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ACOUSTIC ANALYSIS OF PROLONGED VOWELS IN ADOLESCENTS AND YOUNG ADULTS WITH FRIEDREICH'S ATAXIA

by

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B.S. University of Central Florida, 2012

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ABSTRACT

This study employed spectral analyses for acoustic measures of sustained vowel productions from a group of 20 adolescents and young adults with Friedreich’s Ataxia (FA) and compared findings with a group of 20 age-equivalent and gender-matched normal control participants. State-of-the art spectral analyses from the Analysis of Dysphonia in Speech and Voice (ADSV) program, developed for various voice disorders from Kay Elemetrics, were applied to initial 2 second sustained vowel segments of the vowels /a/, /i/, and /o/. Spectral analyses included averages and standard deviations of 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), Cepstral/Spectral Index of Dysphonia (CSID), and Mean Cepstral Peak Prominence Fundamental Frequency (Mean CPP F0).

Statistical analyses revealed significant differences between the spectral analyses of voice characteristics of individuals with FA and those of normal controls for all measures except for CPP SD. The aim of these analyses was to determine spectral differences evident in vowel productions of individuals with FA using new cepstral-derived measures that characterize the phonatory instability and dis-coordination present in this disorder. Such research may not only help develop early non-invasive indicators of ataxia and track disease progression, but also serve to stimulate research into alleviating the symptoms of this devastating disease.

This work is dedicated to my mother and father, Anna and Bobby Hardin, whose abundant support, love, and encouragement made this document possible. May this manuscript represent the hard-work and perseverance they have modeled for me and my brother.

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CHAPTER 1: 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 (Blaney & Hewlett, 2007). Friedreich's 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, autosomal recessive disorder that affects the central and peripheral nervous systems, heart, skeleton, and pancreas.

FA is an autosomal recessive genetic disorder in which a mutation on chromosome 9 causes multiple repetitions of the guanine-adenine-adenine (GAA) trinucleotide creating a defect in the frataxine 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 out of 50,000 individuals (Eigentler et al., 2012). It is estimated that 9000 individuals have FA in the United States (Koeppen, 2011). Although FA is uncommon, it is the most prevalent hereditary ataxic disorder. Prevalence does not differ significantly between genders (Koeppen, 2011). 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 (Koeppen, 2011).

There are several core symptoms related to FA that distinguish it from other dysarthric disorders of which are essential for professionals to be aware. This degenerative disease can cause negative visual effects, nystagmus, uncoordinated gait and ataxic movements (Sheth, 2010), cardiomyopathy, lower body sensory impairment, diabetes, and possible hand-wasting (Koeppen, 2011). A salient feature of FA is mixed dysarthria, with a prominent ataxic component, and usually indicates a lesion of the cerebellum and its connections (Blaney & Hewlett, 2007).

Speech symptoms include imprecise consonant productions, irregular articulatory breakdowns, reduced voice onset time, duration reduction, and overall reduced intelligibility (Koeppen, 2011). Breakdowns in speech production can be attributed to loss of coordination and muscle weakness, which are common characteristics of FA (Sheth, 2010). Hypotonia may be present due to damage to the lower motor neurons in the brainstem, causing reduction of speech duration (Blaney & Hewlett, 2007). Individuals with FA can be categorized into subgroups based on phonetic profiles of differing severity in speech abnormalities, and clusters of specific speech errors (Blaney & Hewlett, 2007). A phonetic profile was developed through analyzing patterns of minimal pair contrast errors and associating these errors with certain etiologies of specific speakers. An example of a high frequency phonetic error found in speech production of those with FA is word final plosive voicing contrast on consonants (e.g., ‘bat’ vs. ‘bad’).

To identify this disorder, intelligibility assessments, speech analysis, and quality of life measures should be assessed by a trained Speech-Language Pathologist. Neurogenic subsystem disorders cause distinct acoustic voice characteristics that can be represented through acoustic analysis (Kent, Vorperian, & Duffy, 1999). Significant differences found in vocal quality result in communication problems for FA patients and should be accurately measured by assessment tools. It is important to remember that FA is degenerative and changes over time; therefore, continued assessment is necessary to provide effective treatment (Koeppen, 2011). Perceptual analysis, the gold-standard of voice evaluation, should be conducted in conjunction with acoustic measures to verify findings. In order to confirm that acoustic and perceptual abnormalities are related to FA and to identify the location of lesion in the brain, the following tests can be utilized: electrocardiogram (ECG), electromyography (EMG), genetic testing, muscle biopsy, x-ray, computed tomography (CT scan), and magnetic resonance imaging (MRI) of the head (Sheth, 2010). Spectrographic analysis of speech can provide necessary information for treatment goals on an individual’s prosody, voice, respiration, resonance, articulation, and intelligibility (Koeppen, 2011).

Acoustic analysis may provide valuable quantitative data on specific differences of FA speech production compared to normal speaking individuals. A vocal sample from an individual can be analyzed to provide objective correlates for perceptually distinguishable motor speech deficits (Ackermann & Hertrich, 2000). Formerly, it was difficult to obtain good reliability on acoustic measures because analysis was time-consuming and required voice laboratory expertise (Carding, Wilson, MacKenzie, & Deary, 2009). However, presently there are a number of speech analysis software programs that allow rapid estimation of a number of acoustic signal characteristics. The use of these objective measures combined with perceptual characteristics should contribute significantly to data employed for evidence-based practice. In regard to dysarthria, the prominent speech disorder of FA, a small number of published studies exist on the acoustic characteristics that correlate with the obvious perceptual errors produced by disordered individuals (Bunton & Weismer, 2001).

A Review of the Literature

Friedreich's Ataxia

FA presents with unique speech characteristics that reflect the various subsystems affected. Speech features change or worsen as the disease progresses; however, studies of the association between acoustic signal modifications with the stages of disease progression within the individuals with FA are only now beginning to appear (Rosen et al., 2012). A common aspect that is found in the literature on FA is that this progressive disease creates a complex variation of symptoms dependent on the stage of degeneration. Research in 1863 on FA identified loss of cells in the hypoglossal nuclei and degenerative damage of the spinal cord extending to the medulla (Blaney & Hewlett, 2007). Due to the diffuse involvement of the hypoglossal, vestibular, glossopharyngeal, and vagus cranial nerves and the trigeminal root, dysarthria associated with FA is a mixed ataxic/spastic/flaccid type (Blaney & Hewlett, 2007). Although cranial nerve involvement suggests a mixed dysarthria, FA is often classified as an ataxic dysarthria.

Speech Symptoms

In the first two years of disease onset, dysarthria typically emerges as a salient feature of FA (Levee, 2011). Overall, it is agreed that ataxic dysarthria renders disordered individuals unintelligible, causing several communication problems. Severity of unintelligibility varies and has been found to range from mild to severe (Blaney & Hewlett, 2007). Reduced intelligibility in FA individuals has been reported to be associated with word final voicing, vocal harshness, and inconsistent articulation breakdowns, combined with word final voicing contrasts involving plosives yielding the highest error rate (Blaney & Hewlett, 2007a). In a follow-up study on word final voicing, listeners' misperceptions on minimal pairs of single words produced by individuals with FA, differing only by voicing of the final plosive consonant, were related to vowel duration, voicing in the closure phase, formant 1 (F1) frequency at mid-vowel, and a drop in F1 at the termination point of the vowel (Blaney & Hewlett, 2007b). Ouellon, Ryalls, Lebeuf, & Joanette (1991) found higher standard deviations in VOT productions in French speaking patients with FA compared to normal peers, although average VOT values were similar.

Although there are controversies over dysarthria severity rating scales and underlying factors affecting intelligibility, distinct abnormal speech characteristics have been reported for individuals with FA. Ackermann and Hertrich (2000) found dysphonia to be a salient feature of cerebellar ataxia, and the most common speech features to be irregular articulation errors, imprecise consonants and vowels, and a perceptually 'harsh' voice. The most prominent aspects of speech affected by ataxic dysarthria are articulation, prosody and phonation (Folker et al., 2012). In addition to articulatory breakdowns, Eigentler *et al.* (2012) described a decrease in loudness over long periods of phonation, and a slower speech rate in analysis of speech samples from individuals with FA. Perceptually, listeners have identified hoarseness, breathiness, and nasality in the voices of individuals with ataxia (Wolfe & Steinfatt, 1987). Several authors believe that individuals with FA can be classified into subgroups, based on common speech symptoms categorized as phonetic profiles (e.g., Folker et al., 2012).

