



Temporal acoustic measures distinguish primary progressive apraxia of speech from primary progressive aphasia



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ABSTRACT

The purpose of this study was to determine if acoustic measures of duration and syllable rate during word and sentence repetition, and a measure of within-word lexical stress, distinguish speakers with primary progressive apraxia of speech (PPAOS) from nonapraxic speakers with the agrammatic or logopenic variants of primary progressive aphasia (PPA), and control speakers. Results revealed that the PPAOS group had longer durations and reduced rate of syllable production for most words and sentences, and the measure of lexical stress. Sensitivity and specificity indices for the PPAOS versus the other groups were highest for longer multisyllabic words and sentences. For the PPAOS group, correlations between acoustic measures and perceptual ratings of AOS were moderately high to high. Several temporal measures used in this study may aid differential diagnosis and help quantify features of PPAOS that are distinct from those associated with PPA in which AOS is not present.

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1. Introduction

Apraxia of speech (AOS) is a neurological speech disorder that has been of considerable theoretical and clinical interest for many years. Studies of AOS have contributed to the understanding of speech programming and the neural network that undergirds that process. Refinements in defining its clinical features have helped sharpen the diagnostic boundaries among AOS, aphasia, and dysarthria. Although AOS is usually stroke-induced, it can be the primary sign of neurodegenerative disease, in which case it has been termed primary progressive AOS (PPAOS, Duffy, 2006). Neuroimaging correlates of PPAOS include the left or left-greater-than-right superior premotor cortex and supplementary motor areas (Josephs et al., 2012; Whitwell et al., 2013). Pathologically, it most often is associated with tau biochemistry (4R tau) and progressive supranuclear palsy or corticobasal degeneration (Josephs et al., 2006). PPAOS was the primary focus of this study.

Because there is considerable variability in terminology and criteria for identifying different subtypes of primary progressive aphasia (PPA), particularly PPA subtypes that may be associated

with AOS, it is important to provide at the outset the terminology we will adopt in this paper. The designation PPAOS will be used for speakers who have AOS, but no evidence of aphasia. Although the terms agrammatic PPA (agPPA) and nonfluent PPA (nfPPA) are often used together or synonymously in many studies, the term agPPA in this paper will refer to speakers who met criteria for agPPA but had no evidence of AOS. We will use the designation nfPPA when referring to the literature on the nonfluent/agrammatic variant of PPA in which AOS and agrammatism are considered core features (Gorno-Tempini et al., 2011) and one or both problems can be present. Although AOS occurs very frequently in nfPPA (Duffy, Strand, & Josephs, 2014; Ogar, Dronkers, Brambati, Miller, & Gorno-Tempini, 2007), in this study we did not examine individuals who had both AOS and agrammatism. It is generally accepted that AOS does not occur in the semantic variant of PPA and that it is uncommon in the logopenic variant of PPA (lvPPA), although AOS errors can be difficult to distinguish from the phonological errors associated with lvPPA (Croot, Ballard, Leyton, & Hodges, 2012).

Diagnosis of nfPPA may be less reliable than for other PPA variants, possibly because of reduced reliability of AOS diagnosis (Harris et al., 2013). Some suggest that AOS be abandoned as a classification feature in PPA because its diagnosis relies on expert

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clinical judgment (Sajjadi, Patterson, Arnold, Watson, & Nestor, 2012). These concerns can be raised about many PPA features (e.g., judgments of “fluency”) but they do point to a need for greater diagnostic reliability.

Acoustic measures have potential as markers to improve diagnostic reliability and quantify features of AOS, as demonstrated in studies of stroke-induced AOS in which acoustic measures have quantified slow rate, syllable segmentation, equalized stress, and voicing abnormalities (e.g., Ballard et al., 2016; Haley, Jacks, de Riesthal, Abou-Khalil, & Roth, 2012; Kent & McNeil, 1987; Rogers & Storkel, 1999; Vergis et al., 2014; Ziegler & von Cramon, 1986). Acoustic measures have also been sensitive to temporal abnormalities in speakers with nPPA, and to differences between groups with nPPA versus lvPPA in which the nPPA group included at least some speakers with AOS (Ash et al., 2009; Ballard et al., 2014; Code, Ball, Tree, & Dawe, 2013; Knibb, Woollams, Hodges, & Patterson, 2009). Wilson et al. (2009) concluded that reduced maximum speech rate in their nPPA group likely reflected effects of AOS, but such conclusions are qualified by the possible confounding influence of co-occurring aphasia.

Acoustic correlates of PPAOS have received little attention but they recently have documented progressive slowing of speech rate in a few cases (Duffy et al., 2015, $n = 2$; Laganaro, Croisier, Bagou, & Assal, 2012, $n = 1$). More convincing evidence would come from comparing speakers with PPAOS to neurologically normal speakers and to speakers with PPA without AOS. Inclusion of groups with agPPA without AOS, and lvPPA without AOS, would capture PPA variants that most often co-occur with AOS (nPPA), or whose phonological errors can be difficult to distinguish from AOS sound-level errors (lvPPA and, probably to a lesser degree, nPPA). The degree to which acoustic measures distinguish PPAOS from those comparison groups would help gauge their value as diagnostic markers of PPAOS.

