Symptom Expression Across Voiced Speech Sounds in Adductor Laryngeal Dystonia

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**Summary:** **Objectives.** Differential diagnosis for adductor laryngeal dystonia (AdLD) is often carried out by comparing symptom expression during sentences with either all voiced or voiced and voiceless consonants. However, empirical research examining the effects of phonetic context on symptoms is sparse. The purpose of this study was to examine whether symptom probabilities varied across voiced speech segments in an all-voiced sentence, and whether this variability was systematic with respect to phonetic features.

**Methods.** Eighteen speakers with AdLD read aloud a sentence comprised entirely of voiced speech sounds. Speech segment boundaries and AdLD symptoms (phonatory breaks, frequency shifts, and creak) were labeled separately, and speech segments were coded as symptomatic or asymptomatic based on their temporal overlap. Generalized linear mixed effects models with a binomial outcome variable were used to compare the probability of symptom expression across: 1) all speech segments in the sentence, and 2) four speech sound classes (vowels, approximants, nasals, and obstruents).

**Results.** Significant symptom variability was found across voiced speech segments in the sentence. Furthermore, the estimated probability of a symptom occurring on vowels and approximants was significantly greater than that of nasals and obstruents.

**Conclusion.** These results indicate that AdLD symptoms are not uniformly distributed across voiced speech segments with systematic variation across speech sound classes. To explain these findings, future work should investigate how the complex interactions between the vocal tract articulators and glottal configurations may influence symptom expression in this population.

**Key words:** Adductor laryngeal dystonia—Phonetic context—Differential diagnosis—Source-filter interaction.

**INTRODUCTION**

Laryngeal Dystonia (LD; also known as Spasmodic Dysphonia) is a focal dystonia of neurological origin affecting the laryngeal musculature during vocalization. Adductor LD (AdLD) comprises 85% of LD cases and is characterized by a harsh, strained-strangled vocal quality and intermittent phonatory breaks due to overadduction of the vocal folds. In contrast, abductor LD (AbLD) accounts for about 10% of cases and is typified by a breathy vocal quality and breaks in phonation caused by excessive abduction of the vocal folds. Although the exact pathophysiology underlying LD is unknown, it is associated with altered neural structure and function, as well as abnormal sensory processing. Typically emerging around age 45-50, this disorder has a significant negative impact on communication, mental health, and quality of life. Due to the rarity of LD (estimates are as low as 1 in 100,000), individual variability and overlap in features with other voice disorders, diagnostic reliability is often low, even among experienced clinicians. Consequently, the delay between the initial clinician interaction and accurate diagnosis is about 5 years, which can prolong communication challenges.

Differential diagnosis is typically carried out by speech-language pathologists, otolaryngologists, and neurologists using a combination of auditory-perceptual, laryngoscopic, and neurological evaluation. Auditory-perceptual speech evaluation is usually based on two key features of LD—task-specificity and variation across phonetic contexts. LD is task-specific in nature, presenting most prominently during volitional speech production, with notable reductions in severity during whispered speech, singing/humming, non-modal phonation, and innate vocalization like laughing and crying. This criterion can be useful for distinguishing LD from muscle tension dysphonia (MTD), which is expressed more consistently across vocalization types.

In addition, symptoms in LD vary across phonetic contexts, which may help to distinguish between AdLD, AbLD, and MTD. Specifically, speakers with AdLD tend to be more symptomatic on voiced-loaded phrases, speakers with AbLD show increased speech sound durations on voiceless consonants, and those with MTD tend to be consistent across contexts. The general voiced/voiceless distinction has provided meaningful guidance for developing diagnostic stimuli and points to the link between intrinsic laryngeal muscle use (ie, adduction during voiced sounds and abduction during voiceless sounds) and symptom expression. The simple theory that emerges is that more voicing and, thus, more vocal fold adduction will lead to increased symptom expression in AdLD. However, a clear characterization of how other phonetic features may interact with voicing can inform the development of novel diagnostic stimuli that can maximally...
elicited symptoms to aid in reliable diagnosis. Furthermore, understanding the effects of phonetic context on voicing has the potential to provide insight on the physiological manifestation of symptoms. Therefore, research aimed at defining the phonetic contexts associated with symptom expression in LD is warranted.

