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Spatiotemporal Coupling of the Tongue in Amyotrophic Lateral Sclerosis

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Abstract

Purpose: The primary aim of the investigation was to identify deficits in spatiotemporal coupling between tongue regions in amyotrophic lateral sclerosis (ALS). The relations between disease-related changes in tongue movement patterns and speech intelligibility were also determined.

Methods: The authors recorded word productions from 11 individuals with ALS with mild, moderate, and severe dysarthria using an x-ray microbeam during word productions. A coupling index based on sliding window covariance was used to determine disease-related changes in the coupling between the tongue regions across each word.

Results: The results indicated decreased spatiotemporal coupling of mid-posterior tongue regions and reduced tongue speed in the ALS-moderate subgroup. Changes in the range of tongue coupling relations and speed of movement were highly correlated with speech intelligibility.

Conclusions: These results provide new insights into the loss of lingual motor control due to ALS and suggest that measures of tongue performance may provide useful indicators of bulbar disease severity and progression.

Keywords: tongue coupling, coupling index, tongue kinematics, speech intelligibility, dysarthria

Tongue movements during speech and swallowing are accomplished through the spatial and temporal coupling of quasi-independent tongue regions, which are commonly referred to as the *tip, blade, body,* and *dorsum* (Mermelstein, 1973). Coupling among different tongue regions is imperative to speech because it determines the shapes that the tongue assumes for different speech sounds and allows for smooth transitions between sounds in connected speech (Green & Wang, 2003; Stone, Epstein, & Iskarous, 2004). Tongue-region coupling is delimited by the biomechanical properties, muscle composition, and neuronal innervation of the tongue, as well as by the linguistic composition of speech (DePaul, Waclawik, Abbs, & Brooks, 1998; Kelso, Saltzman, & Tuller, 1986; Perkell, 1969; Slaughter, Li, & Sokoloff, 2005; Stål, Marklund, Thornell, DePaul, & Eriksson, 2003). In amyotrophic lateral sclerosis (ALS), motoneuron loss causes morphological and functional changes to the tongue. Morphological changes alter the shape, size, position, and internal structure of the tongue (Cha & Patten, 1989; DePaul et al., 1998). Functional changes include weak, slow, and reduced tongue movements (Cha & Patten, 1989; Darley, Aronson, & Brown, 1975; DePaul & Brooks, 1993; Hirose, Kiritani, & Sawashima, 1982; Yunusova, Weismer, Westbury, & Lindstrom, 2008). These changes may limit the coordinative options that are used by talkers with ALS to produce clear and intelligible speech.

One way to characterize tongue coupling is to quantify the spatiotemporal relations among the movements of different tongue regions (i.e., tongue tip, anterior tongue body, posterior tongue body, and tongue dorsum) during speech. Using this approach, Green and Wang (2003) identified the unique coupling patterns that distinguished different consonants. They observed that movement coupling was significantly stronger between adjacent tongue regions (i.e., tongue tip and anterior tongue body) than between nonadjacent regions (i.e., tongue tip and tongue dorsum), suggesting constraints on movement independence because of tissue linkages and other biomechanical properties of the tongue. These results provide an empirical basis for the

current investigation of the effects of ALS on the spatiotemporal coupling of different tongue regions during speech.

The majority of information on tongue impairment due to ALS has come from perceptual and acoustic studies of affected speech (Darley, Aronson, & Brown, 1975; Hirose et al., 1982; Hirose, Kiritani, Ushijima, & Sawashima, 1978; Kent et al., 1989, 1990, 1992; Tjaden & Turner, 1997; Turner & Tjaden, 2000; Turner & Weismer, 1993; Weismer, Jeng, Laures, Kent, & Kent, 2001; Weismer, Kent, Hodge, & Martin, 1988; Weismer, Martin, Kent, & Kent, 1992). These studies have documented how speech sounds become slowed and less distinct over the course of the disease. Specific changes to tongue movements have also been observed, including reduced movement extents and speeds (Kent et al., 1975; Weismer, Yunusova, & Westbury, 2003; Yunusova et al., 2008). The effect of these kinematic changes on lingual coupling in persons with ALS, however, has not been investigated.

Motivation for investigating ALS-related changes in tongue movement coupling comes from histopathologic findings showing a disproportionate degeneration of muscle fiber groups and muscle fiber types in the anterior part of the tongue compared to the posterior tongue (DePaul et al., 1998). Increased muscle atrophy, fat replacement, and fibrosis were also observed in the anterior part of the tongue compared to the posterior part (Atsumi & Miyatake, 1987; Cha & Patten, 1989; Stålberg, Schwartz, Schiller, & Thiele, 1976). The differential structural changes to tongue regions could lead to changes in spatiotemporal coupling patterns between these regions.

Furthermore, ALS is a disease that affects both upper and lower motoneurons, resulting in changes in muscle tone, strength, and reflex activity. Affected individuals could exhibit both spastic and flaccid changes in tongue muscles. Both conditions are hypothesized to restrict the normal range of movement coupling observed among different tongue regions. Spasticity is expected to result in increased tongue muscle stiffness, and, possibly, increased coupling, whereas muscle flaccidity may result in decreased coupling. In comparison, healthy talkers are known to produce a wide range of coupling relations among different tongue regions to accommodate the variety of lingual shapes needed to produce English consonants (Green & Wang, 2003).

The primary aim of this investigation was to determine whether the tongue impairments due to ALS compromise the spatiotemporal coupling of the tongue during speech. More specifically, we hypothesized that persons with ALS present with a restricted range of movement-coupling relations between tongue regions. A secondary aim of this investigation was to determine the relation between tongue kinematic measures and speech intelligibility.

Method

Participants

Tongue-movement data were obtained from participants included in the x-ray microbeam (XRMB) dysarthria database, which was collected at the University of Wisconsin—Madison. Participants in the experimental group included 11 individuals diagnosed with ALS. Talkers with ALS (six male, five female) had a mean chronological age of 48.63 years (*SD* = 8.70, range = 39–68). The participants with ALS were further divided into subgroups on the basis of perceptual severity judgments of their speech by a speech-language pathologist. These ratings were used to stratify affected participants instead of sentence intelligibly scores. The ALS-mild category included four individuals, ALS-moderate had two participants, and ALS-severe had five participants. Three experienced speech-language pathologists classified the dysarthria type (Darley, Aronson, & Brown, 1969a, 1969b). The majority of the talkers with impairments exhibited a mixed spastic-flaccid type of dysarthria (see Table 1).