Assessment and Diagnosis

Speech language pathologists (SLPs) lack appropriate information concerning the neurological, physiological, perceptual, and acoustic features required for intervention decisions because of the limited number of studies on FA and incomplete data on speech symptoms (Blaney & Hewlett, 2007). In order to assess the intelligibility of dysarthria associated with FA, Blaney and Hewlett (2007) utilized an intelligibility rating scale to perceptually determine the severity of 11 adult males who had been diagnosed with FA, who were between the ages of 22-45 years. The majority of FA participants were judged to have mild dysarthria. Blaney and Hewlett (2007) stated that acoustic analysis of dysarthria, in conjunction with perceptual ratings, is beneficial in providing insight into the clinical presentation of speech symptoms that remains poorly understood. As previously mentioned, phonetic profiling of clusters of speech errors can be applied with physiological and acoustic analysis in diagnosing FA (Blaney & Hewlett, 2007). One means to further delineate clinical speech symptoms is to isolate specific acoustic correlates of the disease that can be measured across time.

Results from a study (Eigentler et al., 2012) provided data to support a correlation between the Frenchay Dysarthria Assessment (FDA) scores and GAA repetition length, but not with disease duration; revealing an early appearance of effects on speech production in FA. The speech scale employed found a decrease of loudness, articulation problems, and a lower speech rate amongst the FA participants.

Assessments such as the FDA are time-consuming and somewhat subjective; whereas, instrumental techniques programmed to calculate acoustic measures quickly provide essential data for the development of interventions and to document change across disease progression (Folker et al., 2012). In Folker *et al.*'s study, a perceptual analysis of connected speech, acoustic analysis of speech and voice, accelerometric assessment of nasality, and spirometric and kinematic assessment of breathing during speech were employed as assessment tools. The overall goal of the study was to find measures that would be useful to track the progression of dysarthria and measure speech symptoms. Results revealed that instrumental techniques were helpful for tracking disease progression and subsystem changes. In order to measure intervention outcomes of FA treatment, Folker et al. suggested that acoustic measures are effective because of their greater accessibility over other evaluation measures such as physiological instrumentation.

Acoustic Analysis

Acoustic analysis provides objective measures of voice quality and speech production that are perceptually distinguishable and are helpful for collecting baseline measures, tracking the progress of intervention, and for quantifying voice disorder symptoms. Bunton and Weismer (2001) found that acoustic measures of vowel articulation are highly correlated with intelligibility of speakers with dysarthria caused by diseases such as amyotrophic lateral sclerosis (ALS), Parkinson's disease, and cerebral palsy.

Acoustic Measures

Common acoustic measures include fundamental frequency (F0), jitter, shimmer, duration of phonation, harmonics-to-noise ratio, formant frequencies, signal amplitude, speech rate, and voice onset time (VOT). A review of the literature by Ackermann and Hertrich (2000) reported inconsistent findings regarding F0 in those with ataxic dysarthria, with some studies results indicating a decreased mean F0, some with no significant differences in F0, and others signifying a higher fundamental frequency for those with ataxic dysarthria when compared to results from normal participants. Jitter, shimmer, and noise-to-harmonics ratio represent the most widely recognized and investigated parameters of voice quality and are frequently employed in clinical and research settings (Ackermann & Hertrich, 2000; Gelfer, 1995; Maryn et al., 2009).

For the data available on acoustic correlates of speech common in FA, segmental durations of phonation represent the majority of this data (Ackermann & Hertrich, 2000). Analysis of speaking rate assessed by oral diadochokinesis is a popular articulatory test and provides useful information about the precision and speed of the speech mechanism. Consistent reports have revealed a reduced speaking rate in individuals with FA (Ackermann & Hertrich, 2000). VOT differs among speakers depending on the language spoken and represents the time between the onset of voicing and the release of a stop consonant (Ryalls & Behrens, 2000). Ackermann and Hertrich (2000) found that in many studies, those with ataxic dysarthria displayed voicing problems due to prolonged or variability of the VOT in voiceless stops (Ryalls & Behrens, 2000).

Although all of the acoustic measures previously described are valuable for quantifying progress throughout therapy and perhaps disease progression (Kent, Vorperian, & Duffy, 1999), these measures provide a univariate, isolated method of voice analysis (Awan, 2011). The Analysis of Dysphonic Speech and Voice (ADSV) provides more in-depth and comprehensive measures to analyze the quality of voice during sustained vowel phonation with a multivariate approach. Voice production is a multidimensional process requiring the control of frequency, intensity, vocal quality along with the coordination and implementation of aerodynamic aspects (Awan, 2011). In order to obtain adequate measures that properly represent this complex process, a multivariate approach should be employed. 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 (Awan, 2011). Unlike univariate methods, a multivariate approach considers several variables (i.e. frequency, intensity, and aerodynamic components) and provides information that is useful to determine normal vs. abnormal behaviors by combining variables despite the level of the individual value (Awan, 2011). A separate multivariate formula is automatically calculated through the ADSV program to produce a multidimensional representation of voice, termed Cepstral/Spectral Index of Dysphonia (CSID). This unique and valuable multivariate estimate of severity is a combination of Cepstral Peak Prominence (CPP), CPP SD, Low/High Spectral Ratio (L/H Ratio), and L/H Ratio Standard Deviation (L/H Ratio SD). The CSID value has been directly correlated with the perceptual measures from the Consensus Auditory-Perceptual Evaluation of Voice (CAPE-V) (Awan, 2011). Correlating acoustic measures with the traditionally viewed "gold-standard" of perceptual characteristics provides a comprehensive evaluation of individual voice production. This information can provide valuable insight into the severity of the individual's vocal quality, an adequate baseline of performance, and lead to proper decisions for effective treatment options.

In order to predict the severity of a disordered voice, the mean of CPP has been shown to be an effective measure (Awan, 2011). This mean value does not provide information on the stability of CPP over the length of voice production, therefore ADSV employs spectral/cepstral calculations across the phonation duration to produce a standard deviation (SD) of the amount of variability in the voice signal (Awan, 2011). An average of CPP (mean CPP F0), determined by comparing the amplitude of the cepstral peak to the average cepstral amplitude, may provide a strong correlation to the severity of a voice disorder and help distinguish between normal and abnormal vocal productions (Awan, 2011). Awan and colleagues have also shown CPP SD to provide data that may distinguish normal from disordered voices (Awan, 2011). Due to the typical steadiness or stability of a normal voice, CPP SD is expected to be lower, reflecting a small degree of variability (Awan, 2011). Therefore, a dysphonic voice is expected to demonstrate a high volume of variability and a larger CPP SD value (Awan, 2011).

L/H Ratio represents the ratio of low- versus high-frequency spectral energy in a voice sample and has been utilized to provide information about the severity of dysphonic voices (Awan, 2011). A normal voice tends to have a high L/H Ratio since the majority of the spectrum energy is collected in the F0 (Awan, 2011). The opposite is found in dysphonic voices which reflect a low L/H Ratio because of an increased amount of high-frequency spectral noise that often results in the perception of breathiness (Awan, 2011).

L/H Ratio SD indicates the stability or steadiness of the L/H Ratio across the duration of a vocal sample (Awan, 2011). Normal vowel prolongations are expected to result in a low L/H Ratio SD due to the consistency typically found in unaffected voices (Awan, 2011). Greater variability is anticipated in a consistent and an intermittent dysphonic voice, which is reflected in a higher L/H Ratio SD value (Awan, 2011).

Vowel Characteristics

Although there are conflicting views between authors about the use of sustained vowel samples versus connected speech, it is generally agreed that the analysis of connected speech is more complex and difficult to analyze although valuable acoustic measures can be obtained from vowel production (Carding, Wilson, MacKenzie, & Deary, 2009). Connected speech is influenced by the glottal and subglottal mechanisms and contains voiceless phonemes, rapid voice onset and offset times, and prosodic and amplitude changes (Maryn et al., 2009). Vowels are typically not affected by speech rate, vocal pauses, phonetic context, and stress the way that continuous speech is affected (Maryn et al., 2009). In order to improve accuracy of acoustic measurements, most researchers have chosen sustained vowel samples because of the more clearly time-based cycle demarcations than that in continuous speech (Lowell, 2012), in addition to standardized speech tasks. Difficulty with the stability of phonation during vowel production and tremor has been found to affect the intelligibility of dysarthric individuals and are related to changes in perturbation measures (Ackermann & Ziegler, 1991; Gelfer, 1995; Keller, Vigneux, & Laframboise, 1991; Levee, 2011).

