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**The association between longitudinal declines in speech sound accuracy and speech intelligibility in speakers with amyotrophic lateral sclerosis**

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27 **ABSTRACT**

28           The purpose of this study was to examine how neurodegeneration secondary to  
29 amyotrophic lateral sclerosis (ALS) impacts speech sound accuracy over time and how speech  
30 sound accuracy, in turn, is related to speech intelligibility. Twenty-one participants with ALS  
31 read the Bamboo Passage over multiple data collection sessions across several months.  
32 Phonemic and orthographic transcriptions were completed for all speech samples. The  
33 percentage of phonemes accurately produced was calculated across each phoneme, sound class  
34 (i.e., consonants versus vowels), and distinctive feature (i.e., features involved in Manner of  
35 Articulation, Place of Articulation, Laryngeal Voicing, Tongue Height, and Tongue  
36 Advancement). Intelligibility was determined by calculating the percentage of words correctly  
37 transcribed orthographically by naive listeners. Linear mixed effects models were conducted to  
38 assess the decline of each distinctive feature over time and their impact on intelligibility. The  
39 results demonstrated that overall phonemic production accuracy had a nonlinear relationship  
40 with speech intelligibility and that a subset of features (i.e., those dependent on precise lingual  
41 and labial constriction and/or extensive lingual and labial movement) were more important for  
42 intelligibility and were more impacted over time than other features. Furthermore, findings  
43 revealed that consonants were more strongly associated with intelligibility than vowels, but  
44 consonants did not significantly differ from vowels in their decline over time. These findings  
45 have the potential to (1) strengthen mechanistic understanding of the physiological constraints  
46 imposed by neuronal degeneration on speech production and (2) inform the timing and selection  
47 of treatment and assessment targets for individuals with ALS.

48 **Keywords:** phonemic analysis, intelligibility, neurodegenerative disease, speech impairment

49

50 **WORD COUNT:** 7090 words

51

52 **Introduction**

53 Amyotrophic lateral sclerosis (ALS) is a neurodegenerative disease that results in the  
54 degeneration of both upper and lower motor neurons, which causes progressive weakness and  
55 atrophy of muscles (Strong et al., 2017). Speech deficits are a common initial symptom of the  
56 disease and lead to loss in intelligibility and, eventually, of all verbal communication in 75-95%  
57 of individuals (Brownlee & Bruening, 2012; Carpenter et al., 1978). In progressive dysarthrias,  
58 such as those associated with ALS, deficits in intelligibility have been linked to a reduction in  
59 articulatory performance (De Bodt et al., 2002; Lee et al., 2014; Rong et al., 2015; Sidtis et al.,  
60 2011; Yunusova et al., 2008). Although clinicians use perceptual judgment to assess the state of  
61 bulbar disease, researchers have typically characterized the decline in articulatory precision using  
62 speech acoustics or kinematics (Kuruvilla et al., 2012; Rong & Green, 2019; Wang et al., 2018;  
63 Yunusova et al., 2012). Few studies have examined how these articulatory impairments constrain  
64 speech sound production (i.e., based on perceptual judgment) and how the resulting speech errors  
65 degrade intelligibility over time. This knowledge is needed to (1) optimize the timing and  
66 selection of treatment goals focused on prolonging oral communication and (2) strengthen our  
67 understanding of the physiological constraints secondary to neuronal degeneration.

68 To effectively manage progressive dysarthria, there is a pressing need to understand how  
69 speech sound accuracy deteriorates over time. The few studies that examined speech sound  
70 errors in progressive dysarthria secondary to ALS were conducted two to three decades ago  
71 (Bunton & Weismer, 2001; DePaul & Kent, 2000; Kent et al., 1989, 1990, 1991; Riddel et al.,  
72 1995). These studies found a high proportion of stop-nasal contrast errors (Kent et al., 1989,  
73 1991) and a higher error rate for voiceless sounds than for voiced sounds (Riddel et al., 1995).

74 However, this research was largely cross-sectional (Bunton & Weismer, 2001; Kent et al., 1989,  
75 1990; Riddell et al., 1995), with the exception of a few longitudinal case studies (DePaul & Kent,  
76 2000; Kent et al., 1991). Furthermore, these studies primarily examined productions of isolated  
77 words rather than connected speech (Bunton & Weismer, 2001; DePaul & Kent, 2000; Kent et  
78 al., 1989, 1990, 1991; Riddell et al., 1995); although single-word speech batteries are efficient to  
79 administer and easier to experimentally control for confounds such as phonetic environment,  
80 longer utterances provide a more representative assessment of speech function (Allison et al.,  
81 2019).

82 A number of studies have examined the relative impact of different speech sound errors  
83 on intelligibility, which revealed a large effect of impaired consonants and, specifically, impaired  
84 fricatives and affricates (Ansel & Kent, 1992; Bunton et al., 2007; Hodge et al., 2013; Kim et al.,  
85 2010; McLeod et al., 2012; Platt et al., 1980). Many of these studies, however, have been  
86 conducted with speakers with non-progressive dysarthria secondary to cerebral palsy (CP),  
87 which precludes examining how declines in speech sound accuracy affect intelligibility *over*  
88 *time*. The need to assess longitudinal change is crucial because the capabilities and needs of  
89 patients with neurodegeneration change significantly as the disease progresses (Yorkston &  
90 Beukelman, 1991).

91 Moreover, prior work suggests that the spread of motor deterioration is not uniform  
92 across cranial nerve nuclei. That is, ALS may result in differential impairment of the speech  
93 mechanism over time (e.g., lingual muscles may be more impaired than labial muscles at  
94 different points across the disease) (DePaul et al., 1988; Kent et al., 1990; Langmore & Lehman,  
95 1994; Lawyer & Netsky, 1953; Stipancic et al., 2021; Yunusova et al., 2008). Indeed, tongue  
96 weakness has been shown to be more prominent than weakness in other articulators (DePaul &

97 Brooks, 1993; Solomon et al., 2017; Wood et al., 1992). This differential impairment can affect  
98 different speech sounds to varying degrees and therefore can be evidenced, in part, by patterns of  
99 speech sound accuracy. For example, difficulty producing alveolars but not bilabials may  
100 indicate tongue tip weakness with spared upper and lower lip function. Similarly, difficulty  
101 producing velars but not alveolars could indicate tongue back weakness with spared tongue tip  
102 function.

103 In the current study, we investigated (1) the impact of speech errors on intelligibility in a  
104 paragraph-reading task and (2) the changes in speech error patterns over time in a cohort of 21  
105 speakers with ALS. This study differs from prior work on speech error patterns in ALS (Bunton  
106 & Weismer, 2001; DePaul & Kent, 2000; Kent et al., 1989, 1990, 1991; Riddel et al., 1995) in  
107 that our focus was to characterize distinctive feature accuracy rather than the *types* of sound error  
108 (e.g., distortions, substitutions, deletions). Distinctive feature errors were of particular interest  
109 because they can provide insights into underlying physiological impairments. If we observe a  
110 preponderance of errors in alveolars but not in bilabials, we might posit that speakers are losing  
111 function over their tongue tip muscles prior to lip muscles; or if we observe a preponderance of  
112 errors in fricatives but not glides, we might posit that speakers are losing fine force control but  
113 can still achieve more coarse, low velocity movements.

114 To this end, the following research questions were addressed:

- 115 1. What is the overall effect of phonemic accuracy on speech intelligibility in speakers with  
116 ALS?
- 117 2. Which sound class (i.e., consonants versus vowels) and distinctive feature (e.g.,  
118 fricatives) errors have the greatest impact on speech intelligibility in speakers with ALS?

119 3. Do different sound classes and distinctive features degrade similarly over time (as  
120 represented by data collection sessions) in a given speaker with ALS?