Ten healthy individuals were age and gender matched to participants with ALS and formed the control group. The mean age of the healthy participants (six male, four female) was 52 years (*SD* = 7.35, range: 46–69). The majority of the participants spoke with an Upper Midwest dialect of American English. All healthy talkers had a negative history of speech, language, and hearing problems. Participants were selected from the XRMB database only if acoustic and tongue-pellet-movement data relevant to the study were available. Specifically, participants were only included if tongue data were present for at least two tongue pellets and the two pellets affixed to the mandible.

Table 1. Demographic and dysarthria details of participants with amyotrophic lateral sclerosis (ALS).

ID	Gender	Age	Subgroup	Type of dysarthria	Scaled sentence intelligibility
10	M	55	ALS-severe	Spastic-flaccid	13.6
41	F	41	ALS-severe	Spastic-flaccid, ataxic	33.4
33	F	47	ALS-severe	Spastic-flaccid, ataxic	40.3
20	M	44	ALS-severe	Spastic-flaccid, flaccid	50.5
24	M	43	ALS-severe	Spastic-flaccid, flaccid	51.2
40	F	53	ALS-moderate	Spastic, mixed	51.5
42	M	68	ALS-moderate	Flaccid, mixed	55.0
43	F	45	ALS-mild	Ataxic, flaccid	97.6
38	M	57	ALS-mild	Spastic-mixed	128.4
02	F	39	ALS-mild	Ataxic	145.5
09	M	43	ALS-mild	Flaccid, hypokinetic	158.7

Experimental Tasks

The XRMB database includes an inventory of speaking tasks including words, sentences, and paragraphs recorded from every speaker. We restricted the speech data for this particular study to several target words embedded in sentences. To assess the functional independence of different tongue regions, we selected words that elicited quasi-independent movement of the anterior tongue blade and tongue dorsum. The chosen words were *cat*, *dog*, and *took*, which were produced in the following sentences, respectively: (a) "To feed the ***cat***, one must shoo the ***dog***," and (b) "I ***took*** a spoon and dish." We chose these sentences from the Hunter passage (Crystal & House, 1982). All of the selected sentences were read at a self-selected, comfortable speech rate. We recorded 10 repetitions of each sentence from each speaker, and only sentences with intact acoustic and kinematic data were used for analysis. A majority of the talkers with dysarthria and healthy talkers spoke at least five repetitions of each word. Throughout each recording session, participants were seated in a comfortable reclining examination chair that provided head support. Natural head movements were otherwise unrestrained.

Kinematic Data Acquisition and Processing

Kinematic data were acquired with x-ray microbeam (Westbury, 1994). Eight gold pellets (2–3 mm in diameter) were attached to the tongue (four pellets), lips (two pellets), and jaw (two pellets). The tongue pellets were glued to regions commonly classified as tongue blade (T1, T2), tongue body (T3), and tongue dorsum (T4). T1 and T4 were placed approximately 10 mm and 60 mm from the apex of the extended tongue, respectively. T2 and T3 were placed equidistant from each other and from T1 and T4 such that the distance between any two pellets was never less than 1 cm. Of the two mandibular pellets, MANi was attached to the buccal surface of the central incisor and MANm was placed at the junction between the first and second mandibular molars. Three pellets formed a reference triangle; one was attached to the buccal surface above the maxillary incisors and two were attached on the bridge of the nose.

Various sampling rates were used for the tongue and mandibular pellets. T1 was sampled at 160 Hz; T2, T3, and T4 were sampled at 80 Hz; and mandibular pellets were sampled at 40 Hz. All signals were subsequently smoothed and resampled at a uniform rate of 145 Hz. The reference pellets were used to correct for head movements. Additionally, the jaw was decoupled from the tongue so that tongue positions could be studied independent of the jaw. For this procedure, the translational and rotational components of mandibular movements were computed and subtracted from movements of the tongue (Westbury, Lindstrom, & McClean, 2002).

Data Segmentation

Prior to analysis, vertical position–time histories of the pellets were segmented from the peak displacement of one opening gesture to the peak displacement of the next closing gesture within a word. Analysis was limited to the vertical motion of the tongue because movement in the vertical dimension is an essential kinematic goal for the speech utterances included in the study. For the word *took*, the segment for covariance analysis included the opening gesture for the vowel /ʊ/ and the closing gesture for /k/. The pellet marker representative of the place of articulation for the target consonant was used to segment the opening and closing gestures. For example, the peak of pellet marker T1 was used to mark the beginning of the opening gesture for /ʃ/ in *took*. T4 was used to mark the end of the closing gesture of /k/ (see Figure 1).

Measurements

Average covariance within pellet pairs (see Green & Wang, 2003) provided an index of coupling strength between adjacent and nonadjacent tongue regions. It was calculated for the Y-position–time histories of all possible pellet pairs (i.e., T1 × T2, T1 × T3, T2 × T3, T2 × T4, T3 × T4, T1 × T4) using a sliding window. A window size of 50 points per 350 milliseconds (approximately 2.9 windows within 1 second) was determined to be the optimal size for capturing the major changes in movement coupling across each utterance. The covariance formula is given below (see Green & Wang, 2003, for complete details):

$$\mathrm{Cov}_{T1,T2} = \mathrm{Corr}_{T1,T2} \times SD_{T1} \times SD_{T2} \quad (1)$$

For each utterance, pairwise correlations were computed on the vertical time histories of pellet pairs T1 × T2, T2 × T3, T3 × T4, T1 × T3, T2 × T4, and T1 × T4. The average of the correlation coefficients among pellet pairs and the *SD* of movement across sliding window intervals represent the average covariance, which is a local estimate of spatiotemporal coupling. Similar sliding window approaches have been used to examine the range of coupling patterns between the movements of the rib cage and abdomen during speech breathing (Moore, Caulfield, & Green, 2001).

