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A First Investigation of Tongue, Lip, and Jaw Movements in Persons with Dysarthria due to Multiple Sclerosis

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Abstract

Background: Multiple sclerosis can affect the speech motor system and result in dysarthria.

Objectives: This pilot study sought to identify tongue, lip, and jaw motor deficits in persons with dysarthria due to multiple sclerosis (PwDMS) to better understand the speech motor mechanisms that underlie their aberrant speech.

Methods: Tongue and jaw movements during /ai/ and lower lip and jaw movements during /bob/ were examined in eleven PwDMS and fourteen age- and sex-matched controls using three-dimensional electromagnetic articulography. Movement duration, maximum displacement, peak speed, stiffness (i.e., peak speed/displacement ratio), and jaw contribution to lower lip and tongue displacements were of particular interest.

Results: Whereas most kinematic measures yielded significant between-group differences for tongue and jaw motor performance during /ai/, lower lip and jaw motor performance during /bob/ were mostly comparable between groups.

Conclusion: Findings suggest that speech movements of the tongue are differentially more impaired than those of the lower lip in PwDMS. Particularly the ability to move the tongue with adequate speed during speech was significantly impaired in PwDMS, which may explain, in part, their slowed speech rate. Aberrant jaw kinematics during /ai/ may be a compensatory strategy to maximize speech clarity in the presence of the impaired tongue motor performance.

INTRODUCTION

Approximately 40-50% of patients with multiple sclerosis (MS) develop dysarthria over the course of the disease^{1,2}. In these patients, cerebellar and cortico-bulbar tract lesions can result in a mixed type of dysarthria with spastic and ataxic components¹⁻⁴. However,

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individuals with MS can develop virtually any type of dysarthria depending on disease-related lesion locations³. The most common speech characteristics of persons with dysarthria due to MS (PwDMS) are imprecise articulation and deviant prosodic stress patterns, which are often the result of a slowed speech rate, phoneme prolongations, and impaired loudness control^{1,2,5}. A harsh or strained/strangled voice and abnormal pitch levels can also be observed^{1,2,5}.

Speech intelligibility, or the proportion of spoken words that can be understood by the listener, remains relatively intact in many PwDMS³. Listeners, however, perceive the speech of affected individuals as unnatural and impaired^{3,6}. Particularly the slow speech rate and impaired pitch control as well as the irregular articulatory breakdowns have been shown to contribute to the perceived speech inadequacy in these individuals^{1,2}. Although disease-related cognitive impairments have been shown to contribute to the abnormally slow speech rate of persons with MS^{7,8}, the disease-related decline in orofacial motor control is also known to contribute to their disordered speech^{3,4}. However, studies that have systematically investigated the speech motor deficits in individuals with MS are currently lacking.

The few studies that are currently available on orofacial muscle performance in persons with MS investigated tongue and lip muscle strength and endurance as well as diadochokinetic task performance^{9,10}. Study outcomes suggested that tongue muscle performance was impaired in persons with MS whereas lip muscle performance was either unaffected in these individuals or less affected than tongue muscle performance. However, orofacial muscle strength and endurance as well as performance on diadochokinetic tasks are not necessarily predictive of speech motor function¹¹⁻¹⁴. Therefore, it is difficult to draw conclusion about tongue- and lip-specific motor deficits that may underlie the impaired speech of PwDMS.

Deviant speech motor performance patterns may further occur in the jaw, a structure that is known to support lip and tongue movements during speech. For example, in individuals with a tongue-specific impairment, jaw movements were found to be abnormally large¹⁵⁻¹⁷. The increased jaw support to tongue movements during speech is thought to be a compensatory movement strategy that these speakers implement to maintain speech function in the presence of the tongue impairment¹⁸. Insights in speech motor performance patterns of persons with dysarthria can be used to optimize speech interventions that aim to maximize speech function (e.g., specifically targeting the impaired structure, training structure-specific compensatory behaviors). They can also lead to the development of new treatment approaches for persons with dysarthria.