121

## 122 **Materials and methods**

### 123 *Participants*

124 Data were collected from twenty-one participants who were diagnosed with ALS by a  
125 neurologist using El Escorial criteria (Brooks et al., 2000) at the University of Texas, Dallas  
126 (UTD); MGH Institute of Health Professions (MGH IHP); and the University of Nebraska,  
127 Lincoln (UNL). This data was part of a multi-site, longitudinal study on bulbar degeneration in  
128 ALS. Table 1 provides demographic data for each participant, including age at the first data  
129 collection session, sex, the number of sessions each participant participated in, and the number of  
130 days between the first and last sessions. Intelligibility at the first session ranged from 37 to 100%  
131 ( $M = 84.14$ ,  $SD = 18.54$ ), while intelligibility at the last session ranged from 0 to 100% ( $M =$   
132  $48.99$ ,  $SD = 47.40$ ). All participants spoke English as their native language, passed a bilateral  
133 hearing screening at 25 dB at 1000, 2000, and 4000 Hz, reported normal or correct-to-normal  
134 vision, and passed the Montreal Cognitive Assessment (MoCA).

135

**Table 1.** Demographic and speech characteristics of the ALS participants.

Subject	Sex	Age at First Session	Number of Data Collection Sessions	Days between First and Last Session (Timepoint 1 and Timepoint 2)	Intelligibility at First Session	Intelligibility at Last Session	Database
ALS01	M	46	3	243	83%	0%	UNL
ALS02	M	44	5	148	87%	0%	UNL
ALS03	M	51	6	133	90%	77%	UNL
ALS04	M	54	9	238	89%	0%	UNL
ALS05	F	64	8	245	90%	1%	UNL
ALS06	F	48	5	226	58%	0%	UNL
ALS07	M	77	4	196	90%	0%	UNL
ALS08	F	62	4	101	68%	0%	UNL

ALS09	F	62	7	238	37%	33%	UNL
ALS10	M	44	3	81	38%	33%	UNL
ALS11	M	53	4	542	98%	96%	UTD
ALS12	M	61	4	591	90%	100%	UTD
ALS13	M	52	3	428	92%	0%	UTD
ALS14	F	62	4	367	93%	0%	UTD
ALS15	M	44	3	444	100%	100%	UTD
ALS16	F	54	4	547	92%	100%	UTD
ALS17	M	42	3	386	100%	94%	UTD
ALS18	M	52	3	393	86%	100%	UTD
ALS19	F	57	3	295	100%	98%	UTD
ALS20	F	52	3	504	86%	100%	MGH IHP
ALS21	M	56	3	591	100%	98%	MGH IHP

*Note.* UNL = University of Nebraska, Lincoln; UTD = University of Texas, Dallas; MGH IHP = MGH Institute of Health Professions.

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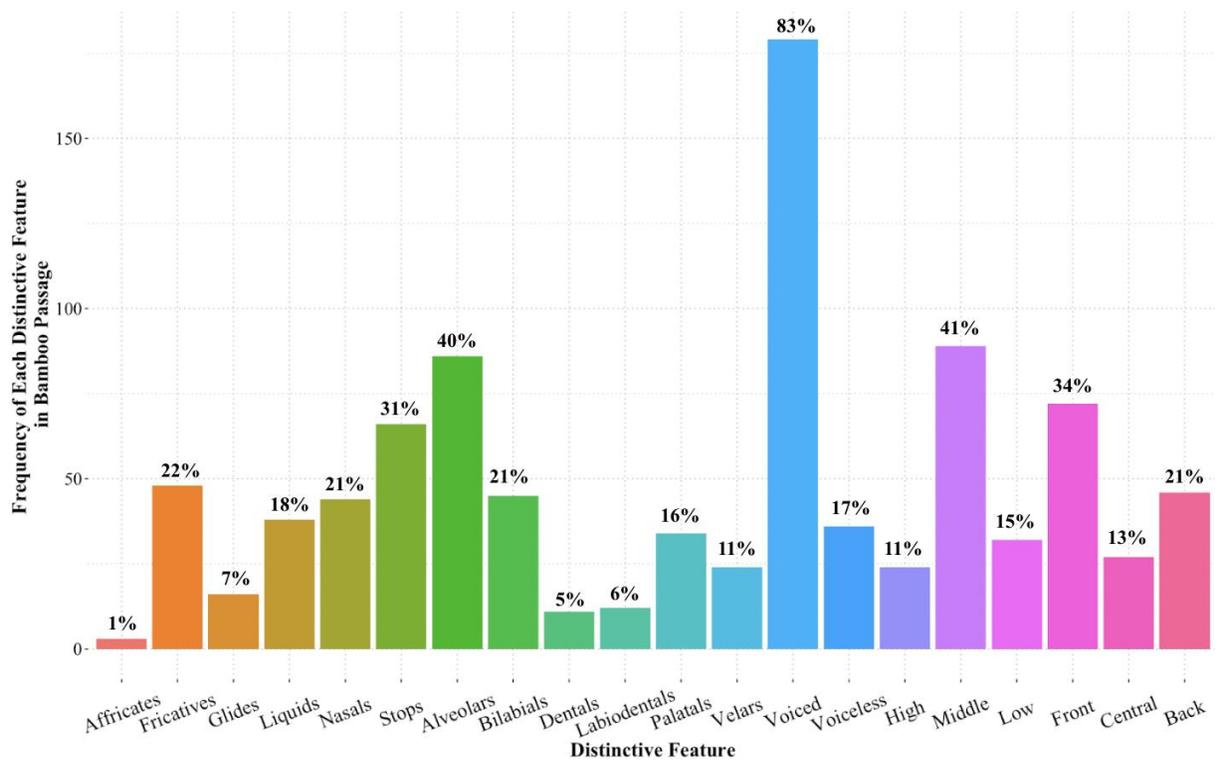
137 ***Procedures***

138 *Data collection*

139 Participants came to the clinic at the respective institution (UTD, MGH IHP, or UNL) for  
140 multiple data collection sessions, separated by three months on average. The study protocol was  
141 approved by the institutional review boards of all institutions involved in the study, and written  
142 informed consent was obtained from all participants. The number of days between the first and  
143 last sessions varied from 81 to 591 days ( $M = 330.33$ ,  $SD = 163.97$ ). See Table 1 for the number  
144 of sessions and days between the first and last session for each subject. Data from all sessions  
145 were of interest for Research Questions 1 and 2, but only data from the first and last sessions  
146 were of interest for Research Question 3 (more detail is provided below). As part of a larger  
147 experimental testing protocol, participants read the Bamboo Passage (Green et al., 2004) (see  
148 Appendix) at each session, which included eight sentences and 97 words (215 total phonemes).  
149 Participants were instructed to read the passage at a comfortable speaking rate and loudness. See  
150 Appendix for the complete Bamboo Passage and a bar chart illustrating the frequency (and  
151 percentage of total phonemes in the passage) of each distinctive feature in the passage.

152 **Appendix**

153 Bamboo walls are getting to be very popular. They are strong, easy to use, and good-looking.  
154 They provide a good background and can create a look of a Japanese garden. Bamboo is one of  
155 the largest and most rapidly growing grasses all over the world. Many varieties of bamboo are  
156 grown in Asia, although it is also grown in America. Last year we bought a new home and have  
157 been working on the flower garden. In a few more days, we will be done with the bamboo wall  
158 in our garden. We have really enjoyed the project.