Figure 2 illustrates the effectiveness of the covariance analysis for capturing the effect of lingual motor decline for the word *took*. Figure 2A shows the move-

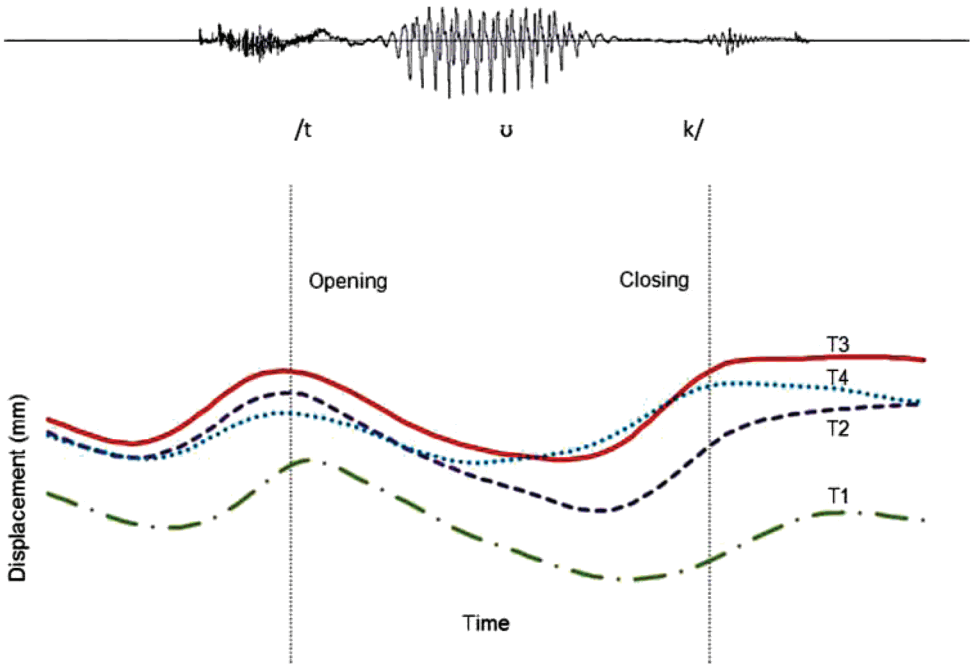

Figure 1. Top panel: Acoustic waveform of the word *took*. Bottom panel: vertical tongue displacement for the pellet pairs T1, T2, T3, and T4. In this example, the word *took* was segmented using the peak displacement of T2 and the peak displacement of T4 as indicated by dotted lines.

ment path for each pellet in the mid-sagittal plane for the word *took*. The top two images in Figure 2B display the vertical time histories as the tongue transitions from the alveolar to the velar position for *took* in the healthy talkers and talker with dysarthria, respectively. For the healthy talker, visual inspection reveals high coupling between adjacent and nonadjacent regions for /t/ but more independent movement between nonadjacent regions for /k/. In comparison to the healthy talker, the participant with ALS and with severe dysarthria exhibits a restricted range of coupling relations between adjacent and nonadjacent tongue regions from the alveolar to velar position.

The average covariance estimated from the average of pellet-pair correlations and *SD* across sliding window intervals for *took* is displayed in the bottom panel of Figure 2B. The healthy talker showed a high positive coupling for pellet-pairs T2 × T3, T2 × T4, and T3 × T4; and negative coupling for T1 × T3, and T1 × T4. The talker with severe ALS showed near-zero coupling for all pellet pairs. A high positive covariance value indicates highly coupled movements, whereas negative covariance values indicate movement in opposite directions between regions of the tongue. A near-zero covariance suggests limited movement between regions of the tongue.

For the words *took, cat,* and *dog,* we would expect functionally independent movements (negative covariance) between tongue regions T1 and T4 for movement from the alveolar to velar positions and vice versa. For these utterances, adjacent tongue regions T1 × T2, T2 × T3, and T3 × T4 should display functional dependence and highly coupled movements resulting in a high positive covariance.

Within-pair interquartile range (IQR) of covariance was computed for each pellet pair, word, and group. The IQR measure quantified the range of coupling relations within pellet pairs observed during the production of each word. We calculated the IQR for each of the six pellet pairs.

Across-pair range of covariance of tongue coupling was used to determine the extent of coupling relations across the entire tongue. The overall range was calculated for each word as the difference between the maximum and minimum covariance values pooled across all pellet pairs.

The measures of covariance between different parts of the tongue may depend on the degree to which the tongue movement is affected by the disease. Kinematic measures have been used to directly quantify movement-related changes to the tongue. Therefore, we included these measures in the current investigation to determine underlying tongue movement changes that might influence spatiotemporal coupling of the tongue.

Displacement was calculated as the straight-line path between the maximum and minimum vertical position for each pellet from the beginning to the end of the parsed target word for each tongue pellet.

