Acoustic Analyses of Prolonged Vowels in Young Adults With Friedreich Ataxia

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Summary: Objectives. Finding measures that track disease progression and determine treatment efficacy is vital for appropriate management in Friedreich ataxia (FA). The purpose of this study was to determine which cepstral- and spectral-based measures extracted from prolonged vowels using Analysis of Dysphonia in Speech and Voice (ADSV) program discriminate between those who have FA and normal voice (NV) peers.

Study Design. This is a descriptive, prospective study.

Methods. Initial 2 seconds of prolonged /a/, /i/, and /o/ were analyzed through ADSV from 20 individuals diagnosed with FA and 20 NV individuals. ADSV measures used were cepstral peak prominence (CPP), cepstral peak prominence standard deviation (CPP SD), low/high spectral ratio (L/H ratio), low/high spectral ratio standard deviation (L/H ratio SD), and the Cepstral/Spectral Index of Dysphonia (CSID).

Results. L/H ratio SD was the only measure where significant differences were found across all vowels between groups. Comparing measures per vowel, the vowel /o/ was significantly different between groups on four of five measures. Discrimination analysis revealed 100% of those in the FA group were classified correctly (sensitivity), whereas 95% of NV members were correctly identified (specificity) when all ADSV measures, with the exception of L/H ratio, were entered.

Conclusions. Unstable periods of phonation, such as initiations of voice production in vowels, may yield robust acoustic cues in the FA population. ADSV provides measures that, when considered together, have excellent sensitivity and very good specificity. Vowels yielded differing results on ADSV measures; analysis of different vowel types is recommended.

Key Words: Friedreich ataxia–Acoustic analysis–Spectral-cepstral analysis–Vowels–ADSV.

INTRODUCTION

Ataxia typically occurs because of damage to the cerebellum and is characterized by a lack of muscle coordination and timing of movements. An ataxia changes the ability and extent of voluntary movement and alters the prolonged reflex muscle contractions needed for posture and equilibrium maintenance.[1] Friedreich ataxia (FA) is a specific ataxia caused by lesions in the cerebellum and in cortical, bulbar, and spinal pathways of the nervous system. FA is a hereditary, degenerative, disorder that affects the central and peripheral nervous systems, heart, skeleton, and pancreas. It is an autosomal recessive genetic disorder in which a mutation on chromosome 9q13 causes multiple repetitions of the guanine-adenine-adenine trinucleotide creating a defect in the frataxin gene characteristic of the disease. It is typical of nucleotides on DNA strands to repeat, but mutations occur when certain genes contain more than the typical amount of repetitions. This rare disorder occurs in approximately 1 of 50 000 individuals.[2] It is estimated that 9000 individuals have FA in the United States.[3] Although FA is uncommon, it is the most prevalent and most frequently occurring of young-onset hereditary ataxic disorders.[4] Prevalence does not differ significantly between genders. The average age of onset for this progressive disease is as young as 10 years of age, and the mean age of death is approximately 32 years of age.

Dysarthria, which occurs in approximately 84–100% of those with FA,[5] usually begins to become noticeable in the first 2 years after disease onset,[6] and is a mixture of ataxic, spastic, and flaccid features.[1,7] Speech symptoms include, but are not limited to, imprecise consonant productions, irregular articulatory breakdowns, increased variance in voice-onset time, vocal instability, dysphonic voice (strained-strangled, rough, breathy), shorter utterance duration per breath, reduced differences in voice-onset time between voice and voiceless stops, and overall reduced intelligibility.[1,3,8–11] Breakdowns in speech production can be attributed to loss of coordination and muscle weakness due to damage to the cerebellum, corticospinal tracts, and cranial nerves.[12] Reduced intelligibility in FA individuals has been reported to be associated with word-final plosive voicing, vocal harshness, and inconsistent articulation breakdowns.[13] In addition to articulatory breakdowns, Eigentler et al[2] described a decrease in loudness over long periods of phonation and a slower speech rate in analysis of speech samples from individuals with FA. Ackermann and Hertrich[14] found dysphonia to be a salient feature of cerebellar ataxia; the most common speech features reported were irregular articulation errors, imprecise consonants and vowels, and a perceptually "harsh" voice. In sum, the most prominent aspects of speech affected in FA are articulation, prosody, and phonation.[10]

Acoustic analysis is a popular method of quantifying speech and voice attributes[15] and thus may provide valuable quantitative data on specific differences of FA speech production

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compared with normal speaking individuals. An advantage that acoustic analysis offers is the provision of objective correlates for perceptually distinguishable motor speech deficits.[5] Acoustic analysis measures provide estimates of voice quality and speech production that may or may not be perceptually distinguishable by listeners and are helpful for collecting baseline measures, tracking the progress of intervention, and for quantifying voice disorder symptoms. It has been suggested that acoustic measures are necessary for detecting subtle changes in speech that occur with disease progression in those with FA,[9] in contrast to perceptual estimates that can be problematic due to listener/rater reliability issues.[16,17]

