensors attached to the same locations of the participants' face as described above at a sampling rate of 100 Hz. Based on the recordings, we calculated the maximum speed of the composite movement of LL and jaw (*maxspd:LL+J*; measured by the marker on LL because lower lip rides on the jaw) and averaged across the 10 repetitions for each participant to serve as an indicator of articulatory performance. To assess velopharyngeal performance, a Nasometer (Model 6400, KAYPentax) was used to obtain nasalance, which is defined as the ratio of $nasal/nasal + oral$ acoustic energy, while the participants repeated "Buy Bobby a puppy" for 3 times. We calculated the median nasalance (*med:naso*) for each sentence and averaged across the 3 repetitions for each participant to serve as an indicator of velopharyngeal performance.

2.3. Changes of velopharyngeal and articulatory performance during disease progression

To assess the changes of velopharyngeal and articulatory performance during the disease progression, we first applied a nonparametric approach (*ggplot*, *stat_summary*, R 3.2.3) to derive the average pattern of change in *med:naso* and *maxspd:LL+J* relative to the change in speaking rate across all participants with ALS. Speaking rate was used as a metric indicative of the primary disease effect on speech because previous studies of ALS suggested that, as the disease progresses over time, speaking rate declines in a roughly linear manner [12]. Based on the nonparametric pattern of change in *med:naso* and *maxspd:LL+J*, we selected either a linear or nonlinear mixed effects (LME/NLME) model (*lmer*, *nlmer*, R 3.2.3), whichever provided a better visual match with the nonparametric pattern, to quantitatively model the longitudinal changes in *med:naso* and *maxspd:LL+J* as a function of speaking rate, while accounted for inter-subject variability.

2.4. Relationship between velopharyngeal and articulatory performance during early and late stages of disease

To test the hypothesis of stage-dependent articulatory compensation for VPD, we first defined the early and late stages of ALS based on *maxspd:LL+J* using a cutoff of 138 mm/s (see Results for more details about how this cutoff was determined). Following the assignment of disease stages, we compared *maxspd:LL+J* and *med:naso* among the early ALS group (i.e., $maxspd : LL + J > 138mm/s$), the late ALS group (i.e., $maxspd : LL + J <= 138mm/s$), and the control group using ANOVAs. Post-hoc pairwise comparisons were made between each two groups using Cohen's d for any measure that showed a significant group effect.

Because the relationship between *maxspd:LL+J* and *med:naso* as found by the ANOVA and post-hoc tests above might reflect a general disease effect that underlies the co-occurring changes in articulatory and velopharyngeal performance, this confounding effect needed to be considered while interpreting the relationship between *maxspd:LL+J* and *med:naso*. Therefore, we compared speaking rate among the three groups (i.e., early ALS, late ALS, and control) using ANOVA. If the relationship between *maxspd:LL+J* and *med:naso* was due to a general disease effect, the change in speaking rate (used as an indicator of disease effect) across the three groups should follow a consistent pattern as the changes in *maxspd:LL+J* and *med:naso*.

2.5. Predicting two stages of speech intelligibility decline based on velopharyngeal and articulatory performance

To determine whether velopharyngeal and articulatory performance predict the two stages of speech intelligibility decline in ALS (i.e., early stage - slow decline; late stage - rapid decline) as found by previous studies [12, 13], we applied a bi-phasic nonlinear mixed effects (NLME) model (*nlmer*, R 3.2.3) to estimate the speech intelligibility of the 142 participants with ALS using *maxspd:LL+J* and *med:naso* as predictors, while accounted for a random effect of subject. The transition across the two stages was defined consistently with Section 2.4 by a cutoff of *maxspd:LL+J* at 138 mm/s.

As discussed in Section 2.4, changes in *maxspd:LL+J* and *med:naso* as well as the change in speech intelligibility could be partially driven by the underlying disease effect. To determine the contributions of velopharyngeal and articulatory performance to speech intelligibility apart from the disease effect, we applied another bi-phasic NLME model to predict the speech intelligibility of the participants with ALS using *maxspd:LL+J* and *med:naso* as predictors and included speaking rate as a covariate to control for disease effect.

3. Results

3.1. Changes of velopharyngeal and articulatory performance during disease progression

Figure 1 shows the change of *med:naso* with respect to speaking rate. The nonparametric approach indicated that *med:naso* increased in a roughly linear pattern as speaking rate slowed down. Therefore, an LME model was selected to predict the

changes in *med:naso* as a function of speaking rate, which accounted for 62% of the total variance in the participants with ALS ($p < 0.001$).

