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Investigating Perceptual Subgroups of Ataxic Dysarthria

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Abstract

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Ataxic dysarthria can present with considerable variation of symptoms, bringing into question whether it is a single entity. Recent evidence suggests that subgroups of ataxic dysarthria may exist and likely emerge from motor control patterns of inflexibility and/or instability (Hartelius et al., 2000). To further examine this theory, eleven speakers with ataxic dysarthria completed speech diadochokinetic and polysyllabic word repetition tasks. Ten listeners perceptually rated speech samples using an online visual analog scale to identify the presence of subgroups in speakers with ataxic dysarthria. Based on the diadochokinetic tasks, namely the alternating motion rate (AMR) task, nine of the eleven speakers were found to align with the instability profile. The results complement converging evidence of the existence of subgroups and provide a potential link between the instability/inflexibility theory and the relative contribution of cerebellar feedforward/feedback mechanisms in ataxic dysarthria.

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Introduction

Ataxic dysarthria, a motor speech disorder, is a form of speech impairment that can result from cerebellar disruption. People with this dysarthria frequently face daily challenges being understood at home and in the community, and often have to cope with the stigma of sounding intoxicated. These communication difficulties can be socially isolating, have a harmful impact on a person's functioning, and thus can lead to decreased quality of life (Vogel, Folker, and Poole, 2013).

The features of ataxic dysarthria can span all subsystems of speech, but are most evident in articulation and prosody. Some of the traditionally described characteristics include excess loudness variation, irregular articulatory breakdowns, excess and equal stress, prolonged phonemes, monopitch, monoloudness, and slow rate (Duffy, 2013). Given the cerebellar control circuit's role in coordination, these speech characteristics primarily reflect errors of force, timing, speed, and direction of movement.

Ataxic dysarthria has historically been viewed as a single entity. However, studies throughout the past few decades have reported a wide range of characteristics that are often contradictory. For example, one speaker may display monopitch and monoloudness while another has highly varied pitch and loudness. This heterogeneity has prompted consideration of subgroups of ataxic dysarthria which is the focus of the proposed study.

Multiple theories have been postulated to account for the wide variation in presentation of ataxic dysarthria. One explanation posits that the speech variation might be due to *lesion location* as certain regions of the cerebellum may have specialized function for speech. For example, bilateral, superior areas appear to mediate speech motor control and the right cerebellar hemisphere seems to contribute to the planning and processing of speech (Spencer & Slocumb, 2007). However, deviant speech characteristics can result from a wide range of possible lesion sites within the cerebellum. As discussed by Kent, Kent, Weismer, and Duffy (2000), there is considerable breadth of cerebellar lesion sites that can lead to ataxic dysarthria, including the superior cerebellar vermis, both cerebellar hemispheres, paravermal and lateral aspects of the hemispheres, and the left paravermal area. To date, lesion site has not been reliably used to understand varied speech presentation in individuals with cerebellar damage. Additionally, research examining speech characteristics among patients with the same etiology have shown

varied results. For example, wide ranging motor speech outcomes have been reported for resections of comparable cerebellar tumors (e.g., Cornwell, Murdoch, and Ward, 2005) and for speakers with Friedreich's ataxia (Folker et al., 2012).

Clusters of deviant speech characteristics (subsystem hypothesis)

An alternative theory suggests that the subgroups of ataxic dysarthria can be categorized according to the *subsystems of speech* that are affected. A formative study by Darley et al. (1969) found that the speech characteristics of those with ataxic dysarthria appear to “cluster” by subsystem impairment. This study examined the speech of those with ataxic dysarthria resulting from seven different neurological disorders. The results evidenced three different clusters, termed “articulatory inaccuracy”, “prosodic excess”, and “phonatory-prosodic insufficiency”. Articulatory inaccuracy refers to imprecise consonants, irregular articulatory breakdowns, and distorted vowels. Prosodic excess refers to excess and equal stress, prolonged phonemes, prolonged intervals, and a slow rate of speech. Lastly, the third cluster of phonatory-prosodic insufficiency involves a harsh vocal quality, monopitch, and monoloudness. It was noted that some speech dimensions appeared in more than one cluster or were correlated with speech dimensions in other clusters (e.g. hypernasality correlated with imprecise consonants), thus there were substantial interrelations between the three clusters. Additionally, the contradictory nature of the prosodic excess and phonatory-prosodic insufficiency clusters in particular supports the idea that the variation within ataxic dysarthria cannot be understood simply through the lens of individual subsystems.

