Comparison of Acoustic Measures in Discriminating Between Those With Friedreich's Ataxia and Neurologically Normal Peers

Sophia Luna-Webb
University of Central Florida

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COMPARISON OF ACOUSTIC MEASURES
IN DISCRIMINATING BETWEEN THOSE WITH
FRIEDREICH’S ATAXIA AND NEUROLOGICALLY NORMAL PEERS

by

SOPHIA L. LUNA-WEBB & CECYLE CARSON, PH. D.

A thesis submitted in partial fulfillment of the requirements
for the Honors in the Major Program in Communication Sciences and Disorders
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Abstract

Background: Technological advancements in speech acoustic analysis have led to the development of spectral/cepstral analyses due to questions regarding the validity of traditional time-based measures (i.e., Jitter, Shimmer, and Harmonics-to-Noise-Ratio) in objectifying perturbations in dysphonic voices.

Aim: This study investigated the validity of time-based measures in discriminating those with Friedreich’s ataxia (FA) from normal voiced (NV) peers when compared to cepstral-spectral measures.

Method: A total of 120 sustained vowel phonations from an existing database of 40 participants (20 FA; 20 NV) of the vowels /a/, /i/, and /o/ were analyzed to determine which set of variables (i.e., time-based vs. cepstral-spectral) better predicted group membership. Four variables of time-based measures (Jitter Local %, Jitter RAP, Shimmer Local %, Shimmer APQ11, and HNR) were analyzed via the freeware program PRAAT and compared to four cepstral-spectral measures (Cepstral Peak Prominence, Cepstral Peak Prominence Standard Deviation, Low/High Ratio Standard Deviation, and the Cepstral/ Spectral Index of Dysphonia) extracted from the Analysis of Dysphonia in Speech and Voice (ADSV) software program.

Results: Findings from a discriminant analysis showed sensitivity and specificity results to be better for ADSV measures; 100% of those in the FA group were classified correctly (sensitivity), and 95% of members in the NV group were correctly identified (specificity) as compared to PRAAT (70% sensitivity and 85% specificity).
Conclusions: Cepstral-spectral measures are much more accurate in discriminating between those with FA and NV peers as compared to time-based estimates.

Keywords: acoustic measures, Friedreich’s ataxia, acoustic analysis, time-based measures, cepstral-spectral measures, dysphonia
This work is dedicated to my sons, Aldo and Emilio Webb, whom I aspire to be their motivation in continuing their education. May you always remember the infinite love and sacrifice of your mother.
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Chapter 1: Introduction

Friedreich’s ataxia (FA) is a multisystem degenerative disease of the nervous system affecting many qualities of life. Dysarthria, a pervasive symptom of FA, is a prominent early indicator of the disease that increases in severity as the disease progresses. Further, dysarthria is also a complicated multidimensional motor speech disorder that can have dysphonia as a characteristic, with related frequency perturbations and noise occurring in the voice signal. With technological advancements in the field of communication sciences and disorders, traditional methods that were commonly used in acoustic analysis of voice are being replaced by more innovative advancements in acoustic analysis. With the development of Cepstral Peak Prominence (CPP) analysis, traditional acoustic measures (i.e., jitter, shimmer, harmonic-to-noise ratios) which have been the basis in objectifying perturbations in normal and pathological voices have been heavily criticized for their lack of reliability and validity (e.g., Bielamowicz, Kreiman, Gerratt, Dauer, & Berke, 1996; Heman-Ackah, et al., 2003; Rabinov, Kreiman, Gerratt, & Bielamowicz, 1995). This Fourier transformation algorithm, first described by A. Michael Noll (1964), has been growing in popularity as a reliable quantitative method for use in a variety of voice disorders and diagnoses. Analysis of Dysphonia in Speech and Voice (ADSV™; Kay Elementrics), a recently developed and marketed acoustic analysis software program allows users to extract measures of fluctuations in the voice signal via cepstral and spectral analysis. Therefore, the intent of this study is to compare traditional acoustic measures to cepstral/spectral analysis measures in the ability to discriminate between those with normal voices and those with dysphonic voices who have the diagnosis of Friedrich’s ataxia.
Chapter 2: Literature Review

Friedrich’s Ataxia

Ataxia is a result of damage to the cerebellum in which the control circuits responsible for the timing and coordination of muscle movements are disrupted. Ataxia impedes the smoothness and effectiveness of volunteer muscle contraction, negatively affecting the person’s stance and balance. While ataxia can be considered a functional neurological disorder caused by strokes, multiple sclerosis, tumors, alcoholism, peripheral neuropathy, metabolic disorders, and vitamin deficiencies, the National Institute of Neurological Disorders and Stroke (NINDS) also describes the condition to be organic in nature (2014).

Friedrich’s ataxia (FA) is an autosomal recessive degenerative disease caused by a genetic abnormality involving the inheritance of an unstable guanine-adenine-adenine (GAA) repeat expansion on the gene transcription; FA is the most common hereditary disorder of the nervous system. As first described by Nikolaus Friedreich (1863), it is a degenerative disorder associated with ataxia, dysarthria, pyramidal tract deficiencies, sensory loss, cardiomyopathy and diabetes.

The typical age of onset is reported between the ages of 7 and 25 years, with symptoms occurring in early adolescence years (Anheim, Tranchant, & Koenig, 2012; Delatycki & Corben, 2012). Although FA commonly affects individuals of European descent, evidence of the GAA expansion have also been found in North African, Middle-Eastern and Indian populations
(Anheim, Tranch, & Koenig, 2012; Ashley, Hoang, Lynch, Perlman, & Maria, 2012; Parkinson, Boesch, Nachbauer, Mariotti, & Giunti, 2013). It is estimated that the incidence of FDRA is about 1 in 29,000 (Delatycki & Corben, 2012) to 50,000 in the Caucasian population (Ashley et al., 2012), with an estimated 9,000 individuals presently affected in the United States (Koeppen, 2011). The devastating disease also does not differ according to gender, affecting both males and females alike.