Relatively few studies have examined the specific phonetic components of stimuli that are associated with speech features in AdLD. In one of the earliest studies to do so, Ludlow and Connor found that speakers with AdLD were notably slower to repeat /a/ than /pa/ or /pata/, with a greater amount of time spent on the laryngeal closure between repetitions. Although this was not strictly related to AdLD symptoms, it indicated a dissociation between the effects of voiced and voiceless sounds on laryngeal movement control in this population. Erickson provided the first empirical evidence that voicing environment can affect AdLD symptom expression using controlled stimuli. More specifically, Erickson found that sentences with a high ratio of voiced to voiceless consonants evoked significantly more symptoms than sentences with a low ratio. They further singled out voiced plosives as particularly related to the number of AdLD symptoms across sentence categories and “The Rainbow Passage,” although this observation was not statistically verified. To better assess this suggestion, Cannito et al. asked speakers with AdLD to read sentences contrasted by consonant voicing (voiced vs. voiceless) and manner of articulation (continuant vs. plosive). Although the voiced sentences were judged as more dysphonic than the voiceless sentences in AdLD, there were no significant main effects of, or interactions with, manner of articulation, indicating that symptoms do not vary across these classes of consonants. The careful control of stimuli used in these studies was critical in uncovering phonetic patterns that influence symptom expression in AdLD. However, because symptom expression was aggregated across sets of phrases with variable phonetic content or auditory-perceptual ratings were based on whole phrases, these studies did not provide information regarding the phonetic contexts in which symptoms actually occurred or the probability and variability of symptoms occurring in these contexts. This fine-grained linking of symptom expression and context is necessary for determining what phonetic factors contribute to increased symptom expression, as well as how these factors can be exploited to develop maximally informative diagnostic speech samples.

To date, only one study has examined symptom expression related to specific phonetic contexts in AdLD. Lorch and Whurr analyzed portions of conversational speech samples from six native French speaking participants with AdLD. Their data provide some evidence that syllables with consonant clusters are likely to yield phonatory breaks, and that harsh voice quality was found more on open and mid-vowels (eg, /a/, /e/) than closed and neutral vowels (eg, /u/, /y/, /a/). They also found that harsh vowels, compared to vowels not judged to be harsh, were more likely to be preceded by voiced consonants. However, the small sample size and lack of controlled stimuli in this study make it difficult to draw strong conclusions, especially cross-linguistically. Thus, in order to infer the relationship between phonetic context and symptoms across speakers with AdLD, statistical analysis of a larger sample is necessary. Furthermore, although it is clear that voiced speech sound-loaded and voiceless speech sound-loaded sentences lead to different levels of symptom expression in AdLD, it is unclear how symptoms vary across sounds within these utterances.

The purpose of the present study was to examine the probability of AdLD symptom expression at the level of individual speech sounds and classes of speech sounds when speakers with AdLD read an all-voiced sentence aloud. Following detailed labeling of symptoms and speech segment boundaries in recordings of 18 speakers with AdLD, generalized linear mixed effects models were used to investigate: 1) whether symptom probabilities varied over the course of the sentence, and 2) whether this variability was systematic with respect to speech sound classes. It was hypothesized that AdLD symptoms would be expressed with higher frequency during the production of vowels and sonorant consonants (ie, nasals and approximants), which require continuous phonation, and lower frequency during obstruents (ie, stops and fricatives) which typically include gaps in phonation.

METHODS

Participants

Eighteen speakers (13 female, 5 male) participated in this study*. Their ages ranged from 55 to 69 years (females: mean = 63.3, SD = 4.06; males: mean = 59.0, SD = 2.91). All speakers were diagnosed with AdLD by the same board-certified laryngologist. At the time speech samples were collected, the disorder had been present in all participants for at least 6 months. Speakers did not exhibit any coexisting vocal tremor or other sites of dystonia and, with exception of the diagnosis of AdLD, self-reported that they were in good general health. All speakers exhibited a range of AdLD severity and were receiving ongoing Botox treatment for their AdLD. To ensure that speakers were symptomatic at the time of testing, all recordings took place before their scheduled Botox injection. Participants provided informed consent to voluntarily participate in accordance with Western University’s Research Ethics Board (REB #18588E). Participants were not compensated for their time.