Instability/inflexibility hypothesis

Most germane to the current study is the theory that the presentation of ataxic dysarthria may align with a pattern of *instability or inflexibility*. Hartelius et al. (2000) first posited this theory and suggested that the deviant speech characteristics in an individual with ataxic dysarthria may reflect acoustic patterns of isochrony (inflexibility) and variability (instability). In relation to motor control, instability refers to the tendency for the output to be more variable in contexts where it is not expected. Conversely, inflexibility refers to abnormal invariance in the output. As the output relates to speech, inflexibility and instability can be used to describe patterns of perceptual and acoustic speech characteristics in individuals with ataxic dysarthria.

The type of speech associated with the inflexible pattern has often been referred to as “scanning speech”. In typical speech, some prosodic characteristics are expected to have a certain level of variability and isochrony. For example, variability is expected in syllable durations, while isochrony in syllable lengths is expected intrautterance during repetitions of the same sentence (Hartelius et al., 2000). Hartelius and colleagues examined the two patterns at the level of temporal programming, specifically testing syllabic duration, intrautterance syllable lengths, and interstress intervals. When compared to the control group, 14 speakers with multiple sclerosis and ataxic dysarthria showed: a) increased syllable durations and decreased variability, b) decreased isochrony in syllable length interutterance, and c) increased duration and variability in interstress intervals. These results pointed to patterns of both inflexibility and instability of temporal control in speakers with ataxic dysarthria. In a study by Ackermann et al. (1992), 12 patients with cerebellar infarction underwent perceptual examination for speech deficits. When considering articulation, phonetic analysis of speech samples revealed irregularly distributed articulatory deficits and slowed speech tempo to be the most common speech features. These results could point to a possible alignment with a pattern of instability as well, as irregularly distributed articulatory deficits reflect high variability. There is also some evidence for the possibility of a third subgroup that aligns with characteristics of both inflexible and unstable motor patterns, or a mixed presentation. That is to say, some individuals may demonstrate features of inflexibility in one subsystem of speech and features of instability in another, dependent on the context or speech task. For example, in a study of speech profiles of individuals with FRDA, one case was found to have speech characterized by both excess pitch variation (instability) as well as equal and excess stress and reduced pitch variation (inflexibility) (Folker et al., 2012).

Other studies consider *disease progression* as a possible source of the variation demonstrated within ataxic dysarthria. Some evidence suggests that individuals who present with instability eventually evolve into a pattern of isochrony or inflexibility in their speech profile. In a study by Ackermann et al. (2007), it was hypothesized that cerebellar dysfunction occurs in two stages, each characterized by a different pattern of speech characteristics. In the first stage, the authors suggested that speech is characterized by a pattern of instability (including highly variable syllable timing and syllable length). In the second stage, the speech purportedly evolves into “scanning speech” that is characterized by a pattern of inflexibility (“slowed and

isochronous syllable pacing”). 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.

Cerebellar control circuit theory

Finally, it has been suggested that the relative contribution of *cerebellar feedforward versus feedback control* may explain some of the variation in speech patterns. Drawing from the limb motor control literature, feedback and feedforward control models within the cerebellum work in conjunction to help train and improve accuracy of motor control. The feedforward control loop, or inverse model, refers to the neural representation of the transformation from a desired movement trajectory (of a limb, for example) to the motor commands needed to carry out the action. On the other hand, the feedback control loop is based on the trajectory error after the planned movement has been executed. It transforms the trajectory error in sensory coordinates into motor command coordinates, which are then used to train the inverse model (Wolpert et al., 1998). In this process, known as the cerebellar feedback-error-learning model (CBFELM), the sum of the feedforward and feedback control helps to produce smooth, accurate, and well-timed movements. With respect to speech, information about the intended utterance passes through the cerebellum, which helps prepare, initiate, and revise the motor plan (Kent, Kent, Weismer, & Duffy, 2000; Spencer & Slocumb, 2007). Feedforward and feedback control are involved in this process, but the extent to which the cerebellum functionally affects feedforward and feedback control in speech is unclear. In a typical speaker, it is thought that speech relies more heavily on feedforward control due to the rapid nature (Tourville & Guenther, 2011).