**Ataxic Dysarthria and Speech Symptoms**

Dysarthria is a primary clinical feature in Friedreich’s ataxia; characterized as the most common and early clinical symptom (Eigentler, Rhomberg, Nachbauer, Ritzer, Poewe, & Boesch, 2012) of this progressive disease; and present in more than 90% of individuals (Delatycki & Corben, 2012; Rosen, Folker, Vogel, Corben, Murdoch, & Delatycki, 2012). Further, ataxic dysarthria is a motor speech disorder characterized by the scanning pattern of speech, disturbed articulation of both consonants and vowels, and abnormal voice quality (Kent, Kent, Duffy, Thomas, Weismer, & Stuntebeck, 2000). Scanning speech is described by broken up or nonfluent speech in which words are separated by multiple pauses. Abnormal voice qualities may include: mono-pitch, mono-loudness, pitch breaks, harshness, breathiness, voice tremors, and/or strained/struggled voice (Kent et al., 2000).

While the characteristics of ataxic dysarthria are well defined, speech symptoms that relate to FA are still poorly understood (Blaney & Hewlett, 2007a). In fact, the speech disorder of FA has been described by some as a “mixed dysarthria” (Blaney & Hewlett, 2007a; Folker, Murdoch, Rosen, Delatycki, Corben, & Vogel, 2012), suggesting that speech symptoms vary
according to neurological components that are affected. In a review of the literature, Blaney and Hewlett (2007a) reported that FA often results in a mixture of ataxic/spastic/flaccid dysarthria components. Other prominent and common features include reduced articulation rate and reduced respiratory function (Folker et al., 2012). In contrast, Eigentler et al. (2012) found that while voice is considered to be most compromised, respiration was a component found to be less affected in FA and had no significant correlation to the disease duration compared to healthy individuals.

In general, FA compromises the respiratory, velopharyngeal, laryngeal, and articulatory subsystems to some degree (Folker, et al., 2012). Noticeable speech symptoms may include: deviations of voice quality (strained-strangled, roughness, breathiness, glottal fry, phonation breaks, pitch breaks, excess or reduced pitch variations, decrease in loudness); articulatory breakdowns (imprecision of consonants and vowels productions, reduction of phrase/phoneme length, distorted vowels, prolonged phonemes); and deviations in prosody (increase of equal and excess stress, reduction in speech rate, and prolonged intervals). Overall, ataxic dysarthria results in a significant reduction in intelligibility thus compromising the individual’s speech communication.

**Perceptual Measures**

The perceptual method in detecting disordered voice and speech features is considered to be the “gold standard” in clinical practice (Rabinov, Kreiman, Gerratt, & Bielamowicz, 1995). Perceptual ratings from listeners are used to give an estimate of the overall severity of the dysarthria and the associated speech patterns in voice quality, intelligibility, and prosody (Kent,
et al., 2000). Speaking tasks, such as sustained vowel phonation, syllable repetition, sentence recitation, and conversation can be used in perceptual analysis. Sustained vowel and syllable alternation motion rate (AMR) are two commonly used tasks that are sensitive to motor disruptions and insensitive to language (Kent, et al., 2000). Standardized perceptual tests include, but are not limited to, the Assessment of Intelligibility of Dysarthric Speech (Yorkston, Beukelman, & Traynor, 1984), PaTA, Frenchay Dysarthria Assessment (Enderby & Palmer, 2008), and the Consensus Auditory-Perceptual Evaluation of Voice (CAPE-V; Kempster, Gerratt, Verdolini, Barkmeier-Kraemer, & Hillman, 2009, & Zraick, Kempster, Connor, Thibeault, Klaben, Bursac, Thrush, & Glaze, 2011).

However, clinical limitations exist in perceptual analysis. One major limitation with this method is the reliability of the listener’s judgment. Since perceptual analysis weighs heavily on listener skill, despite training and experience, a lack of reliability between and among listeners (listener biases) may still arise, particularly in voices with severe dysphonia (Blaney & Hewlett, 2007a). Blaney and Hewlett found that listeners respond to severe dysphonia by using their own range of listening strategies in an attempt to decode the signal.

The lack of consistency in establishing intelligibility scores is another shortcoming of perceptual analysis. Numerous researchers have proposed their own method and criteria of classifying severity and rating intelligibly in dysarthria, making it difficult to compare results and accurately define the voice disorder. Blaney and Hewlett (2007b) stated that careful consideration should be given to intelligibility-based severity ratings of dysarthria.
Acoustic Measures

Traditional Measures

While perceptual analysis is considered to be the “gold standard” in diagnosing pathological voice qualities, clinicians have gained more interest in obtaining acoustic analyses to objectively validate their perceptual judgments. In an online survey of voice diagnostic procedures used by Speech-Language Pathologists who had experience using or interpreting stroboscopic evaluations, Behrman (2005) reported that around 75% of respondents (n = 41) considered acoustic measures important for certain diagnostic tasks (helping to define overall and specific treatment goals).

In order to acoustically measure dysphonia in FA, we first need to identify the aspects of the voice signal to distinguish typical from atypical patterns. Phonation is the release of energy, beginning at the source, as the air expires from the lungs, creating subglottal air pressure below the vocal folds and consequently setting them into vibration, thus creating an acoustic signal, or a frequency on a spectrum. Because the human voice is a complex wave, the harmonics of the frequency, typically sinusoidal waves, create a complex wave. The first harmonic (H1) is considered to be the fundamental frequency in the waveform. The fundamental frequency, or f0, is the rate at which the vocal folds vibrate or the number of times the glottis opens/closes in a period of time measured in Hertz (Hz).

To analyze the cycle-to-cycle variations in the vocal fold vibrations and to identify noise components in the waveform (non-harmonic elements), two general measures were employed, one quantifying perturbations (jitter and shimmer) and the other computing the harmonics-to-
noise ratio (HNR). Well defined in the literature, these measures have been the widely accepted approaches in the diagnostic and evaluation of voice disorders (Brockmann, Drinnan, Storck, & Carding, 2011). Numerous voice analyses software and hardware systems have adopted these measures to quantify features of dysphonic voices (e.g., CSpeech, Computerized Speech Lab (KayPentax), Multi-Dimensional Voice Program, and Praat).