The purpose of this study was to gain initial insights in tongue, lower lip, and jaw motor performance during speech in PwDMS. This study specifically investigated speech kinematic parameters that are commonly associated with deviations in speech rate (i.e., movement duration, peak speed, ratio of peak speed and displacement) and speech imprecision (i.e., maximum displacement, jaw contribution to overall lower lip and tongue movements). Based on previous studies on persons with MS^{9,10}, tongue kinematic measures were hypothesized to show significant differences between PwDMS and controls; however, no hypotheses could be formulated with regards to the potential between-group differences in lip and jaw kinematics.

METHODS

Participants

The study was approved by the Institutional Review Board of the Vanderbilt University Medical Center and informed consent was obtained for each participant prior to data collection. The dataset presented is a subset of a larger research project on speech motor performance in patients with MS. Only patients with MS who exhibited symptoms of dysarthria as determined by a certified speech-language pathologist with expertise in motor speech disorders were included in the current study. Thus, the current dataset consists of eleven patients with MS (five females, six males, mean age: 53.1, range: 40 - 61) and 14 controls (six females, eight males, mean age: 54.0 range: 45 - 63). Table 1 provides demographic and disease-related information of the participants with MS. Patients presented with primary progressive (PP), relapsing remitting (RR), and secondary progressive (SP) MS^{19,20}, were 3 month free from clinical relapses, steroids administration, and changes in the Expanded Disability Status Scale (EDSS)²¹. All SPMS patients who were on medications had some signs of active disease.

A standard speech evaluation was administered during the study to obtain speech severity ratings, speech intelligibility scores, and speech perceptual characteristics. A summary of each participant's speech evaluation can be found in Table 2. Specifically, sentence intelligibility scores were calculated based on a standardized reading test consisting of eleven sentences that vary in length (Sentence Intelligibility Test, SIT)²². A certified speech-language pathologist with expertise in motor speech disorders provided speech severity ratings, dysarthria type, as well as the speech perceptual characteristics based on the audio recordings of the SIT test. All participants were required to pass a standard hearing screening and achieve 25/30 on the Mini Mental Status Examination (MMSE)²³. This MMSE cutoff is commonly used in speech studies that involve patients with dysarthria to ensure that these patients are able to follow directions and complete the required speech tasks correctly.

Experimental Tasks

All participants repeated the sentences “*She saw a boy with a kite hiding behind the house*” and “*Say Bobby again*” five times. For the purpose of this study the diphthong /ai/ embedded in the word “*kite*” and the opening and closing movement associated with /bob/ in the word “*Bobby*” were of specific interest. These target utterances were chosen because previous research showed that speech deficits are best observable during speech segments that require large and rapid changes in vocal tract configurations such as those during diphthongs (i.e., /ai/) and stops²⁴. Further, the words “kite” and “Bobby” were selected because the phonetic context provided reliable onsets and offsets for the kinematic analyses.

Data Acquisition and Processing

Speech kinematic data were recorded using 3D electromagnetic articulography (AG501, Medizintechnik Carstens). As in previous studies^{25,26} sensors were attached at midline to the upper and lower lips, tongue tip and tongue back, and lower incisors (to record jaw movement). Three head reference sensors were placed on goggles, which each participant

wore during the data collection. All kinematic data was corrected for head movement and transposed into a head-based coordinate system using the *Normpos* software²⁷ and low-pass filtered at 15 Hz in *SMASH*²⁸.

For posterior tongue and jaw kinematic measures the 3D position of the posterior tongue and jaw sensors were extracted at the onset and offset of the diphthong /ai/. The onset and offset of the diphthong were defined by the positional minimum and maximum of the posterior tongue in the vertical dimension²⁵. Finally, for lower lip and jaw kinematic measures the 3D position of the lower lip and jaw sensors were extracted at the onset and offset of the closing movement during “*Bobby*”. The onset and offset of the closing movement were defined by the lower lip positional minimum and maximum in the vertical dimension. Displacements and speed of the posterior tongue, lower lip, and jaw sensors were calculated using *SMASH*.

