Do Perceptual Subgroups Exist in Ataxic Dysarthria?

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Abstract

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When the cerebellar circuit is damaged, the resulting speech disturbance is known as ataxic dysarthria. Features of this disorder include irregular articulatory breakdowns, distorted vowels, excess and equal stress, prolonged phonemes, and abnormal pitch/loudness variation. However, heterogeneity within this disorder has been documented by clinicians and researchers for decades, suggesting that subgroups of ataxic dysarthria may exist. The purpose of this study was to extend this line of research to determine a feasible, reliable perceptual method for identifying and differentiating these proposed subgroups. Two hypotheses were tested based on existing theoretical frameworks: (1) speakers be classified into instability/inflexibility (or mixed presentation) subgroups, and/or (2) speakers be classified into subgroups based on differential speech subsystems involvement. Four dysarthria experts listened to speech samples of adults with ataxic dysarthria and completed rating forms of the speech features. Results suggested that five speakers fit the pattern of instability, two speakers aligned with inflexibility, and one speaker had a mixed presentation. Patterns did not emerge according to differential subsystem involvement. The emergence of subgroups has implications for clinical and research practice; additional research is warranted.

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Introduction

Ataxic dysarthria refers to speech disturbances caused by disorders of the cerebellum. Decades of acoustic and perceptual studies have attempted to describe the features associated with this motor speech disorder, which span across multiple speech subsystems, including articulation, phonation, and prosody. Traditionally reported perceptual characteristics of ataxic dysarthria include irregular articulatory breakdowns, excess and equal stress, prolonged phonemes, slow rate, monopitch, monoloudness, and excess loudness variations (Duffy, 2013). This list is merely a sample of possible characteristics – ataxic dysarthria can present with highly varied symptoms (Sidtis et al., 2011). In fact, seminal studies by Darley et al. (1969) and ensuing investigations have characterized this dysarthria as having a wide range of speech characteristics, some of which appear contradictory (e.g., excessive prosody as well as insufficient prosody). This long-standing heterogeneous description of ataxic dysarthria brings into question whether it is a single entity.

The wide range of characteristics of ataxic dysarthria has led researchers to attempt to explain why such diversity exists within one speech diagnosis. One such explanation posits **lesion locus**. There is a wide range of possible lesion sites within the cerebellum that can lead to speech disturbances. While certain regions of the cerebellum are thought to be more highly correlated with certain speech subsystems (e.g., superior regions are heavily involved in articulatory control), researchers have not been able to reliably use lesion locus to predict speech characteristics (Spencer & Slocomb, 2007). Additionally, speakers with the same neurological etiology show highly heterogeneous speech symptoms. Folker et al. (2012) examined the speech profiles of seven individuals with Friedrich's ataxia (a congenital disorder which leads to spinocerebellar degeneration). The only speech characteristic that all seven participants shared was articulatory imprecision. Speech symptoms were highly variable, despite identical etiologies, showing that variability within ataxic dysarthria cannot be fully explained by differences in lesion locus.

Other studies on heterogeneity within ataxic dysarthria point to **disease progression** as an explanation for the wide range of symptoms. Ackermann et al. (2007) hypothesized that cerebellar dysfunction happens in two stages, where ataxic speech first exhibits temporal instability (e.g., highly variable syllable timing) and then evolves into 'scanning' speech, defined as "slowed and isochronous syllable pacing." Additionally, consonant imprecision and excess and equal stress were found to be predictors of severity in spinocerebellar ataxia (Sidtis et al., 2011). That is, with increased overall severity, speakers tended to exhibit more severe consonant imprecision and equalized stress. However, a similar study found severity predictions to be highly inconsistent among speakers despite similar speech symptom variability (Schalling et al., 2008). Additionally, when speakers with spinocerebellar ataxia were matched for genotype, disease severity, and disease duration, variable speech profiles were still found (Brendel et al., 2015). The differences in impairment could not be accounted for by the progression of the disorder. Thus, the evidence connecting speech characteristics to severity and disease progression is limited and inconclusive, and is not sufficient to explain the wide range of features present in ataxic speech.

In an attempt to explain the diversity of ataxic dysarthria, evidence has emerged regarding the possibility of subgroups. Thus far, studies tend to align with one of two subgroup theories: (1)

speakers exhibit clusters of deviant speech characteristics according to speech subsystem or (2) speakers tend toward either inflexibility or instability.

Clusters of deviant speech characteristics (subsystem hypothesis)

Formative research by Darley and colleagues (1969) noted that speech characteristics of ataxic dysarthria appear to cluster in a way that reflects differential subsystem involvement. These clusters were labeled "articulatory inaccuracy," "prosodic excess," and "phonatory-prosodic insufficiency". Articulatory inaccuracy refers to irregular articulatory breakdowns, consonant imprecision, and distorted vowels. The second cluster, prosodic excess, refers to slow rate, excess and equal stress, and prolonged phonemes and intervals. Scanning speech, which refers to a tendency toward temporal isochrony at the syllable level, is also included in this cluster. The final cluster, phonatory-prosodic insufficiency, includes harshness, monopitch, and monoloudness. The existence of these clusters, and the seemingly contradictory nature of prosodic excess and prosodic insufficiency, supports the notion that variation within the disorder may be indicative of differential subsystem impairment.

Joanette and Dudley (1980) built upon Darley and colleagues' subsystem hypothesis with a perceptual study examining 22 speakers with Friedreich's ataxia. Speech samples were rated on 16 dimensions encompassing vocal intensity, vocal quality, prosody, and articulation. Through an intercorrelation matrix analysis, the authors found that speakers could be grouped according to two "factors" rather than the three dimensions proposed by Darley and colleagues. The two factors were: (1) general dysarthric factor – highest ratings in the dimensions of imprecise consonants and prolonged phonemes, and (2) phonatory-stenosis factor – highest ratings in the dimensions of harshness, pitch breaks, and pitch level. These findings revealed two dichotomous groups within Friedreich's ataxia, which aligns with the hypothesis that distinct speech profiles in ataxic dysarthria may reflect speech characteristics that cluster according to differential subsystem involvement.

Additional support for the subsystem hypothesis comes from the differential impairment of respiration in some speakers with ataxic dysarthria. For example, Ebert et al. (1995) analyzed breathing rhythm and coordination of breath in six patients with cerebellar damage. The authors found "ataxic breathing" patterns, including (1) breathing arrests during spontaneous breathing, and (2) incoordination between breathing and other motor systems. Ataxic breathing during speech is a form of respiratory instability, which may underlie speech symptoms, including prosody and phonation. Irregularities in chest wall movements have also been documented in speakers with ataxic dysarthria during sustained phonation and syllable repetition (Murdoch et al., 1999). Thus, differential impairment in the respiratory subsystem is a possible factor in the heterogeneity of ataxic speech, and provides additional evidence for the speech subsystem hypothesis.

Kent and colleagues (2000) found high intersubject variability of syllable durations and pause durations during syllable repetitions. Syllable AMRs (alternating motion rates) were used to acoustically examine prosody, and high variability among speakers was found even within this one speech task. There was also significant intersubject variability in level of phonatory impairment, and evidence of sex differences in regards to jitter (only female subjects showed abnormality in this dimension). The authors posited that their findings, which show heterogeneity in speech disturbances at multiple levels of production, could reflect possible subgroupings according to combinations of subsystem impairments rather than distinct neuropathologies. This is in agreement with Darley and colleagues' original subsystem hypothesis, but Kent and colleagues argue that a detailed subsystem description of the speech disorder is required before subgroups can be fully understood. Kent and colleagues (1998) also suggest that disturbances in neural control do not uniformly affect the subsystems. Instead, they theorize that speakers show potential global effects related to timing and positioning, with other individualized abnormalities on top of those general dysarthric features. Understanding each individual patient's profile requires a detailed understanding of each subsystem, which is also required in order to classify subgroups within the overall dysarthric population (Kent et al., 1998).

Instability/inflexibility hypothesis

In contrast to the subsystem hypothesis, Hartelius and colleagues (2000) proposed that features of ataxic dysarthria may be linked to patterns of *instability* or *inflexibility*. Generally, the principle of instability would be associated with unusually increased variability, such as poorly modulated pitch and loudness variations. In contrast, the principle of inflexibility would be associated with speech symptoms that are abnormally invariant and equalized, such as scanning speech where equal emphasis is given to each syllable. If subgroups of ataxic dysarthria exist, they might reflect the degree to which instability versus inflexibility of motor control predominates (Duffy, 2013). There is acoustic and perceptual evidence to support this theoretical subgrouping, as summarized below.

Instability

With respect to characteristics reflecting *instability*, there are numerous studies supporting the presence of variability in the speech patterns of individuals with ataxic dysarthria, across subsystems and speech tasks. For instance, with respect to articulation, motor incoordination (e.g., overshoot and undershoot) causes articulatory variation in the form of slowness, inaccuracy, and poor timing of articulatory gestures (Duffy, 2013). One study found that perceptual variability occurs as a result of the same articulatory target being produced multiple different ways (e.g., on-target, distorted, omitted) within one speaker and one task (Ackermann et al., 1992). Similarly, acoustic studies of prosody have suggested that ataxic dysprosody occurs in the form of variable intensity, variable fundamental frequency, and poor control of stress assignment and syllable timing (Boutsen et al., 1997; Boutsen et al., 2011; Hartelius et al. 2000; Ackermann et al., 2007). Additionally, respiratory incoordination is seen in some ataxic speakers, which may manifest as a lack of coordination between breathing and speaking, causing perceptual variability in the speech signal, such as audible inspiration at syntactically inappropriate times or excess loudness variations (lack of synchrony of respiration and phonation, Duffy, 2013).

To summarize, speech features that fall into the *instability* category include variable and inconsistent speech rate; articulatory variability; abnormally variable prosody as shown by poorly modulated intensity, variable fundamental frequency, and poorly controlled stress patterns; dysrhythmia; and respiratory incoordination (Hartelius et al., 2000; Duffy, 2013; Kent et al., 2000). Therefore, an *instability* subgroup would reflect the tendency of some speakers with ataxic dysarthria to speak with more variation, resulting in inconsistent articulation errors, variable rate and rhythm, and fluctuations in pitch and loudness.

Inflexibility

Conversely, there are several reports that reflect *inflexibility* in the speech patterns of speakers with ataxic dysarthria. With regard to prosody, research supports the presence of reduced rate from an equalization pattern across syllables (Kent et al., 1979) as well as abnormally reduced variation in intensity, pitch, and stress patterns (Duffy, 2013). This prosodic inflexibility is sometimes referred to as scanning speech. This type of ataxic speech is perceptually isochronous, or evenly timed and metered, rather than variable or unpredictable. When considering imprecise articulation– a hallmark feature of ataxic dysarthria – relatively consistent articulatory imprecision has been found in some ataxic speakers (Duffy, 2013; Hartelius et al., 2000; Ackermann, 1994). Odell et al. (1991) reported that relatively consistent vowel distortions, rather than variable errors, were seen due to overshoot or undershoot. Thus, an *inflexibility* subgroup would reflect the tendency of some speakers with ataxic dysarthria to speak with less variation, resulting in a staccato, isochronous speech rhythm, monopitch, monoloudness, and a reduction in rate. Moreover, articulation would be consistently distorted. Table 1 (below; France, 2014) summarizes the perceptual characteristics within each speech subsystem that are proposed to align with either *instability* or *inflexibility*.