_Jitter_ refers to the measurement of voice frequency perturbation; the timing variation between cycle-to-cycle, whereas _shimmer_ refers to voice amplitude perturbation; the amplitude variation between period-to-period. While it is typical to find small irregularities in the acoustic wave (Brockmann et al., 2011), jitter and shimmer are important measures of the cycle-to-cycle variations related to fundamental frequency (Ryalls & Behrens, 2000). Similarly, _harmonics_, expressed in decibels (dB) are the multiples of the fundamental frequency. Through a ratio algorithm, scientists were able to extract and distinguish noise components of a waveform from the harmonic components, called _harmonics-to-noise ratio_.

These time-based measures are characterized by their dependence on accurate identification of periodic cycle boundaries and calculation of the cycle-to-cycle perturbations in a waveform (i.e., jitter, shimmer). Jitter represents the variability in irregular vibratory patterns of the fundamental frequency, resulting in an increase percentage of jitter. While shimmer describes varying loudness in the voice, it is also affected by intensity; or sound pressure level (SPL), which is commonly not controlled or considered (Brockmann et al., 2011). The problem emerges in dysphonic voices which are noisy and aperiodic; the noise can mask cycle-to-cycle boundaries.
and randomness in cycles can make accurate determination of when one cycle ends and the other begins extremely difficult, thus resulting in estimates that may be invalid and unreliable.

Another major limitation of time-based measures is the inability to effectively analyze continuous speech. An established protocol for use with those who have dysarthria has not been agreed upon. Some have suggested that there may be a better correlation between perceptual ratings and continuous speech, particularly in individuals with dysphonia (Awan & Roy, 2009) in contrast to prolonged vowels. While both speech tasks are essential to describing pathological voices, Hillenbrand and Hounde (1996) found that sustained vowel productions (/ɑ/) more accurately predicted perceived breathiness when compared to connected speech (second sentence from the Rainbow Passage).

**Praat: A traditional-based approach voice analysis**

In 1992, phoneticians Paul Boersma and David Weenink developed a popular freeware computer program for use when analyzing, synthesizing, and manipulating speech, called Praat (http://www.praat.org/). Its acoustic measures are based on estimating cycle lengths using a waveform-matching algorithm and searching for the best match between consecutive cycles. In Praat, a linear signal processing algorithm estimates the extent of match between presumed cycles (Boersma & Weenink, 2011). Pratt’s speech analysis offers five different calculation measures of jitter and six different measures of shimmer, along with noise perturbation (HNR).

**Analysis of Dysphonia in Speech and Voice (ADSV)**

The Analysis of Dysphonia in Speech and Voice (ADSV) is a recently developed software program developed by Shaheen N. Awan (2011) and is commercially available from
Kay Pentax. ADSV uses spectral/cepstral analysis, rather than time-based, and can analyze both sustained vowel and continuous speech samples. It provides a multi-variable analysis consisting of the Cepstral Peak Prominence (CPP), Low/High spectral ratio (L/H Ratio) and their corresponding standard deviations; and the Cepstral Spectral Index of Dysphonia (CSID). Awan and colleagues have evaluated and validated the ability of cepstral and spectral measures to quantify the presence and severity of dysphonia in sustained vowel and connected speech productions, and have reported moderate to high associations with listener perceptions (e.g., Awan & Roy, 2005; Awan, Roy, Jette, Meltzner, & Hillman, 2010; Awan, Solomon, Helou, and Stojadinovic, 2013; Lowell, Kelley, Awan, Colton, & Chan, 2012; Peterson, Roy, Awan, Merrill, Banks, & Tanner, 2013).

**Purpose**

The use of acoustic estimates to track the progression of the complicated disease process associated with FA has recently been recommended, although, to date, few have added acoustic analysis of voice into estimates of disease progression and clinical outcomes (e.g., Folker, Rosen, Murdoch, Delatycki, Corben, Vogel, 2011; Rosen, et al., 2012). As Delatycki stated (2009), the development of sensitive indicators capable of detecting small changes in performance are needed. Acoustic measures may be able to provide clinically relevant insights about changes that aren’t detectable through other means (Rosen, et al., 2012).

The purpose of this research was to determine the effectiveness of cepstral/spectral measures yielded from ASDV as compared to time-based measures from Praat in discriminating those with FA from gender matched and age equivalent normal voiced peers. No study, to our
knowledge, has evaluated the use of cepstral and spectral versus time-based measures with this population. Results from this study should be considered clinically valuable because it is assumed that some clinicians may be using available time-based freeware programs to supplement and objectify subjective (perceptual) measures of voice in clientele, so it is important to know how accurate these measures are in lending themselves to disease tracking and treatment efficacy in FA.

Chapter 3: Method

Research Design

The following study was designed as a retrospective design. ADSV measures have been identified which distinguish between individuals with FA as compared to normal voiced peers from a previous study conducted by Hardin (2012). Acoustic estimates obtained from Hardin were available and used in this study to determine the effectiveness of traditional time-based acoustic measures yielded from Praat as compared to the newer cepstral/spectral measures extracted from ADSV in discriminating between individuals with FA and those with normal voices.

Participants:

The 20 adolescents and young adults who were diagnosed with FA were primarily recruited by a pharmaceutical company and the Institut National de la Santé et de la Recherche Médicale (INSERM) in France. FA participants were native French speakers, with a mean age of 18.5 years (SD= 3.7 years), and age range of 10 to 25 years; there were 10 males and 10 females.
Similarly, the 20 normal voice (NV) group representing gender-matched and age-equivalent peers were recruited from the University of Central Florida in the United States. The mean age of those in the NV group was 20.7 years (SD= 2.2 years) with an age range of 18 to 25 years; there were also 10 males and 10 females (See Table 1).

Inclusion criteria for those in 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 disordered participant in the FA group, and (6) speak English as their first language. These participants were blinded to the intent of the study.

Recording

In France, the FA participants were recorded using the Marantz digital audio tape-recorder (DAT), set at 22.5 kHz sampling rate, in a quiet office. Recordings of stimuli were always conducted in the same sequence; specifically, each participant sustained the vowels /a/, /i/, and /o/ at their normal fundamental frequency for the longest duration possible.