Particular vowels have provided somewhat different results for acoustic measures of dysarthria and ataxia vocal quality. Wolfe and Steinfatt (1987) found that tongue height during vowel production was correlated with vocal disorder severity. The vowels /a/ and /i/ were frequently employed in studies throughout the literature for vowel samples investigating dysarthric and ataxic voices (i.e. Ackermann, & Ziegler, 1991;Eigentler, Rhomberg, Nachbauer, Ritzer, Poewe, & Boesch, 2012; Folker, Murdoch, Rosen, Cahill, Delatycki, Corben, & Vogel, 2012; Kent, Vorperian, & Duffy, 1999; Qi & Hillman, 1997). Limited data is provided for vowels such as /o/ but there is evidence of contrast errors at a segmental level between high versus low vowels due to dysarthria (Blaney & Hewlett, 2007a). Therefore, a variety of vowels at differing tongue heights should be considered when analyzing sustained vowel phonations of individuals with dysarthria. The errors found in high versus low vowel contrasts can be analyzed as pattern profiles to be used for assessment and specific treatment goals for affected individuals (Blaney & Hewlett, 2007a).

Data Collection and Analysis

A variety of methods are found across the literature in terms of type of data collection and analytic systems utilized to measure acoustic parameters. Gelfer (1995) found that the existing data on phonatory stability has primarily been focused on adult males, the elderly, or children. Specifically for FA, most studies have included a participant sample with a large age range or with only one gender represented (Blaney & Hewlett, 2007; Bunton & Weismer, 2001; Eigentler et al., 2012; Folker et al., 2012). Maryn et al. (2009) described how different acoustic measures, such as pitch perturbation, can be affected by recording equipment, algorithms, sampling rate, and period extraction during data collection and analysis. Overall, acoustic analysis has been employed for many reasons such as differentiating normal and abnormal phonation, quantifying severity of dysphonia, providing objective information on speech mechanisms, and to measure a client's progression throughout treatment (Folker et al., 2012; Maryn et al., 2009; Wolfe & Steinfatt, 1987). Keller, Vigneux, and Laframnboise (1991) found acoustic analysis to be the least expensive and intrusive instrumental approach which provides detailed measures about disordered behavior.

Current analysis systems include the Dysphonia Severity Index, the Hoarseness Diagram, the Multidimensional Voice Program (MDVP; Kay Elemetrics, 1993) and the Acoustic Voice Quality Index (Kreiman & Gerratt, 2010). These algorithms do not provide an easy way to understand the changes in measures extracted because, as Kreiman and Gerratt (2010) stated, they are not linked to any unique vocal quality model. Most acoustic algorithms use time-based analysis requiring the algorithm to distinguish cycle boundaries. Acoustic analysis based on detection of the signal period can be affected by cycle-to-cycle variability and becomes unreliable when tracking cycles for jitter, shimmer, and harmonics-to-noise ratio from an aperiodic voice signal (Lowell, 2012). Cepstral-based measures are more reliable because they are not based on the detection of period boundaries within the acoustic signal or time-based signal components and have an exceptional discriminative capacity in differentiating dysphonic from normal voices (Lowell, 2012). Studies often report moderate reliability of acoustic findings, but cepstral analysis has proven to be accurate and reliable in determining acoustic measures for dysphonic voices (Lowell, 2012). The Computerized Speech Laboratory and SpeechTool analyze cepstral peaks but do not use linear regression analysis like the Analysis of Dysphonic Speech and Voice (ADSV) (KayPENTAX, 2011) in its spectral- and cepstral-based analysis. ADSV is the first commercial program using cepstral-based and linear regression analysis for voice quality assessment of sustained vowels and continuous speech samples in dysphonic and normal voices (KayPENTAX, 2011; Lowell, 2012). This newly developed software algorithm has been employed recently in studies analyzing continuous speech samples (Awan, Roy, & Dromey, 2009; Lowell, 2012) and sustained vowel phonation of dysphonic voices (Awan, Helou, Stojadinovic, & Solomon, 2011; Lowell, Kelly, Awan, Colton, & Chan, 2012; Solomon, Awan, Helou, & Stojadinovic, 2012) but has not been utilized with specific dysphonic voice disorders, such as ataxic dysarthria.

The use of the Cepstral/Spectral Index of Dysphonia (CSID) creates a strong correlation between acoustic measures and perceptually rated voice characteristics from voice quality assessments such as the Consensus Auditory Perceputal Evaluation of Voice (CAPE-V) (Lowell, 2012). This is relevant because perceptual ratings have traditionally been viewed as the “gold-standard” of rating voice disorder severity. To have a quality and reliable acoustic measure that is strongly correlated with perceptual measures is extremely valuable for the quantification of voice characteristics and for clinical purposes, such as determining treatment goals (Levee, 2011) and for tracking disease and intervention progression.

Purpose

The aim of the following study is to compare objective acoustic analysis measures of prolonged vowels from adolescents and young adults with FA to normal voices of young, age-equivalent and gender-matched college students in order to determine if significant differences exist between 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), the Cepstral/Spectral Index of Dysphonia (CSID), and Mean Cepstral Peak Prominence Fundamental Frequency (Mean CPP F0) calculated by ADSV.

Justification

Note that this proposed study differs from previously cited studies in several important aspects. Currently, no studies have been conducted with 20 FA participants in a relatively restricted age range. The age homogeneity of the sample is unparalleled in recent studies. Various results have been found for acoustic parameters in those with FA using time-based measures. To obtain more reliable data, a cepstral based analytic program (ADSV) was selected to compute acoustic measures.

Hypotheses

It was hypothesized that acoustic measures, specifically (1) CPP, (2) CPP SD, (3) L/H Ratio, (4) L/H Ratio SD, (5) CSID, and (6) Mean CPP F0, were expected to be significantly different in the dysphonic samples as compared to the NV samples. In FA participants, 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 (Awan, 2011); whereas no hypothesis was made for Mean CPP F0 changes in dysphonic voices, since research results on F0 have been contradictory.

CHAPTER 2: METHOD

Research Design

The following study was designed as a descriptive, prospective series design to identify any relationships between acoustic measures and FA that differ from the acoustic measures identified from the NV group. The acoustic measures obtained provide a combination of prospective and retrospective data on the distinct voice characteristics perceived in individuals with FA.

Variables

This study included 6 dependent variables for measurement of acoustic characteristics: (1) Cepstral Peak Prominence (CPP), (2) Cepstral Peak Prominence Standard Deviation (CPP SD), (3) Low/High Spectral Ratio (L/H Ratio), (4) Low/High Spectral Ratio Standard Deviation (L/H Ratio SD), (5) the Cepstral/Spectral Index of Dysphonia (CSID), and (6) the Mean Cepstral Peak Prominence Fundamental Frequency (Mean CPP F0); quantified from the vocal samples of the FA and NV group. The independent variable was defined as the presence of FA.

Participants

Twenty adolescents and young adults who were diagnosed with FA from France and twenty college students with normal voices 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 that was supported by a pharmaceutical company and the Institut National de la Santé et de la Recherche Médicale (INSERM) (the equivalent of France's National Institute of Health). The pharmaceutical company and health institute recruited patients for this medical study in order to confirm that treatment was only being provided for individuals who had FA. All 20 of these individuals were native French speakers, with a mean age of 18.5 years, and age range of 10-25 years; there were 10 males and 10 females (See Table 1). In order to provide the most age comparable sample possible, 20 age-equivalent and gender-matched college students were recruited, through various methods, including word of mouth, flyers in the University of Central Florida's (UCF) Communication Sciences and Disorders (CSD) Department, direct appeal to students enrolled in CSD undergraduate and graduate classrooms, and through online social networks from UCF. Being the second largest public university in the United States, UCF provided a large selection of individuals who met the criteria for recruitment.

Inclusion criteria 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 placed in a group called normal voice (NV).

Table 1

Participant Characteristics

	Friedreich's Ataxia		Normal Voice	
	Gender	Age	Gender	Age
1	Male	10	Male	18
2	Male	13	Male	18
3	Male	16	Male	18
4	Male	16	Male	19
5	Male	16	Male	20
6	Male	17	Male	20
7	Male	18	Male	21
8	Male	19	Male	21
9	Male	20	Male	21
10	Male	21	Male	23
11	Female	16	Female	18
12	Female	17	Female	19
13	Female	17	Female	19
14	Female	19	Female	20
15	Female	20	Female	21
16	Female	21	Female	22
17	Female	22	Female	23
18	Female	23	Female	24
19	Female	24	Female	24
20	Female	25	Female	25

Institutional Review Board (IRB)

Prior to enrolling in the study, each participant read an informed consent document approved by the IRB at UCF (Appendix A). The IRB recommended that, since the study presents no risks, signatures on the consent forms were not required but that copies of the form should be available for the participants to keep. Researchers accepted a verbal consent in order to proceed with the collection of data.