Overall tongue and lower lip displacements were decoupled from jaw displacements by subtracting jaw displacements from the overall tongue and lower lip displacements. The decoupling was done in a linear fashion for the lower lip movements^{29,30}; however, for the posterior tongue, the rotational movement of the jaw was taken into account^{26,31}. Finally, the percent jaw contribution to overall tongue and lower lip displacement was calculated for each participant.

Statistical analysis

To determine between-group differences, each kinematic measure was submitted to a linear mixed model analysis with group and gesture (when applicable) as the fixed effects and participants as random effects. Group x gesture interactions were also examined (when applicable). The critical p -value was set to $p < .05$. Because routine statistical diagnostic procedures revealed that the percent jaw contributions of overall tongue and lower lip displacements were normally distributed, no log-transformation was needed for this measure. Data are presented as group means ($\pm$ standard error, *SE*) unless otherwise specified.

RESULTS

Tongue and jaw kinematic measures associated with the diphthong /ai/

As can be seen in Figure 1, movement duration was longer in PwDMS than controls [$F(1,21.4) = 14.910$, $p = .001$, *Mean difference* = 0.069 s, *SE* = 0.018 s]. In addition, the upper panels of Figure 2 show group means for tongue kinematic measures and the lower panels of Figure 2 show the group means for the jaw kinematic measures for the diphthong /ai/. As can be seen, PwDMS produced lower tongue peak speeds [$F(1, 22.970) = 5.573$, $p = .027$, *Mean difference* = 31.976 mm/s, *SE* = 13.545 mm/s, Panel B of Figure 2] and larger jaw displacements [$F(1, 22.713) = 7.225$, $p = .013$, *Mean difference* = 3.151 mm, *SE* = 1.172 mm, Panel D of Figure 2]. Furthermore, the peak speed/displacement ratios were lower in PwDMS than controls for the tongue [$F(1, 21.908) = 7.895$, $p < .001$, *Mean difference* = 3.335, *SE* = 0.788, Panel C of Figure 2] and for the jaw [$F(1, 20.268) = 19.007$, $p < .001$, *Mean difference* = 3.917, *SE* = 0.898, Panel F of Figure 2]. Finally, Figure 3 shows the relative contribution of the jaw to the overall tongue displacement. A significant group effect

was found with PwDMS producing the diphthong with greater relative contribution of the jaw than controls [$F(1, 22.895) = 5.632, p = .026, \text{Mean difference} = 12.54\%, SE = 5.28\%$].

Lower lip and jaw kinematic measures associated with /bob/

Figure 4 displays the mean movement durations for the opening and the closing gesture for each group. There was a significant main effect of group and a significant main effect of gesture on movement duration. The group x gesture interaction was not significant. Thus, movement durations were generally longer in PwDMS than in controls [$F(1, 24.773) = 7.219, p = .013, \text{Mean difference} = 0.027\text{s}, SE = 0.010\text{s}$]. Further, in both groups the closing gesture was associated with significantly shorter movement durations than the opening gesture [$F(1, 115.154) = 187.493, p < .001, \text{Mean difference} = 0.040\text{s}, SE = 0.003\text{s}$].