Data collection procedures involving those in the NV group were conducted in a quiet room the UCF and were comparable to those used in France. Sustained vowel voice samples of the NV participants were recorded on a Roland Edirol digital recorder (R-09HR), set at a 44 kHz sampling rate. Samples were then down-sampled through Multispeech (KayPentax) to a rate of 22.5 kHz. The down-sampling allowed recordings to have the same sample rate for further
acoustic analysis. The mouth-to-microphone distance during recording was held constant as 12 inches using the internal microphone on the Edirol (20-40 KHz frequency response).

**Acoustic Measures**

This study included a total of 10 dependent variables (acoustic measures) divided into two sets. One set included time-based measures extracted from Praat, namely: (1) Jitter Local %, (JLocal) (2) Jitter Relative Average Perturbation (JRAP), (3) Shimmer Local % (SLocal) (4) Shimmer-APQ11 (S-APQ11), and (5) Harmonics-to-Noise Ratio (HNR); while the second set was composed of cepstral-spectral measures obtained from ADSV, including: (6) Cepstral Peak Prominence (CPP), (7) CPP Standard Deviation (CPP SD), (8) Low/High Spectral Ratio (L/H Ratio), (9) L/H Spectral Ratio Standard Deviation (L/H Ratio SD), and (10) Cepstral/Spectral Index of Dysphonia (CSID). These measures were extracted from the three sustained vowels.

As stated earlier, Praat software provides five different measures of jitter and six for shimmer. For this research two were selected for jitter, two for shimmer, and one for HNR. JLocal is the average difference between peaks of consecutive cycles, divided by the mean frequency; whereas, SLocal is similar to JLocal, with the difference involving the measurement of amplitude of consecutive peaks, rather than time difference between peaks and divided by average amplitude. JRAP is calculated by averaging the difference between a period or cycle and the period prior to and following the cycle of interest, then dividing by the average period. S-APQ11 is based on comparing the absolute difference between amplitudes of the period of interest and 10 surrounding periods, then dividing by the average amplitude. Finally, HNR is an estimate of the periodicity of the waveform. The amount of energy in the harmonic (periodic)
portion is contrasted with noise in the waveform (Boersma & Weenink, 2011). Measures of jitter and shimmer are predicted to be higher while HNR values should be lower in dysphonic voices (Williamson, n.d.).

Five acoustic estimates provided by ADSV were recorded. Their definitions follow. CPP is a measure representing the amplitude of the most prominent cepstral peak and compared to an expected amplitude using linear regression. The CPP value is predicted to be lower in those who have dysphonic voices. The stability or variability of CPP over time is measured by a standard deviation measure, CPP SD, and should be higher from dysphonic voice samples because of probably increased inconsistency across the sample (Awan, 2011).

L/H Ratio represents the ratio of low- versus high-frequency spectral energy in a voice sample and is sensitive to the occurrence of high-frequency noise, especially above 2-3 kHz. Dysphonic voices would be expected to display a low L/H Ratio because of an increased amount of high-frequency spectral noise. L/H Ratio SD is a measure of the steadiness of the L/H Ratio across the duration of a sample. Greater variability would be expected in a dysphonic voice, as compared to normal voices, and would be displayed as a higher L/H Ratio SD value (Awan, 2011).

CSID is a combination of CPP, L/H Ratio, and associated standard deviations; thus, it is considered as multidimensional (Awan, 2011). CSID has been referred to as an estimate of the severity of dysphonia and has been reported to correlate highly with listener perceptions from the CAPE-V (Awan, Roy, Jette, Meltzer, & Hillman, 2010). Larger CSID scores would be expected from dysphonic voice samples as compared to normal voice samples.
**Sustained Vowel Phonation and Parameters of Voice Samples**

Traditionally, sustained vowel phonation has been the cornerstone in the clinical assessment of voice and speech. Moreover, prolonged vowel analysis has been found to be sensitive to motor disruptions (Kent et al., 2000). To provide an accurate representation of voice features affected by FA, the first 2 seconds (2000ms) of each prolonged vowel was selected based on the unstable nature of voicing onsets which might be even more unstable in those with FA.

Each participant sustained the vowels /ɑ/, /i/, and /o/ at their normal fundamental frequency maintaining phonation as long as possible on a single breath, and at a comfortable vocal loudness level. A total of 120 voice samples were obtained from those in the FA (60 samples; 3 vowels X 20 participants) and NV groups (60 samples).

**Statistical Analysis**

Acoustic estimates produced from Praat and ADSV were entered into an analysis of covariance (ANCOVA) to test for significant differences between groups using SPSS (Version 22.0; SPSS, Inc., Chicago, IL). Age was found to be significantly different between groups (t (38) = -2.29, p <.05), in that participants in the FA group were younger than those in the NV group; therefore, age was controlled statistically as a covariate. Independent t-tests were conducted on variables when Levene’s test of homogeneity of variance demonstrated significantly different or unequal variances. Further, Cohen’s D formula was used to determine the effect sizes between groups (i.e., FA and NV peers) for those acoustic estimates with significant F-values. Lastly, a Discriminate analysis, followed by a Stepwise procedure, was
employed to determine the specificity and sensitivity of Praat and ADSV measures in predicting group membership. An alpha level of $p \geq .05$ was selected.

**Chapter 4: Results**

ANCOVA outcomes on the time-based measures of JRAP, JLocal, SLocal, S-APQ11, and HNR among sustained vowel phonations /ɑ/, /i/, and /o/ are summarized on Table 2. In general, only shimmer measures and HNR values for the vowel /ɑ/ were found to be significantly different between groups. Neither of the jitter measures were statistically different between the groups, nor among the vowels. Cohen’s D values were in the large range for SLocal /ɑ/, S-APQ11 /ɑ/, and HNR /ɑ/ (Cohen, 1988). Average values were in the direction predicted, with the exception of jitter and shimmer measures for /i/. Therefore, JRAP, JLocal, SLocal and S-APQ11 results for /i/ were not entered into the Discriminate analysis.

Findings from the Discriminate analysis for Praat measures revealed six misclassifications in the FA group (P# 3, 8, 10, 13,16, 19), yielding 70% sensitivity (true positive rate). Three participants were incorrectly identified in the NV group (P# 26, 27, 36), resulting in a specificity of 85% (true negative rate). Overall, 77.5% of the 40 participants were correctly classified.