Table 2. Summary of salient speech features that are consistent with patterns of instability versus inflexibility (France, 2014).

	Instability	**Inflexibility**
Respiration	• Incoordination of breathing with speaking • Paradoxical breath patterning	• Consistently reduced breath support
Phonation	(see pitch description under Prosody)	
Articulation	• Variable; possible undershoot and overshoot	• Relatively consistent articulatory imprecision
Resonance	• Intermittent instances of hypernasality and hyponasality	
Prosody	• Inconsistent rate with variable syllable length • Uncontrolled loudness variation • Variable pitch • Variable stress patterns • Variable rhythm (especially in AMRs/SMRs)	• Decreased rate with equal pattern across syllables • Monoloudness • Monopitch • Isochronous, metered stress patterns • Steady rhythm (especially in AMRs/SMRs)

Note. AMRs = alternating motion rates, SMRs = sequential motion rates (Duffy, 2013).

Mixed Instability/Inflexibility

It is possible that individuals will share features across the two subgroups of inflexibility and instability. Indeed, according to Duffy (2013; personal communication), the *combination of subgroups* may be most common. Prior to the pilot study described below, there was no literature to directly substantiate this notion. Indirect support stems from the clinical presentation of some speakers with ataxic dysarthria. That is, elements of instability, such as poorly modulated prosody, may intermingle with the attributes of inflexibility, such as a relatively consistent disruption to articulation.

Some studies emerged prior to the theory by Hartelius and colleagues (2000) that aligned with this notion of instability/inflexibility. For example, Boutsen and colleagues (1997) examined durational variability (DV) during syllable AMRs by utterance type (i.e., repetitions of /pʌ/, /tʌ/, and /kʌ/ were compared to one another). Three subgroups emerged. Speakers exhibited either (1) similar DV across all three utterance types, (2) higher DV during repetitions of /pʌ/, or (3) higher DV during repetitions of /kʌ/. While the authors did not connect these results to notions of instability/inflexibility, this study appears to fit within the framework. That is, subgroup 1 showed a tendency toward *stable* durational variability, while subgroups 2 and 3 tended towards *variability* across utterance type. Thus, subgroups could be formed according to whether they exhibited *unstable* durational variability or *inflexible* durational variability.

Testing the instability/inflexibility hypothesis: A pilot study (Spencer & France, 2015)

Preliminary research by Spencer & France (2015) used perceptual ratings of ataxic dysarthria to group speakers into one of three subgroups: (1) tendency towards instability, (2) tendency towards inflexibility, and (3) mixed presentation. Audio files were compiled from Aronson (1993), Duffy (2013), and Freed (2000) – pre-recorded patient samples used in educational programs as exemplary presentations of pure ataxic dysarthria. Speakers represented diverse forms of cerebellar compromise, including tumor, stroke, cerebellitis, sporadic cerebellar degeneration, and Friedreich's ataxia. The sample included 6 female and 4 male speakers. All audio speaker samples contained an example of connected speech (reading or conversation) and speech diadochokinetics, a maximum performance task where the speaker rapidly produces alternating or sequential syllables. Eight of ten speaker samples also contained an example of sustained phonation. Samples ranged in duration from 52 – 140 seconds, with an average of 76.5 seconds.

A perceptual rating form (Figure 1, below) was developed based on the Hartelius et al. framework. Ten experienced speech-language pathologists with more than five years of clinical experience working with speakers with dysarthria rated each sample according to 12 speech features across three speaking tasks (diadochokinetics, sustained phonation, and connected speech). Membership in a given subgroup was based on the following criterion: samples needed greater than a 20% difference between inflexibility/instability criteria in a given speaking task, with the second speaking task demonstrating a minimum of a 10% difference in the same direction. Results suggested that five speakers fit the pattern of **instability**, one speaker aligned with **inflexibility**, and four speakers had a **mixed presentation** (combined instability/inflexibility). Inter-rater reliability was measured using point-to-point agreement and was deemed sufficient.

Figure 1: Rating sheet for perceptual features per speech task with color coding to reflect hypothetical subgroups of Instability (red) and Inflexibility (green).

Listener # Sample #

DDKs				
Articulation	☐ Variable errors	☐ Consistent errors		☐ Normal
Voicing	☐ Variable errors	☐ Consistent errors		☐ Normal
Rate	☐ Variable	☐ Consistently slow		☐ Normal
Rhythm	☐ Variable			☐ Normal
Sustained Phonation				
Loudness	☐ Variable	☐ Consistently too loud	☐ Consistently too quiet	☐ Normal
Pitch	☐ Variable			☐ Normal
Connected Speech				
Articulation	☐ Variable errors	☐ Consistent errors		☐ Normal
Stress	☐ Irregular placement	☐ Equalized across syllables		☐ Normal
Rate	☐ Variable	☐ Consistently slow		☐ Normal
Pitch	☐ Variable	☐ Monopitch		☐ Normal
Respiration	☐ Incoordination of breath with speech	☐ Speech on residual air	☐ Short phrases	☐ Normal
Loudness	☐ Uncontrolled	☐ Consistently too loud	☐ Consistently too quiet	☐ Normal

Figure 2: Sample of speakers who fit criteria for the Instability subgroup (n=5).

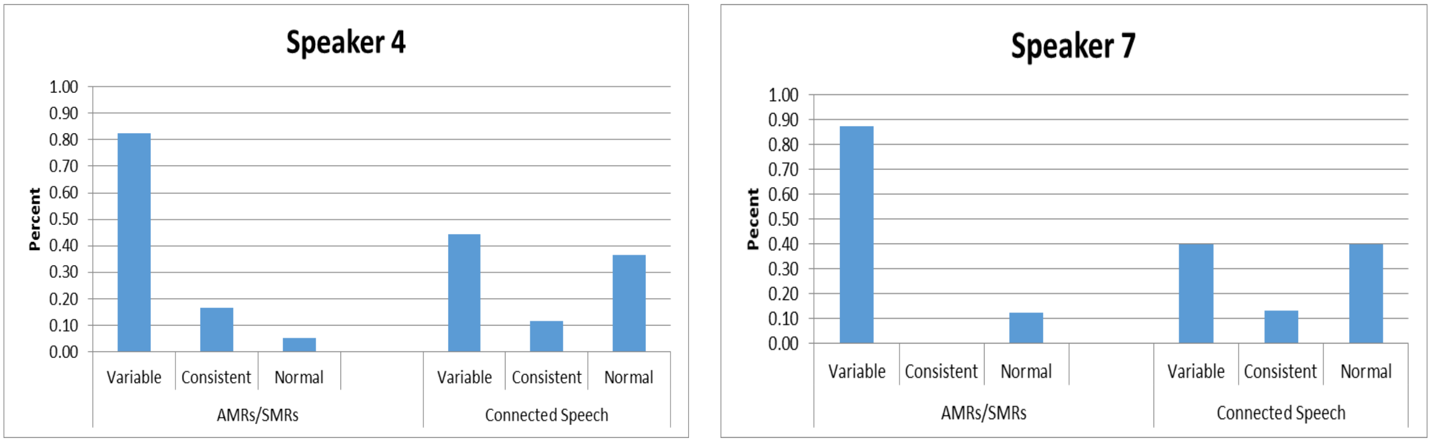


Figure 3: Speaker fitting the criteria for the Inflexibility profile (n=1).

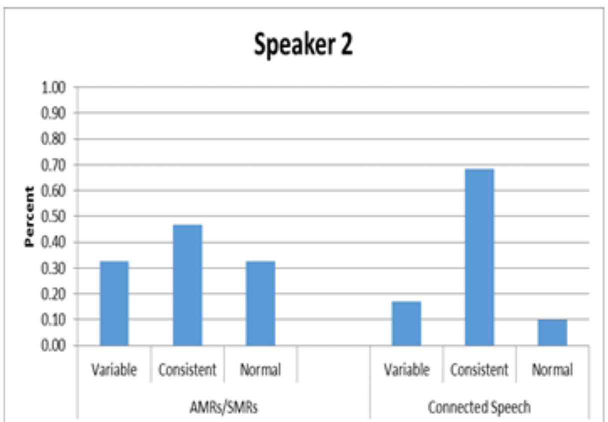
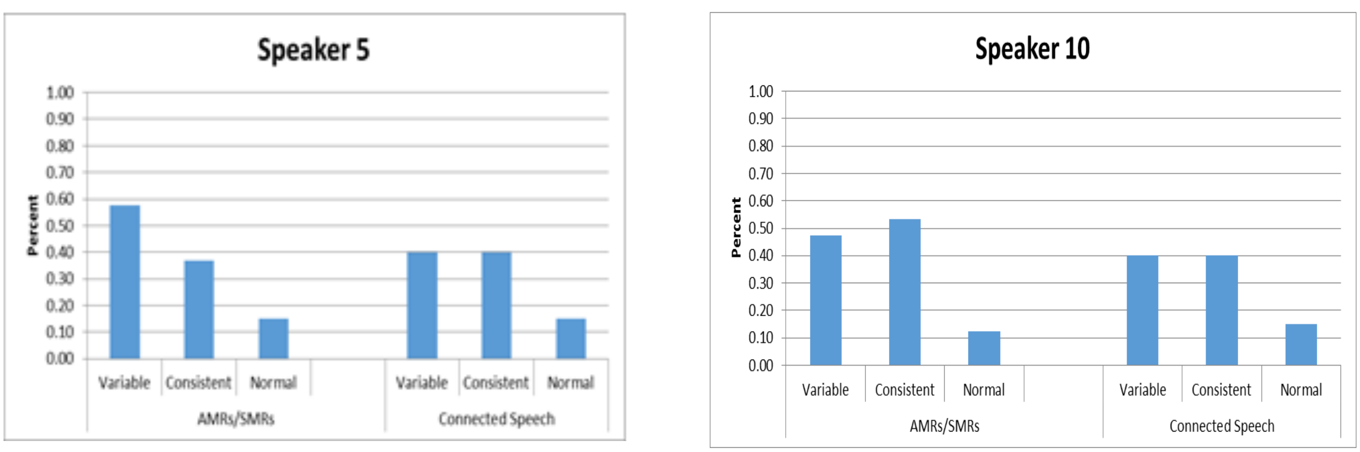


Figure 4: Sample of speakers who fit criteria for the Mixed subgroup (n=4).



Note. Figures reflect the percent of features chosen across categories of variable (instability), consistent (inflexibility) or normal, per speaking task, averaged across the ten listeners.

This study yielded preliminary evidence that speakers with ataxic dysarthria may present with speech characteristics that cluster toward a pattern of instability, inflexibility, or a combination of the two subgroup profiles. To highlight, Figure 2 (Speakers 4 and 7) shows a pattern of variability across multiple speaking tasks, based on the higher percentage of variability ratings when compared with ratings of consistency or normalcy. This pattern was evident for both speaking tasks: Alternating/Sequential Motion Rates (AMRs/SMRs) and connected speech. Figure 3 (Speaker 2) illustrates a pattern of consistency across speaking tasks. Finally, Figure 4 shows a pattern of combined instability/inflexibility based on a bias toward instability/inflexibility for only one speaking task (Speaker 5) and similar ratings of variability and consistency across both tasks (Speaker 10). This pilot study was the first to use perceptual judgment (the gold standard in dysarthria diagnosis) to group ataxic speakers, and the current study will be a continuation and extension of this line of research.