159 Bar chart illustrating the frequency (and percentage of total phonemes in the passage) of each  
160 distinctive feature in the Bamboo Passage.  
161  
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163 *Recording and phonemic transcription*

164 Speech samples were recorded using a high-quality Shure Microflex microphone (at 22  
165 kHz sampling rate) placed approximately 15 mm from the participant's mouth. One research  
166 assistant, who had completed graduate-level coursework and advanced training in phonemic

167 transcription, identified and phonemically transcribed each word produced by participants using  
168 the transcription consensus procedures described by Shriberg and colleagues (Shriberg et al.,  
169 1984). The research assistant spoke English as their native language and had normal hearing. The  
170 phonemic transcriptions were then entered into the open-source software Programs to Examine  
171 Phonetic and Phonological Evaluation Records (PEPPER) (Shriberg et al., 2000). To assess  
172 interrater reliability for the phonemic transcriptions, a second research assistant with the same  
173 qualifications as the first transcribed a randomly selected 10% of the Bamboo Passage samples.  
174 The intraclass correlation coefficient for absolute agreement (i.e., ICC[2,1]) was used to  
175 calculate reliability between the two raters and demonstrated strong phoneme-by-phoneme  
176 agreement between the two transcribers ( $ICC = .91, p < .001$ ).

177

### 178 *Speech intelligibility ratings*

179 Undergraduate and graduate students at UTD were recruited through advertisements via  
180 flyers, emails, and verbal announcements to participate as listeners for the study. There was one  
181 listener per each unique participant-timepoint pair. Thus, each listener transcribed only one  
182 Bamboo passage read by a participant with ALS to remove the effect of familiarity (Beukelman  
183 & Yorkston, 1980). All listeners spoke English as their native language, passed hearing  
184 screenings, and were not familiar with the Bamboo Passage nor with the goals of the study. Each  
185 listener listened to the Bamboo passage produced by one speaker with ALS and was instructed to  
186 orthographically transcribe each word across the eight sentences. Each passage was presented to  
187 the listeners using professional-grade headphones. The following instructions were given to each  
188 listener: *You will hear a speaker reading a short passage. We would like you to type each word*  
189 *the speaker produces in the order that you hear them. If a word is unintelligible (can't be*

190 *understood as a real word), please type an 'X' where the word is in the sentence. You are*  
191 *allowed to go back and re-listen to any portion of the audio that you feel is necessary.*

192 Intelligibility for each speaker was determined by calculating the percentage of words correctly  
193 transcribed by the listener. To calculate interrater reliability for the intelligibility transcriptions,  
194 10% of all Bamboo samples recorded were randomly selected, transcribed independently by a  
195 second blinded naive listener, and intelligibility was calculated. ICC(2,1) was again used to  
196 calculate reliability between the two intelligibility scores obtained from the two raters and  
197 demonstrated strong agreement ( $ICC = .92, p < .001$ ).

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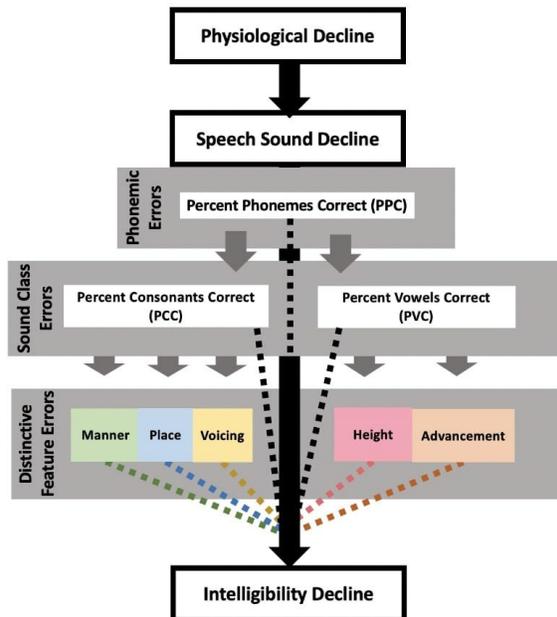
### 199 ***Phoneme, sound class, and distinctive feature analysis***

200 The PEPPER software was used to assess the speech production of individual speakers.  
201 The phonemic transcriptions provided by the research assistants and the target sentences (i.e., the  
202 eight sentences as written in the Bamboo Passage) were entered into PEPPER, which provided  
203 target phonemic transcriptions of each sentence. PEPPER then completed the following three  
204 analyses by comparing the phonemic transcriptions (i.e., those provided by the research  
205 assistants and the target transcriptions provided by the software program): (1) *phoneme*-level  
206 analysis (Phonemic Errors in Figure 1), which consisted of percent phonemes correct (PPC); (2)  
207 *sound class*-level analysis (Sound Class Errors in Figure 1), which consisted of percent  
208 consonants correct (PCC) and percent vowels correct (PVC); and (3) *distinctive feature*-level  
209 analysis (Distinctive Feature Errors in Figure 1), which consisted of the percent accuracy of  
210 features involved in Manner of Articulation (affricates, fricatives, glides, liquids, nasals, stops),  
211 Place of Articulation (alveolar, bilabial, dentals, labiodental, palatal, velar), Laryngeal Voicing  
212 (voiced consonants and voiceless consonants), Tongue Height (high, middle, low), and Tongue

213 Advancement (front, central, back). For example, a /p/ that was produced as an /f/ would count  
 214 as an incorrect phoneme at the Phonemic Error level, incorrect consonant at the Sound Class  
 215 Error level, and incorrect Manner (stop -> fricative) and Place (bilabial -> labiodental) but not  
 216 Voicing (i.e., it was correctly produced as a voiceless consonant) at the Distinctive Feature Error  
 217 level. Figure 1 illustrates the connections between the three levels and their position in the causal  
 218 pathway from physiological decline to intelligibility decline.

219

220 **Figure 1.** Flow chart illustrating the connection between physiological decline, speech sound  
 221 decline, and intelligibility decline. Neurologic abnormalities due to neurodegenerative diseases,  
 222 such as amyotrophic lateral sclerosis (ALS), lead to Physiological Decline, including that of the  
 223 speech subsystems. Physiological Decline of speech subsystem function leads to Speech Sound  
 224 Decline, or the inability to produce correct speech sounds, which then results in Intelligibility  
 225 Decline. The dotted lines indicate the relationships we examined in this study between each level  
 226 of analysis and intelligibility.



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228

## 229 *Statistical analysis*

230 *Effect of phoneme accuracy on intelligibility (Research Question 1).* For the first research  
 231 question, we sought to determine the relationship between PPC and intelligibility. Because we

232 used data from all sessions and each participant had multiple sessions, a linear mixed effects  
233 (LME) model was conducted by regressing intelligibility on PPC with subject as a random  
234 intercept.

235 *Sound class and distinctive feature errors with the greatest impact on intelligibility*  
236 *(Research Question 2)*. For the second research question, we examined the effects of sound class  
237 accuracy (i.e., accuracy of consonants [PCC] and vowels [PVC]) and distinctive feature accuracy  
238 (i.e., accuracy of Manner features, Place features, Voicing features, Height features, and  
239 Advancement features) on intelligibility. Again, because we used data from all sessions and each  
240 participant had multiple sessions, LME models were conducted by regressing intelligibility on  
241 sound class (i.e., consonants and vowels) or distinctive features (e.g., affricates) with subject as a  
242 random intercept.