A Stepwise Discriminant analysis was employed to determine which set of acoustic measures from Praat were most effective in predicting group membership (i.e., FA and NV). Only SLocal /ɑ/ entered into the discriminate function. Results indicated that, of the 20 FA
participants, only 12 were classified correctly (60% sensitivity); whereas, 16 of those in the NV group were classified correctly (80% specificity).

ANCOVA results for ADSV measures revealed that all acoustic estimates were significantly different between the groups, dependent on the vowel. In general, results for most cepstral-spectral based measures were found to be significantly different between groups. L/H Ratio SD values were the only estimates that were significantly different across all three vowels between the groups. All values were in the direction predicted with the exception of L/H Ratio /o/ (see Table 3). It was predicted that L/H ratio values would be higher for those in the NV group because their voices should have more low frequency components and less noise (high frequency components) than those in the FA group who presumably had dysphonic voices; however, the opposite was found. Thus, the Discriminant analysis using cepstral-spectral based measures from ADSV was based on CPP, CPP SD, L/H Ratio SD, and CSID for all three vowels, and excluded L/H Ratio.

Results from the Discriminant analysis revealed 100% sensitivity in correctly identifying those in the FA group, while 95% of participants in the NV group were correctly identified (# 37 was misclassified) (i.e., one false positive). Twelve variables entered into the Stepwise discriminate function in the following order: L/H Ratio SD /i/, CPP /o/, L/H Ratio SD /a/, L/H Ratio SD /o/, CSID /i/, CPP SD /o/, CSID /a/, CPP /a/, CSID /o/, CPP /i/, CPP SD /i/, and CPP SD /a/. Sensitivity and specificity results from the Stepwise procedure were identical to the findings when all 12 ADSV measures were included.
Chapter 5: Discussion

The purpose of this study was to compare traditional acoustic measures of voice to newer measures that do not rely on the accurate identification of periodic cycle boundaries to calculate perturbations and noise in a waveform. To date, no other study has applied acoustic measures of voice to discriminate between those with FA and neurologically normal peers, despite suggestions that speech diagnostic measures may be clinically useful in tracking change in FA (Folker, et al., 2012; Rosen, et al., 2012; Singh, Epstein, Myers, Farmer, & Lynch, 2010) and in demonstrating response to therapy (Rosen, et al., 2012; Blaney & Hewlett, 2007a). In the present study, cepstral-spectral measures extracted from ADSV were 100% accurate in correctly classifying those who had FA, and 95% accurate in classifying gender-matched and age-equivalent normal voiced peers. Cepstral/spectral measures from ADSV were more accurate in predicting group membership than time-based measures from Praat, and thus are recommended for use with those diagnosed with FA.

The use of acoustic measures in analyzing speech and voice with this disease has an advantage over some physiological measures because of the accessibility and ease of collection of speech and voice samples across time, and their possible association with neurological decline (Folker, et al., 2012). As reported by Delatycki (2009), effects of FA on the nervous system have mainly been determined through use of rating scales (including a speech subscale that provides a general estimate of dysarthria or “speech”), impairment patient report, and functional composites such as the Multiple Sclerosis Functional Composite (Cutter, et al., 1999). These general or broad assessments are important in documenting global declines in motor control, but may not be sensitive to small changes to specific skills that are needed to evaluate the
effectiveness of therapies on FA. Thus, it would seem specific performance measures related to
dysarthria, a key feature of FA, may hold promise and prove useful as indicators of disease
progression due to the muscle activation and coordination needed from a variety of speech
subsystems (articulation, resonation, phonation, and respiration). An array of measures of
speech performance (including voice) has not been determined or recommended for assessing
this population, which may be one reason why, as of yet, no effective pharmacological treatment
has been found that demonstrates positive changes which results in improved speech of those
with FA (Vogel, Folker, & Poole, 2014).

Although not part of the research question, our results support findings by Brockmann et
al. (2011) that recommend the use of /a/ as the vowel of choice when using Praat for time-based
acoustic analyses. This idea is based on the finding that both shimmer estimates and the HNR
measure for the vowel /a/ were significantly different between the groups, and all had large
effect sizes (Cohen, 1988). It should be noted that all participants in the Brockmann et al. study
had normal voices, unlike half of our participants. When analyzing dysphonic voice signals with
Praat, the user must take into account the limitations imposed by time-based measures, especially
in regard to more severely dysphonic voices (Awan, 2011; Carding, Steen, Webb, MacKinzie,
Deary, & Wilson, 2004).

The inclusion of several vowels that differ in terms of features such as tongue height (high,
mid, low), horizontal position of the tongue (front-central-back), and tenseness (tense, lax) is
recommended for use when collecting samples from individuals with FA for analysis, in addition to
speech samples. The measures that entered the ADSV stepwise analysis included all three vowels
sampled.
**Limitations**

Several limitations in the methodology of this study should be noted as considerations and suggestions for future studies:

1. Gender was not included as a variable in this study. It is unknown whether gender may have influenced statistical results related to predicting group membership, although ADSV measures classified both males and females in the FA group correctly. While gender was found to be a less important factor in the reliability of acoustic measures than control of vocal loudness during vowel prolongation, it was found to have an effect on jitter and shimmer using Praat (Brockman, et al., 2011). Also, Garrett (2013) reported gender differences in CPP and L/H spectral ratio estimated using ADSV from a stable mid-section of the vowels /i/ and /a/ produced by normal voiced adult males and females. CPP and L/H spectral ratio results indicated that male participants (20-30 and 40-50; n = 30) had higher CPP and L/H spectral ratio values than female participants (Garrett, 2013). Future studies of dysphonic voices should include gender as a potential variable in statistical analyses.

2. This study only obtained sustained vowel phonations and did not include continuous speech tasks. Since irregular speech patterns are more discernable in conversation, future studies should analyze the impact of vowel phonations when compared to prolonged vowel phonation and within the context of continuous speech.

3. This study has attempted to predict group membership solely from acoustic methods. Typically, a complete diagnostic battery for medical evaluations is combined by many other measures, including perceptual and physiological in addition to acoustic
parameters. Determining the combination of acoustic variables that best predict a disease process, along with perceptual ratings, and physiological examinations may provide the most clinically valid results.