Rationale for current study

The study by Spencer & France (2015) provided preliminary evidence of subgroups of ataxic dysarthria. Further investigation was warranted. The current study implements modifications in three primary areas: (1) speech sample collection, (2) rating form and judges, and (3) inclusion of an alternative hypothesis.

First, speech samples were collected from live participants with ataxic dysarthria rather than pre-recorded educational samples. This standardized the collection of speech samples and improved upon the collection of demographic and disease-related information. Additionally, speakers were asked to volitionally vary their DDK rates and the clarity of their conversational speech, in order to rule out certain speech characteristics (i.e., slow rate) as existing not as a product of their impairment but as a compensatory strategy for increased intelligibility.

Rationale for non-habitual speech tasks

Recent work by Lowit and colleagues (2014) looked at ataxic speakers with excess and equal stress patterns, and whether they were able to vary fundamental frequency to create pitch contrasts and signal sentence-level stress. Two speakers in particular stood out because while they were unable to vary pitch due to limited F0 control, they showed compensatory use of durational and intensity contrasts to signal stress position. This relates to the inflexibility/instability framework (Hartelius et al., 2000) because it indicates that ataxic speakers with features aligning with inflexibility (excess and equal stress; monopitch) may be able to compensate by varying other speech dimensions.

There is more robust literature showing that those with hypokinetic dysarthria from Parkinson's disease are able to vary speech patterns (i.e., loudness) in response to explicit and implicit cues (Ho et al., 1999; Darling & Huber, 2011). Additionally, work by Tjaden and colleagues (2013) showed significant changes in rate, vocal intensity, and vowel spectral characteristics in the speech of adults with Parkinson's disease and Multiple Sclerosis when asked to read sentences under 4 speaking conditions: (1) habitual, (2) loud, (3) slow, (4) clear. The "clear speech" condition elicited the greatest changes, in response only to the verbal instruction to speak "how you might talk to someone in a noisy environment or with a person who has hearing loss. Exaggerate the movements of your mouth. You may also be louder and slower than usual. If your regular speech corresponds to a clearness of 100, you should aim for a clearness twice as loud, or a clearness of 200." Based on this literature, speakers were asked to modulate rate and loudness during DDKs and conversational speech samples.

Next, the perceptual rating form were modified to reflect additional speech dimensions and allow for responses to be plotted on rating scales rather than forcing binary choices. Additionally, perceptual ratings were completed by four dysarthria experts rather than ten experienced speech-language pathologists, in order to enhance the validity and reliability of the perceptual judgments. Finally, the current study contrasts the instability/inflexibility hypothesis with an alternative hypothesis (i.e., the subsystem hypothesis).

In sum, the present study investigated whether subgroups of ataxic dysarthria can be identified based on perceptual ratings across four speaking tasks (two maximum performance and two connected speech). Specifically, there are three central research questions:

(1) Can speakers be classified into instability/inflexibility (or mixed presentation) subgroups based on Visual Analog Scale patterns?

(2) Can speakers be classified into subgroups based on presence/severity ratings across speech subsystems of articulation, phonation, respiration, and prosody?

(3) Can speakers be classified into mixed subgroups based on both Visual Analog Scale patterns and presence/severity ratings across speech subsystems?

Based on the extant literature and preliminary findings from Spencer and France (2015), it is hypothesized that speakers will be classifiable based on instability/inflexibility/mixed subgroups. Table 2 summarizes the proposed hypotheses.

Table 2: Proposed null hypothesis and two alternate hypotheses, with related interpretations.

	Hypothesis	**Interpretation**
H_0	No clusters emerge from VAS ratings related to inflexibility and instability. No clusters emerge from Mayo Clinic Scale ratings pertaining to articulation or prosody.	Fail to reject null hypothesis.
H_1	Instability/Inflexibility Hypothesis	Visual Analogue Scale ratings align with subgroups of instability, inflexibility, or mixed instability/inflexibility, but no clusters emerge for differential subsystems.
H_2	Differential Subsystem Hypothesis	Mayo Clinic Scale ratings align with differential impairment of articulation, phonation, respiration or prosody, but no patterns emerge from VAS.

Methods

Participants

Speakers

Eight participants with an existing speech diagnosis of ataxic dysarthria were included in the study. Participants were screened and excluded for: (1) overt signs of extracerebellar pathology (e.g., Parkinson-like symptoms), (2) disease duration of less than 1 year, (3) presence of another neurological disorder and/or history of head trauma, (4) severe or uncontrolled psychiatric disorder, (5) presence of dementia, based on *Mini Mental State Examination* (MMSE; Folstein et al., 1975) score less than 26/30, (5) moderate-severe depression based on *Beck Depression Inventory II* (Beck et al., 1996) score greater than 19, and (6) alcohol or drug dependency. All participants were native speakers of American English with intact hearing (thresholds of 50dB or less at 500, 1000, and 2000Hz) and typical developmental history with regards to speech, language, and cognition. One participant was tested at the University of Washington Motor

Speech & Cognitive Disorders Laboratory, while the other seven were tested in their respective homes. See Tables 3-5 for detailed participant information.

Table 3: Participant demographic information.

	Age	Sex	Race/Ethnicity	Years of Education	Diagnosis	Symptom Duration in Years
A1	55	F	African American	14	SCA3	15
A2	35	M	Asian	18	SCA3	3
A3	39	F	White	12	SCA2	18
A4	76	F	White	13.5	SCA6	10
A5	41	F	White	12	Friedreich's Ataxia	27
A6	75	M	White	14	SCA6	21
A7	36	F	Asian	16	SCA3	17
A8	43	F	White	18	Friedreich's Ataxia	24
	M = 50 *SD = 16.9*	*6 female*	*5 White*	*M = 14.7* *SD = 2.4*	*8 SCA*	*M = 16.9* *SD = 7.7*

Note. SCA = spinocerebellar ataxia

Table 4: Primary neurologic symptoms reports by each participant.

	Primary Characteristics
A1	Nystagmus; blurred vision; unsteady gait
A2	Unsteady gait; mildly imprecise articulation
A3	Difficulty walking; incoordination; slow, imprecise speech
A4	Incoordination; double vision; slow, imprecise speech
A5	Incoordination; difficulty walking; heart problems; slow, imprecise speech
A6	Incoordination; difficulty walking; slow, imprecise speech (worsens throughout the day)
A7	Balance and gait disturbances; double vision; dysphagia (PEG tube since 2014); severe dysarthria
A8	Incoordination; dysarthria; nystagmus; dysphagia; incontinence

Table 5: Participant screening measures and mean intelligibility scores.

	BDI-II	**MMSE**	**Sentence Intelligibility**
A1	4	30	95.68
A2	5	29	94.32
A3	5	30	97.27
A4	2	30	96.82
A5	11	29	34.87
A6	13	30	84.77
A7	12	29	14.32
A8	4	30	95.68
	M = 7 *SD = 4.3*	*M = 29.6* *SD = 0.5*	*M = 76.7* *SD = 32.9*

Note. BDI-II = Beck Depression Inventory-II; MMSE = Mini-Mental State Examination; Sentence Intelligibility based on average of four raters using the Speech Intelligibility Test.

Listeners

Perceptual rating of the speech samples was performed independently by four internationally recognized dysarthria experts (Drs. Miller, Spencer, Strand, and Yorkston). Additionally, speech samples collected for intelligibility testing were rated independently by four speech-language pathology master's students.

Tasks

Speech Recordings and Preparation

All participants were informed about the aim of the study and gave written informed consent. The computerized version of the Speech Intelligibility Test (Yorkston et al., 1996) was administered for a quantifiable index of the speaker's sentence-level intelligibility. Speech sampling consisted of three tasks: one-minute monologue, sustained phonation, and speech diadochokinetics (AMRs/SMRs). The one-minute monologue was elicited twice, once with no additional instructions, and once with instructions for "clear speech," adapted from Tjaden and colleagues (2013). AMRs/SMRs were produced in four conditions: (1) habitual, (2) as fast as possible, (3) slow and steady, and (4) twice as loud. Speech tasks were administered in a randomized order with loud samples grouped for optimal recording conditions. See Appendix A for a protocol summary.

Participants were recorded using a high quality head-mounted microphone (AKG C520) with a constant mouth-to-microphone distance of 2 in. The microphone was connected to a portable digital speech recorder (Zoom H6, GU-ZOOMH6). All speech samples were recorded in a quiet environment with low ambient noise.

Speech samples were edited using acoustic software (Adobe Audition). Audio files were clipped to include: (1) one trial each of AMRs (/pa/, /ta/, /ka/) and SMR (/pa-ta-ka/) in each speaking condition, (2) one trial of sustained vowel phonation, and 3) one 30-second clip of connected speech from each monologue. The criteria used for extracting the sample from the monologue were based on Bunton et al. (2007) and included: exclusion of the first 10 seconds and last 10 seconds for all; selection from a period of continuous speech.

Perceptual ratings of habitual speech tasks

Based on the audio presentation of the three habitual speech tasks (AMR/SMRs; connected speech; sustained phonation), expert listeners first indicated the presence and type of dysarthria. Following detailed rating instructions, listeners were exposed to two brief familiarization audio samples: one exemplifying a tendency toward irregular and variable errors and one exemplifying a tendency toward consistent and stable errors. Listeners then completed perceptual rating forms to identify the presence and severity of speech characteristics on the Mayo Clinic 5-point scale (Duffy, 2013) and to indicate on a 100 mm Visual Analogue Scale (VAS) where a given characteristic falls on a continuum of consistency/variability (Spencer and France, 2016). Consistency/variability rating anchors were specific to each speech characteristic. The sustained phonation task does not lend itself to Visual Analogue Scale ratings due to the inherent inflexibility that this task imposes. Therefore, only Mayo Clinic presence/severity ratings were given to listeners for the sustained phonation task. Figures 5-7 show the forms used for the task-specific ratings. Order of presentation of the tasks (diadochokinetics, sustained phonation, connected speech) within each subject was counterbalanced, with random presentation of participants.

Rater: ________________________ Participant Code: ________

Diadochokinetic Task (AMRs/SMRs)

Articulation errors	**0 Absent**	**1 Mild**	**2 Moderate**	**3 Marked**	**4 Severe**

Errors are irregular and variably present ______________________________ Errors are consistently present

Rate of repetition (slow)	**0 Normal**	**1 Mild**	**2 Moderate**	**3 Marked**	**4 Severe**

Variable rate ______________________________ Rate is consistently slow

Voicing Errors	**0 Absent**	**1 Mild**	**2 Moderate**	**3 Marked**	**4 Severe**

Errors are irregular and variably present ______________________________ Errors are consistently present

Figure 5: Perceptual rating form specific to the diadochokinetic task (AMRs/SMRs).

Note. A "0" rating in the presence/severity scale would preclude a rating with the Visual Analog Scale.