This study examined temporal acoustic features that are relevant to rate and syllabic stress abnormalities commonly associated with PPAOS in order to determine their sensitivity to abnormality and their specificity relative to normal speech and variants of PPA with which PPAOS may be confused. Assuming that some acoustic measures are sensitive to perceived rate and prosodic abnormalities associated with speech programming difficulties, and not to language impairment, we hypothesized that they would not differ among normal speakers and speakers with agPPA and lvPPA without AOS, and that those groups would have shorter utterance durations, more rapid syllable production rates, and greater differences between an unstressed and stressed vowel within a multisyllabic word than PPAOS speakers. A primary goal was to identify an easily measured acoustic marker(s) that can confirm the diagnosis and quantification of PPAOS and support its distinction from PPA without AOS.

2. Methods

2.1. Participants

The participants with PPAOS or PPA were among a cohort of 169 people who participated in IRB-approved studies of PPA and PPAOS. The criteria and methods for diagnosing PPAOS and PPA and its variants have been described elsewhere (Botha et al., 2015; Josephs et al., 2012; Josephs et al., 2013).

2.1.1. Primary progressive apraxia of speech (PPAOS)

Within the 169 participant cohort, 65 individuals had AOS based on the presence of speech features currently accepted as either distinctive of AOS (e.g., distorted sound substitutions or additions; increased sound distortions with increased length, com-

plexity, or rate), or commonly associated with AOS but possibly overlapping with aphasia (e.g., audible or visible articulatory groping) or dysarthria (e.g., slow rate; sound distortions) (Ballard et al., 2015; McNeil, Robin, & Schmidt, 2009; Strand, Duffy, Clark, & Josephs, 2014). After excluding those who unequivocally also had aphasia, 31 received a diagnosis of PPAOS. Among them, nine had unequivocal dysarthria and thus were excluded because of the possible influence of dysarthria on the acoustic measures. One additional individual was excluded because of a prominent non-English accent that might have biased acoustic measures. The remaining 21 individuals were the primary focus of this study (two of them were described in detail by Duffy et al., 2015). All of them had speech features distinctive of AOS. Many of them also had speech features that can overlap with aphasia or dysarthria but, as stated, none had unequivocal evidence of aphasia or dysarthria by any other measure. That is, their performance on several measures of language ability/aphasia was normal (see Results for details) and their motor speech was not characterized by any unequivocal speech features associated with dysarthria that do not overlap with AOS (e.g., strained voice quality, reduced loudness, hypernasality, rapid rate). We believe this is the largest cohort of individuals with “pure” AOS, regardless of etiology, for whom speech-language and acoustic data have been reported in a single study.

2.1.2. Agrammatic variant of primary progressive aphasia (agPPA)

Within the 169 participant cohort were 38 individuals who were classified as agPPA at initial assessment and one individual who initially was unclassifiable but was classified as agPPA at second assessment. The agPPA diagnosis was consistent with consensus criteria for nPPA/agPPA diagnosis (Gorno-Tempini et al., 2011); that is, there was evidence of agrammatism and/or AOS, plus at least two of three additional features (impaired complex sentence comprehension, spared single word comprehension, spared object knowledge). Among those 39 individuals, only six had no AOS or dysarthria, which qualified them for this study. As documented in the Results, their overall aphasia severity was mild but ranged from near normal to moderate. In addition to evidence of agrammatism in spoken or written language, they had mild to marked impairment in sentence comprehension and normal to moderate impairment on confrontation naming. Their inclusion allowed us to examine if agPPA without AOS is associated with acoustic temporal abnormalities, and if any such abnormalities are distinguishable from those that might be associated with PPAOS. We hypothesized that the acoustic measures would be sensitive to PPAOS but not to deficits associated with agPPA in the absence of AOS.

2.1.3. Logopenic variant of primary progressive aphasia (lvPPA)

Within the 169 participant cohort were 65 individuals who were classified as lvPPA. The lvPPA diagnosis was consistent with consensus criteria; that is, there was evidence of impaired word retrieval in spontaneous speech and naming, and at least three of four additional features (phonologic speech errors, spared single word comprehension and object knowledge, absence of frank agrammatism, spared motor speech). Twenty of them were selected for study because none had dysarthria or features of AOS that do not overlap with features of phonologic paraphasias (e.g., syllable segmentation) and because a previous study (Petroi, Duffy, Strand, & Josephs, 2014) established that they made few-to-many phonological errors. As documented in the Results, their overall aphasia severity was mild-moderate but ranged from near normal to severe. Their sentence comprehension and naming ability ranged from normal to severely impaired. Their inclusion allowed us to examine if lvPPA without AOS or dysarthria, but with varying degrees of phonologic errors, is associated with acoustic

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