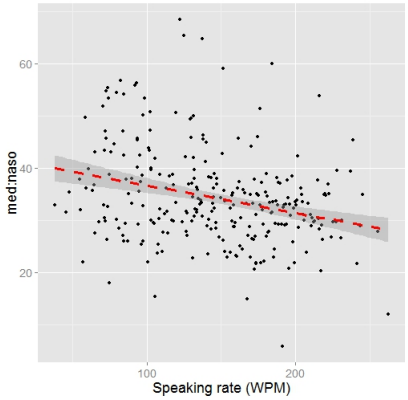

Figure 1: *Change of med:naso with respect to speaking rate in participants with ALS. The red dashed line corresponds to the fit of the LME model. The shaded area corresponds to the 95% confidence interval of the LME fit.*

Figure 2 shows the change of *maxspd:LL+J* with respect to speaking rate. The nonparametric approach derived a nonlinear pattern (blue solid line in Figure 2), which roughly corresponded to two phases with a breakpoint at speaking rate = 160 WPM (the sudden changes at the low and high boundaries of speaking rate were considered as artifacts and disregarded). Based on this pattern, we selected a bi-phasic NLME model to simulate the changes in *maxspd:LL+J* as a function of speaking rate (red dashed line in Figure 2), which accounted for 70% of the total variance in the participants with ALS. When speaking rate was above 160 WPM, *maxspd:LL+J* did not change significantly ($p = 0.218$ for β coefficient) and the average of *maxspd:LL+J* remained above 138 mm/s; and when speaking rate was below 160 WPM, the average of *maxspd:LL+J* dropped below 138 mm/s and continued decreasing as speaking rate slowed down ($p = 0.018$ for β coefficient).

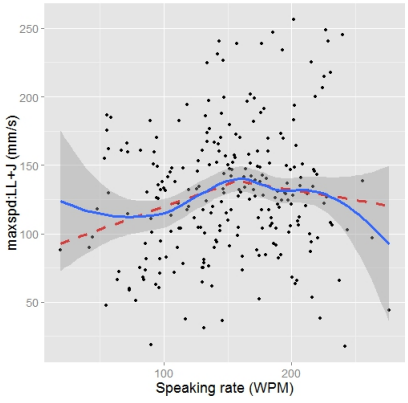

Figure 2: *Change of maxspd:LL+J with respect to speaking rate in participants with ALS. The blue solid and red dashed lines correspond to the fit of the nonparametric and NLME approaches, respectively. The shaded area corresponds to the 95% confidence interval of the nonparametric fit.*

3.2. Relationship between velopharyngeal and articulatory performance

Based on the bi-phasic model of *maxspd:LL+J* (i.e., early stages: *maxspd:LL+J*>138 mm/s; late stages: *maxspd:LL+J*<=138 mm/s), the participants in the early and late stages of ALS were stratified as the early and late ALS groups, respectively. Comparisons across the early ALS, late ALS, and control groups showed significant group effects for *maxspd:LL+J* ($p < 0.001$), *med:naso* ($p = 0.046$), and speaking rate ($p = 0.017$) (see Figure 3). Post-hoc pairwise comparisons suggested that (1) during the early stages of ALS, *maxspd:LL+J* was increased ($d = 1.2$), *med:naso* was slightly decreased ($d = -0.28$), and speaking rate was decreased ($d = -0.38$) relative to that of controls; and (2) during the late stages of ALS, *maxspd:LL+J* was decreased ($d = -1.33$), *med:naso* was slightly increased ($d = 0.14$), and speaking rate was decreased ($d = -0.41$) relative to that of controls. A comparison of the early versus late stages of ALS indicated a large increase of *maxspd:LL+J* ($d = 2.67$), moderate decrease of *med:naso* ($d = -0.45$), and no change in speaking rate ($d = 0.09$).

To verify the increase in lower lip and jaw movement speed as an articulatory strategy to compensate for VPD, we simulated the effect of increasing articulatory movement speed on the aerodynamic features of syllables /pʌ/ and /ba/ given an open velopharyngeal port, using a custom aerodynamic model of the vocal tract. This model was implemented in MATLAB, which was based on Sprouse's model [15] with a number of improvements including simulation of a more realistic vocal tract shape and refined time scale to better simulate dynamic speech. Based on the simulation, we found that the increase of articulatory movement speed by 50% resulted in (1) shorter nasal emission and (2) faster and increased oral airflow. Both changes led to reductions in nasalance and thus, verified our hypothesis.

3.3. A predictive model of speech intelligibility decline during early and late stages of ALS

A two-phase NLME model using *maxspd:LL+J* and *med:naso* as predictors accounted for 64% of the variance in the speech intelligibility of the participants with ALS. *med:naso* was significantly correlated with speech intelligibility throughout the entire disease course (β coefficient= -0.2124, $p = 0.001$). During the late stages (*maxspd:LL+J*<=138 mm/s), *maxspd:LL+J* was significantly correlated with speech intelligibility (β coefficient= 0.043, $p = 0.041$).

The two-phase NLME model that used *maxspd:LL+J* and *med:naso* as predictors and included speaking rate as a covariate to control for disease effect accounted for 65% of the variance in the speech intelligibility of the participants with ALS. Disease effect, as indicated by the slowing of speaking rate, had a significant effect on speech intelligibility throughout the disease course (β coefficient= 0.079, $p < 0.001$). Apart from the disease effect, *med:naso* was significantly correlated with speech intelligibility during the early stages of the disease (β coefficient= -0.169, $p = 0.005$).