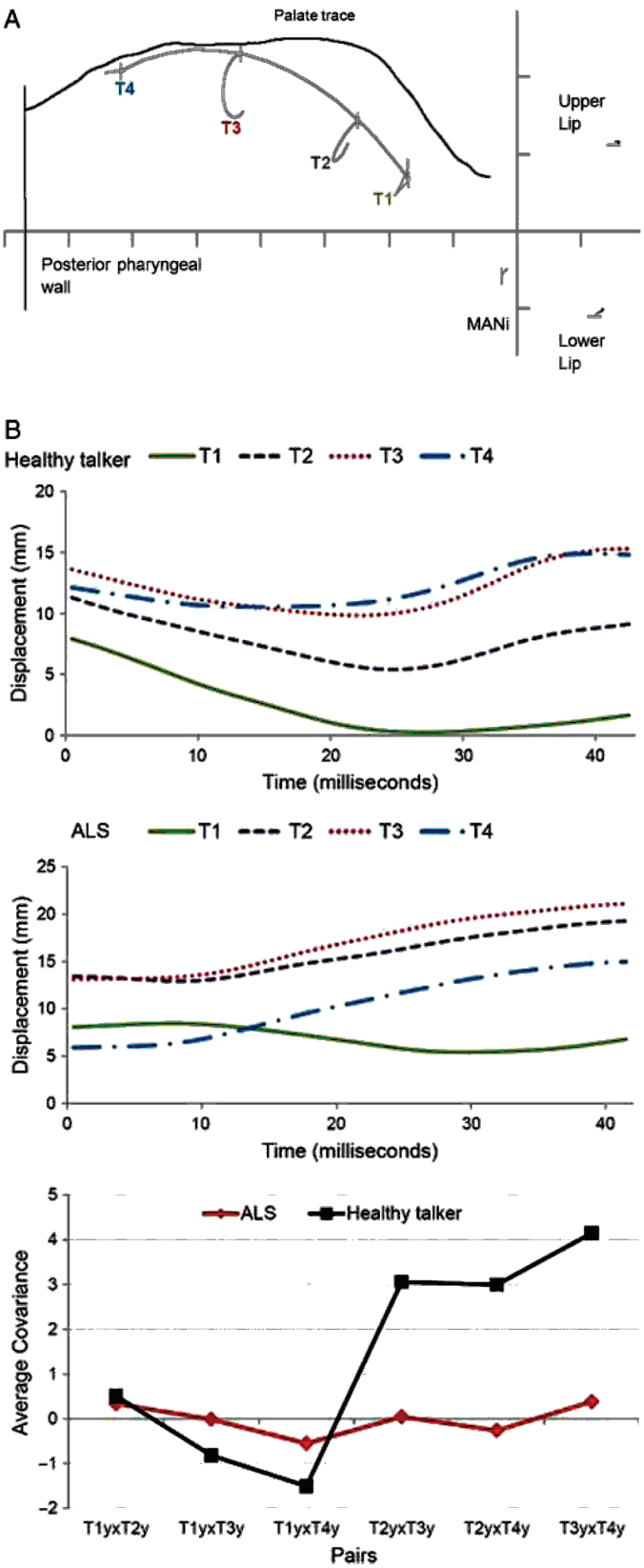

Figure 2. A: The movement path for each tongue pellet in the mid-sagittal plane during production of the word *took* by a healthy talker. **B.** Top panel: Displacement-time history waveforms of pellets on the anterior tongue blade (T1), posterior tongue blade (T2), tongue body (T3), and tongue dorsum (T4) in healthy talker. Middle panel: Talker with ALS. Bottom panel: Pairwise covariance between groups for the word *took* for the same healthy talker and talker with ALS.

Maximum speed was calculated as the maximum value of the first derivative of the vertical positional time-history for the whole parsed target word. As a result, we included the maximum value from both the initial release and the final constriction. Only the greater of the two maximum values was chosen for analysis because of our interest in the speed-generating capacity of the talkers regardless of opening and closing movements.

Sentence Intelligibility Estimation

Scaled sentence intelligibility was determined on the basis of direct magnitude estimation (DME). Ten listeners were required to rate the intelligibility of two sentences from the Hunter passage ("Tom Brooks was such a man"; and "Once he thought he saw a bird, but it was a large leaf that had failed to drop to the ground during winter") and an additional sentence ("To feed the *cat* one must shoo the *dog*") relative to a sentence that was considered to have mid-level intelligibility and was the fixed modulus with a score of 100. An average of the DME scores for each sentence across the 10 listeners and across the three sentences for each participant was calculated to obtain the sentence intelligibility score (for more details, see Yunusova, Weismer, Kent, & Rusche, 2005).

Statistical Analysis

We analyzed all data using the GLIMMIX procedure (SAS Institute, Cary, NC), with family-wise level of significance at $p \leq .05$. The Kenwood-Rodgers degrees of freedom adjustment was used for pairwise comparisons. We performed a rank transformation (Conover & Iman, 1981) on the dependent variables mean covariance, IQR covariance, and range of covariance prior to analyses because these values did not follow a normal distribution. The linear mixed-effect model used for the rank-transformed mean covariance, rank-transformed IQR covariance, maximum speed, and displacement included the fixed effects of group, pairs, word, and all two-way-interactions effects. The model also included random effects of subject nested within group and subject by pairs by word nested within group.

Results

Average Covariance

For the within-pair average covariance measure, a significant main effect was observed for word, $F(2, 237) = 20.68$, $p < .0001$, and pairs, $F(5, 239) = 15.48$, $p < .0001$. The analysis also revealed a significant interaction effect for Group × Pairs, $F(15, 229) = 2.06$, $p < .05$. Post hoc pairwise analyses revealed that the control group exhibited significantly greater average covariance for the adjacent tongue pairs T2 × T3 and T3 × T4 as compared to the ALS-moderate subgroup. The ALS-mild subgroup also showed significantly greater average covariance compared to the ALS-moderate subgroup for the pairs T2 × T4 and T3 × T4. In addition, the ALS-severe subgroup had significantly greater average covariance compared to the ALS-moderate subgroup for the tongue back pair T3 × T4 (see Figure 3). A significant Group × Word interaction effect, $F(6, 230) = 2.43$, $p < .05$, was also observed for the average covariance measure. The control group had significantly greater average covariance compared to the ALS-moderate subgroup for the word *cat*. The word *dog* showed significant interactions between subgroups whereby the ALS-mild subgroup had significantly greater average covariance compared to the ALS-moderate subgroup.

Within-Pair IQR of Covariance

The within-pair IQR showed a significant main effect for group, $F(3, 15) = 3.88$, $p < .05$; pairs, $F(5, 241) = 2.56$, $p < .05$; and word, $F(2, 239) = 34.73$, $p < .0001$. A significant interaction effect was observed for Group × Word, $F(6, 227) = 5.84$, $p < .0001$, for the IQR measure. Post hoc pairwise analyses revealed that the IQR for the words *dog* and *took* were significantly greater for the control group as compared to the ALS-moderate and ALS-severe subgroups. Similarly, the ALS-mild subgroup also demonstrated significantly greater IQR as compared to the ALS-moderate and ALS-severe subgroups for the word *dog*.

Across-Pair Range of Covariance

The across-pair covariance range showed a significant main effect for group, $F(3, 14) = 5.12$, $p < .05$. Post hoc analyses showed that the healthy talkers exhibited a significantly greater range of tongue-coupling patterns compared to the ALS-moderate and ALS-severe subgroups (see Figure 4).

Displacement

A significant main effect was observed for group, $F(3, 12) = 4.75$, $p < .05$; marker, $F(3, 173) = 79.96$, $p < .0001$, and word, $F(2, 174) = 16.56$, $p < .0001$, for the displacement measure. Post hoc pairwise comparisons of groups revealed that the ALS-mild subgroup had significantly greater displacements compared to the ALS-severe subgroup (see Figure 5). Post hoc pairwise marker comparisons revealed that the displacements of all tongue markers were significantly different from each other except T2 and T4 (T1 < T2 = T4 < T3). Within each group, the tongue blade (T1) and tongue body (T3) pellets had the smallest and largest displacements, respectively. Post hoc analyses on the main effect for word revealed that the displacements for the words *cat* and *took* were significantly greater than for *dog*.