243 *Extent of decline over time for each sound class and distinctive feature (Research*  
244 *Question 3)*. For the third research question, we examined change over time for the accuracy of  
245 each sound class and distinctive feature. In contrast to Research Questions 1 and 2, we only used  
246 data from the first and last sessions. The data were thus stratified into two timepoints: ‘Timepoint  
247 1’, which was the first session, and ‘Timepoint 2’, which was the last session. We then created a  
248 new variable (i.e., ‘Difference’), which consisted of the difference in accuracy for each sound  
249 class or distinctive feature from Timepoint 1 to Timepoint 2 (e.g., accuracy of Fricatives at  
250 Timepoint 1 minus accuracy of Fricatives at Timepoint 2). Importantly, because the date of  
251 disease onset was not available for all participants, we were unable to determine the extent of  
252 decline *from initial diagnosis*. Instead, we captured the extent of decline from each participant’s  
253 initial session. LME models were conducted by regressing the difference in accuracy on sound

254 class or distinctive feature with subject as a random intercept. The contrasts were then assessed  
255 for each model.

256 For the analyses for Research Questions 2 and 3, six separate LME were conducted (i.e.,  
257 sound classes, Manner features, Place features, Voicing features, Height features, and  
258 Advancement features). We conducted individual models because the small sample size reduced  
259 the power necessary to combine all predictors into one model. Additionally, conducting  
260 individual models allowed us to include only between-group predictors (and no within-group  
261 predictors) in a single model. For example, because Manner and Place features overlap with  
262 Voicing features (e.g., a fricative or an alveolar can be voiced or voiceless), it was advantageous  
263 to examine Manner and Place separately from Voicing.

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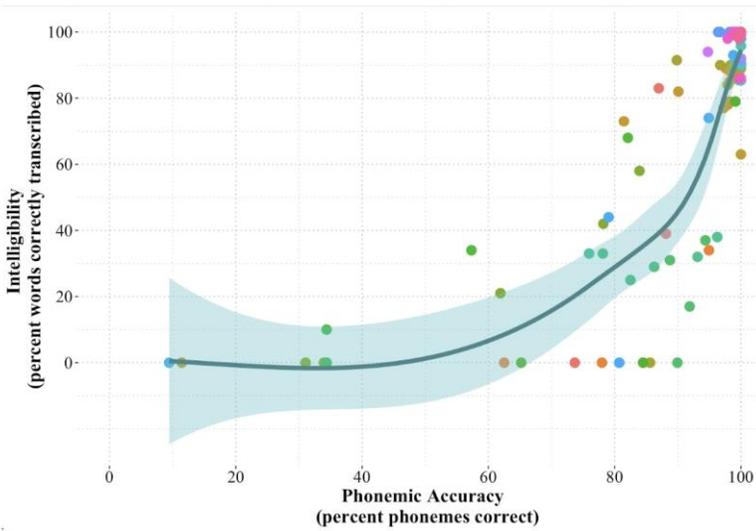
## 265 **Results**

### 266 *Effect of phoneme accuracy on intelligibility (Research Question 1)*

267 Average intelligibility across all participants was 84.14% words correct ( $SD = 18.54$ ) in  
268 the first session and 48.99% words correct ( $SD = 47.40$ ) in the last session. The overall LME  
269 analysis revealed a significant effect of PPC on intelligibility ( $b = 1.39$ ,  $SE = .11$ ,  $t(88.86) =$   
270  $12.27$ ,  $p < .001$ ). The strength of the association between the two variables was also examined  
271 using the standardized beta coefficient, which indicated a strong association ( $B = .77$ ). This  
272 relationship was determined to be non-linear, as small declines in PPC initially corresponded  
273 with large declines in intelligibility, but once PPC was lower than 75%, even large declines in  
274 PPC did not result in noticeable declines in intelligibility (see Figure 2).

275 **Figure 2.** Scatterplot illustrating the relationship between percent phonemes correct [PPC] and  
276 intelligibility. The colored dots represent each participant, with multiple sessions per participant  
277 (and therefore multiple dots of the same color). Although an arcsine transformation was  
278 performed on the two variables for the statistical analyses, no transformation was performed on

279 the data for this visualization to ease interpretability. A lowess (locally weighted scatterplot  
280 smoothing) regression line was also used to illustrate the relationship between the two variables.



281

282

283 ***Sound class and distinctive feature errors with the greatest impact on intelligibility (Research***

284 ***Question 2)***

285 The results of the LME analyses examining the effects of sound class accuracy (i.e.,  
286 accuracy of consonants [PCC] and vowels [PVC]) and distinctive feature accuracy (i.e., accuracy  
287 of all Manner features, Place features, Voicing features, Height features, and Advancement  
288 features) on intelligibility are summarized in Table 2 and Figure 3. Most notably, for the sound  
289 class model, consonants were significantly associated with intelligibility, whereas vowels were  
290 not. For the distinctive feature models, the following features were significantly associated with  
291 intelligibility: fricatives and affricates in the Manner model, alveolars and labiodentals in the  
292 Place model, voiced consonants in the Voicing model, low and high vowels in the Height model,  
293 and front and back vowels in the Advancement model.

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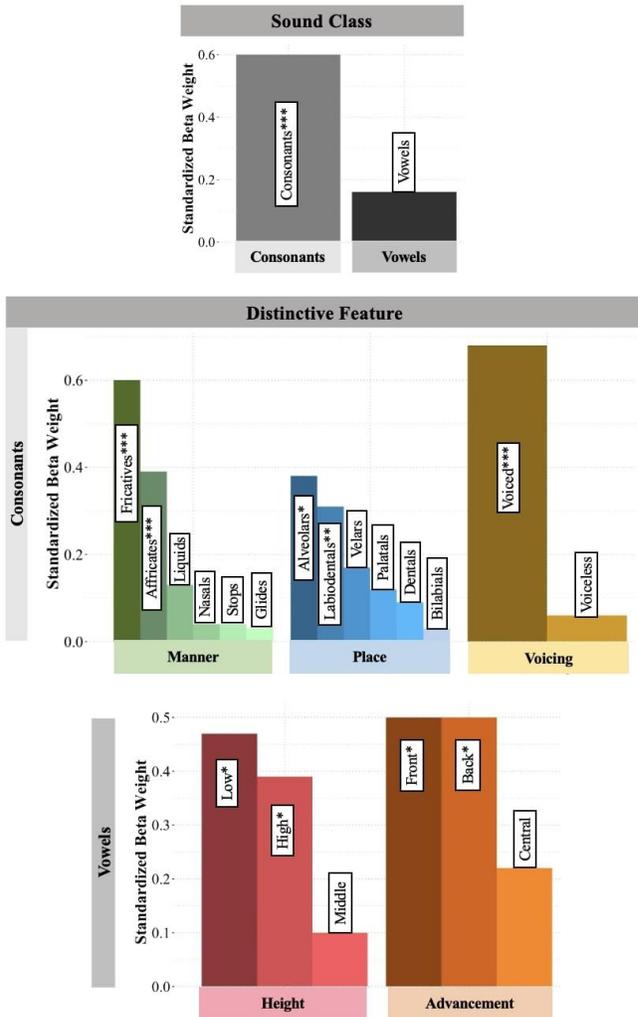
295 **Table 2.** Linear mixed effects (LME) models examining the association between intelligibility  
296 and each sound class and each distinctive feature.