Recording procedure

Recordings took place in a sound-attenuated recording environment under consistent conditions using a cardioid condenser microphone (SHURE PG81) positioned 15 cm from the mouth, preamplifier, and Kay-Pentax Sona Speech

*Gender information was unavailable for this sample.
II software (Pine Brook, NJ). Samples were digitized at a 44.1 kHz sampling rate with all samples collected by the same clinician. Experimental stimuli were displayed to speakers in large print. Speakers were seated comfortably during all speech tasks. Participants were asked to read two productions of a sentence comprised solely of voiced phonemes (‘Early one morning a man and a woman were ambulating along a one-mile lane running near Rainy Island Avenue’)

using their typical pitch and loudness. This sentence was examined because several publications have previously used it to elicit AdLD symptoms,32−38 and it contains multiple instances of the voiced speech sound classes under investigation.

Speech segment boundary marking
Speech segment boundaries were identified using an open-source forced alignment toolkit (P2FA).39 This toolkit uses acoustic models of the phonemes in American English to find the best match between the recorded speech sample and a pre-defined text string. The text string for each recording was modified to exactly match what was said (eg, clinician talking in the background, reading errors). Boundary timing information from each recording was then imported as a textgrid into Praat (Version 6.2.09,40) and manual adjustments were made by the first author based on correspondence with the waveform, spectrogram, and audio. Across all recordings, there were two instances of minor reading errors, and adjustments for these errors are described as follows. One participant produced a repetition (“a one-mile lane” → “a one- a one-mile lane”) on the second reading of the sentence; the first “a one-” was marked as silence and not included in the analysis. A second participant omitted the word “a” (“a man and a woman” → “a man and woman”) during the first reading of the sentence; this segment was removed from the textgrid and was treated as missing data in the statistical model (see Statistical analysis below). Speech segment labels were kept consistent across all speakers; no adjustments were made for dialectical and phonetic variation.

Symptom labeling
As reported in an earlier study,41 two trained technicians independently identified and labeled AdLD symptoms from the acoustic waveforms and spectrograms in Praat for each sentence repetition. AdLD symptoms were defined using the criteria from Sapienza et al.23 Phonatory breaks were defined as absence of voicing that typically occurs during a phonetic segment. Frequency shifts were defined as a change of 50 Hz or more in a F0 that occurs within 50 ms. Other irregularities in F0 were labeled as “creak,” which was used as an umbrella term following Keating et al.32 In instances in which the original two technicians did not agree (approximately 16% of all identified symptoms), a third technician (a voice specialized speech-language pathologist) worked with the original two technicians to derive consensus judgments.

Data extraction
Following independent speech segment and AdLD symptom labeling, the determination of whether a symptom occurred during each produced speech segment was carried out. The presence of any symptom (ie, a phonatory break, a frequency shift, or creak) occurring during a given segment was coded as a binary outcome — 1 if the symptom was present during a segment, 0 if it was not. Because of this binary coding strategy, it was possible that a minor segment boundary labeling error could lead to erroneous identification of a symptom occurring on a given segment (eg, symptom onset occurring within a few milliseconds of a segment boundary, erroneously leading to coding both adjacent segments as symptomatic). Therefore, symptoms had to either occur within the time-boundaries of the segment or overlap in time with the segment by at least 15 ms.

Statistical analyses
All statistical analyses were carried out in R (version 4.1.2). To test whether AdLD symptoms varied across the all-voiced sentence (as opposed to the null hypothesis that they occurred uniformly across speech segments), a generalized linear mixed effects analysis with a binomial outcome variable was performed using the glmer function (package lme4).43 The outcome variable in this analysis was the presence of any symptom occurring during the segment. In this analysis, segment position was modeled as a categorical fixed effect with 69 levels (for each of the 69 segments in the sentence) with random intercepts allowed for each speaker. Because segments vary in duration and, therefore, in the amount of time during which a symptom can occur, duration was included as an additional fixed effect control covariate. Type III analysis of deviance Wald chi-square tests were then used to assess the main effect of segment position.