4. Samples were not recorded in a sound treated environment, so it is likely that some external room noise was captured along with the signal. This issue could have an effect on ADSV measures, although it is common in clinical settings to record voice productions from clients in a quiet room.

**Conclusion**

FA is a complex, degenerative disease that has dysarthria as one of its cardinal features, which impacts the ability to be understood by listeners, and in turn can affect quality of life. To date, no pharmacological treatment has proven to be effective in improving the speech of those with FA (Vogel & Folker, Poole, 2014), possible due to the use of global measures to indicate change. Our results support the use of cepstral/spectral measures from ADSV to analyze the prolonged vowels of /a/, /i/, and /o/. Findings verify the sensitivity and specificity of these measures, as compared to traditional time-based measures. Further research is needed to create a set of measures with the ability, combined with general assessments, to capture the slow decline in motor function and validate pharmacological and behavior treatments for FA to help those affected live a fuller life, despite the disease.
### Table 1

Participant Characteristics

<table>
<thead>
<tr>
<th>P#</th>
<th>Gender</th>
<th>Age</th>
<th>P#</th>
<th>Gender</th>
<th>Age</th>
</tr>
</thead>
<tbody>
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<td>1</td>
<td>Male</td>
<td>10</td>
<td>21</td>
<td>Male</td>
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</tr>
<tr>
<td>2</td>
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<td>Male</td>
<td>18</td>
</tr>
<tr>
<td>3</td>
<td>Male</td>
<td>16</td>
<td>23</td>
<td>Male</td>
<td>18</td>
</tr>
<tr>
<td>4</td>
<td>Male</td>
<td>16</td>
<td>24</td>
<td>Male</td>
<td>19</td>
</tr>
<tr>
<td>5</td>
<td>Male</td>
<td>16</td>
<td>25</td>
<td>Male</td>
<td>20</td>
</tr>
<tr>
<td>6</td>
<td>Male</td>
<td>17</td>
<td>26</td>
<td>Male</td>
<td>20</td>
</tr>
<tr>
<td>7</td>
<td>Male</td>
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<td>27</td>
<td>Male</td>
<td>21</td>
</tr>
<tr>
<td>8</td>
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<td>28</td>
<td>Male</td>
<td>21</td>
</tr>
<tr>
<td>9</td>
<td>Male</td>
<td>20</td>
<td>29</td>
<td>Male</td>
<td>21</td>
</tr>
<tr>
<td>10</td>
<td>Male</td>
<td>21</td>
<td>30</td>
<td>Male</td>
<td>23</td>
</tr>
<tr>
<td>11</td>
<td>Female</td>
<td>16</td>
<td>31</td>
<td>Female</td>
<td>18</td>
</tr>
<tr>
<td>12</td>
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<td>32</td>
<td>Female</td>
<td>19</td>
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<td>Female</td>
<td>19</td>
</tr>
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<td>14</td>
<td>Female</td>
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<td>34</td>
<td>Female</td>
<td>20</td>
</tr>
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<td>15</td>
<td>Female</td>
<td>20</td>
<td>35</td>
<td>Female</td>
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</tr>
<tr>
<td>16</td>
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</tr>
<tr>
<td>19</td>
<td>Female</td>
<td>24</td>
<td>39</td>
<td>Female</td>
<td>24</td>
</tr>
<tr>
<td>20</td>
<td>Female</td>
<td>25</td>
<td>40</td>
<td>Female</td>
<td>25</td>
</tr>
</tbody>
</table>

*Note 1. P# = participant number.*
Table 2

Analysis of Covariance (ANCOVA) of Adolescents and Young Adults with Friedreich’s Ataxia (FA) and Normal Voices (NV) on Acoustic Measures from Praat.

<table>
<thead>
<tr>
<th>Measures</th>
<th>Vowels</th>
<th>FA</th>
<th>NV</th>
<th>F- value</th>
<th>Cohen's</th>
</tr>
</thead>
<tbody>
<tr>
<td>Jitter (RAP)</td>
<td>/a/</td>
<td>.26 (.11)</td>
<td>.24 (.08)</td>
<td>0.66</td>
<td></td>
</tr>
<tr>
<td></td>
<td>/i/</td>
<td>.29 (.18)</td>
<td>.33 (.21)</td>
<td>1.95</td>
<td></td>
</tr>
<tr>
<td></td>
<td>/o/</td>
<td>.24 (.12)</td>
<td>.19 (.12)</td>
<td>0.8</td>
<td></td>
</tr>
<tr>
<td>Jitter (%)</td>
<td>/a/</td>
<td>.49 (.18)</td>
<td>.45 (.16)</td>
<td>0.28</td>
<td></td>
</tr>
<tr>
<td></td>
<td>/i/</td>
<td>.54 (.29)</td>
<td>.59 (.35)</td>
<td>1.71</td>
<td></td>
</tr>
<tr>
<td></td>
<td>/o/</td>
<td>.45 (.19)</td>
<td>.36 (.22)</td>
<td>0.77</td>
<td></td>
</tr>
<tr>
<td>Shimmer (%)</td>
<td>/a/</td>
<td>8.26 (2.89)</td>
<td>5.36 (2.23)</td>
<td>10.38**</td>
<td>1.12</td>
</tr>
<tr>
<td></td>
<td>/i/</td>
<td>6.16 (3.30)</td>
<td>6.94 (3.56)</td>
<td>-1.70†</td>
<td>-0.23</td>
</tr>
<tr>
<td></td>
<td>/o/</td>
<td>6.19 (2.27)</td>
<td>5.54 (3.04)</td>
<td>0.18</td>
<td></td>
</tr>
<tr>
<td>Shimmer (APQ)</td>
<td>/a/</td>
<td>7.91 (3.17)</td>
<td>5.07 (2.25)</td>
<td>8.96**</td>
<td>1.03</td>
</tr>
<tr>
<td></td>
<td>/i/</td>
<td>4.92 (2.71)</td>
<td>6.92 (4.55)</td>
<td>4.12*</td>
<td>-0.53</td>
</tr>
<tr>
<td></td>
<td>/o/</td>
<td>6.02 (2.69)</td>
<td>5.37 (3.13)</td>
<td>0.22</td>
<td></td>
</tr>
<tr>
<td>HNR</td>
<td>/a/</td>
<td>14.71 (3.0)</td>
<td>17.92 (3.34)</td>
<td>8.22**</td>
<td>-1.01</td>
</tr>
<tr>
<td></td>
<td>/i/</td>
<td>17.64 (4.63)</td>
<td>19.87 (3.47)</td>
<td>1.18</td>
<td></td>
</tr>
<tr>
<td></td>
<td>/o/</td>
<td>20.23 (3.02)</td>
<td>20.66 (4.73)</td>
<td>-0.34†</td>
<td>-0.11</td>
</tr>
</tbody>
</table>