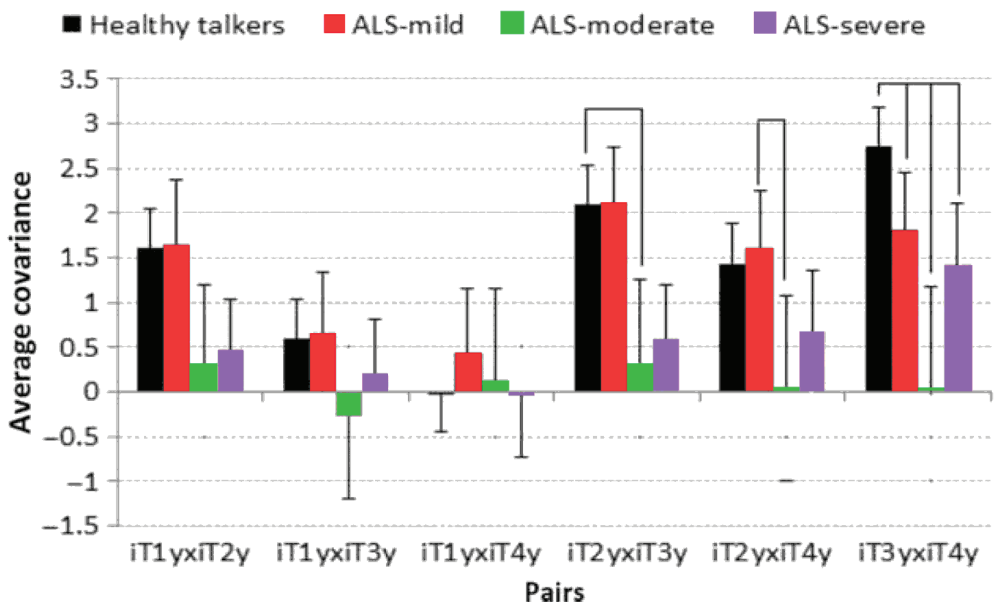

Figure 3. Pairwise average covariance across words for healthy talkers and talkers with ALS. Line for T2 × T3 denotes significant difference between healthy talkers and ALS-moderate subgroup; line for T2 × T4 denotes significant difference between ALS-mild and ALS-moderate subgroups; line for T3 × T4 denotes significant difference between ALS-moderate subgroup and healthy talkers, as well as ALS-mild and ALS-severe subgroups.

Maximum Speed

A significant main effect was observed for group, $F(3, 12) = 6.84$, $p < .01$, and word, $F(2, 183) = 5.39$, $p < .01$, for the dependent measure, maximum speed. A significant interaction effect was observed for Subgroup × Marker, $F(9, 176) = 2.09$, $p < .05$. The healthy talkers had significantly greater maximum speed for all tongue pellets compared to the ALS-moderate and ALS-severe subgroups. The ALS-mild subgroup had greater maximum speed compared to the ALS-moderate subgroup for T3 and greater

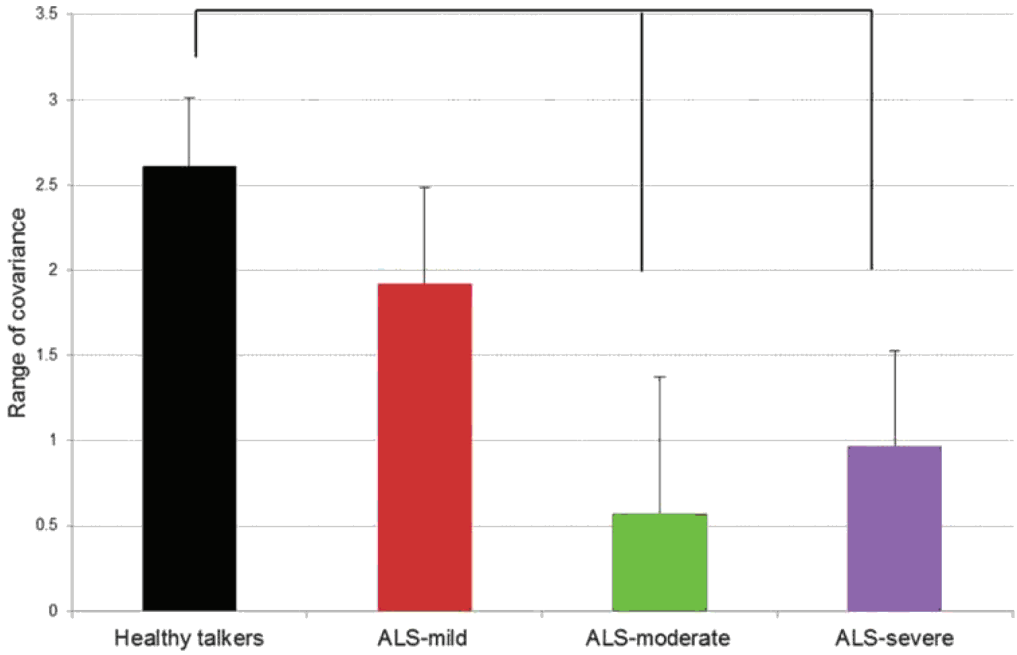

Figure 4. Across-pair covariance range in healthy talkers and talkers with ALS. Line denotes significant differences between the healthy talkers and the ALS-moderate and ALS-severe subgroups.

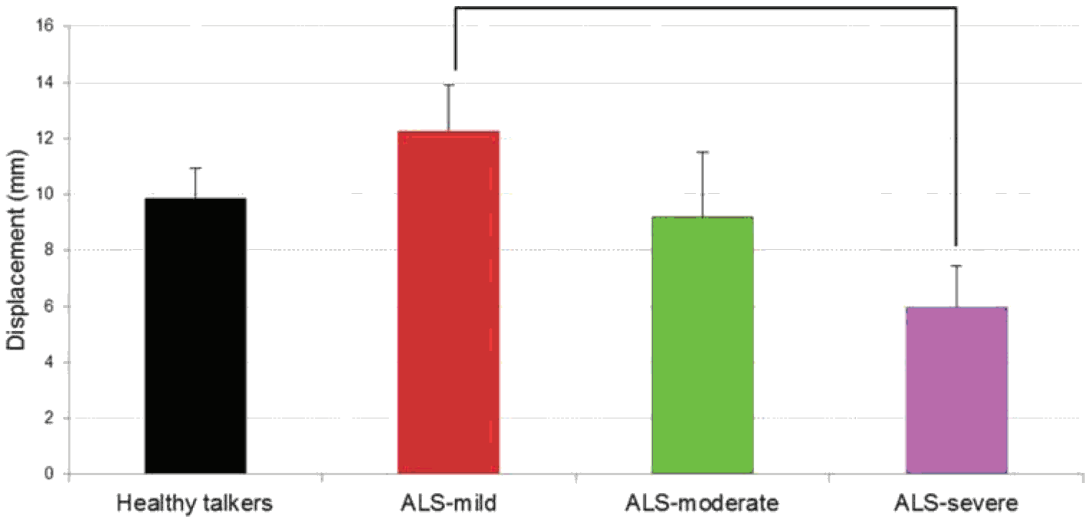

Figure 5. Mean values and *SE* of displacement for healthy talkers and ALS subgroups across all words and markers. Lines indicate significant main effects between the ALS-mild and ALS-severe subgroups.