Rater: ________________________ Participant Code: ________

Sustained Phonation Task

Duration of sustained /a/	**0** **Normal**	**1** **Mildly Reduced**	**2** **Moderately Reduced**	**3** **Markedly Reduced**	**4** **Severely Reduced**
Loudness Fluctuations	**0** **Absent**	**1** **Mild**	**2** **Moderate**	**3** **Marked**	**4** **Severe**
Pitch Fluctuations	**0** **Absent**	**1** **Mild**	**2** **Moderate**	**3** **Marked**	**4** **Severe**

Figure 6: Perceptual ratings specific to sustained phonation task.

Rater: ________________________ Participant Code: ________

Connected Speech Task

Articulation errors	**0 Absent**	**1 Mild**	**2 Moderate**	**3 Marked**	**4 Severe**

Errors are irregular and variably present ______________________________ Errors are consistently present

Stress errors	**0 Absent**	**1 Mild**	**2 Moderate**	**3 Marked**	**4 Severe**

Irregular and variable stress ______________________________ Excess and equal stress

Rate of speech (slow)	**0 Normal**	**1 Mild**	**2 Moderate**	**3 Marked**	**4 Severe**

Variable rate ______________________________ Rate is consistently slow

Abnormal Pitch	**0 Absent**	**1 Mild**	**2 Moderate**	**3 Marked**	**4 Severe**

Variable pitch (abnormal pitch fluctuations) ______________________________ Monopitch

Abnormal Loudness	**0 Absent**	**1 Mild**	**2 Moderate**	**3 Marked**	**4 Severe**

Variable loudness (abnormal loudness fluctuations) ______________________________ Monoloudness

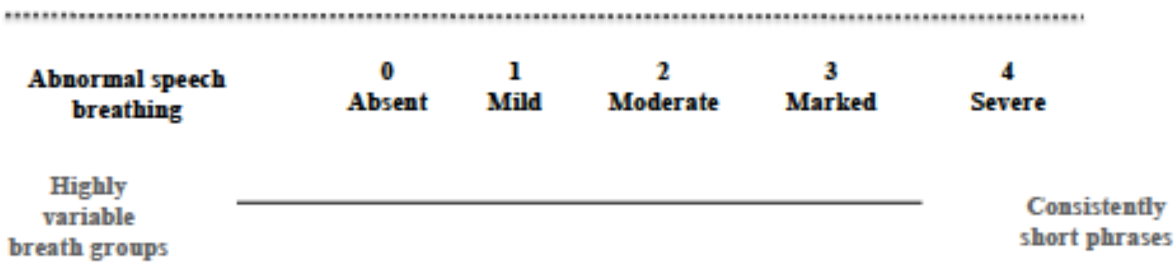


Figure 7: Perceptual ratings specific to connected speech task.

Note. A "0" rating in the presence/severity scale would preclude a rating with the Visual Analog Scale.

Measuring response to instruction

Clear speech monologues were presented in random order to one judge (Dr. Spencer), who rated the samples according to the Connected Speech rating form (Figure 7), weeks after rating habitual monologues. The rater then listened to each speaker's habitual and clear speech monologues back-to-back, and wrote qualitative descriptions of differences in speech characteristics between the two speaking conditions. Standard, fast, and slow AMR/SMRs were compared based on rate, and each speaker's ability to surpass their own variation to establish an alternate rate. AMR/SMRs and sustained phonation in the loud condition were excluded from analysis due to difficulty standardizing loudness across recording environments.

Analysis

Parametric and non-parametric statistics were not implemented for the subgroup analyses, given the small sample size, the manner in which the listeners rated the features, and the inconsistent number of ratings for each feature. Instead, descriptive statistics and visual analyses were employed.

Reliability

To determine intra-rater reliability, 20% of speaker samples were randomly repeated throughout the listening session. See Appendix C for order of presentation for each rater. For VAS ratings, intra-rater reliability of judgments was analyzed using intraclass correlation coefficients (ICC). For Mayo scale ratings, intra-rater and inter-rater reliability of judgments were analyzed using Spearman's rank correlation coefficient. Statistical analysis of inter-rater reliability for VAS ratings was not completed given the nature of the data, but visual analysis is included below.

Results

Intra-rater reliability – VAS ratings

Based on intraclass correlation, VAS ratings were considered reliable (.735) when collapsed across raters ("good" agreement = .60 – .74; Cicchetti, 1994). Intra-rater reliability was variable across raters, with a range of ICC from .339 to .892; see Table 6. The lower ICC (.339; Rater 3) reflects the sensitivity of the test to a shift in variability, as this rater was highly consistently with six out of seven ratings. Visual analysis of intra-rater reliability was also conducted using three levels of agreement. Full agreement was considered when ratings were (1) within 20mm on the VAS and (2) on the same side of midline. Partial agreement was considered when ratings had greater than 20mm difference, but were on the same side of midline. No agreement was defined as ratings that were on the opposite sides of midline, regardless of how close or far apart.

According to percent agreement, 79.06% of all ratings were in full agreement. Please see Table 7 and Figure 8 below.

Table 6: Intraclass correlation coefficients and confidence intervals by rater, for VAS ratings.

			95% Confidence Interval	
Rater	**Observations**	**ICC***	**Lower Bound**	**Upper Bound**
R1	15	.780	.473	.919
R2	5	.892	.407	.988
R3	7	.339	-.433	.840
R4	16	.596	.173	.836
Combined	**43**	**.735**	**.561**	**.847**

**According to Chichetti (1994), ICC from .60 to .74 qualifies as "good" agreement, and .75 – 1.0 qualifies as "excellent" agreement.*

Table 7: Percent intra-rater agreement across tasks and raters, for VAS ratings.

Level of Agreement	**Definition**	**Percentage (across all 4 raters)**
Full agreement	<20 mm and same side of midline	79.06
Partial agreement	>20mm and same side of midline	6.98
No agreement	Opposite sides of midline	13.95

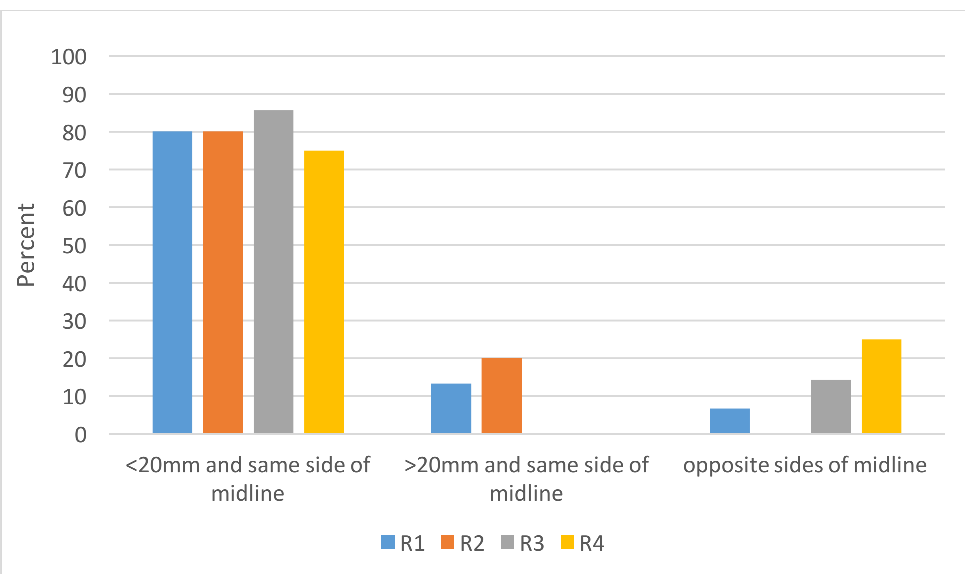


Figure 8: Percent intra-rater agreement for VAS ratings across speaking tasks per individual raters (R1 – R4).

Intra-rater reliability – Mayo scale ratings

As shown in Table 8, Spearman's rank correlation coefficients for all raters were significant and greater than .60. Descriptive analysis was also conducted, as shown in Table 9 and Figure 9. On the 5-point Mayo scale, there was 70.37% full agreement (no difference between time 1 and time 2) across all four raters. For over 96% of ratings, the raters chose the identical severity level, or a rating that was only one level different, between time 1 and time 2.

Table 8: Intra-rater agreement by rater across speaking tasks for Mayo scale severity ratings.

Rater	Observations	Correlation Coefficient	Significance (2-tailed)
R1	21	.680	.001*
R2	21	.791	.000*
R3	18	.951	.000*
R4	21	.864	.000*

**Correlation is significant at the 0.01 level (2-tailed)*

Table 9: Percent intra-rater agreement across tasks and raters, for 5-point Mayo scale ratings.

Difference	**Percentage (across all 4 raters)**
0	70.37
1	25.93
2	3.70
3	0
4	0

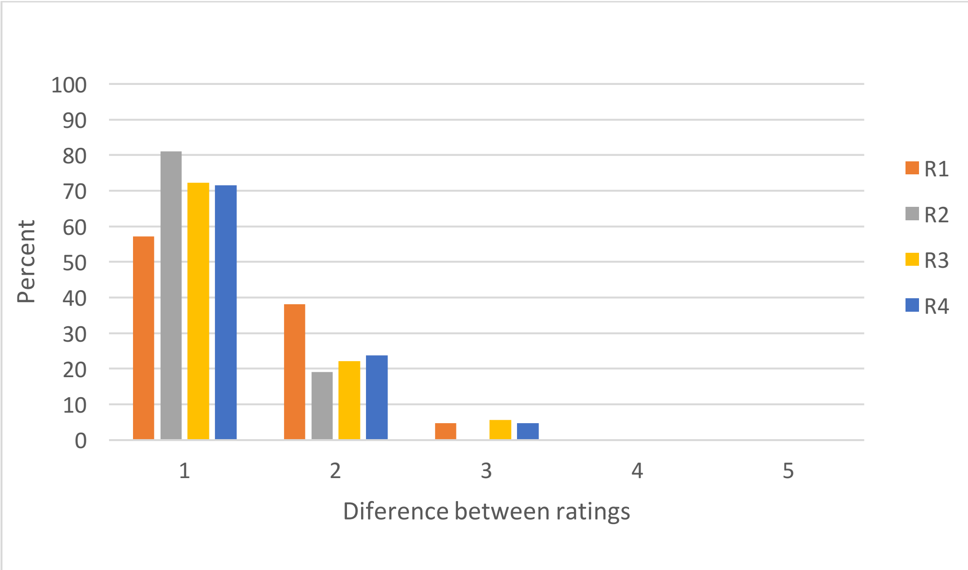


Figure 9: Percent intra-rater agreement for Mayo scale ratings across individual raters, collapsed across speaking tasks.

<u>Inter-rater reliability – VAS ratings</u>

For the DDK ratings, the majority of judges (3-4) were in full agreement 56.5% of the time. Full agreement is defined as a difference < 20 mm and rating on same side of midline (see Figure 10). For the conversation ratings, the majority of judges were in full agreement 67.6% of the time.