The upper panels of Figure 5 present the group means for lower lip maximum displacement (Panel A), lower lip peak speeds (Panel B), and peak speed/displacement ratio (Panel C) and the lower panels of Figure 5 display the group means for jaw maximum displacement (Panel D), jaw peak speed (Panel E), and peak speed/displacement ratio (Panel F). A main effect of gesture was found for lower lip displacement and jaw displacement with the closing gesture being associated with larger lower lip displacements than the opening gesture [$F(1, 154.858) = 98.986, p < .001, \text{Mean difference} = 1.649\text{ mm}, SE = 0.166\text{mm}$]. Further, the closing gesture was associated with larger jaw displacements than the opening gesture [$F(1, 169.222) = 20.835, p < .001, \text{Mean difference} = 0.538\text{ mm}, SE = 0.118\text{ mm}$]. The main effect for group and the group x gesture interactions were not significant.

With regards to peak speed, only the main effect of gesture on lower lip peak speed yielded a significant finding. That is, the closing gesture was associated with greater lower lip peak speeds than the opening gesture [$F(1, 185.229) = 66.466, p < .001, \text{Mean difference} = 22.568, SE = 2.768$]. No main effects or group x gesture interactions were found for jaw peak speed during /bob/.

Finally, a significant main effect of group and gesture was found for peak speed/displacement ratio for the lower lip [group: $F(1, 23.734) = 7.844, p < .001$, gesture: $F(1, 117.653) = 397.721, p < .001$, respectively]. Further the group x gesture interaction was also significant [$F(1, 117.653) = 9.601, p < .001$]. Simple comparisons revealed that in both groups lower lip peak speed/displacement ratios were greater during the closing gesture than during the opening gesture ($p < .001$); however, only during the closing gesture lower lip peak speed/displacement ratios were lower in PwDMS than in controls ($\text{Mean difference} = 2.213, SE = 0.848, p = .016$).

For jaw peak speed/displacement ratios, the main effect of group and gesture were significant. The group x gesture interaction was not significant. Thus, the jaw peak speed/displacement ratio was generally lower in PwDMS than in controls [$F(21.037, 1) = 6.251, p = .021, \text{Mean difference} = 1.870, SE = 0.704$]. Further, overall the closing gesture was associated with a greater jaw peak speed/displacement ratio than the opening gesture [$F(109.365, 1) = 20.801, p < .001, \text{Mean difference} = 1.068, SE = 0.234$].

Finally, group means of the relative contribution of the jaw to the lower lip movement are displayed in Figure 6. A significant effect of gesture on the relative contribution of the jaw to lower lip movements was found. That is, overall the closing gesture was associated with a larger contribution of the jaw than the opening gesture [$F(132.518,1) = 6.730$, $p = .011$, $Mean\ difference = 1.84\%$, $SE = 0.71\%$]. The group effect and the group x gesture interactions were not significant.

DISCUSSION

This study is the first to investigate tongue, lower lip, and jaw motor performance during speech in PwDMS to better understand the physiologic correlates of the aberrant speech perceptual characteristics in these individuals. We focused on speech kinematic parameters that are commonly associated with deviant speech rates and articulatory imprecision. Target utterances were selected to specifically investigate tongue and jaw motor performance (/ai/ in “kite”) and lower lip and jaw motor performance (/bob/ in “Bobby”). Since our study aimed to characterize the speech motor deficits that underlie dysarthria in persons with MS, our cohort of patients was selected based on the presence of a perceivable speech impairment.

Our study outcomes showed that both target utterances were associated with significantly longer movement durations in PwDMS than controls. Significantly reduced tongue peak speeds can explain the prolonged durations of /ai/ in PwDMS. However, prolonged durations during /bob/ cannot be explained by a speed constraint because lower lip and jaw peak speeds were not reduced in these individuals. Such structure-specific speed constraints have been reported previously for dynamic strength tests and diadochokinetic rates in PwDMS^{9,10}.

Prolonged movement durations of /bob/ appear to be the result of reduced peak speed/displacement ratios of the lower lips and jaw; particularly during the closing gesture. Because closing gestures are typically produced with greater peak speeds and shorter durations than opening gestures, they are considered to be more sensitive to disease-related motor performance deficits³³. Indeed, reduced lip and jaw peak speed/displacement ratios have been observed previously in persons with cerebellar atrophy, motor neuron lesions, and caudate nucleus atrophy due to Huntington’s disease^{32, 34-38}. It has been suggested that distorted cerebellar input to the primary motor cortex, impaired upper motor neurons, as well as reduced excitability of the lower motor neurons may result in slowed force development^{32,35}.