Parrell et al. (2017) tested the hypothesis that the cerebellum is more integral in feedforward control for speech compared to feedback control. To assess feedforward control, nineteen speakers with cerebellar degeneration were tested for speech adaption ability across trials when presented with perturbations to sensory feedback. Feedback control was assessed by

examining the patients' online corrective responses to inconsistent perturbations. It was found that patients showed a reduced ability to adapt, which is consistent with feedforward control impairment. They also showed a heightened reactive response to feedback, suggesting an increased reliance on their feedback system. Thus, the varying speech characteristics of ataxic dysarthria may reflect the degree to which feedforward versus feedback mechanism are impaired in individuals with cerebellar impairment. The potential link between theories of cerebellar feedforward/feedback mechanisms and the inflexibility/instability theory of Hartelius and colleagues is interesting and will be addressed in the forthcoming section.

Studies investigating subgroups of Ataxic Dysarthria

Numerous studies have provided perceptual and acoustic evidence for the existence of subgroups in ataxic dysarthria. Ackermann et al. (1994) examined the speech of 14 adults with a pure cerebellar syndrome (CA) as compared to 12 healthy control speakers during a sentence repetition task. It was found that some speakers with CA presented with reduced intrautterance durational variability while others presented with increased interutterance variability of syllable duration, with the latter being more common. The authors interpreted this variability as being due to a difference in speed of articulatory movements (e.g. slower movements results in durational equalization). However, the discrepant speech characteristics noted in this study may also be indicative of a tendency to align with a pattern of instability (increased variability) or inflexibility (reduced durational variability).

Boutsen et al. (1997) provided evidence of subgroups in a study of 27 adults with ataxic dysarthria. In a diadochokinetic task, variable syllable repetition rate, duration, and intensity were noted. Three subgroups emerged based on acoustic measures of durational variability. Speakers had either 1) consistency across the three utterances, 2) higher variability during /pʌ/, or 3) higher variability during /kʌ/. The authors interpreted these subgroups as stemming from the stability of articulatory timing and, to a lesser extent, intensity. Looking through the lens of the instability/inflexibility hypothesis, the first subgroup appears to align with the pattern of inflexibility, while the second and third subgroups appear to align with the pattern of instability.

As discussed previously, Hartelius et al. (2000) also studied the patterns of speech characteristics in 14 speakers with ataxic dysarthria from MS. The theory of inflexibility and

instability arose from this work, based on the variability found across speakers during sentence repetition tasks.

Testing the instability/inflexibility hypothesis: Pilot studies

Two pilot studies from our own laboratory have helped provide further evidence that subgroups of ataxic dysarthria may form based on patterns of inflexibility and instability. A study by Spencer and France (2016) used perceptual ratings of ataxic dysarthria to group speakers into one of three subgroups: (1) tendency towards instability, (2) tendency towards inflexibility, or (3) mixed presentation. The study used pre-recorded audio files from patients with ataxic dysarthria that were considered exemplary presentations. These audio files were compiled from Aronson (1993), Duffy (2013), Freed (2000), & Spencer (2004). The speakers in the sample included 4 males and 6 females, and had ataxic dysarthria resulting from a range of etiologies including cerebellar tumor, cerebellar brainstem ataxia, strokes, TBI, cerebellar degenerative disease, and Friedreich's ataxia. Speech recordings included a diadochokinetic speech task (maximum repetition of sequential and alternating syllables) as well as a connected speech task (conversation or reading). 8 speakers also performed a sustained phonation task. The listeners were 10 experienced speech-language pathologists with over 5 years of clinical experience with speakers with dysarthria. On average, the listeners had 16.3 years of experience with patients with dysarthria. A perceptual rating form was created based on the Hartelius et al. (2000) model of inflexibility and instability and was used to judge the samples (see Appendix A). Each speech sample was rated on 12 speech features among the three speech tasks. Other features not listed (such as features of respiration) were allowed to be written in to capture the listener's perception.