LME Model	Fixed Effects	Estimate	SE	df	t	p
Consonants versus Vowels						
	<b>PCC</b>	<b>1.17</b>	<b>.26</b>	<b>86.67</b>	<b>4.49</b>	<b>&lt; .001***</b>
	PVC	0.24	.22	87.57	1.13	.26
Manner						
	<b>Affricates</b>	<b>0.42</b>	<b>0.10</b>	<b>72.50</b>	<b>4.00</b>	<b>&lt; .001***</b>
	<b>Fricatives</b>	<b>0.89</b>	<b>0.26</b>	<b>82.75</b>	<b>3.48</b>	<b>&lt; .001***</b>
	Glides	0.07	0.23	75.35	0.29	.77
	Liquids	-0.25	0.24	70.94	-1.06	.29
	Nasals	-0.13	0.33	77.94	-0.41	.69
	Stops	-0.06	0.28	75.58	-0.22	.83
Place						
	<b>Alveolars</b>	<b>0.68</b>	<b>0.27</b>	<b>79.96</b>	<b>2.51</b>	<b>.01*</b>
	Bilabials	-0.06	0.30	76.67	-0.21	.84
	Dentals	0.14	0.23	73.98	0.64	.53
	<b>Labiodentals</b>	<b>0.35</b>	<b>0.13</b>	<b>73.96</b>	<b>2.67</b>	<b>&lt; .01**</b>
	Palatals	-0.22	0.26	75.99	-0.87	.39
	Velars	0.27	0.23	83.99	1.21	.23
Voicing						
	<b>Voiced</b>	<b>1.40</b>	<b>0.32</b>	<b>87.93</b>	<b>4.42</b>	<b>&lt; .001***</b>
	Voiceless	0.08	0.20	87.57	0.41	.69
Height						
	<b>Low</b>	<b>0.64</b>	<b>0.29</b>	<b>84.91</b>	<b>2.19</b>	<b>.03*</b>
	Middle	-0.15	0.33	72.80	-0.45	.65
	<b>High</b>	<b>0.52</b>	<b>0.25</b>	<b>83.04</b>	<b>2.05</b>	<b>.04*</b>
Advancement						
	<b>Front</b>	<b>0.71</b>	<b>0.28</b>	<b>85.77</b>	<b>2.53</b>	<b>.01*</b>
	Central	-0.32	0.28	81.24	-1.16	.25
	<b>Back</b>	<b>0.70</b>	<b>0.30</b>	<b>86.90</b>	<b>2.37</b>	<b>.02*</b>

Note. SE = standard error; PCC = percent consonants correct; PVC = percent vowels correct; \* =  $p < .05$ , \*\* =  $p < .01$ , \*\*\* =  $p < .001$ .

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308

309 **Figure 3.** Bar charts of the standardized beta coefficients illustrating the impact of each sound class and distinctive feature on intelligibility (\* =  $p < .05$ , \*\* =  $p < .01$ , \*\*\* =  $p < .001$ ).  
 310



311

312

313 ***Extent of decline over time for each sound class and distinctive feature (Research Question 3)***

314 The results of the LME analyses examining the differences in extent of decline between  
 315 sound classes and between distinctive features within each group (i.e., Manner, Place, Voicing,  
 316 Height, and Advancement) are summarized in Table 3 and Figure 4. Most notably, for the sound  
 317 class model, consonants and vowels did not significantly differ in their extents of decline. For the  
 318 distinctive feature models, fricatives, affricates, and stops declined to a significantly greater  
 319 extent than nasals in the Manner model; labiodentals declined to a significantly greater extent

320 than bilabials and palatals in the Place model; voiceless consonants declined to a significantly  
 321 greater extent than voiced consonants in the Voicing model; and vowels in both Height and  
 322 Advancement models did not significantly differ in their extents of decline.

323  
 324 **Table 3.** Contrasts from linear mixed effects (LME) models examining differences in the extent  
 325 of decline across sessions for each sound class and distinctive feature.

LME Model	Contrast	Estimate	SE	df	t	p
Consonants versus Vowels						
	PCC - PVC	0.02	0.05	20	0.35	.73
Manner						
	Affricates - Fricatives	-0.05	0.08	100	-0.56	.99
	<b>Affricates - Glides</b>	<b>-0.24</b>	<b>0.08</b>	<b>100</b>	<b>-2.96</b>	<b>&lt; .05*</b>
	Affricates - Liquids	-0.15	0.08	100	-1.90	.40
	<b>Affricates - Nasals</b>	<b>-0.35</b>	<b>0.08</b>	<b>100</b>	<b>-4.33</b>	<b>&lt; .001***</b>
	Affricates - Stops	-0.05	0.08	100	-0.66	.99
	Fricatives - Glides	-0.20	0.08	100	-2.40	.17
	Fricatives - Liquids	-0.11	0.08	100	-1.34	.76
	<b>Fricatives - Nasals</b>	<b>-0.31</b>	<b>0.08</b>	<b>100</b>	<b>-3.76</b>	<b>&lt; .01**</b>
	Fricatives - Stops	-0.01	0.08	100	-0.09	1.00
	Glides - Liquids	0.09	0.08	100	1.06	.90
	Glides - Nasals	-0.11	0.08	100	-1.36	.75
	Glides - Stops	0.19	0.08	100	2.30	.20
	Liquids - Nasals	-0.20	0.08	100	-2.42	.16
	Liquids - Stops	0.10	0.08	100	1.23	.81
	<b>Nasals - Stops</b>	<b>0.30</b>	<b>0.08</b>	<b>100</b>	<b>3.67</b>	<b>&lt; .01**</b>
Place						
	Alveolars - Bilabials	-0.06	0.06	100	-0.91	.94
	Alveolars - Dentals	-0.02	0.06	100	-0.39	1.00
	Alveolars - Labiodentals	0.13	0.06	100	2.23	.24
	Alveolars - Palatals	-0.07	0.06	100	-1.22	.83
	Alveolars - Velars	-0.01	0.06	100	-0.11	1.00
	Bilabials - Dentals	0.03	0.06	100	0.52	1.00
	<b>Bilabials - Labiodentals</b>	<b>0.19</b>	<b>0.06</b>	<b>100</b>	<b>3.14</b>	<b>.03*</b>
	Bilabials - Palatals	-0.02	0.06	100	-0.31	1.00
	Bilabials - Velars	0.05	0.06	100	0.80	.97
	Dentals - Labiodentals	0.16	0.06	100	2.62	.10
	Dentals - Palatals	-0.05	0.06	100	-0.83	.96
	Dentals - Velars	0.02	0.06	100	0.28	1.00
	<b>Labiodentals - Palatals</b>	<b>-0.21</b>	<b>0.06</b>	<b>100</b>	<b>-3.44</b>	<b>.01*</b>
	Labiodentals - Velars	-0.14	0.06	100	-2.34	.19
	Palatals - Velars	0.07	0.06	100	1.11	.88
Voicing						
	<b>Voiced - Voiceless</b>	<b>.18</b>	<b>0.05</b>	<b>20</b>	<b>3.62</b>	<b>&lt; .01**</b>

Height

High - Middle	-0.09	0.04	40	-2.17	.09
High - Low	0.01	0.04	40	0.18	.98
Middle - Low	0.09	0.04	40	2.34	.06