It is, however, also conceivable that the impaired ability to generate adequate tongue speed in PwDMS may demand reduced speeds of all other orofacial structures to maintain adequate inter-articulator timing during speech³⁹. Such an adaptive behavior may explain why lower lip and jaw velocity profiles of speech movements in these clinical groups fail to show pathological patterns such as a prolonged acceleration phase³², which is typically seen in limb movements of patients with cerebellar lesions and is indicative of slowed force development⁴⁰. A follow-up study should examine lip, tongue, and jaw velocity profiles of

speech movements in PwDMS to determine if tongue velocity profiles differ from those of the lower lip and jaw in these individuals.

Finally, findings of the current study suggest that persons with MS produce abnormally large jaw displacements, particularly when speech segments require large and rapid tongue movements. Furthermore, lower lip displacements also tended to be larger in PwDMS than controls. Abnormally large lower lip and jaw movements have also been observed in other clinical groups with tongue-specific impairment profiles such as Amyotrophic Lateral Sclerosis^{15,16} and presumably indicate a compensatory strategy to maintain speech clarity in the presence of an impaired tongue¹⁸. Future studies should investigate if direct instructions to exaggerate lip and jaw movements during speech may enhance speech intelligibility in those patients with MS who exhibit reduced speech clarity.

Study limitations and conclusions

To our knowledge, this is the first investigation of tongue, lip, and jaw speech movements in PwDMS in the literature. Thus, this study sought initial insights in the underlying mechanisms of speech abnormalities in PwDMS. Given the small cohort, findings are preliminary and represent an initial exploration; however, they certainly encourage further investigations in larger cohorts of patients with MS. Analyses investigating the associations between speech motor impairment and disease topography as well as associations between speech motor impairment (specifically speed) and cognitive impairment severity, other scales of disability or disease burden, or medications potentially affecting the spastic component of dysarthria (e.g., baclofen) were not performed in the current study because they were outside the scope of this initial work, but they are the focus of ongoing investigations in our group. One may argue that the pathobiological basis of the dysarthria varies in PwDMS and likely also varied in our cohort of participants. Yet, because a tongue-specific speed constraint was found across all PwDMS in this study, the potential heterogeneity of the disease topography was not thought to be a shortcoming of this study.

In conclusion, preliminary findings of this study suggest that PwDMS present with a tongue-specific speech motor impairment. Specifically, these individuals cannot generate sufficient speed when moving the tongue during speech; yet, lip and jaw speed remain unaffected. The tongue-specific speed constraint is reflected in their abnormally slowed speech rate, which contributes to the unnatural sounding speech in PwDMS. These findings suggest that speech treatments should specifically target tongue speed when aiming to reduce speech unnaturalness. Study outcomes further suggest that PwDMS exaggerate their jaw and lower lip movements during speech, which likely explains the relatively preserved speech intelligibility in the presence of an impaired tongue in PwDMS.

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Highlights:

- Increased jaw support for tongue displacement in multiple sclerosis
- Jaw support for lower lip displacements comparable across groups
- Only tongue speed, but not lower lip speed reduced in multiple sclerosis
- Tongue and lip stiffness reduced in multiple sclerosis
- Between group difference greater for tongue stiffness than lower lip stiffness

Figure 1.
Group means ($\pm SE$) for movement durations associated with the diphthong /ai/.

Figure 2. Group means ($\pm$ SE) for tongue and jaw kinematic measures associated with the diphthong /ai/.

Figure 3. The relative contribution of the jaw to the overall tongue displacement associated with the diphthong /ai/.

