



A predictive model for diagnosing stroke-related apraxia of speech



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ABSTRACT

Diagnosis of the speech motor planning/programming disorder, apraxia of speech (AOS), has proven challenging, largely due to its common co-occurrence with the language-based impairment of aphasia. Currently, diagnosis is based on perceptually identifying and rating the severity of several speech features. It is not known whether all, or a subset of the features, are required for a positive diagnosis. The purpose of this study was to assess predictor variables for the presence of AOS after left-hemisphere stroke, with the goal of increasing diagnostic objectivity and efficiency. This population-based case-control study involved a sample of 72 cases, using the outcome measure of expert judgment on presence of AOS and including a large number of independently collected candidate predictors representing behavioral measures of linguistic, cognitive, nonspeech oral motor, and speech motor ability. We constructed a predictive model using multiple imputation to deal with missing data; the Least Absolute Shrinkage and Selection Operator (Lasso) technique for variable selection to define the most relevant predictors, and bootstrapping to check the model stability and quantify the optimism of the developed model. Two measures were sufficient to distinguish between participants with AOS plus aphasia and those with aphasia alone, (1) a measure of speech errors with words of increasing length and (2) a measure of relative vowel duration in three-syllable words with weak–strong stress pattern (e.g., banana, potato). The model has high discriminative ability to distinguish between cases with and without AOS (c -index=0.93) and good agreement between observed and predicted probabilities (calibration slope=0.94). Some caution is warranted, given the relatively small sample specific to left-hemisphere stroke, and the limitations of imputing missing data. These two speech measures are straightforward to collect and analyse, facilitating use in research and clinical settings.

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1. Introduction

Diagnosis of the speech motor planning/programming disorder, apraxia of speech (AOS), has proven challenging, largely due to its common co-occurrence with the language-based impairment of aphasia (McNeil et al., 2004). In fact, Galluzzi et al. (2015) suggested that “the differential diagnosis of AoS and PhI [phonological

aphasic impairment]...is one of the thorniest issues in aphasiology” (p. 66). Currently, diagnosis is based on identifying and rating the severity of up to 16 speech behaviors using checklists. Many of these speech behaviors are not discriminative between AOS and aphasia and it is not known if a subset of the behaviors is required for a positive diagnosis. Unlike previous studies, we apply a statistical modeling approach to identify the key speech measure (s) that support confident diagnosis of AOS when it co-occurs with aphasia, given that pure AOS is rare (Duffy, 2013).

AOS has been defined as a phonetic-motoric disorder of speech

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production characterized by inefficient or impaired translation of intact phonological representations into commands for carrying out intended movements. This translational difficulty leads to articulatory errors in timing and space that are perceived as sound, syllable, and prosodic distortions. Common prosodic distortions include slowed speech rate, syllable segregation, perceived equal stress within multisyllabic words, and prolonged sounds and sound or syllable transitions (Duffy, 2013; McNeil et al., 2009). Acquired AOS is most commonly caused by a left hemisphere stroke (Duffy, 2013) in inferior frontal or premotor regions (Ballard et al., 2014; New et al., 2015). However, a progressive form of AOS also exists that has received considerable attention in recent years (Ballard et al., 2014; Josephs et al., 2006, 2012; Whitwell et al., 2013).

Differentiating between AOS and aphasia, with either stroke or neurodegenerative etiology, has received attention for two primary reasons, which involve research and clinical endeavors. Researchers have sought to understand the nature of sensorimotor control for speech production and how localized damage to the brain affects different aspects of speech and language. Clinically, there is a need to provide more accurate and efficient methods of diagnosis for the purpose of confidently identifying the presence of AOS and then prescribing targeted speech intervention, or increasing awareness of its potential influence on language-focused intervention.

The predominant method of diagnosis of AOS is by perceptual judgment of sound, syllable, and prosodic distortions (Ballard et al., 2000). Researchers have attempted to operationalize criteria for these perceptual judgments using rating scales or frequency counts of speech features (Duffy et al., 2015; Galluzzi et al., 2015; Haley et al., 2012; Staiger and Ziegler, 2008; Strand et al., 2014). Efforts have also been directed toward identifying valid and reliable instrumental measures of orofacial movements and the acoustic speech signal during specific tasks (Ballard and Robin, 2007; Ballard et al., 2014; Duffy et al., 2015; Galluzzi et al., 2015; Haley et al., 2012; Laganaro et al., 2012; Vergis et al., 2014). However, clinicians and researchers currently rely on checklists of perceptual speech features, with little to no adoption of quantitative perceptual or instrumental measures in clinical settings. This is likely due, in part, to limited insight into which behaviors or combination of behaviors give rise to the perception of a specific speech feature, lack of time-efficient or automated analysis routines for instrumental measures, and limited evidence that the reliability and validity of instrumental measures equal or surpass that of perceptual measures.