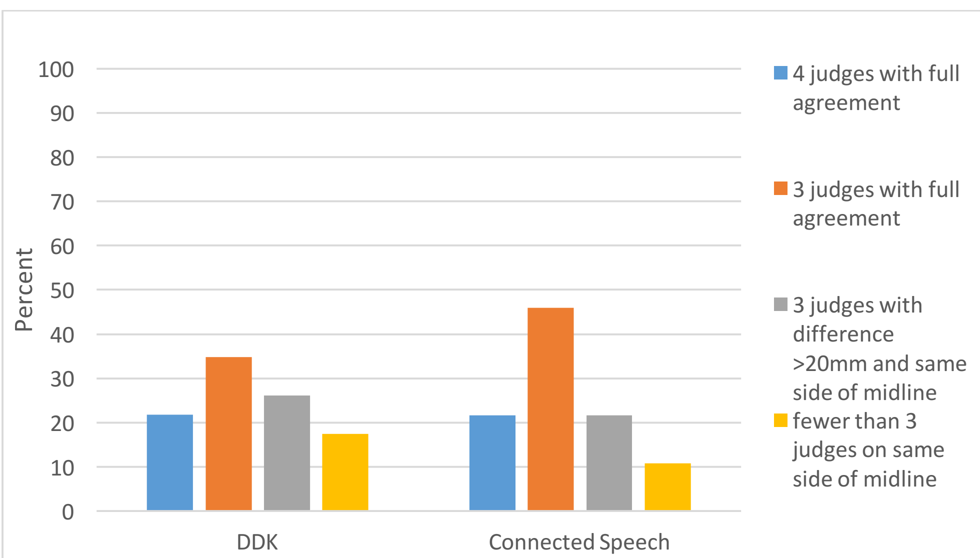


Figure 10: Percent inter-rater agreement for VAS ratings, by speaking task. Full agreement is defined as a difference < 20 mm and rating on same side of midline.

Inter-rater reliability – Mayo scale ratings

For the severity ratings, inter-rater reliability of the 5-point Mayo scale was statistically significant across raters, with correlation coefficients ranging from .699 to .780 (see Table 10). Descriptive analysis of inter-rater reliability is also shown in Figure 11, which indicates that the raters were most often within one severity level across each speaking task.

Table 10: Inter-rater agreement (Spearman's rank correlation coefficient) by rater, collapsed across speaking tasks, for Mayo scale severity ratings.

		R1	R2	R3	R4
R1	Correlation Coefficient	1.000	.737	.699	.780
	Sig. (2-tailed)	n/a	.000*	.000*	.000*
R2	Correlation Coefficient	.737	1.000	.748	.716
	Sig. (2-tailed)	.000*	n/a	.000*	.000*
R3	Correlation Coefficient	.699	.748	1.000	.701
	Sig. (2-tailed)	.000*	.000*	n/a	.000*
R4	Correlation Coefficient	.780	.716	.701	1.000
	Sig. (2-tailed)	.000*	.000*	.000*	n/a

**Correlation is significant at the 0.01 level (2-tailed)*

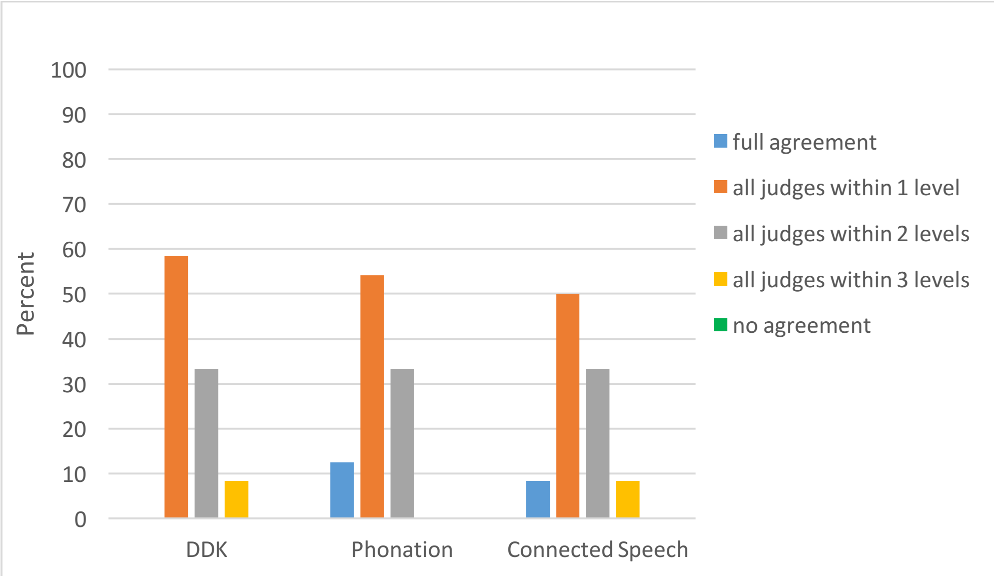


Figure 11: Percent inter-rater agreement for Mayo scale ratings, by speaking task.

<u>Testing the hypotheses</u>

Instability/Inflexibility Hypothesis: VAS ratings

Ratings on the 100mm VAS scale were analyzed according to side of midline. Ratings below 45 reflected items associated with instability, while ratings above 55 reflected items associated with inflexibility. Ratings between 45 and 55 were considered equal or uncertain attribution of instability and inflexibility. Membership in a given subgroup was based on the following criterion: 20% difference between instability/inflexibility in both the DDK and conversational speech ratings. The percent of ratings falling into either theoretical category across all speakers is illustrated in Figure 12.

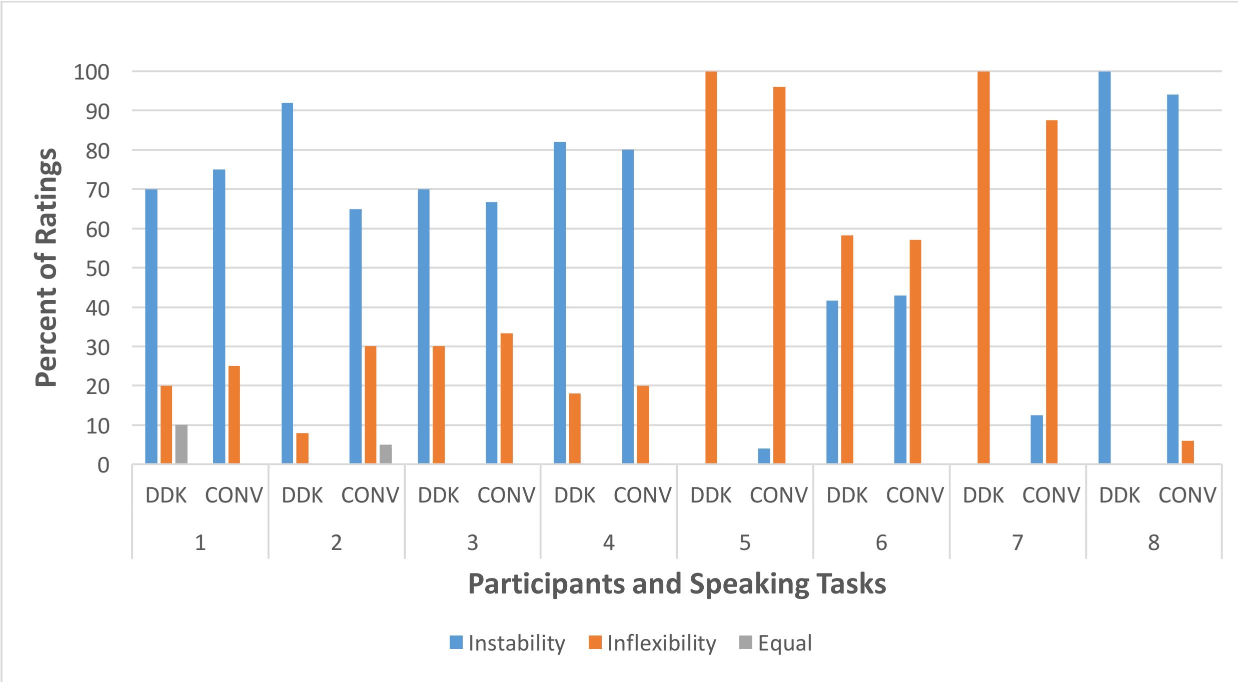
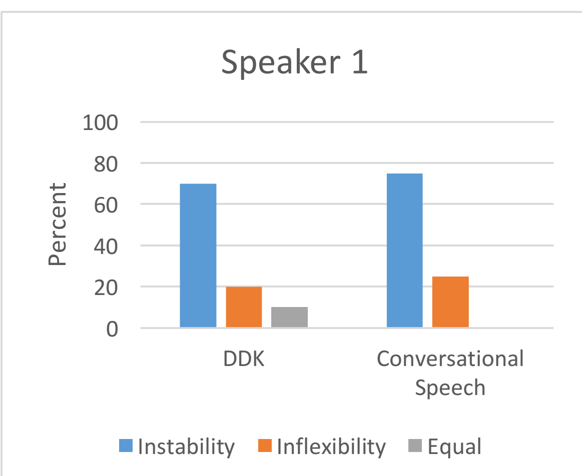
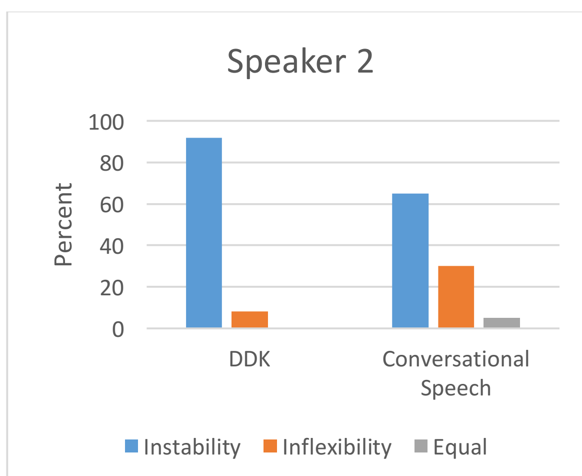


Figure 12: Percent of VAS ratings reflecting instability (<45mm) and inflexibility (>55mm) for each speaker, by speaking task.

<u>Instability</u>

Individual speaker patterns of instability are visually depicted in Figures 13-17. The instability subgroup had five speakers who fit the proposed profile (Speakers 1, 2, 3, 4, and 8). Speaker 8 had the strongest subgrouping, with 100% of DDK ratings and 94% of connected speech ratings aligning with instability.

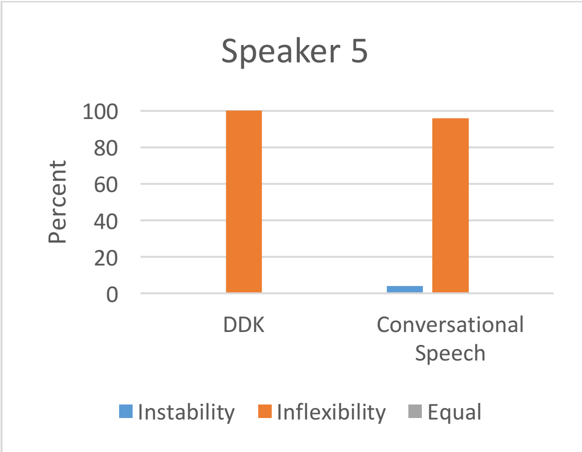


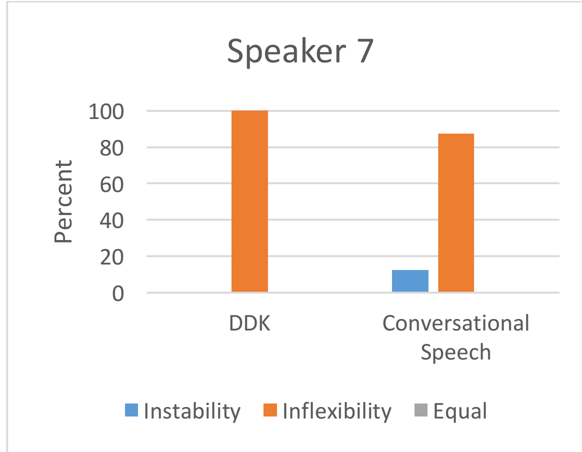


Figures 13-17: Percent of VAS ratings falling on the either side of midline, collapsed across four raters, for speakers fitting the "instability" profile.