Note 1. F0= Fundamental frequency; RAP= Relative Average Perturbation; APQ= Amplitude Perturbation Quotient; HNR= Harmonics-to-Noise Ratio. Values are expressed as means and (standard deviations).

Note 2. Higher mean scores of Jitter and Shimmer measurements are associated with dysphonic voice signals; whereas, higher standard deviations indicate greater variability in the voice signal.

Note 3. * p < .05; ** p < .01, 2-tailed. †t-test; Multivariate test degrees of freedom were 1, 38; t-test degrees of freedom was 35.493 for F0 /a/ vowel; 36.240 for F0 /i/ vowel; 30.995 for Shimmer (APQ) /i/ vowel; and 32.289 for HNR /a/ vowel.

Note 4. Cohen’s D statistic was calculated on only measures in which the F-value was significant.
Table 3

Analysis of Covariance (ANCOVA) Comparisons of Adolescents and Young Adults with Friedreich's Ataxia (FA) and Normal Voices (NV) on Acoustic Measures from the Analysis of Dysphonia in Speech and Voice (ADSV).

<table>
<thead>
<tr>
<th>Measures</th>
<th>Vowels</th>
<th>FA</th>
<th>NV</th>
<th>F-value</th>
<th>Cohen’s D</th>
</tr>
</thead>
<tbody>
<tr>
<td>CPP (dB)</td>
<td>/a/</td>
<td>10.33 (1.86)</td>
<td>11.64 (1.64)</td>
<td>8.09*</td>
<td>0.75</td>
</tr>
<tr>
<td></td>
<td>/i/</td>
<td>6.72 (2.38)</td>
<td>8.04 (1.98)</td>
<td>2.69</td>
<td></td>
</tr>
<tr>
<td></td>
<td>/o/</td>
<td>8.24 (1.99)</td>
<td>10.33 (1.65)</td>
<td>13.43**</td>
<td>1.14</td>
</tr>
<tr>
<td>CPP SD (dB)</td>
<td>/a/</td>
<td>1.10 (.40)</td>
<td>.98 (.68)</td>
<td>0.22</td>
<td></td>
</tr>
<tr>
<td></td>
<td>/i/</td>
<td>.95 (.39)</td>
<td>.79 (.26)</td>
<td>0.66</td>
<td></td>
</tr>
<tr>
<td></td>
<td>/o/</td>
<td>.89 (.24)</td>
<td>.61 (.36)</td>
<td>4.81*</td>
<td>0.91</td>
</tr>
<tr>
<td>L/H Ratio (dB)</td>
<td>/a/</td>
<td>30.81 (5.96)</td>
<td>28.29 (4.49)</td>
<td>1.03</td>
<td></td>
</tr>
<tr>
<td></td>
<td>/i/</td>
<td>27.09 (7.92)</td>
<td>25.52 (4.59)</td>
<td>0.16</td>
<td></td>
</tr>
<tr>
<td></td>
<td>/o/</td>
<td>37.94 (5.78)</td>
<td>30.94 (5.12)</td>
<td>12.79**</td>
<td>-0.128</td>
</tr>
<tr>
<td>L/H Ratio SD (dB)</td>
<td>/a/</td>
<td>2.36 (.90)</td>
<td>1.64 (.44)</td>
<td>9.59*</td>
<td>1.02</td>
</tr>
<tr>
<td></td>
<td>/i/</td>
<td>2.76 (1.05)</td>
<td>1.76 (.46)</td>
<td>-3.88**</td>
<td>1.23</td>
</tr>
<tr>
<td></td>
<td>/o/</td>
<td>2.36 (.67)</td>
<td>1.77 (.52)</td>
<td>7.40*</td>
<td>0.98</td>
</tr>
<tr>
<td>CSID</td>
<td>/a/</td>
<td>30.70 (13.73)</td>
<td>20.83 (9.85)</td>
<td>6.41*</td>
<td>0.83</td>
</tr>
<tr>
<td></td>
<td>/i/</td>
<td>51.89 (17.51)</td>
<td>38.72 (9.08)</td>
<td>5.69*</td>
<td>0.94</td>
</tr>
<tr>
<td></td>
<td>/o/</td>
<td>30.12 (13.45)</td>
<td>20.83 (12.91)</td>
<td>3.19</td>
<td></td>
</tr>
</tbody>
</table>

Note 1. CPP= Cepstral Peak Prominence; CPP SD= Cepstral Peak Prominence Standard Deviation; L/H Ratio= Low/High Spectral Ratio; L/H Ratio SD= Low/High Spectral Ratio Standard Deviation; CSID= Cepstral/Spectral Index of Dysphonia. Values are expressed as means and (standard deviations).

Note 2. Lower mean scores on CPP are suggestive of dysphonic vocal quality; whereas, increased variability as displayed by CPP SD values is indicative of dysphonic voice. A lower L/H Ratio SD is associated with dysphonic voice signals; while a higher L/H Ratio SD is associated with the variability seen in dysphonic voices. Higher CSID estimates are related to increased dysphonia severity.

Note 3. * p < .05; ** p < .01, 2-tailed; † t-test; Multivariate test degrees of freedom were 1, 38; t-test degrees of freedom was 25.94.