maximum speed compared to the ALS-severe subgroup for T4 (see Figure 6). A significant Word × Marker interaction, $F(6, 167) = 2.40$, $p < .05$, was also observed. Post hoc analyses on the Word × Marker interaction revealed a significantly greater maximum speed for the words *cat* and *dog* compared to the word *took* for T1. The maximum speeds for T2 and T3 were significantly greater for the word *cat* compared to the word *dog*.

Relation Between Kinematic Measures and Speech Intelligibility

The correlation between the measures of average covariance, IQR of covariance, and speech intelligibility were small to moderate in magnitude (see Table 2). Range of covariance showed a large correlation with sentence intelligibility (.53; see Figure 7). Similarly, maximum speed, but not displacement, of all four

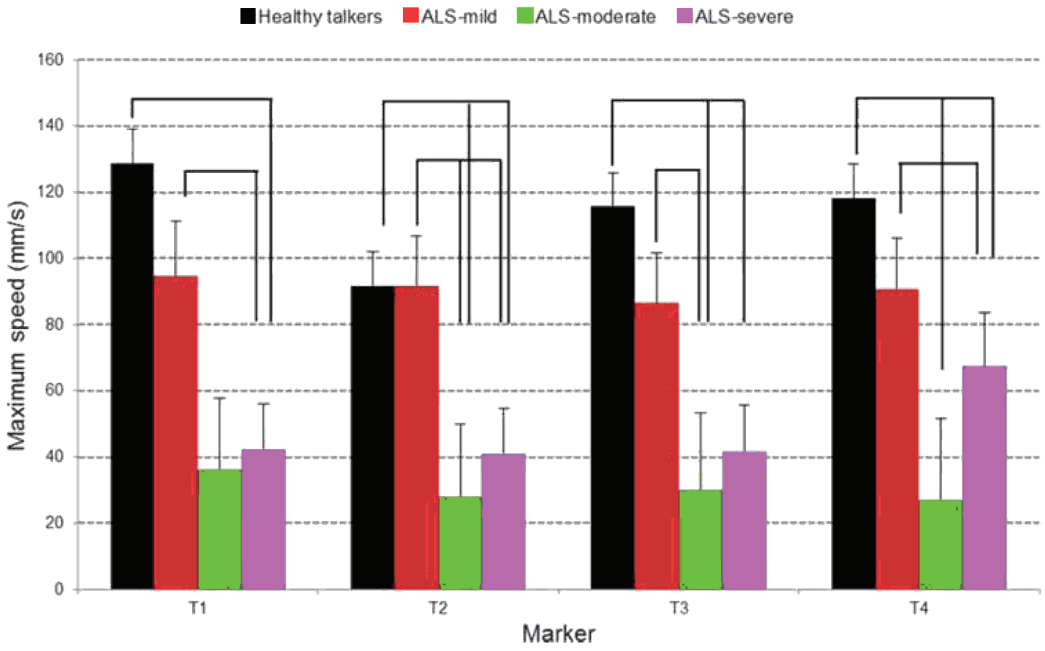

Figure 6. Mean values and *SE* of maximum speed recorded from tongue pellets T1, T2, T3, and T4 for healthy and ALS subgroups across all words. Lines indicate significant interaction effects between healthy talkers, the ALS-mild subgroup and ALS-moderate and severe subgroups for tongue markers.

Table 2. Pearson correlation coefficients for sentence intelligibility and tongue coupling measures across all words.

Measure	T1×T2	T1×T3	T1×T4	T2×T3	T2×T4	T3×T4
Average covariance	.36	.20	–.15	.34	.10	.33
Interquartile range	.31	.40	.36	.34	.18	.33

Large correlation = .5–1.0; medium correlation = .3–.5; small correlation = .1–.3

Table 3. Pearson correlation coefficients for sentence intelligibility and kinematic measures across all words.

Measure	T1	T2	T3	T4
Displacement	.22	.13	.22	–.03
Maximum Speed	.57	.43	.65	.53

Large correlation = .5–1.0; medium correlation = .3–.5; small correlation = .1–.3

tongue markers (T1, T2, T3, and T4) showed a medium to large correlation with speech intelligibility (see Table 3 and Figure 8).

Effect Sizes for Tongue Coupling and Conventional Kinematic Measures

Table 4 displays the effect sizes of the covariance and conventional kinematic measures between the healthy talkers for each ALS subgroup. The ALS-mild subgroup showed small effect sizes for covariance, IQR, and across-pair range but medium effect sizes for maximum speed and displacement measures. The ALS-moderate subgroup had large effect sizes for all measures except displacement. A large effect was observed for all measures except covariance in the ALS-severe subgroup when compared to the healthy talker group; a medium effect size was observed for the covariance measure. Across the ALS subgroups, the ALS-moderate subgroup showed the greatest effect sizes for all measures except displacement.