<u>Inflexibility</u>

Patterns of inflexibility are visually depicted in Figures 18-19. The inflexibility subgroup had two speakers who fit the proposed profile (Speakers 5 and 7). DDK ratings appeared to be more sensitive to inflexibility, as 100% of DDK ratings aligned with inflexibility, for both speakers.





Figures 18-19: Percent of VAS ratings falling on the either side of midline, collapsed across four raters, for speakers fitting the "inflexibility" profile.

<u>Mixed Instability/Inflexibility</u>

One speaker (Speaker 6) did not meet criteria for membership in either subgroup, but rather fits a patterns of mixed instability/inflexibility. In the DDK task, Speaker 6 showed only a 16.7% difference between inflexibility and instability ratings (41.6% instability; 58.3% inflexibility). In conversational speech, this speaker only had a 14.2% difference between inflexibility and instability ratings (42.9% instability; 57.1% inflexibility). This pattern is visually depicted in Figure 20.

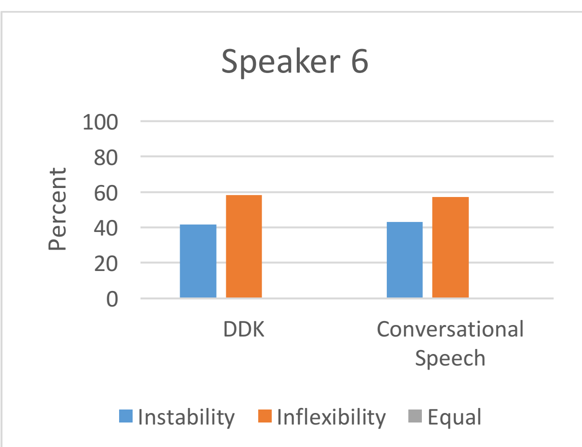


Figure 20: Percent of VAS ratings falling on the either side of midline, across four raters, for speakers fitting the "mixed instability/inflexibility" profile.

Differential Subsystem Hypothesis (Mayo scale ratings)

Mayo Clinic scale severity ratings for DDKs and conversational speech were used to determine whether speakers could be grouped based on differential speech subsystem involvement. Average Mayo scale ratings were calculated for each speech characteristic. For conversational speech, average ratings for stress, rate, and pitch were collapsed into a composite prosody score. This collapsing was justified, as all three scores were within one point or less for all participants. Loudness was not incorporated into this composite or into visual analysis, since it uniformly spans numerous subsystems.

<u>DDKs</u>

As demonstrated in Figure 21, below, none of the speakers showed a significant (greater than one point) difference between the speech subsystems, according to Mayo scale rating for the DDK task.

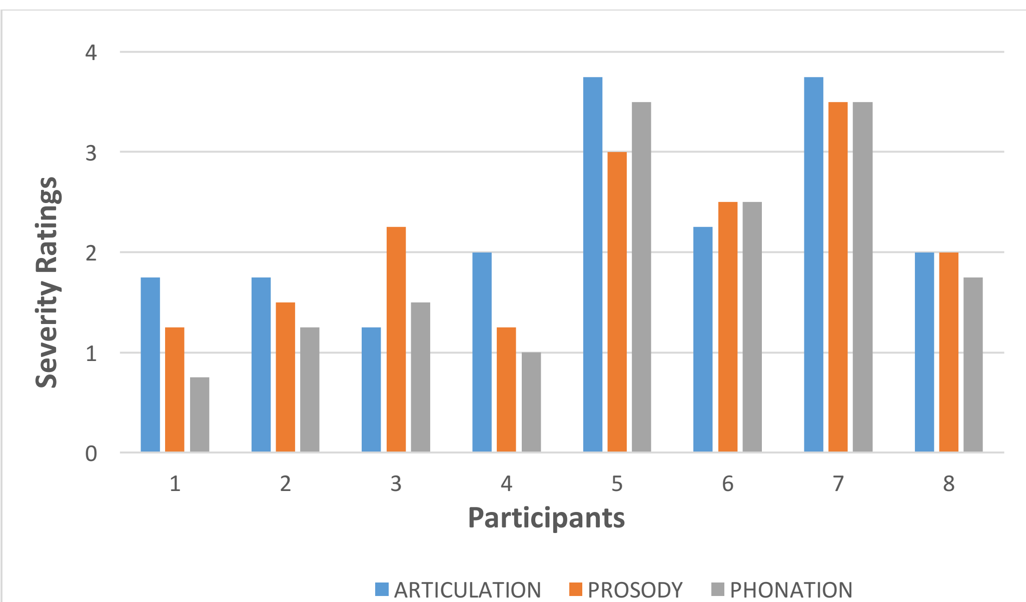


Figure 21: Average severity ratings by speaker for DDK task, according to speech subsystem, based on Mayo scale.

Conversational Speech

Average Mayo scale ratings for articulation, prosody (composite), and respiration during conversational speech are shown in Figure 22, below. Three of eight speakers had a greater than one point difference between two of the subsystems. Speaker 4 showed a 1.25 point difference between articulation and respiration. Speaker 5 showed a 1.08 point difference between articulation and prosody, and a 1.25 point difference between articulation and respiration. Speaker 8 showed a 1.33 point difference between articulation and prosody, and a two point difference between articulation and respiration.

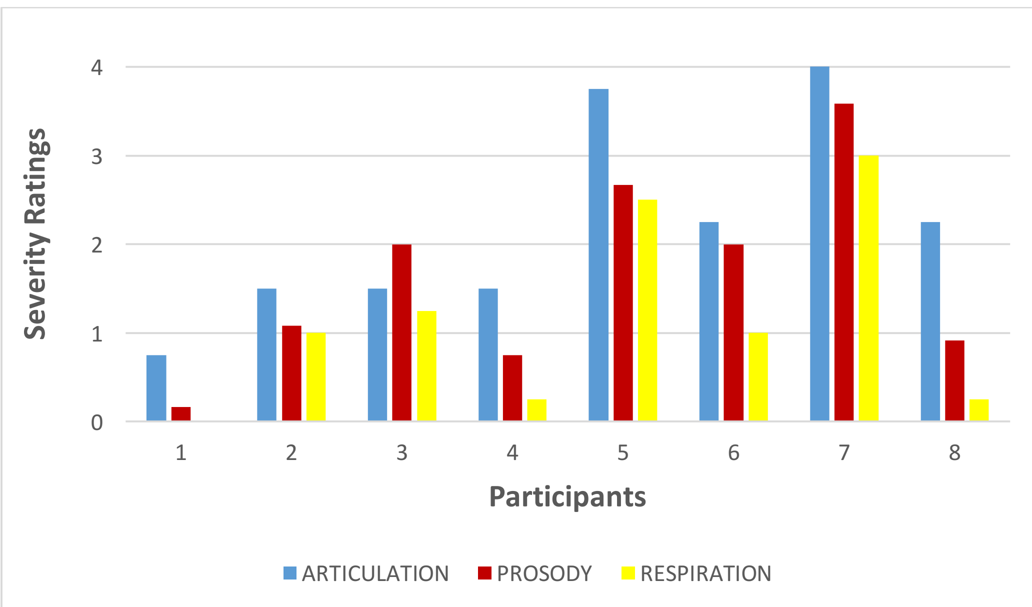


Figure 22: Average Mayo scale ratings by speaker and subsystem, for connected speech.

**Note: Prosody is a composite score combining rate, pitch, and stress. Loudness ratings were excluded from analysis. Mayo Clinic Rating Scale: 0 = absent, 1 = mild, 2 = moderate, 3 = marked, 4 = severe.*

Measuring Response to Instruction

Comparing standard, slow, and fast DDKs

In order to determine whether speakers were able to volitionally increase or decrease their AMR rates, the standard deviation (SD) of rate across syllables (/pa/, /ta/, and /ka/) was calculated for each individual participant. If that speaker's fast/slow productions were within ±1 SD, this was not considered a change from the standard rate, since the speaker was unable to surpass their own variation to establish an alternate rate. Rates either faster or slower by >1 SD were considered "alternate". No participant was able to surpass their standard rate across all three AMR targets (/pa/, /ta/, and /ka/). Two participants were successfully faster across two AMR targets; one participant was successful with one AMR target; and the remaining five participants were unable to markedly increase their rate across all three AMR targets. Speakers were generally more successful in the slow condition: five of eight participants were able to slow their rate >1 SD across all three AMR targets.

Perceptual rating of clear speech monologue

In order to determine whether speakers were able to modulate speech characteristics to increase intelligibility, connected speech was perceptually rated by one judge in both habitual and clear speech conditions. The judge was blind to initial ratings made in the habitual condition. Results of perceptual ratings of habitual vs. loud conversational speech are summarized in Table 11. Overall, Mayo severity ratings were stable, and VAS ratings did not reflect a shift across midline, with the exception of A6 who presented with a mixed profile. Detailed interpretations of these data can be found in Appendix D.

Table 11: Perceptual ratings of habitual monologue compared to monologue with "clear speech" instruction.

		Articulation Errors		Stress Errors		Abnormal Pitch		Abnormal Speech Breathing	
		Mayo	VAS	Mayo	VAS	Mayo	VAS	Mayo	VAS
A1	Habitual	1	0	0	N/A	0	N/A	0	N/A
	Clear	0	N/A	0	N/A	0	N/A	0	N/A
A2	Habitual	1	0	1	21	0	N/A	1	17
	Clear	1	5	0	N/A	0	N/A	0	N/A
A3	Habitual	1	11	2	71	1	54	1	39
	Clear	1	3	2	91	1	55	1	46
A4	Habitual	1	0	2	5	0	N/A	0	N/A
	Clear	2	4	1	41	0	N/A	0	N/A
A5	Habitual	4	100	4	94	3	97	3	94
	Clear	4	100	3	98	3	97	3	65
A6	Habitual	3	46	3	93	1	55	1	15
	Clear	3	78	2	75	1	42	1	2
A7	Habitual	4	100	3	97	3	96	3	99
	Clear	4	100	4	99	3	97	2	94
A8	Habitual	2	3	1	25	1	24	0	N/A
	Clear	1	4	0	N/A	0	N/A	0	N/A

Discussion

The goal of this study was to extend the line of research seeking to understand whether subgroups exist within ataxic dysarthria. Motivated by the pilot study by Spencer & France (2015), the current study sought to determine whether subgroups would emerge based on perceptual ratings of real speakers across three tasks: DDKs, sustained phonation, and connected speech. Two competing hypothesis were tested: (1) Speakers will cluster into instability/inflexibility (or mixed presentation) subgroups based on Visual Analog Scale patterns, or (2) speakers will cluster into subgroups based on Mayo scale severity ratings across speech subsystems of articulation, phonation, respiration, and prosody. According to the results summarized above, subgroups emerged from VAS ratings, and did not emerge from Mayo scale severity ratings. These results are consistent with the findings of Spencer & France (2015) and support the instability/inflexibility hypothesis of Hartelius and colleagues (2000).