Recording

Instrumentation and data collection conducted in this study were comparable to those used in France. Participants in France were recruited and recorded in accord with existing policies regarding human subject participation in that country. Specifically, in France, individuals with FA were recorded on a Marantz digital audio tape-recorder (DAT), set at 22.5kHz sampling rate in a quiet office. Recordings of stimuli were always conducted in the same sequence; namely, each participant sustained the vowels /a/, /i/, and /o/ at their normal fundamental frequency for the longest duration possible. Age-equivalent and gender-matched NV participants from the United States provided voice samples by sustaining the vowels /a/, /i/, and /o/ at their normal fundamental frequency for as long as they could, on a single breath, at a comfortable vocal loudness level. The vowels /a/ /i/, and /o/ were selected for analysis because it has been found that tongue height during vowel production may be related to the severity of a vocal disorder (Wolfe & Steinfatt, 1987). Correlations between tongue height and vocal tract configuration, spectral noise levels of vowels and vowel roughness have been found in the literature (Sansone & Emanuel, 1970). In the U.S., sustained vowel voice samples of the NV participants were recorded on a Roland Edirol digital recorder (R-09HR), set at a 44kHz sampling rate. Samples were then down-sampled to a rate of 22.5kHz, to match the FA samples, for further acoustic analysis. The mouth-to-microphone distance during recording was 12 inches. Recording took place in UCF's CSD conference room; the equivalent of the quiet office used in France for the recording of FA samples. Between the FA and NV groups, 120 vocal samples were collected for the vowels /a/, /i/, and /o/ at a normal fundamental frequency.

Measures

The following acoustic measures were employed: CPP, CPP SD, L/H Ratio, L/H Ratio SD, CSID, and Mean CPP F0. These parameters were computed for the initial 2 seconds of each vowel sample at the normal pitch levels. Due to reports of differences in vowel duration, voicing in the closure phase, F1 frequency at mid-vowel, and a drop in F1 at the end of a vowel causing listeners' misperceptions of FA voice samples, the mid-portion and the terminal portion of vowel samples were not selected for acoustic analysis (Blaney & Hewlett, 2007b). FA significantly impacts the strength of the inspiratory muscles and therefore decreases lung volume and subglottal air pressure (Folker, Murdoch, Cahill, Delatycki, Corben, & Vogel, 2010), causing a reduction in phonation duration (Koeppen, 2011). For these reasons, the initial 2 seconds of each vowel sample was selected to represent the most stable vowel segment and to provide an accurate representation of acoustic measures for comparison of FA and NV participants.

Reliability

To determine inter-rater reliability, 10% of the 120 FA and NV vocal samples (N=12) were transferred from the Edirol recorder to the Dell desktop, cropped from the first recognizable waveform to the initial 2 seconds and then down-sampled to a 22.5kHz sampling rate through MultiSpeech, and entered into the ADSV program for re-analysis. Due to the systematic and consistent manner of segmentation and down-sampling samples in Multispeech and performing acoustic analysis through ADSV, a 100% reliability resulted between the original voice samples and the repeated samples.

CHAPTER 3: RESULTS

The Analysis of Dysphonia in Speech and Voice software program (ADSV) was utilized to provide acoustic data of the chosen parameters for the 120 FA and NV voice samples through programmed formulas and cepstral analysis. Recorded samples on an Edirol were transferred to a Dell desktop, segmented from the first recognizable waveform to the initial 2 seconds and then down-sampled to a 22.5kHz sampling rate on MultiSpeech, and entered into the ADSV program for acoustic analysis. Obtained values were then entered into the SPSS Statistics program for statistical analysis. Six measures were analyzed per participant (N = 40) and included Cepstral Peak Prominence dB (CPP), Cepstral Peak Prominence Standard Deviation dB (CPP SD), Low/High Spectral Ratio dB (L/H Ratio), Low/High Spectral Ratio Standard Deviation dB (L/H Ratio SD), the Cepstral/Spectral Index of Dysphonia (CSID), and Mean Cepstral Peak Prominence Fundamental Frequency Hz (Mean CPP F0). A total of 792 acoustic measures were computed (3 vowels x 40 paricipants x 6 measures + 10% reliability). Data for each participant can be found in Appendix B. These measures were selected to identify acoustic correlates of the FA and NV voice samples.

Descriptive Statistics

Descriptive statistics for five of the dependent variables (CPP, CPP SD, L/H Ratio, L/H Ratio SD, and CSID) combined data for all vowels (/a/, /i/, and /o/) and included means and standard deviations (Table 2). The mean score for CPP-all for the FA participants was lower than the NV group. This is indicative of a decrease in amplitude of the cepstral peak found in dysphonic voice signals with disturbed periodicity (Awan, 2011). Higher scores were found for L/H Ratio-all, CSID-all, CPP SD and L/H Ratio SD scores for the FA participants compared to the NV group. A higher L/H Ratio-all average for FA contradicted what was expected. A higher L/H Ratio is typical of NV voices due to a greater amount of low-frequency energy than high-frequency energy in the signal, whereas the opposite is expected in dysphonic voices (Awan, 2011). This finding suggests a difference between the effects of ataxic dysarthria and dysphonia on vocal production and is further addressed in the Discussion section. The larger CSID-all score reflected the variability, inconsistency, and instability of the dysphonic vocal quality in the FA group through a multivariate approach. More variability and inconsistency would be expected in dysphonic voices, and the standard deviation values yielded for CPP (SD) and L/H Ratio (SD) lend support to this association. Awan (2011) stated that severely dysphonic voices demonstrate constant "noise", which is reflected in low L/H Ratio SD scores. Since the average L/H Ratio SD was higher in the FA group than the NV group in the current study, the majority of FA voices may have not been severely dysphonic; however, they did demonstrate a larger variability of high frequency noise in the acoustic signal (i.e., vocal unsteadiness) than NV samples indicating dysphonia. Thus, hypotheses for all measures, except L/H Ratio-all, were verified.

Inferential Statistics

Inferential statistics of the five dependent variables were employed to determine significant differences across variables. Analysis of variance (ANOVA) was selected to test for significant differences; results are summarized in (Table 2). Overall, all measures (except CPP SD) were significantly different between the groups. Findings from the measures CPP all, L/H Ratio SD, and CSID-all indicated that those in the FA group had voices that were more dysphonic/less normal than those in the NV group. Those individuals with FA had voices that had significantly higher L/H Ratio values as compared to NV peers, which signified that FA voices contained greater low frequency noise as compared to high frequencies than did voices in the NV group. As indicated earlier, this finding was unexpected as dysphonic voices usually contain a greater amount of noise that competes with and partially obscures low frequency energy. It was felt, however, that those in the NV group may not have had excessively "noisy" voices. It has been reported that low L/H Ratios are a prominent predictor of breathy voices (e.g. Awan & Roy, 2006, 2009). No significant differences were found between the experimental groups on CPP SD values.

Table 2

ANOVA 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).

Measures	FA	NV	df	*F* value
CPP-all (dB)	8.43 (1.87)	10.0 (1.61)	1, 38	8.13**
CPP SD (dB)	.98 (.25)	.79 (.36)	1, 38	3.60
L/H Ratio-all (dB)	31.95 (5.95)	28.25 (4.38)	1, 38	5.03*
L/H Ratio SD (dB)	2.49 (.50)	1.72 (.26)	1, 38	37.66**
CSID-all	37.57 (12.03)	26.73 (8.70)	1, 38	10.67**

Note 1. CPP-all= Cepstral Peak Prominence all vowels; CPP SD= Cepstral Peak Prominence Standard Deviation; L/H Ratio-all= Low/High Spectral Ratio all vowels; L/H Ratio SD= Low/High Spectral Ratio Standard Deviation; CSID-all= The Cepstral/Spectral Index of Dysphonia all vowels; df= degrees of freedom. Values are expressed as means and (standard deviations). All measures, except CSID-all, are expressed in decibels (dB). CSID-all has no unit of measurement because it is derived from a combination of other measures (CPP, CPP SD, etc.)

Note 2. A lower mean score on CPP-all, L/H Ratio-all, and CSID-all are suggestive of dysphonic vocal quality. A higher L/H Ratio-all is associated with normal voice signals. A higher CPP SD and L/H Ratio SD score indicates less phonatory stability on the measures.
Note 3. $^*p \leq .05$; $^{**}p \leq .001$, 2-tailed.

Descriptive Statistics – Mean CPP F0

For Mean CPP F0, descriptive statistics were employed to analyze each vowel separately (Table 3). As expected, NV samples demonstrated higher cepstral peaks across all vowels, as reported in the descriptive statistics, whereas FA samples demonstrated lower cepstral peaks.