Dysarthria characteristics that aligned with instability were considered "unusually variable" while characteristics that aligned with inflexibility were considered "unusually consistent". Membership in a given subgroup was based on the following criterion: samples needed to have a difference greater than 20% 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 indicated that five speakers fit the pattern of instability, one speaker fit the pattern of inflexibility, and four speakers demonstrated a mixed presentation (combined instability/inflexibility).

A subsequent study by Spencer and Dawson (2018) tested the inflexibility/instability framework against the competing prediction of differential subsystem involvement. The question of interest was whether perceptual ratings across three speech tasks could be used to understand the variability of speech patterns in speakers with ataxic dysarthria. More specifically, the investigators examined if speakers would be classified into the instability/inflexibility patterns based on visual analog scale (VAS) ratings or into subsystem profiles based on presence/severity ratings of features across speech subsystems (see Appendix B for rating forms). Speech samples were audio recorded from 8 speakers (2 male, 6 female) with ataxic dysarthria from hereditary ataxia (Friedreich's ataxia or spinocerebellar ataxia). Speech elicitation tasks included a one-minute monologue, sustained phonation, and a speech diadochokinetic task (alternating and sequential motion rates). The listeners for the perceptual ratings were four internationally recognized dysarthria experts. Listeners rated the participant's speech recordings using the scales, with variability/consistency anchors specific to each speech characteristic. Membership in a given subgroup was based on the criterion discussed above (Spencer & France, 2015). Speech profiles were found to align with the instability/inflexibility framework, with the pattern of instability being most prevalent (5 speakers). Speech profiles did not emerge for the speakers using the differential subsystem framework.

Thus, evidence is emerging regarding potential subgroups of ataxic dysarthria. The most recent studies (Spencer & France, 2016; Spencer & Dawson, 2018) further support the possibility of the instability/inflexibility theory as a framework to account for the heterogeneity of ataxic dysarthria presentation. The current investigation was a partial replication and expansion of these studies. Modifications centered on 1) sample size and diversity, 2) a focus on the DDK task, and 3) inclusion of a word stress task. Studies of ataxic dysarthria would benefit from a larger sample size with a range of participants across severity, disease duration, and etiology in order to allow for a more comprehensive examination of the perceptual subgroups. Additionally, the perceptual ratings focused on the DDK tasks as the two pilot studies suggested that the DDK task was best able to highlight patterns of instability and, to some extent inflexibility, in an efficient manner. Finally, a word stress task (production of polysyllabic words) was added to underscore patterns of inflexibility (and instability) in an efficient manner. That is, patterns of equalized stress or variable stress would be amplified during production of polysyllabic words.

Rationale for current study

Thus, the present study examined whether subgroups of ataxic dysarthria can be identified based on perceptual ratings across two speaking tasks (a maximum performance diadochokinetic task and a word repetition task). The following research question was addressed: Will speakers with ataxic dysarthria present with speech profiles that align with instability/inflexibility (or mixed presentation) subgroups based on perceptual rating patterns? Based on previous literature and the preliminary studies by Spencer and France (2016) and Spencer and Dawson (2018), it is hypothesized that listeners will identify perceptual patterns of instability/inflexibility across both speech tasks.