Instability/Inflexibility hypothesis (Visual Analog Scale ratings)

Patterns emerged according to VAS ratings for all speakers – seven speakers were judged to align with either instability or inflexibility groups, while one speaker showed equal categorization and was therefore categorized as "mixed instability/inflexibility" (along similar guidelines as Spencer & France, 2015).

Instability Subgroup

Speakers 1, 2, 3, 4, and 8 demonstrated speech characteristics rated by the judges to align with a pattern of instability (or unusual variability). Both the maximum performance task (DDKs) as well as connected speech task were able to capture variability within speech characteristics. However, the DDK ratings were more sensitive to variability than connected speech for all but one of these five speakers. Given the constraints on rate, rhythm, and articulatory precision imposed by the DDK task, this aligns with expectations, as well as corroborates similar findings from Spencer & France (2015). Speech DDKs require a speaker to produce stable, equally timed syllable repetitions, so this task highlights any uncontrollable variability within articulation (variable imprecision), voicing (variable voicing errors), and rate (variable rate of repetition). By its very nature, connected speech is less sensitive to variability due to the innate variability of rate and rhythm that occur within normal conversational speech. Additionally, perceptual judgments of specific speech characteristics are naturally more difficult in the context of minimally structured conversation. Despite this, abnormal variability was still perceived in the connected speech of these five speakers.

This group was the most populated, with over half of the participants aligning with instability. This finding is similar to that of Spencer & France (2015), and reinforces the importance of the cerebellar control circuit for the timing of movements. The instability profile most clearly reflects the most salient and classically observed characteristic of ataxic dysarthria – irregular articulatory breakdowns. Breakdowns in timing and coordination result in irregular errors. In connected speech, raters were most likely to perceive instability in regards to articulation errors than characteristics related to stress, pitch, loudness, and speech breathing. Speaker 2 was an exception to this pattern, with the majority of their instability ratings falling under abnormal loudness (excess loudness fluctuations) and abnormal speech breathing (highly variable speech groups).

While no statistical tests were performed, there was no apparent relationship between membership in the instability group and etiology, symptom duration, speaker age, or form of spinocerebellar degeneration. However, there does appear to be a relationship between instability and overall dysarthria severity (as measured by the Sentence Intelligibility Test). The five speakers within this subgroup had the five highest mean SIT scores (all above 94%). This finding provides partial support for the hypothesis by Ackermann et al. (2007) – that cerebellar dysfunction happens in two stages, where ataxic speech first exhibits temporal instability and then evolves into inflexibility, or 'scanning' speech. However, these speakers ranged in time post-onset of symptomatic cerebellar degeneration from 3 years to 24 years. While they could all potentially progress to a more inflexible speech profile as disease progression ensues, it is also possible that this speaking pattern will maintain for the duration of the disease. Alternately, speech characteristics aligning with instability (irregular articulatory breakdowns, variable rate and stress, etc.) may be less impactful on overall intelligibility, rather than the reverse. For example, variable and inconsistent articulatory imprecision may lead to higher intelligibility than pervasive and constant articulatory imprecision. The relationship between instability/inflexibility and intelligibility is further discussed below.

Inflexibility Subgroup

Speakers 5 and 7 demonstrated speech characteristics rated by the judges to align with a pattern of inflexibility (or unusual stability). While only two speakers met criteria for membership in this group, their membership can be considered relatively strong – with 100% of DDK ratings reflecting inflexibility for both speakers, and greater than 87% of connected speech ratings reflecting inflexibility for both speakers. The DDK task was also more sensitive to inflexibility for these speakers. Perceptual correlates of inflexibility were judged to be excess and equal stress, consistently slow rate, consistent articulatory breakdowns, monopitch, and monoloudness. Additionally, both speakers were judged to have abnormal speech breathing, resulting in consistently short phrases.

Both speakers in the inflexibility subgroups were noted by all four judges to exhibit spasticity in regards to vocal quality (mixed ataxic-spastic dysarthria). Spasticity may have influenced these ratings, as it can result in consistently short phrases, as well as the perceptual correlates mentioned above. However, other speakers, who were classified by the judges to align with instability, exhibited spasticity as well. Additional research should be done to fully examine the influence of spasticity on perceptual ratings in instability and inflexibility.

There was no apparent relationship between membership in the inflexibility group and etiology, symptom duration, speaker age, or form of spinocerebellar degeneration. However, there does appear to be a relationship between inflexibility and overall dysarthria severity (as measured by the Sentence Intelligibility Test). The two speakers within this subgroup had the two lowest mean SIT scores (14% and 35%). Perceptual rating of many speech characteristics became more difficult as intelligibility decreased, according to anecdotal reports from the raters. For example, rating the consistency or variability of articulatory errors became problematic when a speaker had highly reduced intelligibility in connected speech, as these two speakers did. Therefore, the rating forms may not have been as sensitive to instability for these speakers, due to their overall severity. Alternately, the features than align with inflexibility (e.g., consistently short phrases, excess and equal stress, and consistent articulatory breakdowns) may lead to reduced intelligibility in a more salient manner than those aligning with instability.

Mixed Instability/Inflexibility Subgroup

Speaker 6 did not meet criteria for membership in either subgroup, but rather showed fairly equalized ratings across instability and inflexibility. While Duffy (2013; personal communication) proposed this as the most common subgroup, it was the least populated for this study. For this speaker, ratings tended not to cluster at either end of the VAS scale (near 0 or near 100) but rather closer to midline; 75% of DDK VAS ratings and 62% of connected speech

VAS ratings were between 25 and 75. Additionally, certain features were perceived as variable while others were perceived as stable. For example, the majority of rate and stress judgments aligned with inflexibility (consistently slow rate and excess and equal stress), while the majority of articulation judgments aligned with instability (irregular articulatory breakdowns). These perceptual characteristics led to a mixed profile in both DDKs and connected speech.

As with the above mentioned instability and inflexibility subgroups, no apparent relationship was seen between the mixed profile and etiology, symptom duration, or speaker age. This speaker had a mean SIT score of 85% (closest to the mean across speakers; 77%). With only one member in this group, conclusions cannot be drawn about the relationship between the mixed profile and overall dysarthria severity.

Subsystem hypothesis (Mayo Clinic Scale ratings)

The hypothesis that subgroups would emerge based on differential subsystem involvement, as measured by Mayo scale severity ratings across speech characteristics in all three speech tasks, was not substantiated. While there was some indication of differential subsystem involvement for two speakers (Speakers 5 and 8 showed relatively more severe ratings in articulation over prosody across tasks), no pattern emerged for the majority of participants. This finding is counter to that of Joanette and Dudley (1980), whose participants were also comprised of speakers with spinocerebellar ataxias. Additionally, Darley and colleagues' (1969) clusters ("articulatory inaccuracy," "prosodic excess," and "phonatory-prosodic insufficiency") did not sufficiently capture the speaking patterns of these participants, based on the Mayo scale severity ratings. This may reflect a lack of subgroupings based on these subsystems, or a lack of sensitivity of our rating forms to successfully capture differential subsystem involvement.

Response to instruction

Some of the articulatory features in ataxic dysarthria may be the consequence of compensatory strategies employed to overcome faulty coordination of multi-articulator movement sequences (Kent, Kent, Weismer, & Duffy, 2000). These compensations are difficult to distinguish from the disorder itself. One method for better understanding the contribution of compensation versus the core characteristics of the dysarthria is to ask the speaker to intentionally vary their speech pattern. In the present study, when cued to volitionally vary their speech (increase/decrease rate during DDKs; increase clarity during connected speech), most speakers were unable to do so. Exceptions were found, however. Importantly, these exceptions did not align with particular subgroupings.

There was no apparent relationship between a speaker's ability to increase DDK rate and their membership to a certain subgroup. The two speakers (Speaker 2 and Speaker 5) who were able to increase DDK rate in two out of three syllable contexts belonged to different subgroups. Five out of eight speakers (Speakers 3, 4, 5, 7, and 8) were able to slow their DDK rates in all three syllable contexts. Again, these speakers belonged to both instability and inflexibility subgroups. These results demonstrate the unlikelihood of the perceptual existence of the inflexibility subgroup being a result of compensation rather than due to physiologic impairment. If speakers were compensating for variability by slowing their rate and imposing an isochronous rhythm in order to increase intelligibility, they should be able to subsequently increase rate upon instruction, compromising stability; however, this pattern was not seen.

Similarly, there was no apparent relationship between responsiveness to “clear speech” instruction and membership in a given subgroup. Only one speaker (Speaker 6) demonstrated a shift between subgroups upon “clear speech” instruction. However, this subject was classified as a mixed profile, so a tendency to shift between subgroups in connected speech ratings is unsurprising. These results further corroborate the notion that the instability subgroup cannot be compensated for through volitional speaker control (conscious or unconscious).

Clinical and Research Implications

The “gold standard” for the diagnosis of dysarthria is perceptual judgment. That is, speech-language pathologists and researchers are trained to recognize the patterns of speech disturbance that arise when different motor systems are compromised (e.g., cerebellar, basal ganglia, upper motor neuron, etc). Yet, the heterogeneity of symptoms related to ataxic dysarthria creates confusion among clinicians and likely contributes to the wide variability of treatment outcomes.

Identifying subgroups of ataxic dysarthria would have several important outcomes. First, it would facilitate differential speech diagnosis which is particularly important as dysarthria can be the initial symptom of cerebellar degeneration (Whaley et al., 2011), and can be differentially susceptible to cerebellar pathology than limb/trunk function (Brendel et al., 2013). Second, it would lead to more refined selection of participants in research on ataxic dysarthria as opposed to inclusion of speakers with disparate speech patterns. Third, treatment for ataxic dysarthria could be better tailored. For example, the treatment approach for someone fitting the inflexibility profile may preferentially focus on activities that bring variability to speech, such as contrastive stress or flexible breath groups. Conversely, the treatment approach for someone fitting the instability profile may target activities to bring stability to speech, such as attenuated use of pitch/loudness for emphasis or regulated breath groups. Finally, identification of subgroups could inform theoretical and clinical models of cerebellar functioning and disease, which remain limited.

This study provides additional support for the notion that heterogeneity exists within ataxic dysarthria. Clustering of speakers into patterns of instability or inflexibility was also supported, providing further evidence on which perceptual characteristics indicate membership in a given subgroup. As mentioned above, understanding subgroups can guide clinicians in modifying treatment, to either target a decrease in excess speech variability (for those in the instability group), or target the use of more typical speech variability (for those in the inflexibility group).

The findings related to response to instruction also have potential for clinical application. For example, while volitional control over speech features was limited for most participants, the majority of speakers were able to slow rate during DDKs and some speakers were able to increase clarity through increased loudness and exaggerated articulation. Additionally, while subgroup shifts were not seen during "clear speech," five speakers were noted by the rater to have increased intelligibility in this condition, with only the brief instruction described above (Tjaden et al., 2013). This may be applied in a clinical context as a behavioral strategy for increasing intelligibility of some speakers with ataxic dysarthria. In addition to these clinical applications, the results of the current study inform the direction of future research and move the literature one step closer to reliably understanding and defining the proposed subgroups.