Figure 4. Group means ($\pm$ SE) for movement duration associated with the opening and closing gesture of */bob/*.

Figure 5. Group means ($\pm$ SE) for lower lip and jaw kinematic measures associated with the opening and closing gesture of /bob/.

Figure 6.
The relative contribution of the jaw to the overall lower lip displacement associated with the opening and closing gesture of /bob/.

Table 1.

Demographic and disease-related information for participants with MS.

Code	Age	Yrs Dx	MS Type	EDSS	Disease Modifying Agents
MSF02	40	20	SP	7.0	Natalizumab
MSF03	51	27	SP	7.5	None
MSF04	49	19	SP	6.5	Fingolimod
MSF06	61	12	RR	0	Natalizumab
MSF07	56	3	SP	6.5	None
MSM02	45	13	SP	3.5	Interferon beta 1a
MSM04	60	7	SP	7.5	None
MSM05	53	14	PP	7.5	None
MSM06	55	22	RR	6.0	Dimethylfumarate
MSM08	59	20	SP	7.5	Alemtuzumab
MSM12	55	14	PP	6.5	Ocrelizumab

Note: MS = multiple sclerosis, Yrs Dx = years since diagnosis, EDSS = expanded disability status scale, PP = primary progressive, RR = relapsing remitting, SP = secondary progressive

Table 2.

Speech characteristics of participants with MS.

Code	Dysarthria Type	Speech Severity	SIT	Speech Characteristics
MSF02	Ataxic	Mild-moderate	93	slow rate, excessive and equal stress, articulatory impression, irregular breakdowns, vowel distortions, phoneme prolongations, excessive loudness, monopitch, strained-strangled voice, harsh voice
MSF03	Ataxic-spastic	Moderate	100	slow rate, excessive and equal stress, imprecise consonants, phoneme prolongations, irregular articulatory breakdowns, vowel distortions, low pitch, monopitch, loudness decay, harsh voice, wet voice, strained-strangled voice, hyponasality
MSF04	Ataxic-spastic	Mild	100	slow rate, excess and equal stress, imprecise consonants, phoneme prolongations, irregular articulatory breakdowns, vowel distortions, low pitch, monopitch, monoloudness, harsh voice
MSF06	Ataxic-spastic	Mild	99	slow rate, irregular articulatory breakdowns, phoneme prolongations, monopitch, reduced volume, monoloudness
MSF07	Ataxic-spastic	Mild	96	slow rate, phoneme prolongations, imprecise consonants, breathy voice
MSM02	Ataxic-flaccid	Mild	98	slow rate, excessive and equal stress, irregular articulatory breakdown, phoneme prolongations, low pitch, monopitch, low volume, strained-strangled, hypernasality
MSM04	Spastic-ataxic	Moderate	78	slow rate, inappropriate silences, excessive and equal stress, imprecise consonants, phoneme prolongations, irregular articulatory breakdowns, vowel distortions, monopitch, hypernasality
MSM05	Ataxic-spastic	Moderate	99	slow rate, excessive and equal stress, phoneme prolongations, low pitch, monopitch, monoloudness, harsh voice, strained-strangled, hypernasality
MSM06	Ataxic	Mild	98	vowel distortion, irregular articulatory breakdowns, monopitch, harsh voice
MSM08	Flaccid-ataxic-hyperkinetic	Moderate-severe	92	slow rate, inappropriate silences, short breath groups, imprecise consonants, phoneme repetitions, phoneme prolongations, irregular articulatory breakdowns, vowel distortions, monopitch, voice tremor, low volume, breathy voice, voice stoppage, nasal emissions, forced inspiration-expiration
MSM12	Spastic-ataxic	Mild	98	slow rate, excessive and equal stress, phoneme prolongations, vowel distortions, strained-strangled voice, harsh voice, monoloudness, monopitch

Note: SIT = sentence intelligibility test score in % words understood correctly