Methods

Participants

Speakers:

Eleven adult participants, age 19-68, with an established diagnosis of ataxia and dysarthria were included in the study. Recruitment was open to individuals with acquired forms of ataxia (e.g., stroke, TBI) however, all participants presented with hereditary forms of ataxia. Participants were excluded for: a) disease duration of less than 1 year, b) presence of another neurological disorder and/or history of head trauma, c) severe or uncontrolled psychiatric disorder, d) presence of dementia based on the Mini-Mental State Examination (Folstein, Folstein, & McHugh, 1975) score of $< 26/30$, e) moderate-severe depression based on the Beck Depression Inventory-II (Beck, Steer, & Brown, 1996) score of > 19 , and f) alcohol or drug dependency. All participants were native speakers of American English with typical developmental history with respect to speech, language, and cognition, and had hearing/vision adequate for study participation. Speaker characteristics are summarized in Table 1 and Appendix C (neurologic manifestations of ataxia).

Table 1: Demographic and screening information for speakers with ataxia.

Speaker	Age	Sex	Medical Diagnosis	Years Post Diagnosis	Years of Education	BDI-II Score	MMSE Score
A1	52	Male	SCA3	14	15	6	30
A2	59	Female	SCA14	6	16	8	29
A3	57	Female	SCA7	3	15	12	30
A4	37	Male	FRDA	5	15	15	30
A5	48	Male	SCA3	3	15	5	30
A6	29	Female	Unspecified ataxia	3	16	17	30
A7	58	Female	SCA3	17	14	4	29
A8	68	Male	SCA3	10	16	1	29
A9	60	Female	SCA3	5	16	6	30
A10	19	Male	SCA2	3	13.5	2	30
A11	38	Male	SCA1	2	15	14	30
Mean	47.7			6.45	15.13	8.18	29.73
Range	19-68			2-17	13.5-16	1-17	29-30

Note. SCA: Spinocerebellar Ataxia; FRDA = Friedreich's Ataxia; BDI-II = Beck Depression Inventory II; MMSE = Mini Mental State Examination.

Dysarthria characteristics of the speakers are highlighted in Table 2. Two expert listeners independently determined dysarthria type based on recordings of the Caterpillar passage (Patel et al., 2013). There was 95.5% agreement that the speakers presented with ataxic dysarthria, with 100% consensus agreement following discussion of mixed versus single dysarthria presentation for Speaker A6. Individual speech characteristics were identified by one expert listener. Sentence intelligibility ratings were determined using the Speech Intelligibility Test (Yorkston, Beukelman, & Hakel, 1996); scores reflect the average of three independent (student) raters.

Table 2. Dysarthria characteristics of speakers with ataxia.

Speaker	Speech characteristics	Average Speech Intelligibility [†]
A1	Irregular articulatory errors, telescoping, distorted vowels	96%
A2	Irregular articulatory errors, telescoping, equal stress	96%

A3	Irregular articulation errors, decreased pitch variation, telescoping, equal stress, difficulty coordinating breathing and speech	98%
A4	Irregular articulation errors, slow rate, equal stress, prolonged phonemes	99%
A5	Irregular articulation errors, telescoping	82%
A6	Irregular articulation errors, harsh vocal quality, equal stress, distorted vowels, difficulty coordinating breathing and speech	94%
A7	Irregular articulation errors, mild telescoping, mild equal stress	97%
A8	Irregular articulation errors, telescoping, distorted vowels	89%
A9	Irregular articulation errors, telescoping	96%
A10	Irregular articulation errors, excess loudness variations, telescoping, distorted vowels, prolonged phonemes	91%
A11	Irregular articulation errors, excess and equal stress, telescoping, distorted vowels	95%

†Speech Intelligibility based on average of three raters using the Speech Intelligibility Test.

Note. Telescoping reflects syllables that are run together, giving speech an accelerated character (Duffy, 2020).

Listeners:

Perceptual ratings of the speech samples were performed independently by 10 female speech-language pathology graduate students from the University of Washington.¹ All listeners had taken the Motor Speech Disorders graduate course and were familiar with the perceptual diagnostic criteria of dysarthria and the rating of perceptual characteristics. Listeners varied in their clinical exposure to adults with dysarthria. All listeners were native speakers of American English with typical developmental history with respect to speech, language, and cognition, and adequate hearing per self-report.