Strand et al. (2014) provide a recent example of a perceptual rating protocol for AOS diagnosis. This group developed the Apraxia of Speech Rating Scale (ASRS), which uses a perceptual rating scale to judge speech behaviors across connected speech, word and sentence repetition, and alternating and sequential motion (i.e., diadochokinesis) tasks. Ratings address frequency and severity of observed behaviors, ranging from 'not present', to 'detectable but infrequent', 'frequent but not pervasive', 'pervasive across all/most tasks but intelligibility not markedly affected', and 'pervasive with marked reduction in intelligibility'. The ASRS requires rating of 16 speech behaviors, which have traditionally populated AOS feature checklists (Duffy, 2013; McNeil et al., 2009), including phoneme distortions, increased distortion errors with increased utterance length or speech rate, inaccurate productions in the sequential motion rate task (SMR, i.e., repeating 'pataka'), syllable segmentation, lengthening of speech segments, sound or syllable repetitions, and groping. Each feature is rated on a 0–4 scale, with a total possible score of 64 (0=no detectable speech abnormality). Findings to date suggest a score > 8 predicts the presence of clinically-judged AOS (Strand et al., 2014). Strand and colleagues tested the scale against consensus diagnosis by two

experts with a sample of 133 individuals, 33 with stroke-related AOS plus aphasia, 23 with primary progressive AOS with or without aphasia, and 78 with primary progressive aphasia alone. They reported high inter-rater reliability ($r > 0.87$) on presence and severity level for the 16 speech features. Currently, there are limited definitions and examples of the 16 behaviors and the reliability of the tool for less experienced clinicians has yet to be tested. Further, while it may be useful to measure all of these features to obtain a profile that can guide intervention, the optimal subset of features and their thresholds of severity necessary for positive AOS diagnosis are unknown.

Haley et al. (2012) explored perceptual scaling as well as quantitative measures. The latter were frequency counts of perceptual speech features, similar to those of Strand et al. (2014), and acoustic measurements of speech such as word and utterance durations and a scanning index capturing uniformity of syllable duration within utterances. They tested 39 individuals with aphasia with or without a diagnosis of AOS. Consistent with previous work on reliability of perceptual measurements outside the Mayo Clinic (Mumby et al., 2007; Zyski and Weisiger, 1987), expert raters' inter-rater reliability within 1 point on the scale varied from 46 to 93% agreement. Haley and colleagues found that four quantitative measures differentiated the individuals with AOS, or possible AOS, from the no-AOS group: percentages of segmental distortions, substitutions and prolongations, and inconsistent productions over five sequential repetitions of multisyllabic words. Inter-rater reliability for the quantitative perceptual measures was acceptable (Cunningham et al., in press). Unfortunately, no acoustic measures in Haley et al.'s study were informative, limiting the possibility of rapid automated measurements to support routine clinical diagnosis. Also, multivariate analyses were not conducted to identify whether a combination of measures accurately predicted expert AOS diagnosis.

Despite no acoustic measures correlating with expert diagnosis in Haley et al.'s study (2012), there has been a long history of exploration into acoustic measurements of speech in AOS (Ballard et al., 2000; McNeil et al., 2009). These have included, among others, measures of segment and intersegment durations, variability of segmental durations (Kent and McNeil, 1987; Seddoh et al., 1996; Square-Storer and Apeldoorn, 1991; Zaretsky and Velleman, 2011) variability of stop gap and voice onset time measures (Seddoh et al., 1996), and co-articulation (Ziegler and von Cramon, 1986). More recently, Ballard and colleagues have reported a measure that appears to capture the lexical stress changes in AOS and that correlates well with perceptual judgment of equal stress (Ballard et al., 2010). This measure is the pairwise variability index (PVI), which quantifies the relative duration of adjacent vowels in polysyllabic words, normalized for individual speech rate. Using discriminant function analysis, (Ballard et al., 2014) reported that the PVI measure of vowel duration by itself was the strongest predictor of presence of AOS, showing 88% agreement with expert judges in a sample of 40 individuals with primary progressive aphasia. Duffy et al. (2015) and Vergis et al. (2014) also noted that this measure has diagnostic potential. However, the studies by Ballard and colleagues (Ballard et al., 2014; Vergis et al., 2014) explored a small set of perceptual and acoustic measures and it is possible that other measures could prove as informative or moderate the explanatory power of the PVI.

1.1. Purpose

While predictive modeling is being used in other fields, it has not yet been applied to neurogenic speech disorders. Hence, the current study aimed to identify variables that most strongly predict the presence of AOS in a cohort of individuals with aphasia

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