4. Discussion

4.1. Increase of articulatory movement speed to compensate for VPD during early stages of ALS

As shown in Figures 1 & 2, the time courses of changes in nasalance and articulatory speed were distinct. Specifically, nasalance increased linearly as speaking rate slowed down (Fig-

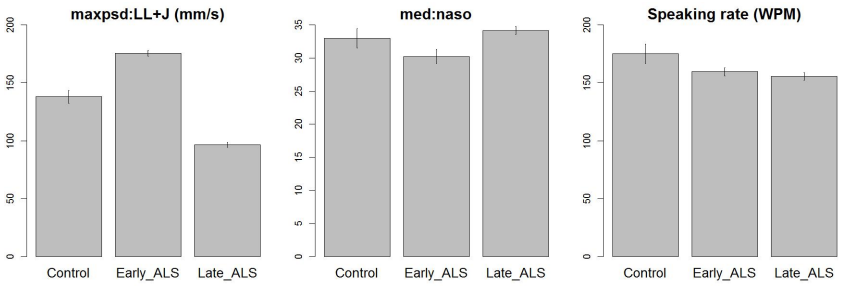

Figure 3: Bar graphs that show the mean and standard deviation of *maxspd:LL+J*, *med:naso*, and speaking rate for controls and participants in early and late stages of ALS, respectively.

ure 1). Because speaking rate changes in healthy speakers appear to have no effect on nasalance [14], the association between the changes in nasalance and speaking rate as shown in Figure 1 was interpreted to be disease-related, suggesting that nasalance increased at a relatively constant rate throughout the disease course. This finding provided evidence against the argument that velopharyngeal function only starts to decline when muscle weakness progresses to a critical level.

Throughout the early stages of ALS, the speed of lower lip and jaw movement remained high (i.e., $> 138mm/s$) (Figures 2) and exceeded that of controls (Figure 3). As shown in Figure 3, the increase in lower lip and jaw movement speed during the early stages coincided with a decrease in nasalance. Furthermore, this negative association between nasalance and articulatory speed was not a consequence of the co-occurring disease effects on articulatory and velopharyngeal functions because speaking rate, which was used as an indicator of disease effect on speech, did not change across the two stages (Figure 3). Therefore, it is most likely that the increase in lower lip and jaw movement speed during the early stages served as a behavioral compensation for VPD. Prior studies have established the general effect of articulatory adjustment for reducing hypernasality related to VPD [7, 8]. Our results based on the aerodynamic simulations provided further evidence to support the increase of articulatory speed as a compensation strategy. This strategy was, however, no longer available during the late stages, possibly due to the loss of capacity for further increasing of articulatory movement speed. As a result of disease effect on the articulatory function, articulatory speed decreased progressively during the late stages of the disease (Figure 2).

4.2. Predicting early and late speech intelligibility decline based on velopharyngeal and articulatory performance

The overall decline of speech intelligibility was modeled by a two-phase NLME model based on nasalance and the speed of lower lip and jaw, which accounted for 64% of the total variance in intelligibility. This finding is consistent with Rong et al. [5], who suggested that articulatory and resonatory impairments showed the most substantial contribution to speech intelligibility decline in ALS among all of the four speech subsystems (i.e., articulation, resonance, phonation, and respiration).

The underlying mechanisms that drive the longitudinal change of speech intelligibility were, however, different across the early and late stages. The relatively slow decline of speech intelligibility during the early stages was found to be associated with nasalance. Because the changes in nasalance during the early stages reflected a combination of disease effect on velopharyngeal function and articulatory compensation for VPD, the contribution of these two components to speech intelligibility needed to be assessed independently. Specifically, the disease effect led to a decline of speech intelligibility, as demonstrated by the positive correlation between speaking rate and intelligibility in the NLME model with speaking rate as a control covariate to represent disease effect. Articulatory compensation, on the other hand, led to a decrease of nasalance (Figure 3) and consequently, an increase of speech intelligibility, as illustrated by the negative correlation between nasalance and intelligibility when the disease effect was controlled. Combining of the disease effect and articulatory compensation resulted in overall slow declines of speech intelligibility.

The rapid decline of speech intelligibility during the late stages was associated with both increased nasalance and decreased speed of lower lip and jaw. This association was, however, lost when speaking rate was included as a control covariate in the model, suggesting that the changes in nasalance and articulatory speed during the late stages were primarily attributed to the disease effects on velopharyngeal and articulatory functions. Because articulatory compensation became unavailable during the late stages, nasalance increased as a result of disease effect, which co-occurred with the decrease of articulatory speed. Both changes led to rapid declines of speech intelligibility.

5. Conclusions

In this study, we explored the mechanisms that underlie the effects of VPD on speech intelligibility during the early and late stages of ALS. Our finding of articulatory compensation for VPD during the early stages may help explain why some individuals with ALS are capable of maintaining intelligible speech despite VPD. It also sheds light on the potential therapeutic application of articulatory compensation for the effect of VPD on dysarthric speech, which will be assessed using expanded speech samples in future studies. Our finding also suggests the loss of articulatory compensation was characterized by the drop of the maximum speed of the composite lower lip and jaw movement below 138 mm/s, which serves as the onset of precipitous speech decline and thus, might provide important timing information that can assist clinicians for planning of interventions (e.g., assistive communication).

6. Acknowledgements

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