Table 3

Comparison of Fundamental Frequency (F0) of Adolescents and Young Adults with Friedreich's Ataxia (FA) and Normal Voices (NV) from the Analysis of Dysphonia in Speech and Voice (ADSV).

Measures	FA	NV
Mean CPP F0a (Hz)	168 (48)	176 (63)
Mean CPP F0i (Hz)	179 (57)	192 (69)
Mean CPP F0o (Hz)	174 (57)	181 (66)

Note 1. Mean CPP F0a= mean Cepstral Peak Prominence Fundamental Frequency vowel /a/; Mean CPP F0i= mean Cepstral Peak Prominence Fundamental Frequency vowel /i/; mean Cepstral Peak Prominence Fundamental Frequency vowel /o/. Values expressed as means and (standard deviations). All measures are expressed in Hertz (Hz).

Note 2. Lower scores on all measures indicate dysphonic vocal quality.

Correlations

Pearson correlations were employed to determine significant correlations between the five dependent variables (Table 4). A negative correlation was found for CPP-all and L/H Ratio SD which indicated that as cepstral peaks increased, the variance in spectral energy decreased for normal voices. For dysphonic voices, the opposite would be expected to occur; namely, cepstral peak would decrease as the variance in spectral energy increased. A negative correlation was also found between CPP-all and CSID which signified that the lower the cepstral peak, the more severe the dysphonic voice. CPP SD and L/H Ratio SD had a positive correlation and was illustrated as higher variances in cepstral and spectral energies in dysphonic voices and lower variability in normal voice samples. A positive correlation occurred between CPP SD and CSID; indicating that the more variability in cepstral energy present in a voice sample, the greater dysphonia severity. A positive correlation was also demonstrated between LH Ratio SD and CSID which indicated that as variance in spectral energy increased, dysphonia severity increased. The direction of these correlations reflected what was expected of dysphonic and normal voice samples.

Table 4

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

	CPP-all	CPP SD	L/H Ratio-all	L/H Ratio SD	CSID-all
CPP-all (dB)					
CPP SD (dB)	.02				
L/H Ratio-all (dB)	.18	.26			
L/H Ratio SD (dB)	-.32*	.40*	.19		
CSID-all	-.70**	54**	-.18	.68**	

<u>Note 1.</u> CPP-all= Cepstral Peak Prominence all vowels; CPP SD= Cepstral Peak Prominence Standard Deviation; L/H Ratio-all= Low/High Spectral Ratio all vowels; L/H Ratio SD= Low/High Spectral Ratio Standard Deviation; CSID-all= The Cepstral/Spectral Index of Dysphonia all vowels. All measures, except CSID-all, are expressed in decibels (dB). CSID-all has no unit of measurement because it is derived from the other measures (CPP, CPP SD, etc.)

<u>Note 2.</u> * Correlation is significant at the 0.05 level, 2-tailed; ** Correlation is significant at the 0.01 level, 2-tailed.

CHAPTER 4: DISCUSSION

The current study primarily focused on quantifying spectral/cepstral acoustic measures of sustained vowels produced by adolescents and young adults with FA to age-equivalent and gender-matched unimpaired college students and compared the findings to determine significant differences between the dependent variables. The majority of the past literature has focused on traditional, time-based acoustic features such as duration, fundamental frequency (F0), jitter, shimmer, and harmonics-to-noise ratio (HNR). Although these measures provide adequate and valuable information about vocal quality, the data is univariate and does not reflect the complex interactions of subsystems required for functional voice production. Multivariate approaches, such as spectral/cepstral analysis, provide comprehensive information about vocal quality and represent the performance of subsystems by including measures of vocal stability and consistency throughout a voice sample (Awan, 2011). Due to the various subsystems affected in FA and the progressive nature of the disease, a multivariate approach is more appropriate to analyze the interrelated aspects of changes in the acoustic signal and impacted subsystems. The most debilitating aspect of FA is that it is a progressive disease and that voluntary control of fine-motor skills is disrupted. To accurately track disease progression and identify specific breakdowns in voice production in order to provide the most effective treatment possible for an individual with FA, a multivariate acoustic analysis approach is recommended. Appropriate and efficient treatment is essential for this rare and poorly understood disease in order to optimize the affected individual’s communication abilities, occupational success, and social interactions.

Before discussing the findings of the current study, it is important to highlight the differences between normal voice production and the production of individuals with dysphonia versus ataxic dysarthria. The current study utilized the ADSV program which is based on the analysis of dysphonic voices. Understanding the differences between these two disorders and normal voice production is an important foundation for interpreting the results of this study.

Ataxic Dysarthria vs. Dysphonia

A typical individual has the ability to adequately coordinate the subsystems required for voice production with the right timing, energy, and execution. When damage occurs to areas of the brain that control these subsystems, such as the cerebellum, breakdowns occur in the ability to produce voice in a normal manner. Depending on the location and extent of the affected brain area, subsystems such as respiration and control of motor movements required for speech may be impacted.

Ataxic dysarthria is caused from specific damage to the cerebellum and/or cerebellar control circuits and can be classified as a motor speech disorder (Ruddy, 2013). The cerebellar area of the brain is responsible for motor planning and coordination of the timing and force of muscle contractions required for skilled and voluntary movements (Ruddy, 2013). Damage to the cerebellum affects the coordination of the entire body and bodily movements but, more specifically to speech, as it disrupts articulation, voice, and respiration. The coordination of breathing with speech is affected causing reduced duration, increased speech rate, decreased volume and pitch (Legge, 2010; Ruddy, 2013). Articulation and voice symptoms can be grouped into speech clusters. Those with ataxic dysarthria are affected by reduced oral muscular tone and may produce imprecise consonants; prolonged phonemes; irregular articulatory breakdowns (resulting in what is often perceived as “drunken speech”); and vowel distortions (Legge, 2010; Ruddy, 2013). Voice quality is affected by excess and equal stress; prolonged phonemes and intervals; slow rate; harshness; monopitch; and monoloudness (Ruddy, 2013). Treatment for ataxic dysarthria may include exercise to strengthen muscles for improved articulation and/or breathe support for better volume control; articulation drills; and stress and pitch control activities (Legge, 2010).

Dysphonia refers to a voice disorder that impairs the speaking or singing voice (American Speech-Language-Hearing Association, 2005). The etiology of dysphonia is typically multifactorial and usually stems from an abnormality of the structures and/or functions of voice production (American Speech-Language-Hearing Association, 2005). The variables that can cause dysphonia include genetic factors and acute or chronic situations, such as occupational vocal demands, medications, environmental factors, physical trauma, lifestyle choices, and other health issues (American Speech-Language-Hearing Association, 2005). Examples of dysphonia/voice disorders are benign vocal fold lesions (e.g. nodules), unilateral or bilateral vocal fold paralysis, muscle tension dysphonia, and paradoxical vocal fold dysfunction (American Speech-Language-Hearing Association, 2005). Treatment for dysphonia includes voice therapy and/or medical management conducted by a Speech Language Pathologist and/or Otolaryngologist (American Speech-Language-Hearing Association, 2005).

Acoustic Measures

Cepstral measures were quantified for the Cepstral Peak Prominence (CPP) and CPP Standard Deviation (CPP SD) for each vowel in both the FA group and NV group. CPP for all vowels was significantly lower in the FA group as compared to the NV group. This result is in agreement with findings of previous studies that also found a significantly lower CPP value for dysphonic voices (Awan, Helou, Stojadinovic, & Solomon, 2010) as compared to normal voice samples (Awan, Roy, Jette, Meltzner, & Hillman, 2010; Lowell, Kelley, Awan, Colton, & Chan, 2012). The literature also supports a negative correlation between dysphonic severity and CPP, indicating that as the severity of dysphonic voices increased, the CPP value decreased (Awan, Roy, Jette, Meltzner, & Hillman, 2010). This finding in the current study supports the notion that normal voices typically reflect a more prominent cepstral peak, indicated by a distinct fundamental frequency and related harmonics in a periodic signal, whereas an aperiodic signal demonstrates a decrease in amplitude of the cepstral peak (Awan, 2011). According to the literature and the significant difference in CPP values between the FA and NV group, it can be concluded that as disordered voice increases in severity, the cepstral peak of a voice sample will decrease in amplitude. Therefore, CPP can be applied to distinguish between normal and disordered voices, determine the severity of the voice disorder, and track disease progression as it affects voice production.