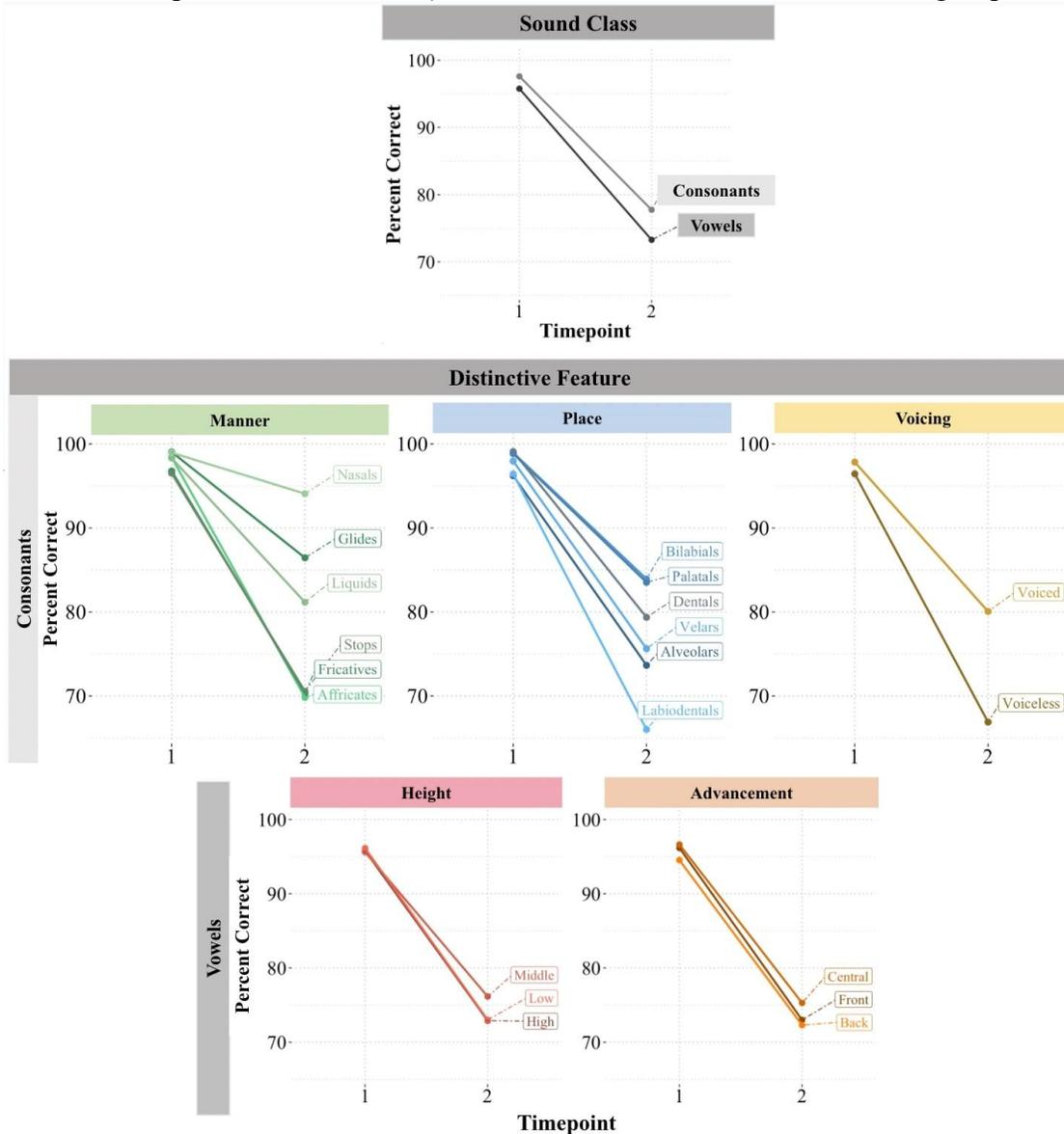
Advancement

Front - Central	-0.03	0.04	40	-0.72	.75
Front - Back	0.01	0.04	40	0.15	.99
Central - Back	0.04	0.04	40	0.87	.66

Note. SE = standard error; \* =  $p < .05$ , \*\* =  $p < .01$ , \*\*\* =  $p < .001$ .

326

327 **Figure 4.** Spaghetti plots illustrating the average extent of decline over time (Timepoint 1 = first  
328 session; Timepoint 2 = last session) for each distinctive feature within each group.



329

330 **Discussion**

331           This work aimed to examine the impact of progressive dysarthria on speech sound  
332 accuracy and how speech errors affect speech intelligibility. The results can be summarized in  
333 three primary findings. First, the impact of phonemic errors on intelligibility was strong but non-  
334 linear. Second, intelligibility was dependent more on the accuracy of consonants than vowels and  
335 on the accuracy of a relatively small number of distinctive features (nine out of 20). Third, the  
336 majority of the consonant types that were most important for intelligibility (i.e., fricatives,  
337 affricates, and labiodentals, which putatively require precise lingual and/or labial constriction)  
338 also declined the most over time. These findings have the potential to (1) strengthen mechanistic  
339 understanding of the physiological constraints imposed by neuronal degeneration in ALS and (2)  
340 inform the timing and selection of treatment and assessment targets for individuals with  
341 progressive dysarthria secondary to ALS.

342

343 ***The impact of phonemic errors on intelligibility is strong but non-linear***

344           The strong association between PPC and intelligibility was expected and consistent with  
345 findings reported in children and individuals with non-progressive dysarthria (Bunton et al.,  
346 2007; Hodge et al., 2013; McLeod et al., 2012). The curvilinear association between these  
347 variables (see Figure 2) suggests that when at least 75% of phonemes were produced correctly,  
348 small declines in PPC resulted in large declines in intelligibility. However, once more than 75%  
349 of phonemes were produced incorrectly, the association between PPC and intelligibility  
350 weakened (i.e., decreases in PPC were not associated with clear decreases in intelligibility).  
351 These findings raise the possibility that the benefits of improving PPC as an intelligibility-  
352 enhancing strategy may be limited for speakers with low PPCs (i.e., lesser than 75%).

353           The sharp decline in intelligibility based on a small decrease in the upper range of PPC  
354 (i.e., 75% and greater) suggests that the phonemes produced incorrectly were important for  
355 intelligibility. This interpretation is supported by our finding that most of the consonant types  
356 that significantly degraded intelligibility (i.e., fricatives, affricates, and labiodentals) also  
357 declined the most over time. The dissociation between PPC and intelligibility in the lower range  
358 of PPC (i.e., 75% and lower) and especially for a subset of individuals with high PPCs was  
359 unexpected. For the individuals in the lower range of PPC, the lack of association with  
360 intelligibility was less surprising, as it is reasonable that without at least 75% of sounds produced  
361 correctly, listeners are very poor at deciphering speech. For the individuals with high PPCs but  
362 low intelligibility, one potential explanation for the lower intelligibility may be that the speakers  
363 had deficits in subsystems other than articulation (i.e., phonation) that impaired their ability to be  
364 understood.

365

366 ***The impact of fricatives, affricates, alveolars, labiodentals, and non-centralized vowels on***  
367 ***intelligibility***

368           To determine the relative impact of different errors on intelligibility, we examined the  
369 association between intelligibility and specific sound classes (i.e., consonants versus vowels) and  
370 distinctive features (e.g., fricatives versus stops). Consistent with prior work, we found a stronger  
371 association between consonant errors (i.e., PCC) and intelligibility than between vowel errors  
372 (i.e., PVC) and intelligibility (Hodge et al., 2013; McLeod et al., 2012). This finding is also  
373 consistent with the hearing-impaired literature, which found that consonants were more  
374 important than vowels for discerning word meaning (Owren & Cardillo, 2006).

375           Furthermore, across all consonants, we found that fricatives, affricates, alveolars, and  
376 labiodentals were most strongly associated with intelligibility. These findings align with prior  
377 work in adults with non-progressive dysarthria, all of which found the strongest associations  
378 between intelligibility and fricatives and affricates compared to other types of speech sounds  
379 (Ansel & Kent, 1992; Kim et al., 2010; Platt et al., 1980). Additionally, previous research has  
380 demonstrated strong associations between acoustic measures of affricate or fricative production  
381 and perceptual impressions of speech, such as intelligibility and articulatory imprecision (Liu et  
382 al., 2016; Maniwa et al., 2008; Tjaden & Turner, 1997). The current findings are inconsistent  
383 with those of Kent and colleagues (1989 and 1991), who observed a high proportion of stop-  
384 nasal contrast errors in speakers with ALS (Kent et al., 1989, 1991). However, stop-nasal  
385 contrasts were most notably impaired for speakers with poor intelligibility (i.e., < 65%), whereas  
386 the current study included speakers with intelligibility across the continuum (i.e., both speakers  
387 with low and high intelligibility), which may have contributed to our discrepant findings. For  
388 example, the types of errors made by speakers with low intelligibility may be distinct from errors  
389 made by more intelligible speakers. We also used different stimuli (i.e., paragraphs versus single  
390 words) and outcome measures (i.e., all possible error types versus certain contrasts) from the  
391 ones used by Kent and colleagues (1989 and 1991), which may be a potential cause of disparate  
392 results.