To test whether the presence of LD discontinuities varied according to speech sound class, an additional binomial linear mixed effects model was constructed. For this analysis, fixed-effects terms were included for speech sound class and segment duration, and random intercepts allowed per speaker. Speech sound class was a categorical variable with four levels for vowels, approximants, nasals, and obstruents (Table 1 for a breakdown of their occurrence in the sentence). Type III analysis of deviance Wald chi-square tests were used to assess the main effect of speech sound class. If this effect was found to be significant, post-hoc pairwise comparisons were calculated with a Tukey correction for multiple comparisons.

RESULTS
For the assessment of the impact of segment position on the presence of AdLD symptoms, the analysis of deviance revealed that there was a significant effect of segment position (χ²68 = 167.4, P < .001) and segment duration (χ²1 = 72.2, P < 0.001) on AdLD symptom expression. The estimated marginal mean and 95% confidence intervals for symptom expression in each segment position (after reverse-
transforming from the logit scale to the probability scale) can be found in Figure 1. Estimated marginal mean probabilities for symptoms occurring on individual segments ranged from 28.8% to 87.4%.

Assessing whether the effect of segment position was systematic across speech sound classes, the analysis of deviance again revealed that there was a significant effect of speech sound class ($\chi^2 = 82.5, P < 0.001$) on AdLD symptom expression. Post-hoc pairwise comparisons with a Tukey correction found significant differences between approximants and nasals ($P < 0.001$, odds ratio [OR] 2.2, 95% CI 1.6-3.1), between approximants and obstruents ($P = 0.03$, OR 1.8, 95% CI 1.0-3.1), between nasals and vowels ($P < 0.001$, OR 0.40, 95% CI 0.31-0.53), and between obstruents and vowels ($P = 0.002$, OR 0.49, 95% CI 0.29-0.83). To summarize, the probability of an LD discontinuity occurring during either a nasal or an obstruent was less than during either a vowel or an approximant (Figure 2).

**DISCUSSION**

In this study we examined the relative probability of symptoms associated with AdLD (ie, phonatory breaks, frequency shifts, or creak) occurring across segments in an all-voiced sentence. In contrast with previous English-language studies that evaluated symptom expression across sentences, the present study used generalized linear mixed effects models to estimate the probability of symptoms occurring at the level of individual segments and speech sound classes. Furthermore, this was the first study to do so using formal statistical analysis in a cohort of English-language speakers. The results showed that: 1) within an all-voiced sentence, symptoms do not occur with an equal probability across all segments, and 2) in particular, nasal consonants (ie, /m/, /n/, /ŋ/) and obstruents (ie, /d/, /b/, /v/) were less likely to contain symptoms than either vowels or approximants (ie, /r/, /l/, /w/). The following sections will discuss these results in relation to current understanding of the interaction between phonetic and laryngeal control in speakers with and without AdLD, as well as limitations and future directions for this research.
Effects of phonetic environment on symptom expression in AdLD

Previous work has sought to explain the effects of phonetic environment on laryngeal control as related to the demands of rapid laryngeal adduction during speech. Ludlow and Connor’s finding that speakers with AdLD have phonation delays when rapid laryngeal control is necessary provided an initial suggestion that certain contexts could facilitate atypical voicing. Following this, Erickson suggested that sentences loaded with voiced consonants were more symptomatic because of the preponderance of voiced stop consonants which require more rapid laryngeal onsets to achieve appropriate voice onset. Subsequent work, however, found that in AdLD, symptoms were not differentially increased in voiced-loaded sentences (compared to voiceless-loaded sentences) when they predominantly featured voiced stop consonants versus approximants. Thus, it was posited that producing an all-voiced sentence requires maintaining the vocal folds in position near the midline, and that this posturing that differs from that of rapid laryngeal control, is the critical feature of speech that facilitates hyperadduction in AdLD.