Although the amplitude of the cepstral peak was found to be significantly different between the FA and NV groups, CPP SD was not found to be statistically significant in the current study. The NV group resulted in a lower CPP SD value and a higher CPP SD was found in the FA group. This finding is consistent with previous studies (Lowell, Kelley, Awan, Colton, & Chan, 2012), indicating that normal voices tend to reflect stability whereas disordered voices display variability across time (Awan, 2011). Awan, Roy, Jette, Meltzner, and Hillman (2010) state that CPP SD combined with other cepstral and spectral measures can strengthen the predictions of voice disorder severity. An interesting finding is that as a disordered voice becomes more severe, the varying stability and unsteadiness of the signal typically becomes more consistent, therefore, reflecting a lower CPP SD (Awan, Roy, Jette, Meltzner, & Hillman, 2010). The affected coordination of the timing and force of muscle contractions required for skilled and voluntary movements in severe FA may cause a reduction in pitch and therefore may result in a lower CPP SD due to lack of variation in fundamental frequency. This finding leads to the speculation that, although there was not a significant difference in CPP SD between the two groups of the current study, the FA participants as a whole were not as severe as they could possibly be since their CPP SD value was higher than the NV group, indicating consistent instability across the vowel duration.

A significant difference between L/H Ratio was found and was higher in the FA group than the NV group. This finding contradicted what was expected. Awan (2011) found that dysphonic voices tend to reflect a low L/H Ratio due the increased high-frequency spectral noise volume, where as normal voices reflect a majority of spectrum energy in the F0 resulting in a high L/H Ratio. The increased amount of high-frequency spectral noise in dysphonic voices is typically perceived as breathiness (Awan, 2011). The current study differs from Awan's study (2011) in that the disordered voices were of those with ataxic dysarthria and not dysphonia. As previously discussed, there are key differences that characterize ataxic versus dysphonic voices. Therefore, the ataxic voice samples did not contain an increased amount of high-frequency spectral noise and may not be as breathy as the voice samples studied by Awan (2011). Ataxic dysarthria is known to effect the coordination of respiratory subsystems with voice production over the duration of phonation creating tension on the vocal cords and subsequent hyperfunction. These physiological changes may have been reflected in the unexpected finding of a higher L/H Ratio in the FA samples. The current study's finding is parallel to the study by Lowell, Kelley, Awan, Colton, and Chan (2012) that also found greater variability in the L/H Ratio of disordered voices as compared to normal voice samples. L/H Ratio, when combined with other cepstral and spectral measures, has the potential to determine severity of vocal function as well as distinguish between abnormal and normal voices.

L/H Ratio SD represents the variability in spectral measures over the duration of a voice sample. Normal voice productions typically remain stable and consistent and would result in a low L/H Ratio, whereas disordered voices tend to contain variability and inconsistencies and therefore a higher L/H Ratio value (Awan, 2011). These expectations were verified in the current study's findings as L/H Ratio SD was lower in normal voices and higher in FA voice signals. This significant difference is parallel with findings in the literature (Awan, Roy, Jette, Meltzner, & Hillman, 2010; Lowell, Kelley, Awan, Colton, & Chan, 2012), except for Awan, Helou, Stojadinovic, and Solomon (2010) who examined cepstral and spectral measures overtime with individuals pre- and post-thyroidectomy. The more severe a disordered voice becomes, the more consistent the variability and instability becomes, therefore resulting in a lower L/H Ratio SD (Awan, Roy, Jette, Meltzner, & Hillman, 2010). The same is true of the cepstral measure, CPP

SD, which also evaluates the variability of a voice signal. This finding leads to the conclusion that the FA participants were not as severe as they could possibly be since the low- and high-frequency ratio SD value was higher than the NV group, indicating inconsistent stability across the vowel duration.

The current study found a significant difference in the CSID value for all vowels between the FA and NV participants. Lowell, Kelley, Awan, Colton, and Chan (2012) also reported a significantly higher CSID value for disordered participants as compared to those with normal voices. This cepstral/spectral measure combines CPP, CPP SD, L/H Ratio, and L/H Ratio measures to produce a multivariate estimate of severity (Awan, 2011). CSID is a unique measurement that reflects the multidimensional aspect of the coordination, execution, and control of aerodynamic aspects, frequency and intensity required for a functional vocal quality (Awan, 2011). Similar to auditory-perceptual dysphonia scales, CSID is based on a scale to provide a summary index of the abnormal vocal quality (Lowell, Kelley, Awan, Colton, & Chan, 2012). This multivariate measure provides insight needed for the proper identification of an abnormal vs. normal voice (Awan, 2011). Based on past and current findings, CSID provides a state-of-the-art representation of vocal quality, based on the multidimensional process required for voice production, in order to accurately determine disordered versus normal voice production.

Descriptive statistics revealed differences between FA participants and NV participants in Mean CPP F0 for all vowels. A higher value was found in the NV group and a lower value in the FA group indicating a reduction in the cepstral peak and fundamental frequency. Due to the effect ataxic dysarthria has on the coordination of subsystems required for voice production (i.e.

respiration and phonation) it can be concluded that a lower Mean CPP F0 value is expected in the disordered voices of individuals with FA.

Summary of Findings

Prior to the current investigation, it was observed that individuals with FA demonstrate perceptually distinguishable differences when compared to individuals with normal voice. Through the utilization of spectral/cepstral analysis, this study found significant differences in spectral/cepstral measures (CPP, L/H Ratio, L/H Ratio SD, and CSID) for all sustained vowels in those with FA. These significant findings are indicative of abnormalities in vocal quality due to the affects of ataxic dysarthria and FA on subsystems required for adequate and functional vocal production. Findings from this study have valuable implications for the evaluation and treatment of individuals with FA and ataxic dysarthria. Spectral/cepstral acoustic findings can be employed to determine if a voice differs from normal and the severity of the abnormal vocal production. If abnormalities are detected and characterize a voice as disordered with a certain level of severity, treatment can be tailored to address specific components that are affected (i.e. vocal stability, amplitude of the cepstral peak/fundamental frequency, coordination of subsystems). Identifying specific vocal components that are affected through spectral/cepstral analysis and defining the severity of the disorder can lead the clinician to begin with the most effective treatment strategy and potentially restore a functional voice production in a shorter amount of time than with the use of time-based measures and/or perceptual analysis alone. Improvements in vocal production would lead to a better quality of life for individuals with FA.

The current study was an initial investigation to acoustically quantify the perceived abnormalities of voice production in individuals with FA through the use of the one-of-a-kind spectral/cepstral analysis of ADSV in order to support the implication of further clinical evaluation and treatment of vocal abnormalities in this population.

Limitations of the Study

A limitation of the current study is the young ages of two of the FA participants (i.e. 10 and 13 year old). Both of the adolescents were males. The age and gender characteristics of these two participants were of concern due to the possible effect of puberty on vocal quality. Although these individuals were as many as 8 years younger than the mean age of FA participants and were adolescent males, their cepstral/spectral measures did not differ greatly from those in the disordered group. Since FA is relatively rare, new FA participants could not be recruited in the time frame of this study. In order to maintain a large sample size of FA and NV participants which has not been conducted in previous research, the younger participants were retained in the current study.

Previous studies on the use of ADSV to quantify spectral/cepstral measures in the vocal samples of individuals with FA do not exist. Due to the recent release of the ADSV program, there is a limited amount of research that includes ADSV as the program for acoustic analysis.

The few studies that include ADSV spectral/cepstral measures are focused on dysphonic voices and not on dysarthric or ataxic voice samples. Comparisons of the results of the current study to past research were limited to studies on dysphonic voices that often compared sustained vowel samples and connected speech segments.

The specificity of the current study that included only acoustic analysis and spectral/cepstral measures on sustained vowel samples is also a limitation of the research. Most of the past literature compares prolonged vowels and connected speech and includes some type of perceptual analysis. Additional temporal acoustic measures, such as duration, voice onset time, and jitter, could also be utilized to analyze connected speech and provide more comprehensive data on the affects of ataxic dysarthria on voice production. The researchers of the current study specifically employed spectral/cepstral acoustic analysis for sustained vowels exclusively and did not include connected speech in order to reduce variables that could affect the methods of the study and results (i.e. participant fatigue). Perceptual analyses of the prolonged vowel samples were included in the current study, but methods and results will be reported in a separate study. Significant findings were quantified for spectral/cepstral measures of the sustained vowel samples and therefore were the focus of the current study.

In order to more closely reflect the methods of data collection for the recorded FA samples, the NV samples were recorded in UCF’s Communication Sciences and Disorders conference room and not in a sound treated room/booth. A null hypothesis was not established regarding the effects of uncontrollable noise in the conference room or in adjacent hallways on the acoustic analysis of voice samples. The effect of environmental noise on FA samples was unable to be determined since the researchers for the current study were not present during data collection in France.