393           When PCC was included in the model, the relationship between PVC and intelligibility  
394 was not statistically significant; however, when vowels were examined in isolation, corner  
395 vowels (i.e., front, back, high, and low) were more significantly associated with intelligibility as  
396 compared to non-corner vowels. This result corroborates prior acoustic evidence that vowel

397 space area, calculated from corner vowels, is an important component of intelligibility in  
398 speakers with ALS (Turner et al., 1995; Weismer et al., 2001).

399

400 *Declines were greatest for distinctive features that may require the greatest articulatory*  
401 *precision*

402 In addition to investigating their associations with intelligibility, we compared the extents  
403 of decline for each sound class and distinctive feature over time. Consonants and vowels  
404 exhibited similar extents of decline across sessions, as did different vowels. For consonants, we  
405 found that fricatives, affricates, and stops declined to a significantly greater extent than nasals,  
406 while labiodentals declined to a significantly greater extent than did bilabials and palatals.

407 These findings can be interpreted within the context of a framework proposed by Kent  
408 (1992), which characterizes different phonemes based on their presumed production complexity  
409 (see Table 4). This framework posits that sounds acquired later in life (e.g., affricates) are more  
410 motorically challenging or phonetically complex than sounds acquired earlier in life (e.g.,  
411 bilabials) (Kent, 1992). Although Kent's framework was intended to explain the acquisition of  
412 speech, the same principals have been applied to patterns of speech loss in adults with  
413 neurodegenerative diseases (van der Merwe, 2009; Wertz et al., 1991). Prior research has  
414 suggested that individuals with dysarthria tend to have difficulty producing sounds with higher  
415 levels of phonetic complexity (Allison & Hustad, 2014; Kim et al., 2010; Kuruvilla-Dugdale et  
416 al., 2018). Speakers with ALS, in particular, are known to have deficits in controlling lingual and  
417 labial movement (Langmore & Lehman, 1994; Perry et al., 2018; Yunusova et al., 2010). These  
418 deficits may underlie the sharp declines in the accuracy of fricatives, affricates, stops, and  
419 labiodentals. Indeed, Manner and Place features that presumably require increased fine motor

420 control and regulated, low velocity vocal tract constrictions (e.g., control of fine force for  
 421 fricatives) declined to a greater extent than those that require less graded, ballistic movements  
 422 (e.g., velopharyngeal port opening for nasals).

423

**Table 4.** An adapted version of Kent's (1992) framework including only sounds relevant to the current study.

Complexity Level (1 = least complex, 6 = most complex)		Distinctive Features			
Vowels		Height	Advancement	Example	
1		<u>Low</u>	<u>Back</u>	/ɑ/	
		Mid	Central	/ə/	
2		<u>High</u>	<u>Back</u>	/u/	
		<u>High</u>	<u>Front</u>	/i/	
		Mid-high	<u>Back</u>	/o/	
3		Mid-low	<u>Front</u>	/ɛ/	
		Mid-low	<u>Back</u>	/ɔ/	
4		Mid-high	<u>Front</u>	/ʌ, /e/	
		Mid-low	<u>Front</u>	/æ/	
		Mid-high	<u>Back</u>	/ɒ/	
Consonants		Voicing	Place	Manner	Example
3		<u>Voiceless</u>	Bilabial	<u>Stop</u>	/p/
		<i>Voiced</i>	Bilabial	Nasal	/m/
		<i>Voiced</i>	<i>Alveolar</i>	Nasal	/n/
		<i>Voiced</i>	Bilabial	Glide	/w/
		<u>Voiceless</u>	Glottal	<i>Fricative</i>	/h/
4		<i>Voiced</i>	Bilabial	<u>Stop</u>	/b/
		<i>Voiced</i>	<i>Alveolar</i>	<u>Stop</u>	/d/
		<u>Voiceless</u>	Velar	<u>Stop</u>	/k/
		<i>Voiced</i>	Velar	<u>Stop</u>	/g/
		<i>Voiced</i>	Palatal	Glide	/j/
		<u>Voiceless</u>	<i>Labiodental</i>	<i>Fricative</i>	/f/
5		<u>Voiceless</u>	<i>Alveolar</i>	<u>Stop</u>	/t/
		<i>Voiced</i>	Velar	Nasal	/ŋ/
		<i>Voiced</i>	<i>Alveolar</i>	Liquid	/ɹ, /l/
6		<u>Voiceless</u>	<i>Alveolar</i>	<i>Fricative</i>	/s/
		<i>Voiced</i>	<i>Alveolar</i>	<i>Fricative</i>	/z/
		<u>Voiceless</u>	Palatal	<i>Fricative</i>	/ʃ/
		<i>Voiced</i>	<i>Labiodental</i>	<i>Fricative</i>	/v/
		<u>Voiceless</u>	Dental	<i>Fricative</i>	/θ/
	<i>Voiced</i>	Dental	<i>Fricative</i>	/ð/	

424

<u>Voiceless</u>	Palatal	<u>Affricate</u>	/tʃ/
<u>Voiced</u>	Palatal	<u>Affricate</u>	/dʒ/

*Note:* Italicized = significant association with intelligibility; underlined = declined to a significantly greater extent than other distinctive features in its group for the current study.

425

426           Prior work has suggested that the time course of degeneration may vary among the  
427 cranial nerves that innervate the speech musculature (DePaul et al., 1988). Specifically, DePaul  
428 and colleagues (1988) found that hypoglossal motor neurons, which innervate tongue muscles,  
429 were affected to a greater extent than trigeminal motor neurons, which innervate jaw muscles  
430 (DePaul et al., 1988). While many studies in dysarthria secondary to other disorders have not  
431 supported a relationship between tongue weakness and impaired speech (McHenry et al., 1994;  
432 Neel et al., 2015; Solomon et al., 1995; Solomon et al., 2000; Theodoros et al., 1995), two recent  
433 studies, one of which focused on ALS, reported strong correlations between tongue weakness  
434 and intelligibility (Searl et al., 2017) or speech impairment (Jones et al., 2015). Based on these  
435 latter studies, we might have expected to find significantly more impaired alveolars (which  
436 involve precise tongue placement) than bilabials (which can be achieved with compensatory jaw  
437 movement even for more severe speakers). Yet, we found that only *labiodentals* declined to a  
438 significantly greater extent than bilabials. This finding should be considered with the knowledge  
439 that all labiodentals are fricatives, which might have driven its significant differences in accuracy  
440 compared to the other Place features. Nevertheless, it is possible that the difference in accuracy  
441 for labiodentals and bilabials may also reflect differential impairment of the lips and jaw, as  
442 labiodentals require the precise regulation of lower lip and jaw muscles to achieve the  
443 appropriate constriction between teeth and lower lip, whereas bilabials can be produced using  
444 more coarsely controlled movements of the lips and/or jaw.

445 Kent's framework also discusses the level of complexity of vowel sounds. Low back  
446 vowels (e.g., /a/) are posited as being the least complex because they involve only slight lingual  
447 elevation, while mid-high back vowels are considered the most complex given the need for  
448 lingual-jaw coordination (Kent, 1992). In contrast to this theory, low vowels have been found to  
449 be *more* impaired than high vowels in speakers with ALS due to the reliance of low vowels on  
450 larger and faster movements (Kent et al., 1990; Kuruvilla et al., 2012; Yunusova et al., 2008).  
451 While our study revealed similar extents of decline across *all* vowels, regardless of their  
452 hypothesized complexity, most of the prior work examined the reduction of tongue movements  
453 rather than phoneme accuracy, which precludes a direct comparison of findings.