Discussion

Tongue movements were recorded in persons with ALS to determine the effect of the disease on the spatiotemporal coupling between tongue regions during speech. When an individual talks, the surface of the tongue assumes many different shapes through the quasi-independent movement of the tongue tip, blade, dorsum, and root. Lingual coupling for speech has been characterized previously by measuring the coupling relations between different regions of the tongue in healthy talkers (Green & Wang, 2003). This work demonstrated that different tongue regions exhibit a wide range of movement-coupling relations to meet the varying demands of different speech sounds. In contrast to those of the healthy talkers, the tongue movements of the talkers with ALS in this study were characterized by a restricted range of movement-coupling relations. The mid-posterior regions of the tongue were more affected than was the anterior re-

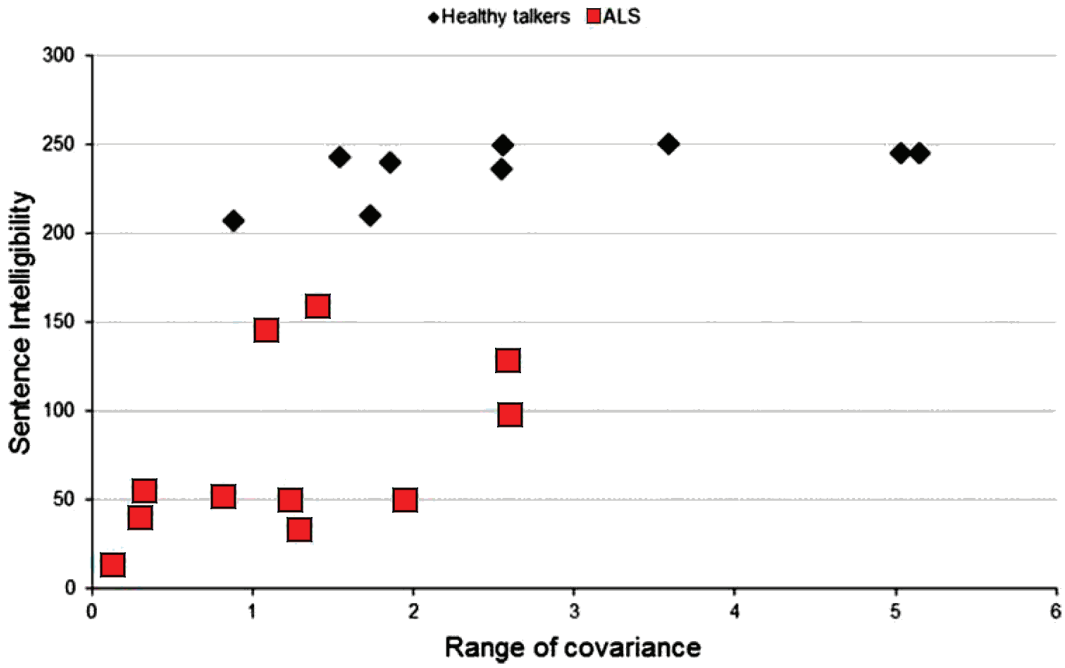

Figure 7. Correlation plot between speech intelligibility and range of covariance across all participants.

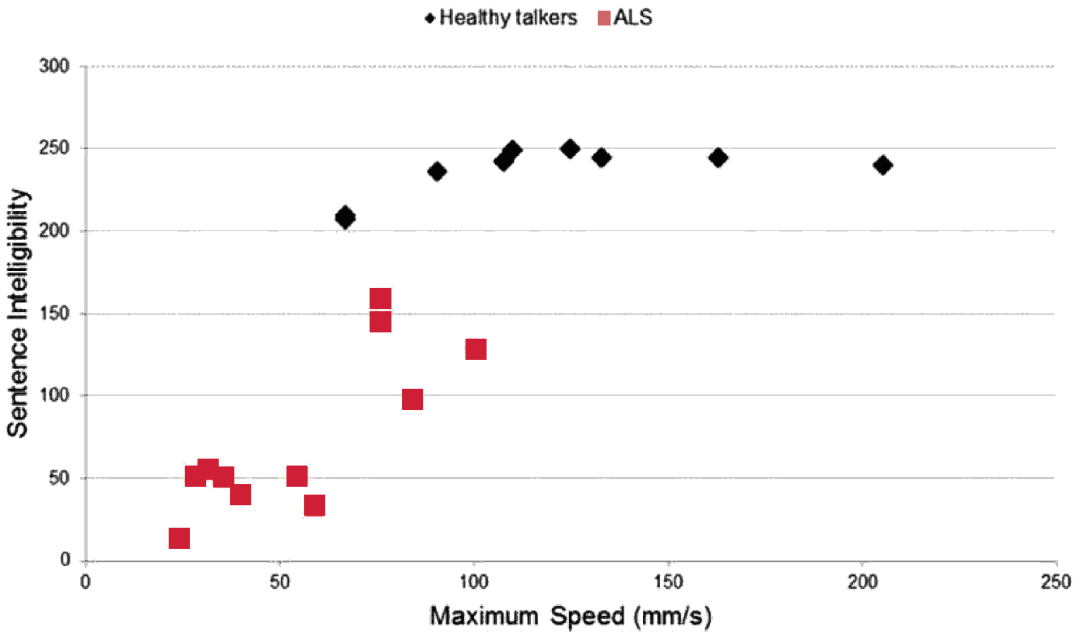

Figure 8. Correlation plot between speech intelligibility and maximum speed for tongue body marker (T3).

gion, and low vowels were more significantly affected than were high vowels (Yunusova et al., 2008). Several findings support the suggestion that lingual coupling is most negatively affected during the active phase of disease progression, which is associated with a rapid decline of the speed of lingual movement.

Altered Spatiotemporal Coupling and Movement Speed During the Moderate Stage of Speech Impairment

One of the most prominent findings of this study was that the largest reduction in both movement speed and movement coupling was between the mild and moderate subgroup. These findings raise the possibility that changes in movement speed rather than slow movement speeds are driving the observed declines in lingual spatiotemporal coupling. That is, if slow movement speed were the driving factor, then we would have expected the spatiotemporal coupling of the ALS-severe subgroup to be even more restricted than that of the ALS-moderate subgroup, which was not the case. Specifically, the results of this investigation show significantly lower maximum tongue movement speeds for the talkers with moderate dysarthria as compared to the talkers with mild dysarthria, and no significant difference in speed between the ALS-moderate and ALS-severe ALS subgroups. Therefore, it is possible that the significant change in movement speed (evident between the ALS-mild and ALS-moderate subgroups but not the ALS-moderate and ALS-severe subgroups) rather than slow movement speed (evident in both the ALS-moderate and ALS-severe subgroups) is driving the observed decline in lingual movement coupling.

Table 4. Effect sizes determined using Cohen's *d* for kinematic measures between healthy participants and ALS subgroups across all words and pairs/markers.