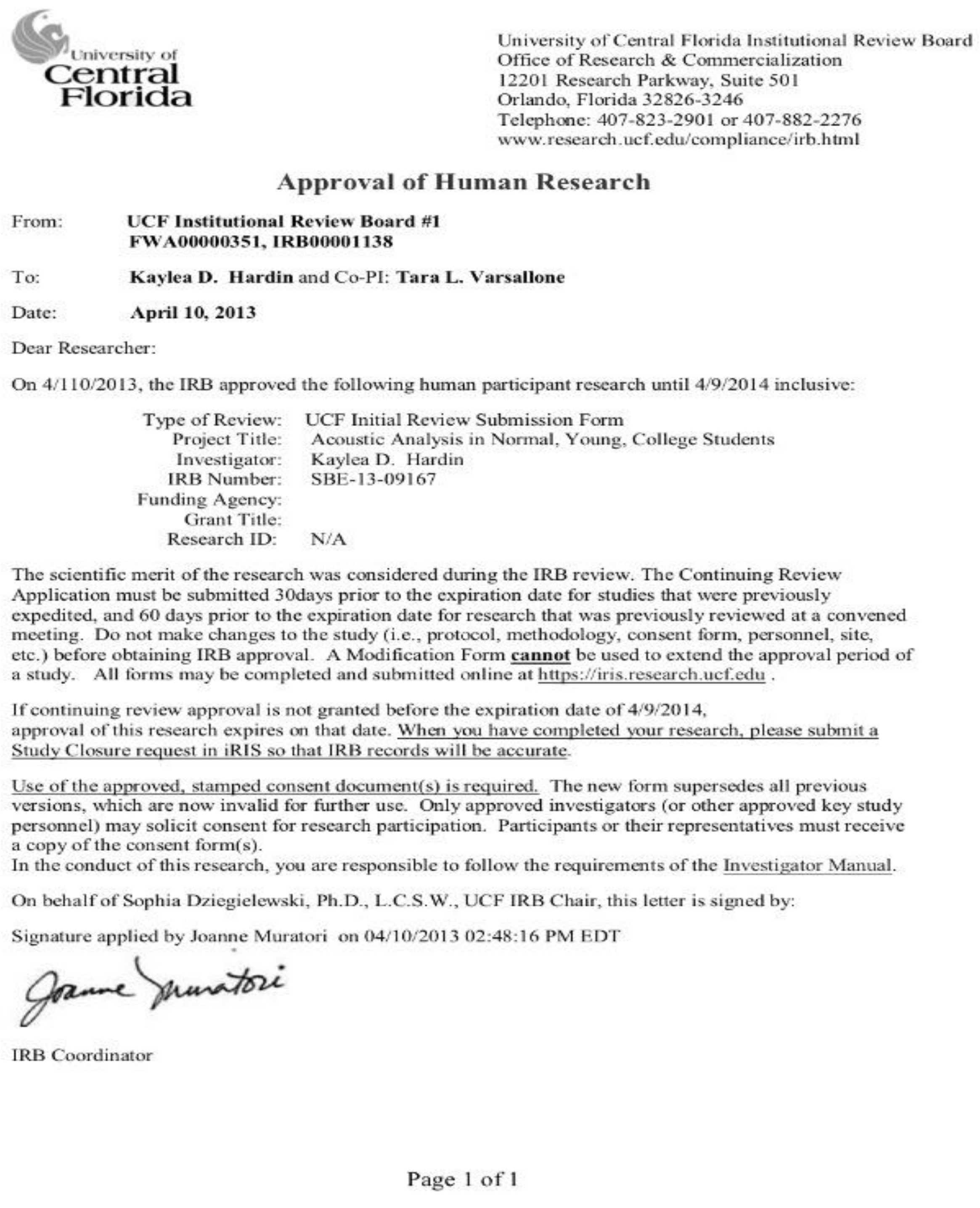
Future Studies

Further investigations into the acoustic features of individuals with severe ataxic dysarthria, with and without FA, using both spectral and cepstral analyses should be conducted. This would provide more information about the affects of ataxic dysarthria on voice production and provide stronger evidence for the use of spectral and cepstral analysis for tracking disease progression, baseline measures, intervention progress, and distinguishing abnormal and normal voice production. For a more comprehensive evaluation of vocal quality, further research should include correlations with auditory-perceptual measures, such as the Consensus Auditory Perceputal Evaluation of Voice (CAPE-V). Combining acoustic and perceptual measures for quantifying vocal characteristics is useful for determining treatment goals and tracking progress (Levee, 2011). Although sustained vowels are not typically affected by aspects that impact connected speech (i.e. speech rate, phonetic context, emphasis, etc.), future studies should consider comparing vowel prolongations to connected speech samples. This combination of sustained vowels and continuous speech could provide further information on an individual's functional communication and voice production abilities. More significant differences may be found in spectral/cepstral measures, such as CPP SD and L/H Ratio SD, in the context of consonant-vowel transitions (Awan, Helou, Stojadinovic, & Solomon, 2010). These suggestions contribute to the evidence needed to provide the most efficient and effective treatment to improve the quality of life for individuals with FA and ataxic dysarthria.

Conclusions

This study is the first to determine spectral and cepstral acoustic differences of sustained vowels between individuals with FA to age-equivalent, gender-matched individuals with normal voices. Significant differences in spectral/cepstral acoustic measures between the two participant groups, supports ADSV as a valid program for evaluating vocal quality of individuals with FA or ataxic dysarthria. The objective measures available through this program demonstrated the ability to distinguish disordered and normal voices and may reflect the severity of abnormal vocal quality. Spectral/cepstral acoustic measures and multivariate approaches, as applied in the current study, have the potential to indicate ataxic dysarthria and track disease progression, through a non-invasive technique. It is hoped that these results stimulate further research about affected voice production in FA and ataxic dysarthria. With more research, improvements in the evaluation and intervention techniques may ultimately lead to the alleviation of the symptoms of this distressing disease and improve quality of life and functional communication abilities.

APPENDIX A:
APPROVAL OF HUMAN RESEARCH



University of
Central
Florida

University of Central Florida Institutional Review Board
Office of Research & Commercialization
12201 Research Parkway, Suite 501
Orlando, Florida 32826-3246
Telephone: 407-823-2901 or 407-882-2276
www.research.ucf.edu/compliance/irb.html

Approval of Human Research

From: **UCF Institutional Review Board #1**
FWA00000351, IRB00001138

To: **Kaylea D. Hardin** and Co-PI: **Tara L. Varsallone**

Date: **April 10, 2013**

Dear Researcher:

On 4/110/2013, the IRB approved the following human participant research until 4/9/2014 inclusive:

Type of Review:	UCF Initial Review Submission Form
Project Title:	Acoustic Analysis in Normal, Young, College Students
Investigator:	Kaylea D. Hardin
IRB Number:	SBE-13-09167
Funding Agency:	
Grant Title:	
Research ID:	N/A

The scientific merit of the research was considered during the IRB review. The Continuing Review Application must be submitted 30days prior to the expiration date for studies that were previously expedited, 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, site, etc.) before obtaining IRB approval. A Modification Form **cannot** be used to extend the approval period of a study. All forms may be completed and submitted online at https://iris.research.ucf.edu .

If continuing review approval is not granted before the expiration date of 4/9/2014, approval of this research expires on that date. When you have completed your research, please submit a Study Closure request in iRIS so that IRB records will be accurate.

Use of the approved, stamped consent document(s) is required. The new form supersedes all previous versions, which are now invalid for further use. Only approved investigators (or other approved key study personnel) may solicit consent for research participation. Participants or their representatives must receive a copy of the consent form(s).
In the conduct of this research, you are responsible to follow the requirements of the Investigator Manual.