Note 4. Cohen’s D statistic was calculated on only measures in which the F-value was significant.
APPENDIX A: Approval of Human Research
Approval of Human Research

From: UCF Institutional Review Board 51
FWA0000381, OHRP0000125E
To: Kaylee D. Hardin and Co-PI: Turel, Vareide
Date: April 16, 2013

Dear Researcher,

On 4/10/2013, the IRB approved the following human participant research until 4/9/2014 inclusive:

Type of Review: UCF Initial Review Submission Form
Project Title: Auditory Analysis in Normal, Young, College Students
Investigator: Kaylee D. Hardin
IRB Number: 13-09161
Funding Agency: N/A
Grant Title: N/A
Research ID: N/A

The nature, extent of the research, and all other factors that are to be studied in the study will be reviewed by the IRB. The Continuing Review Form must be submitted 90 days prior to the expiration date for research that was previously reviewed and 60 days prior to the expiration date for research that was previously reviewed at a convened meeting. Do not make changes to the study, i.e., protocol, methodology, consent form, personnel, etc., until receiving IRB approval. A Modification Form cannot be used to extend the approved period of a study. All forms may be completed and submitted online at https://research.ucf.edu.

If continuing review approval is not granted before the expiration date of 4/9/2014, approval of the research will be lost during that period. If your research is not completed when the approval expires, all participants will be dismissed from participating in the study. If you have any questions about the details of this approval, please contact the IRB Coordinator by phone at 407-823-2851 or fax at 407-822-2275.

Use of the approved, signed consent document(s) is required. The form signed by all participants, which are now valid for further use, is the approved investigator(s) for this study. Participants may securely store the signed copy for future reference. Participants are encouraged to receive a copy of the consent form(s).

In the conduct of this research, you are responsible to follow the requirements of the Investigator's Manual. On behalf of Sophia Dimitriou, Ph.D., L.C.S.W., UCF IRB Chair, this letter is signed by:

Signature: [signature]

IRB Coordinator
APPENDIX B: IRB Informed Consent
Acoustic Analysis in Normal, Young, College Students
Informed Consent

Principal Investigator(s): Kaylea Hardin
Sub-Investigator(s): Tara Varsallone
Faculty Supervisor: Jack Ryalls, PhD

Investigational Site(s): University of Central Florida, Department of Communication Sciences and Disorders University of Central Florida, Communication Disorders Clinic

Introduction: Researchers at the University of Central Florida (UCF) study many topics. To do this we need the help of people who agree to take part in a research study. You are being invited to take part in a research study which will include about 20 people at UCF. You have been asked to take part in this research study because you are a normal speaking UCF speaking. You must be 18 years of age or older to be included in the research study.

The person doing this research is Kaylea Hardin, Masters Student in the Communication Sciences & Disorders graduate program. Because the researcher is a graduate student, she is being guided by Dr. Jack Ryalls, a UCF faculty supervisor in Communication Sciences & Disorders. Tara Varsallone is a Graduate UCF student, who is also participating in this study as part of the research team.

What you should know about a research study:

• Someone will explain this research study to you.
• A research study is something you volunteer for.
• Whether or not you take part is up to you.
• You should take part in this study only because you want to.
• You can choose not to take part in the research study.
• You can agree to take part now and later change your mind.
• Whatever you decide it will not be held against you.
• Feel free to ask all the questions you want before you decide.

Purpose of the research study: The purpose of this study is to establish normative voice measures of speech from young, college students at UCF. Multidimensional Voice Profile (MDVP) quickly and easily provides a revealing snapshot of voice quality. Since its introduction, MDVP has garnered numerous references in peer-reviewed professional journals establishing its reliability, value of multiple parameters, and efficacy. There is an extensive database of normal adult speakers available for the MDVP provided by Kay Elemetrics, but this data is not broken down by age of speaker. We would like to obtain a database and voice parameters for young, normal, healthy, college age students.

What you will be asked to do in the study: Participants will be asked to say various vowel sounds into a microphone on a digital recorder. Speakers will be recorded in a quiet environment.

Location: University of Central Florida, Department of Communication Sciences & Disorders and University of Central Florida, Communication Disorders Clinic.

Time required: We expect that you will be in this research study for 1 session of no more than 15 minutes.

Audio or video taping: You will be audio taped during this study. If you do not want to be audio taped, you will not be able to participate in the study. Discuss this with the researcher or a research team member. If you are audio taped, the tape will be kept in a locked, safe place. The tape will be erased or destroyed after completion of the study.

Risks: There are no reasonably foreseeable risks or discomforts involved in taking part in this study.

Benefits: There are no expected benefits to you for taking part in this study.

Compensation or payment: There is no compensation or other payment to you for taking part in this study.

Confidentiality: We will limit your personal data collected in this study to people who have a need to review this information. We cannot promise complete secrecy.

Study contact for questions about the study or to report a problem: If you have questions, concerns, or complaints, talk to Kaylea Hardin, Graduate Student, Communication Sciences &
Disorders Program, College of Health and Public Affairs, (407) 823-4798 or Dr. Jack Ryalls, Faculty Supervisor, Department of Communication Sciences & Disorders at (407) 823-4798 or by email at ryalls@ucf.edu.

IRB contact about your rights in the study or to report a complaint: Research at the University of Central Florida involving human participants is carried out under the oversight of the Institutional Review Board (UCFIRB). This research has been reviewed and approved by the IRB. For information about the rights of people who take part in research, please contact: Institutional Review Board, University of Central Florida, Office of Research & Commercialization, 12201 Research Parkway, Suite 501, Orlando, FL 32826-3246 or by telephone at (407) 823-2901. You may also talk to them for any of the following:

- Your questions, concerns, or complaints are not being answered by the research team.
- You cannot reach the research team.
- You want to talk to someone besides the research team.

Your signature below indicates your permission to take part in this research.

DO NOT SIGN THIS FORM AFTER THE IRB EXPIRATION DATE BELOW

________________________________________
Name of participant

________________________________________
Signature of participant  Date

________________________________________
Signature of person obtaining consent  Date

________________________________________
Printed name of person obtaining consent
References


 _Journal of Child Neurology_ 27(9), 1095-1120.


 _Journal of Child Neurology_ 27(9), 1095-1120.


Carding, P. N., Steen, I. N., Webb, A., MacKenzie, K., Deary, I. J. & Wilson, J. A.