Limitations and Future Directions

This study was limited by several factors. First, only eight speakers meeting inclusion criteria were found in the timeframe of this project. Ataxia is a relatively rare condition, so additional time and resources should be dedicated towards finding appropriate subjects for future studies. Additionally, due to the lack of individuals with ataxia, inclusion criteria was expanded to include speakers with mixed ataxias, rather than pure ataxic dysarthria. This may have impacted the results, especially in regards to the presence of spasticity and its impact on speech features across subsystems. Furthermore, all eight speakers were those with degenerative spinocerebellar ataxias (SCA2, SCA3, SCA6, and Friedreich's ataxia). Acquired etiologies, such as cerebellar stroke or tumor, were not represented. Future research should seek to include other such speakers in order to determine whether a relationship exists between mechanism of cerebellar damage and subgroup membership.

The small sample size made statistical cluster analysis unfeasible, so the results relied on visual and descriptive statistics, which may be perceived as less powerful. Future studies would ideally perform cluster analyses along with visual and qualitative analyses in order to provide stronger evidence for the existence of perceptual subgroups.

The current study was also limited by the subjective nature of perceptual judgment, and the sensitivity of the rating forms to capture subtle perceptual differences in speech features. While perceptual judgment is the gold standard of dysarthria diagnosis, the addition of corroborative acoustic or instrumental analyses may have been helpful. Even the four internationally recognized dysarthria experts who served as judges expressed difficulty making severity judgments and VAS ratings throughout the rating process. The rating forms should be further improved upon to increase clarity and ease of use.

While overall rater reliability was considered good, there were factors influencing reliability which limit the results of this study. For example, the VAS ratings were variable due to the nature of the 100mm scale. The scale was modified to reflect a different type of judgment than a true VAS (which typically ranges from absent/normal [0] to severe [100]). Our VAS did not reflect a severity range, but two sides of a motor control continuum, which was unfamiliar to the judges. This unfamiliarity may have compromised reliability and therefore decreased strength of the results. Though instructional anchors were given, future research should include additional rater training, or use another method to perceptually capture the distinction between instability and inflexibility within speech characteristics. Additionally, since certain speech features were not present for some speakers within some tasks (therefore precluding VAS rating for that feature), there was a range of the overall number of repeated judgments when assessing intra-rater reliability. A smaller data set may have led to reduced reliability for these raters.

Despite these limitations, this study adds to the current body of work attempting to understand the perceptual subgroups of ataxic dysarthria which have been clinically observed for decades. Based on these results, which are in agreement with Spencer & France (2015), speakers with ataxic dysarthria can be clustered into subgroups based on instability (variability), inflexibility (stability), or a mixed presentation. It is clear from the literature and the present study that pronounced heterogeneity exists within ataxic dysarthria. This heterogeneity must be further understood and classified in order to best serve individuals with ataxia, in diagnosis and treatment of dysarthria, and therefore improve their communication and quality of life.

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Appendix A: Participant Protocol

1. Provide and explain informed consent paperwork and purpose of the study.
2. Hearing Screening (50dB or less at 500, 1000, and 2000 Hz)
3. Participant Intake Interview (Appendix B)
4. Mini Mental State Examination
5. Beck Depression Inventory II
6. Computerized version of Speech Intelligibility Test – sentence level (SIT)

Speech Tasks (7-10) to be administered in random order (clear speech monologue and lloud condition DDKs grouped for recording purposes)

7. Speech diadochokinetic tasks
    a. Standard Condition (all to be administered x2)
        i. "Repeat /pa/ as quickly and as evenly as you can until I tell you to stop" plus direct model.
        ii. "Repeat /ta/ as quickly and as evenly as you can until I tell you to stop" plus direct model.
        iii. "Repeat /ka/ as quickly and as evenly as you can until I tell you to stop" plus direct model.
        iv. "Repeat /pa-ta-ka/ as quickly and as evenly as you can until I tell you to stop" plus direct model.
    b. Fast Condition
        i. "Repeat /pa/ as quickly as you can until I tell you to stop" plus direct model.
        ii. "Repeat /ta/ as quickly as you can until I tell you to stop" plus direct model.
        iii. "Repeat /ka/ as quickly as you can until I tell you to stop" plus direct model.
        iv. "Repeat /pa-ta-ka/ as quickly as you can until I tell you to stop" plus direct model.
    c. Slow and Steady Condition
        i. "Repeat /pa/ slowly and steadily until I tell you to stop" plus direct model.
        ii. "Repeat /ta/ slowly and steadily until I tell you to stop" plus direct model.
        iii. "Repeat /ka/ slowly and steadily until I tell you to stop" plus direct model.
        iv. "Repeat /pa-ta-ka/ slowly and steadily until I tell you to stop" plus direct model.
    d. Loud condition
        i. "Repeat /pa/ as quickly and evenly as you can, at 2x your normal loudness, until I tell you to stop" plus direct model.
        ii. "Repeat /ta/ as quickly and evenly as you can, at 2x your normal loudness, until I tell you to stop" plus direct model.
        iii. "Repeat /ka/ as quickly and evenly as you can, at 2x your normal loudness, until I tell you to stop" plus direct model.
        iv. "Repeat /pa-ta-ka/ as quickly and evenly as you can, at 2x your normal loudness, until I tell you to stop" plus direct model.
8. Sustained phonation task (to be administered x2)
    a. "Take a big breath and say 'ah' for a long as you can."
9. One-minute monologue task
    a. Habitual Condition
        i. "Tell me about a memorable trip you took"
    b. Clear Speech Condition

i. “Tell me about the neighborhood you grew up in. This time, talk how you might talk to someone in a noisy environment or with a person who has hearing loss. Exaggerate the movements of your mouth. You may also be louder and slower than usual. If your regular speech corresponds to a clearness of 100, you should aim for a clearness twice as food, or a clearness of 200.”

Appendix B: Intake Form

Code # ___________

Demographics

Age:

Years of Education:

Career:

1. What is your official diagnosis related to cerebellar disease? When were you given this diagnosis?

2. What are your primary symptoms?

3. How long have you had difficulty with your speech? How would you describe your speech?

4. Did you ever receive speech therapy services for your speech difficulty? Did you ever have a speech problem that wasn't related to your cerebellar disease?

5. Please list the medications you are currently taking.

6. Is English your first language?

7. Have you ever had a stroke, a traumatic brain injury, or any other neurologic disease or condition beyond cerebellar disease?

8. Are you experiencing difficulty managing the use of alcohol or drugs? ([If yes] May I give you information about the local AA/NA group for help?)

9. Do you have significant difficulty seeing or reading things that are within your reach, even when using corrective lenses?

10. What else would you like to let us know?

Appendix C: Audio file order of presentation for each expert rater.

	Rater 1	Rater 2	Rater 3	Rater 4
1	A7 Connected Speech	A6 Sustained Phonation	A6 Sustained Phonation	A7 DDK
2	A2 Sustained Phonation	A3 DDK	A6 Connected Speech	A7 Sustained Phonation
3	A1 DDK	A3 DDK	A6 DDK	A7 Connected Speech
4	A1 Sustained Phonation	A3 Sustained Phonation	A7 DDK	A2 Sustained Phonation
5	A1 Connected Speech	A3 Connected Speech	A7 Sustained Phonation	A2 DDK
6	A6 DDK	A4 Sustained Phonation	A7 Connected Speech	A2 Connected Speech
7	A6 Sustained Phonation	A4 Connected Speech	A7 DDK	A7 Connected Speech
8	A6 Connected Speech	A4 DDK	A2 DDK	A1 DDK
9	A3 Sustained Phonation	A1 Connected Speech	A2 Sustained Phonation	A8 Connected Speech
10	A3 Connected Speech	A1 Connected Speech	A2 Connected Speech	A8 Sustained Phonation
11	A3 DDK	A1 DDK	A4 Sustained Phonation	A8 DDK
12	A6 Connected Speech	A1 Sustained Phonation	A4 Connected Speech	A4 DDK
13	A5 Connected Speech	A4 Sustained Phonation	A4 DDK	A1 DDK
14	A5 DDK	A2 Sustained Phonation	A8 Connected Speech	A1 Connected Speech
15	A5 Sustained Phonation	A2 Connected Speech	A8 DDK	A1 Sustained Phonation
16	A4 DDK	A2 DDK	A8 Sustained Phonation	A5 Sustained Phonation
17	A2 Sustained Phonation	A7 DDK	A1 Sustained Phonation	A5 Connected Speech
18	A2 Connected Speech	A7 Sustained Phonation	A1 Connected Speech	A5 DDK
19	A2 DDK	A7 Connected Speech	A1 DDK	A6 Connected Speech
20	A4 DDK	A5 Connected Speech	A5 DDK	A6 DDK
21	A4 Sustained Phonation	A5 DDK	A8 Sustained Phonation	A6 Sustained Phonation
22	A4 Connected Speech	A5 Sustained Phonation	A1 Connected Speech	A4 Connected Speech
23	A7 Connected Speech	A4 Connected Speech	A3 DDK	A4 DDK
24	A7 DDK	A8 Connected Speech	A3 Sustained Phonation	A4 Sustained Phonation
25	A7 Sustained Phonation	A8 DDK	A3 Connected Speech	A2 Connected Speech
26	A8 Connected Speech	A8 Sustained Phonation	A5 Connected Speech	A3 DDK
27	A8 DDK	A6 Sustained Phonation	A5 DDK	A3 Sustained Phonation
28	A8 Sustained Phonation	A6 Connected Speech	A5 Sustained Phonation	A3 Connected Speech
29	A6 Sustained Phonation	A6 DDK	A5 Sustained Phonation	A2 Sustained Phonation

Note: Samples randomly chosen for intra-rater reliability judgments are highlighted in gray.

Appendix D: Elaboration of differences in perceptual ratings between the two connected speech conditions.

	Summary and interpretation based on Table 11 (pg. 44)
A1	1 point improvement in articulatory precision (from mild to normal). All other categories viewed as typical and stable.
A2	Articulation and pitch unchanged with clear speech. 1 point improvement (mild to normal) in stress and speech breathing. No movement between instability and inflexibility VAS groupings.
A3	No change with clear speech. No movement between instability and inflexibility VAS groupings.
A4	1 point worsening of articulatory precision. 1 point improvement with stress errors. No chance to pitch or speech breathing.
A5	No change to articulation, pitch, or speech breathing. 1 point improvement with stress errors (severe to marked). No movement between instability and inflexibility VAS groupings.
A6	No change to articulation, pitch, or speech breathing. 1 point improvement in stress errors. No movement of VAS grouping for stress or speech breathing. VAS grouping movement for articulation and pitch, but shift direction is not consistent, and scores tend to cluster in the middle (close to 50).
A7	No change in articulation or pitch. 1 point worsening in stress errors. 1 point improvement in speech breathing. No movement between instability and inflexibility VAS groupings.
A8	1 point improvement in articulatory precision (moderate to mild). 1 point improvement (mild to normal) for stress and pitch. No change in speech breathing. No movement between instability and inflexibility VAS groupings.