454

#### 455 *Voiced versus voiceless sounds in the Bamboo Passage*

456 One final notable result was that voiceless sounds declined to a greater extent than voiced  
457 sounds, but errors on voiced sounds were more strongly associated with reduced intelligibility.  
458 The former finding is consistent with prior work demonstrating a higher error rate for voiceless  
459 sounds than for voiced sounds in speakers with ALS (Antolik & Fougeron, 2013; Caruso &  
460 Burton, 1987; Kent et al., 1990; Riddel et al., 1995). While voiceless sounds were  
461 disproportionately impacted compared to voiced sounds, we found that voiced sound accuracy  
462 was more strongly associated with intelligibility than was voiceless sound accuracy. These  
463 findings may be related to phonemic inventory biases in the Bamboo Passage, which was  
464 intentionally loaded with voiced sounds to enhance the automatic detection of word boundaries  
465 in connected speech (Green et al., 2004; Yunusova et al., 2005). The Bamboo Passage may,  
466 therefore, not be the most effective task for comparing the impact of voiced versus voiceless  
467 sounds on intelligibility. Additional research on the impact of speech stimuli is needed.

468 *Clinical implications*

469 Overall, the findings from this study have two central clinical implications for speakers  
470 with ALS: (1) They may help identify potential candidate treatment targets for speakers with  
471 reduced intelligibility; and (2) They may help guide stimuli selection for global measures of  
472 intelligibility or speech severity.

473 First, the strong association between features such as affricates and fricatives and  
474 intelligibility, and the large extents of decline for these same features, suggests that targeting  
475 specific sounds in therapy early on, either directly or indirectly, could benefit speakers with  
476 ALS. This proposal is consistent with recommendations in recent work, which have suggested  
477 that complementing traditional therapy approaches with more specific speech targets (Ansel &  
478 Kent, 1992; Icht, 2021; Kent et al., 1991; Kuruvilla-Dugdale et al., 2018; Platt et al., 1980;  
479 Turner & Tjaden, 2000) or more direct instructions regarding articulatory movements (Mefferd  
480 & Dietrich, 2019) could lead to improved acoustic and perceptual outcomes.

481 Knowledge of which sounds are most impacted by physiologic constraints of ALS over  
482 time may also offer an alternative means of conceptualizing therapy for speakers with lower  
483 intelligibility. The nonlinear relationship between PPC and intelligibility demonstrates that for  
484 speakers who produce less than 75% of all phonemes correctly, increases in PPC (e.g., from 65%  
485 to 70%) have little influence on intelligibility. This finding suggests that targeting specific  
486 sounds in therapy may not be terribly beneficial for this subset of speakers. Thus, to maximize  
487 the functional impact of treatment for these speakers, clinicians could select a list of functional  
488 words for the speaker to use that include less impaired distinctive features (e.g., bilabials, glides,  
489 or nasals).

490           Lastly, our findings may offer further guidance for stimuli selection for assessing  
491 intelligibility. Given the relationships between each impaired distinctive features and  
492 intelligibility, intelligibility ratings may be inflated if the stimuli do not include sounds most  
493 likely to be impacted by ALS. For example, if the stimuli primarily included bilabials and glides,  
494 the speaker may be rated as highly intelligible since those sounds tend to be less affected by  
495 disease progression. Similarly, Kuruvilla and colleagues (2018 and 2020) found that more  
496 phonetically complex stimuli were better able to detect subtle disease-related speech changes in  
497 speakers with ALS. However, the discrepancies between our findings and those of prior work  
498 using single word stimuli suggest that measures of intelligibility may differ depending on the  
499 task. Aside from allowing for more time to achieve appropriate articulatory placement, single  
500 words allow more opportunities to precisely coordinate the speech subsystems (e.g., increase  
501 breath support), which further influences intelligibility (Yunusova et al., 2005). Therefore, the  
502 findings from this study, based on a longer connected speech task, may provide important  
503 knowledge about more functional/ecologically valid impacts of phonemic accuracy on  
504 intelligibility.

505

### 506 *Limitations and future work*

507           There were several limitations in this study. First, although the Bamboo Passage is a  
508 more functional speech task than single words, an even more ecologically valid task would be  
509 spontaneous speech. Indeed, in this study, the same passage was read multiple times by each  
510 participant, which may have led to practice effects. In addition, the wide variation of time  
511 between data collection points may have further influenced our results. Moreover, a potential  
512 reason why we might have found high PPCs for some individuals with low intelligibility is that

513 the phonemic transcribers, too, were familiar with the passage, which could have resulted in  
514 slightly inflated PPC values. Moreover, as noted earlier, the passage is not phonetically balanced  
515 but rather was designed to include more voiced consonants at word boundaries to ease the  
516 identification of pauses (Green et al., 2004; Yunusova et al., 2016). Thus, the unequal frequency  
517 of voiced versus voiceless sounds, in addition to other distinctive features (e.g., fricatives versus  
518 affricates), may have further influenced our findings.

519         Second, while our dataset was, to our knowledge, the largest used to examine  
520 longitudinal changes in speech sound accuracy in ALS, the dataset was still limited by its small  
521 size and unknown timing of disease onset. Additional work is needed to examine the decline of  
522 speech performance from disease onset and whether the same results can be replicated with a  
523 larger sample size.

524         Third, we did not conduct analyses on error types (e.g., distortions, substitutions,  
525 deletions), which may provide useful information about the speaker's physiological constraints.  
526 Future work should examine not only specific error types, but combinations of distinctive  
527 features (e.g., alveolar fricatives) as opposed to in isolation (e.g., alveolars and fricatives). Such  
528 analyses would allow for a more precise examination of the underlying physiology of speech  
529 error patterns.

530         Furthermore, we did not examine the effect of word position (i.e., initial, medial, or final)  
531 on intelligibility or decline over time. Prior work has demonstrated that the highest number of  
532 errors occur in word-final positions (Blaney & Hewlett, 2007). Therefore, our phonemic  
533 accuracy values may have been inflated or deflated if, for example, palatals occurred most  
534 frequently in the word-initial position and alveolar occurred most frequently in the word-final  
535 position, respectively. Moreover, a recent study found that position of a sentence within a longer

536 paragraph influences intelligibility (van Brenk et al., 2022). Given the potential for fatigue across  
537 a lengthy speaking task, further research should investigate the impact of sentence position /  
538 stimuli length on phoneme production. Lastly, future work should consider other features, such  
539 as voice quality and prosody, for a more comprehensive understanding of the different  
540 subsystems' roles in intelligibility.

541

## 542 ***Conclusions***

543 In sum, when examining speech production during a paragraph-reading task over time,  
544 we found a nonlinear relationship between PPC and intelligibility. This relationship suggests that  
545 PPC may have a limited working range and that focusing on increasing phonemic accuracy in  
546 therapy may not be universally helpful for all speakers. We also found that features highly  
547 dependent on precise lingual and labial constriction (i.e., fricatives, affricates, and labiodentals)  
548 and greater lingual and labial movement (i.e., non-centralized vowels) were most important for  
549 intelligibility *and* declined to a greater extent over time. Overall, examining the patterns of  
550 phonemic errors in ALS is integral for better understanding the impacts of physiologic  
551 constraints on functional communication and could, in turn, ultimately inform the selection of  
552 the most effective treatment targets and stimuli for assessing intelligibility.

553

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562

563 **Declaration of interest**

564 The authors report no declarations of interest.

565

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