The present results appear to undermine aspects of both suggestions. In contrast with Cannito et al., data from the present study reveal that different classes of voiced speech sounds are associated with different levels of AdLD symptoms. Furthermore, obstruents (a category that includes stop consonants) were found to be less symptomatic than approximants, which counters the hypothesis offered by Erickson that increased symptoms in voice-loaded sentences are primarily due to increased voiced stop consonants.

There are two factors that could explain these seeming discrepancies. First, the present study relates speech sound classes with symptoms based on whether the symptom occurs within the duration of the speech segment. That is, previous results may have captured information related to the effect of consonant voicing on subsequent voiced productions rather than during the segments themselves. If articulatory events are responsible for triggering (or at least influencing) AdLD symptom expression, there could be an important effect of the phonetic characteristics of surrounding segments. In fact, there may be some additive and interaction effects between these two factors that influence symptom expression. In the present study, stimuli were not controlled with respect to phonetic environment. Indeed, obstruents were particularly limited in their phonetic context due to the small number of tokens. In contrast with Cannito et al. and Erickson, in which plosives were largely in pre- or post-vocalic positions (eg, dog, buried), most plosives were preceded by nasal consonants in the present study. Given that nasal consonants had reduced symptom expression, coarticulatory effects may have led to similar findings in obstruents. Thus, we could not disentangle the effects of phonetic identity and phonetic context. Second, the outcome variables for Cannito et al. included perceptual judgment of overall voice quality and sentence duration which by themselves, may have a non-linear relationship with the carefully labeled symptoms in the present study. Future work that can account for and/or systematically vary phonetic environments, and includes both symptom-level and auditory-perceptual outcomes will be necessary to reconcile these findings.

The results of the present study may best be understood through studies that examine the interaction between the vocal tract and the glottal source. Experimental and modeling work suggests that increased obstruction of the vocal tract (as in fricatives and stop consonants) can facilitate efficient phonation due to increased supraglottal back pressure. This is exemplified by the use of semi-occluded vocal tract exercises in both voice therapy and training for singers. Indeed, recent work using electroglottography (EGG) provides evidence that voiced obstruents are produced with a reduced closed quotient (CQ), indicating a less adducted glottis, a behavior which could help to counter the hyperadduction found in AdLD. Evidence of a similar effect in nasal consonants in speech, however, is more equivocal. Nasal consonants exhibit decreased intensity relative to vowels and approximants, but intraoral pressure and EGG CQ remains virtually the same; this finding is likely due to unimpeded airflow through the nasal passages. Further insight can be gleaned from studies comparing humming (ie, prolonged nasal consonant production) and vowel production in speakers with typical voices and with voice disorders. Consistent with the aforementioned studies explicitly examining speech, these studies found that EGG CQ was not different between humming and vowel production in speakers with typical voices, and that CQ actually increased during humming in speakers with MTD. However, vocal roughness (assessed by experienced laryngologists) and the cycle-to-cycle variation in CQ were significantly decreased in both groups during humming. This also was shown to be the case in speakers with dysphonia secondary to structural changes of the vocal folds. It was suggested that humming has an immediate effect of reducing supraglottic compression in speakers with MTD and typical voice, which in turn can alter glottal airflow and vocal fold vibration. Increased supraglottic constriction is also a common symptom of AdLD and it can contribute to altered vocal quality. Humming has been found to significantly improve phonatory symptoms in speakers with AdLD, however, no attempts have been made to explore this instrumentally. It is plausible that, similar to humming, nasal consonant production in a spoken phrase can reduce AdLD symptoms like vocal roughness or creak.

Finally, despite significant differences in symptom expression between the speech sound classes, the estimated probability of a symptom occurring was greater than 28% on every segment in the phrase. This is likely due to the fact that this experimental phrase contained only voiced sounds.