Measure	ALS-mild	ALS-moderate	ALS-severe
Average covariance	0.01	0.83	0.58
Interquartile range	0.27	1.12	0.87
Range of covariance	0.48	1.43	1.15
Displacement	–0.53	0.15	0.85
Maximum speed	0.56	2.03	1.61

Large effect sizes = 0.8–2.0; medium effect sizes = 0.5–0.7; small effect sizes = 0.2–0.4

The dramatic decline in movement speed and coupling relations observed between the mildly and moderately affected subgroups suggests that the latter subgroup may be experiencing the most active phase of the disease progression, where motor performance deteriorates rapidly. The differences in severity of tongue impairment across the affected group support the prior suggestion that bulbar decline is characterized by at least three major phases postdiagnosis (DePaul, Robbins, Abbs, & Brooks, 1999). During the first phase, speech movements and speaking rate begin to slow, yet speech intelligibility is only minimally or not affected; during the second phase, speed of movement, speaking rate, and speech intelligibility drop precipitously (Yorkston, Strand, Miller, Hillel, & Smith, 1993). It is during this stage in which the rapid declines in the speeds of tongue movements, which were observed in

the moderate-ALS subgroup, may have a destabilizing effect on lingual coupling, as evidenced by the decrease in coupling relations among different tongue regions. During this phase, talkers are expected to alter their speech movements in an attempt to maintain their speech intelligibility as much as possible (i.e., compensate). Compensatory behaviors may change movements of different organs and their interactions in a significant way. During the third phase, the major losses in oromotor control have already occurred, and speaking intelligibly is no longer a viable option for communication.

Speculation about the Potential Effect of Compensation

The reduced-tongue movement coupling in the moderate-ALS subgroup may be due not only to morphological or functional changes in tongue musculature but also due to the behavioral adaptations to rapidly occurring declines in tongue control. Studies in patients with motor deficits such as decreased movement speed, increased movement variability, and discoordination as a result of stroke suggest that fixation of specific body segments may be an adaptive strategy for constraining the motor system (Bernstein, 1967; Vereijken, van Emmerik, Whiting, & Newell, 1992). Even though the compensatory strategy is adapted to improve motor system stability, it has been shown to limit the mobility and the kinematics of the limb in stroke patients (Cirstea & Levin, 2000). To maintain tongue control in the conditions of rapidly progressing disease, the talkers in the moderately affected group of participants with ALS may similarly restrict the motions of the tongue. In contrast, the talkers with severe dysarthria may have been less capable of effectively using this strategy and consequently imposed fewer constraints on the movements of the tongue.

Differences across Tongue Regions

The ALS-moderate subgroup showed significant impairment in spatiotemporal coupling primarily in the mid-posterior tongue. Because coupling relations involving the anterior tongue regions were relatively unaffected, there may be differential impairment across the muscle segments, namely the core and lateral aspects of intrinsic and extrinsic tongue muscles. The morphological organization of tongue musculature may underlie regional differences in tongue function deterioration. The core of the tongue controls elongation, protrusion, and body deformation (Takemoto, 2001). Portions of the extrinsic muscles external to the core control tongue-tip movements. The current findings suggest that these lateral aspects of the tongue may be affected to a lesser extent than muscles that comprise the core. Interestingly, a histopathologic study of the tongue musculature in ALS reported greater degenerative changes to muscle fiber type, muscle fiber group, and connective tissue to the anterior tongue regions compared to the posterior regions (DePaul et al., 1998). This discrepancy might suggest that the behavioral changes occur in the region that might be less structurally impaired; if the mid-posterior tongue is less affected structurally compared to the anterior tongue, then it might be recruited to compensate for a disproportionately affected anterior tongue. A similar response has been observed in the jaw, which remains relatively spared compared to the tongue yet demonstrates significant compensatory behavior, such as greater movement and speed, in order to accommodate a weak and immobile tongue (DePaul & Brook, 1993; Yunusova et al., 2010). The effects of structural changes on behaviors such as speech and swallowing are not well understood and require further investigation.

Kinematic Measures as Early Indicators of Disease Progression and Speech Intelligibility Decline

Sensitive markers of disease progression are needed for improved prognostic accuracy and to serve as outcome measures in clinical trials of therapeutic drugs to treat ALS. To determine whether our various behavioral measures were sensitive to disease progression, we conducted sensitivity/specificity analyses. Maximum speed was the most sensitive (75%) kinematic measure for the mildly affected subgroup, and all kinematic measures were highly sensitive (100%) for the moderately and severely affected subgroups. Across all groups, the specificity of the range of covariance (87.5%) and maximum speed (77.77%) were much higher than that of the other kinematic measures. These findings support the suggestion that tongue kinematics may provide a useful indicator of bulbar disease severity and progression (Yunusova et al., 2010).

Tongue speed and range of spatiotemporal coupling decreased as talkers became less intelligible. A high correlation was evident between speech intelligibility and maximum speed for all tongue markers except the tongue blade marker T2. Similarly, a moderate correlation was evident between speech intelligibility and the range of covariance. Visual examination of the relevant bivariate plots (see Figures 7 and 8) suggest that speech intelligibility dropped when tongue movement speed in the tongue body region (T3) dropped below 100 mm per s and when the range of tongue coupling fell below 2.60. These findings parallel previous reports that showed a precipitous decline in speech intelligibility once speaking rate drops below 120 words per min (Ball, Beukelman, & Pattee, 2002; Yorkston et al., 1993).

Combined, the current findings might suggest that the slowing of tongue movements due to ALS restricts lingual coupling in a way that reduces speech intelligi-

bility. Prior research suggests a potential link between speaking rate and articulatory coordination, with voluntary slowing having a destabilizing effect on articulatory movements (Adams et al., 1993). Additional work is needed, however, to establish causal links between these movement variables and speech intelligibility in a large group of patients with ALS, taking into consideration that articulatory movement decline is just one of many ALS-related changes occurring within the speech system that may negatively impact speech.

Conclusions

The observed reduced spatiotemporal coupling between tongue regions in advanced ALS could be the result of significant structural, functional, and/or behavioral compensatory changes to the mid-posterior tongue. Decreased coupling coincided with significant decreases in movement speed and may be indicative of an active disease phase characterized by significant structural and/or functional deterioration. Both range of tongue coupling and tongue speed correlated with speech intelligibility. Compared to the covariance and conventional kinematic measures, movement speed had the highest sensitivity values early in the disease process. The results of this investigation provide evidence for strong associations between reduced speed, decreased tongue coupling relations, and speech intelligibility decline in talkers with ALS.

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