Common acoustic measures include fundamental frequency, jitter, shimmer, harmonics-to-noise ratio, formant frequencies, signal amplitude, speech rate, and voice-onset time. Jitter, shimmer, and noise-to-harmonics ratio represent acoustical estimates that are commonly included as part of a comprehensive voice evaluation,[18,19] along with perceptual assessment of voice (the gold standard in voice evaluations), videostroboscopy, and possibly aerodynamic measures,[19] and are frequently used in clinical and research settings.[8,14,19–21] Although all these acoustic measures are valuable for quantifying progress throughout therapy and perhaps disease progression,[22] they are based on a univariate, isolated method of voice analysis which has been criticized based on questionable validity that the algorithms have in determining exact cycle boundaries in aperiodic/dysphonic voice signals.[23] Recent advances in the quantification of dysphonic voice samples have led to the commercial release of an automated computer software program, the Analysis of Dysphonic Speech and Voice (ADSV; KayPENTAX, Montvale, NJ), which uses a multivariate approach to analyze the quality of voice during sustained vowel phonation and in speech samples.[24] Voice production is a multidimensional process requiring the control of frequency, intensity, vocal quality along with the coordination, and implementation of aerodynamic aspects. To obtain adequate measures that properly represent this complex process, a multivariate approach has been suggested.[24,25] Multivariate approaches offer a solution to problems that occur with univariate approaches, such as determining if a specific case is actually deviant from what is normal, and provide a multidimensional representation of an individual's voice.[24] Unlike univariate methods, a multivariate approach considers several variables (ie, frequency, intensity, and aerodynamic components) and provides information that is useful to determine normal versus abnormal behaviors by combining variables despite the level of the individual value.[24]

The use of acoustic estimates with FA has recently been recommended to track the progression of the complicated disease process.[9] To date, however, only a few have used acoustic measures in objectively determining the effects of motor speech diseases on the phonatory system, or specifically in using such estimates to assist in the documentation of treatment efficacy, despite findings that respiration, voice quality, voice instability, articulation, and tempo (rate of speech) are affected by FA.[9,13,26,27] It seems important to study the effects of this complex disease acoustically, using cepstral/spectral measures that are not compromised by accuracy issues encountered when using time-based estimates and to estimate accuracy in sorting between FA and normal voice (NV) participants based on cepstral/spectral measures. Therefore, the primary aims of this study were (1) to examine if cepstral/spectral measures of voice differentiate between those diagnosed with FA and a normal voiced comparison group (Is there a difference between FA and NV participants on ADSV measures?) and (2) to determine how accurately participants can be classified into either the FA or NV group based on ADSV measures. (Can group membership be predicted based on ADSV measures and how accurate are those predictions?) To compare ADSV measures between the groups, the effects of gender by vowel type on cepstral/spectral measures were taken into account because differences in these variables have been found to affect cepstral peak prominence (CPP)[27,28] and low/high spectral ratio (L/H ratio)[28] in healthy adults; thus, gender and vowel type were considered when comparing measures between groups. It was hypothesized that the following measures extracted from ADSV analysis would be significantly different in the dysphonic samples as compared to the NV samples, specifically (1) CPP, (2) cepstral peak prominence standard deviation (CPP SD), (3) L/H ratio, (4) low/high spectral ratio standard deviation (L/H ratio SD), and (5) Cepstral/Spectral Index of Dysphonia (CSID).[24] CPP and L/H ratio were hypothesized to be lower in value and CPP SD, L/H ratio SD, and CSID were hypothesized to be higher in value as compared to NV participants.[24] Regarding vowels and gender, information comparing different vowels by gender using cepstral/spectral analysis[27] or ADSV[28] has been limited to healthy adults thus far. Findings have identified greater mean CPP values on low vowels /a/[27,28] and /æ/[27] versus high vowels /i/[27,28] and /u/.[27] In addition, males have been found to have significantly greater mean CPP values than females.[27] For L/H ratio, mean values for /i/ versus /a/ were larger in males as compared to females. Finally, for both genders, higher mean L/H ratios were found on /a/ versus /i/.[28] We predicted that findings from our FA participants would show similar trends.

METHODS

Participants

Twenty adolescents and young adults who were diagnosed with FA from France and 20 college students with NVs from the United States served as participants in this study. The FA patients were recruited from all over the country of France and were referred by physicians for a medical study at the Institut National de la Santé et de la Recherche Médicale. All 20 of these individuals were native French speakers, with a mean age of 18.5 years (SD = 3.7), and age range of 10–25 years; there were 10 males and 10 females (Table 1).

Inclusion criteria for those in the NV group were as follows: (1) must be between the ages of 18–25 years, (2) have no medical history of speech and/or voice disorders, (3) judged to have a perceptually normal vocal quality and be in self-reported good health at the time of the experiment, (4) no history of smoking, (5) be a gender match for an age-equivalent participant in the FA group, and (6) speak English as their first language. This