On behalf of Sophia Dziegielewski, Ph.D., L.C.S.W., UCF IRB Chair, this letter is signed by:

Signature applied by Joanne Muratori on 04/10/2013 02:48:16 PM EDT

Joanne Muratori

IRB Coordinator

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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 (UCF IRB). 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

APPENDIX C:
RAW ACOUSTIC DATA

Participant	Age	CPP-a	CPP Std Dev-a	L/H Spectral Ratio-a	L/H Spectral Ratio Std Dev -a	Mean CPP F0 -a	CSID-(Gender) -a	CPP- i	CPP Std Dev-i	L/H Spectral Ratio-i	L/H Spectral Ratio Std Dev -i	Mean CPP F0 -i	CSID-(Gender) -i	CPP- o	CPP Std Dev-o	L/H Spectral Ratio-o	L/H Spectral Ratio Std Dev -o	Mean CPP F0 -o	CSID-(Gender) -o
fa06F	17	6.9	0.613	29.149	2.226	188.713	35.995	3.994	0.666	33.156	1.915	217.084	42.78	6.296	0.702	38.063	1.895	191.099	27.732
fa06F-2	17	6.9	0.613	29.149	2.226	188.713	35.995												
fa07M	16	9.671	1.404	36.203	2.096	119.122	34.498	5.176	1.231	39.766	4.618	125.81	67.886	8.29	0.912	42.988	3.007	119.99	35.153
fa07M-2	16													8.29	0.912	42.988	3.007	119.99	35.153
fa08F	22	11.128	0.846	20.839	1.87	207.313	25.884	8.354	0.809	12.445	1.665	207.307	44.954	8.022	0.862	31.479	3.265	208.894	39.175
fa09M	13	8.767	0.615	26.005	1.584	199.674	36.911	2.918	1.228	26.9	4.751	187.738	92.318	4.403	1.304	37.738	2.097	261.524	55.007
fa10F	23	9.297	0.73	27.383	3.158	207.491	35.643	4.631	0.911	28.555	1.698	215.465	45.758	5.58	0.875	35.389	2.238	195.853	38.127
fa11F	19	11.397	0.945	26.377	3.619	194.237	33.251	8.729	0.77	23.651	2.115	195.529	34.548	9.136	0.703	34.706	1.449	174.789	15.375
fa12F	25	7.082	0.974	30.796	2.623	228.802	40.338	4.152	0.411	36.848	1.83	256.956	34.851	6.956	0.751	41.092	1.546	233.66	19.506
fa13F	21	9.076	1.069	20.961	2.221	223.314	39.827	6.537	0.62	17.771	2.727	272.858	53.434	7.107	0.864	29.015	2.889	268.082	42.943
fa17M	11	12.787	0.867	36.32	1.633	112.733	11.438	3.99	1.196	30.513	2.216	105.786	64.171	7.797	1.435	37.378	3.707	103.761	54.099
fa18F	20	9.007	0.772	27.342	2.284	193.061	30.755	5.167	0.653	18.123	2.231	195.386	55.666	6.84	0.887	31.786	2.503	192.094	38.518
fa19F	24	8.3	2.052	23.135	1.863	230.937	48.469	6.584	0.974	19.624	3.633	261.51	61.942	6.68	0.509	32.672	2.784	250.216	36.41
fa20M	20	10.75	1.438	34.562	2.577	123.178	35.491	8.133	1.072	30.89	3.43	125.064	53.473	9.752	0.975	40.099	2.138	119.593	25.803
fa20M-2	20													9.752	0.975	40.099	2.138	119.593	25.803
fa22M	19	10.479	1.393	37.796	2.55	156.87	32.612	6.671	1.647	33.949	2.881	146.579	58.617	9.317	0.92	46.486	2.175	146.391	20.72
fa23M	16	9.912	1.277	31.763	2.702	146.742	41.354	5.15	1.073	27.363	4.965	140.179	81.985	9.794	1.048	40.272	2.893	143.72	31.968
fa23M-2	16													9.734	1.048	40.272	2.893	143.72	31.968
fa25F	24	11.867	0.635	42.041	1.931	185.751	-1.403	9.971	0.6	23.067	2.361	221.155	29.767	9.273	0.576	46.286	1.884	198.153	4.568
fa27M	16	12.52	0.995	37.823	1.305	101.526	9.89	9.074	0.717	26.681	1.523	112.719	35.464	10.15	0.985	44.056	1.522	109.3	15.309

fa29M	17	11.865	0.72	35.65	1.99	104.996	17.356	6.373	0.965	40.266	2.813	107.34	45.536	10.43	0.732	47.163	2.352	103.646	14.444
fa29M-2	17	11.865	0.72	35.65	1.99	104.996	17.356												
fa30M	18	11.789	1.486	34.079	1.525	110.709	23.934	8.782	2.06	27.695	3.222	117.174	62.874	6.795	1.269	36.019	1.287	110.892	39.755
fa33M		13.865	1.668	31.304	2.114	108.644	24.264	12.17	0.535	30.628	2.054	117.806	19.796	13.06	0.728	38.986	3.253	113.93	18.302
fa33M-2								12.17	0.535	30.628	2.054	117.806	19.796						
nv01F	24	10.377	0.577	25.205	1.117	237.029	16.019	7.517	0.573	18.281	1.418	257.195	38.126	8.347	0.606	23.192	2.906	219.62	40.994
nv01F-2	24													8.347	0.606	23.192	2.906	219.62	40.994
nv02F	21	9.851	0.915	23.557	1.679	267.491	27.931	6.233	0.87	21.565	2.071	278.941	48.455	8.994	0.674	26.135	1.819	243.378	27.507
nv03F	19	11.471	0.425	26.319	1.904	213.008	14.413	8.907	0.552	21.622	2.001	244.579	32.72	10.5	0.321	32.688	1.862	195.251	10.58
nv03F-2	19													10.5	0.321	32.688	1.862	195.251	10.58
nv04F	24	9.173	0.657	20.957	1.749	261.288	31.441	6.191	0.563	20.933	1.754	294.159	43.629	8.54	0.411	23.761	1.194	296.658	24.442
nv05F	23	9.889	0.444	25.278	1.814	293.108	21.989	7.965	0.657	23.85	2.247	282.085	37.506	8.675	0.215	26.436	1.345	268.444	20.113
nv05F-2	23							7.965	0.657	23.85	2.247	282.085	37.506						
nv06F	20	9.873	0.555	23.795	1.136	201.012	19.627	5.793	0.577	23.184	1.022	200.799	37.598	7.997	0.756	32.208	1.803	187.291	26.258
nv07F	22	10.76	0.636	20.845	1.881	201.352	25.355	5.947	0.887	24.808	1.227	185.394	40.065	11.25	0.477	23.09	1.257	178.744	14.396
nv08F	25	11.304	0.512	29.678	1.464	224.69	9.19	6.647	0.802	24.824	1.265	269.298	36.352	8.089	0.448	30.83	1.698	301.876	23.236
nv08F-2	25													8.089	0.448	30.83	1.698	301.876	23.236
nv09M	21	13.005	0.546	35.11	1.335	129.42	6.064	8.386	0.596	31.057	1.331	139.38	31.154	10.65	0.472	37.018	2.092	133.455	19.429
nv10M	21	10.606	0.802	26.566	1.113	98.178	26.635	6.903	0.812	23.497	1.592	97.848	49.901	10.9	0.703	28.014	1.614	98.278	26.571
nv11M	20	13.744	1.76	34.911	1.199	110.388	14.88	11.49	0.928	30.072	1.502	116.939	23.361	12.68	0.618	36.025	1.364	99.302	7.535

nv11 M-2	20													12.68	0.618	36.025	1.364	99.302	7.535
nv12F	18	10.596	0.324	26.706	2.582	220.677	21.952	8.4	0.512	18.936	1.13	217.913	30.715	10.87	0.256	27.125	1.362	206.898	10.298
nv13F	19	11.402	1.558	27.323	1.182	209.846	20.198	4.722	0.821	26.835	1.971	287.486	48.287	10.3	0.532	30.051	1.309	246.38	12.26
nv14 M	20	14.409	0.961	33.243	1.196	101.521	5.202	10.6	0.811	29.711	1.877	107.302	29.265	12.93	0.895	34.974	2.273	107.65	17.397
nv15 M	18	13.763	0.905	27.552	1.445	116.138	15.318	9.971	1.308	19.832	1.718	142.852	46.465	10.92	0.727	32.461	2.667	141.766	30.072
nv15 M-2	18													10.92	0.727	32.461	2.667	141.766	30.072
nv16 M	21	10.113	1.354	32.803	2.165	107.673	36.113	5.78	1.111	30.671	2.527	126.02	57.591	8.639	1.013	32.928	2.172	110.323	38.894
nv17 M	18	13.202	2.419	28.601	1.769	124.222	35.234	8.454	1.45	26.5	1.793	117.89	48.221	9.526	1.87	28.704	2.544	116.386	51.366
nv18 M	18	13.004	0.763	35.163	2.498	117.392	17.178	10.69	0.774	30.147	1.887	119.859	28.107	11.39	0.365	39.965	1.448	120.063	7.022
nv19 M	19	12.203	2.799	31.974	1.733	147.918	39.844	10.24	0.473	30.691	2.229	186.029	28.891	12.36	0.335	33.509	1.21	202.627	7.406
nv20 M	23	14.022	0.646	30.232	1.903	146.377	12.097	9.894	0.754	33.289	2.658	176.903	33.945	12.96	0.451	39.632	1.381	140.86	0.864

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