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1Note that because symptom probability is determined across subjects and sentence repetitions, the low prevalence of obstruents in the sentence should not affect the probability of a symptom occurring for this category of sounds.
an environment that is most conducive to symptom expression in this population.\textsuperscript{13,24} It is unclear at present how the absolute level or the relative probabilities of symptom occurrence would differ in speech that contained both voiced and voiceless sounds. Phonetic context is sometimes characterized as having an environmental effect on symptoms in AdLD.\textsuperscript{25,29} That is, voiceless consonants are thought to reduce the occurrence of symptoms and the perceived severity of dysphonia during voiced segments by altering laryngeal position during the dynamic transition to these voiced segments.\textsuperscript{25,29} However, as most previous studies have only examined symptom severity at the phrasal level, this has yet to be directly tested. As an alternative, it is possible that in phrases containing both voiced and voiceless speech sounds, the total duration of voiced sounds is simply reduced (compared to all-voiced phrases). Under the theory that vocal fold adduction makes symptoms more likely to occur (or be detected), phrases with less voicing provide fewer opportunities for symptom expression. Consequently, future work comparing symptom expression on stimuli matched phonetically except for the voiced or voiceless quality of the preceding segment is needed to determine how voiceless speech sounds may reduce symptoms in AdLD.

Limitations and future directions
A potential limitation of the present work was that we only analyzed speech from participants with AdLD, and not from speakers with typical voices or with other voice disorders. It is possible that differences found between sound classes are not specific to AdLD but are characteristic of the general population and simply amplified by the severity of dysphonia of the speakers in this population. By including speakers with other voice disorders and with typical voices, a future study would be better equipped to determine whether this effect is specific to AdLD. If it is specific, discrimination analyses could be carried out to assess how well individual speech sound classes discriminate between populations, which could help determine optimal phonetic patterns in diagnostic speech stimuli to maximize efficiency.

Additionally, analyzing speech from a single all-voiced phrase precluded comparing symptom expression between voiced and voiceless speech sound classes and among other sub-classes of speech sounds (ie, stops, fricatives, liquids, and glides). Further, the limited contextual variability and size of the stimulus sentence hindered generalizability of these findings. A more varied and extensive speech sample would allow for detailed analyses using narrower phonetic transcription (rather than using broad phonemic categories). These analyses could better take into account co-articulatory effects and hypoarticulation which occur in natural speech. In addition, a larger speech sample would provide more variability to quantify, and control for, word- and sentence-level prosodic effects (eg, fundamental frequency, voice intensity, and emphatic stress) that may influence symptom expression in AdLD. Finally, because the present study only included the acoustic speech recordings, it was not possible to relate the occurrence of symptoms or the relative expression of symptoms across segments to differences in the physical configuration of the larynx. Thus, it will be important for future studies to include methods like electroglottography, electromyography, and/or high-speed videofluoroscopy to provide a more comprehensive characterization of how different classes of speech sounds impact laryngeal structures in speakers with AdLD and to potentially determine whether somatosensory feedback plays a role in symptom expression. There is a great deal of evidence that somatosensation is altered in dystonia,\textsuperscript{62} including LD,\textsuperscript{8} and that external sensory stimulation can relieve symptoms.\textsuperscript{3,63} More direct instrumental measurement of the larynx has the potential to determine whether phonetic context effects are related to changes in sensory stimulation during the running speech.

CONCLUSION
This study used a new approach to characterize differences in symptom expression across phonetic contexts in AdLD. It is anticipated that further examination of the phonetic contexts that yield increased or reduced symptoms in speakers with AdLD will be helpful in developing novel speech stimuli to improve differential diagnosis. In addition, understanding the relationship between the phonetic context and glottal configuration in this population may provide clearer insights into the pathophysiology of AdLD symptom expression and variability.

DECLARATION OF INTEREST
Cara E. Stepp has received consulting fees from Altec, Inc./Delsys, Inc., companies focused on developing and commercializing technologies related to human movement. Stepp’s interests were reviewed and are managed by Boston University in accordance with their conflict of interest policies. The other authors have declared that no other competing interests existed at the time of publication.

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**SUPPLEMENTARY DATA**

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