Speaker Session

Speech was elicited with two tasks: a speech diadochokinetics task (DDKs; specifically alternating and sequential motion rates) and a polysyllabic word repetition task. For the DDK task, each AMR (/pa/, /ta/, and /ka/) and SMR (/pa-ta-ka/) was elicited by asking the speaker to

¹ The original plan was to recruit experienced speech-language pathologists for the perceptual rating tasks; however, due to COVID-19 setbacks and restrictions, the recruitment plan was shifted to our cohort of first- and second-year graduate students.

produce the items “as quickly and as steadily as you can” following a model by the investigator. DDK items were elicited twice to allow for possible issues such as recording errors; the first trial was used for analysis unless a recording error was present. The word repetition task consisted of three repetitions each of eight polysyllabic words (impossibility, characteristically, indistinguishable, autobiographic, identification, enthusiastically, unreliability, & industrialization) following the investigator model of a novel example word (anticipatory). Speakers were instructed to say the word three times in a typical manner. The words were selected based on polysyllabic word lists by Ma & Tan (2015) and were chosen to represent various stress patterns across 6- and 7-syllable polysyllabic words. The words were intended to sufficiently tax the speakers’ motor system while remaining feasible for any speaker to produce in a repetition task. As above, the task was elicited twice to account for any possible recording error. The two speech tasks were administered in a counterbalanced order. The Speech Intelligibility Test (Yorkston et al., 1996) was also administered as a measure of sentence-level intelligibility, and a reading passage (The Caterpillar passage, Patel et al., 2013) was used for descriptive purposes.

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 (i.e., the laboratory, participant homes, and community centers). Audio files of speech samples were edited using Audacity to include the single trial of each alternating motion rate (/pa/, /ta/, /ka/) and sequential motion rate (/pa-ta-ka/) and one series of each polysyllabic word production.

Listener Session

VAS ratings were used to test the instability/inflexibility theory. The ratings were completed in an online format using the Gorilla Experiment Builder platform. Each listener was randomly assigned to one of two experiment groups (Group A and Group B). Each experiment group contained roughly half of the eleven speakers and their audio samples in random order. See Appendix D for speakers and order of presentation for each group. The listeners were first provided with two brief audio samples for familiarization purposes. One sample exemplified a highly variable speech pattern (aligned with instability) and one sample exemplified an unusually

consistent or isochronous pattern (aligned with inflexibility). Listeners were able to play the familiarization samples as many times as they wished. Instructions for the familiarization task are presented in Appendix E.

Listeners then indicated, using an online-adapted 100-point visual analogue scale (VAS), where a specified speech characteristic fell on the continuum of consistency/variability (Spencer & France, 2016). They were instructed that the middle of the VAS scale represented equal presence of both instability and inflexibility features. They were also instructed to mark “none” in a write-in box if no deficits were noted across all perceptual characteristics being rated on a given sample. Listeners were able to play each sound file 4 times; this number was chosen to allow for an overall impression as well as a replay for each of the three individual rating tasks. See Figures 1 & 2 for the originally intended rating forms as well as the online-adapted rating forms.

Diadochokinetic Task

<p>Articulation Errors</p> <p>Errors are irregular and variably present</p>	<hr style="border: 0; border-top: 1px solid black;"/>	<p>No deficit detected <input type="checkbox"/></p> <p>Errors are consistently present</p>
<hr style="border-top: 1px dotted black;"/>		
<p>Rate of repetition</p> <p>Variable rate</p>	<hr style="border: 0; border-top: 1px solid black;"/>	<p>No deficit detected <input type="checkbox"/></p> <p>Rate is consistently slow</p>
<hr style="border-top: 1px dotted black;"/>		
<p>Voicing Errors</p> <p>Errors are irregular and variably present</p>	<hr style="border: 0; border-top: 1px solid black;"/>	<p>No deficit detected <input type="checkbox"/></p> <p>Errors are consistently present</p>

Figure 1a. Intended visual analog scale (VAS) rating form for diadochokinetic task.

The left side of the VAS scale represents the instability profile (unusual variability) while the right side represents the inflexibility profile (unusual consistency/isochrony). The mid-portion of the scale would represent characteristics of both instability and inflexibility.

DDK: /pa/

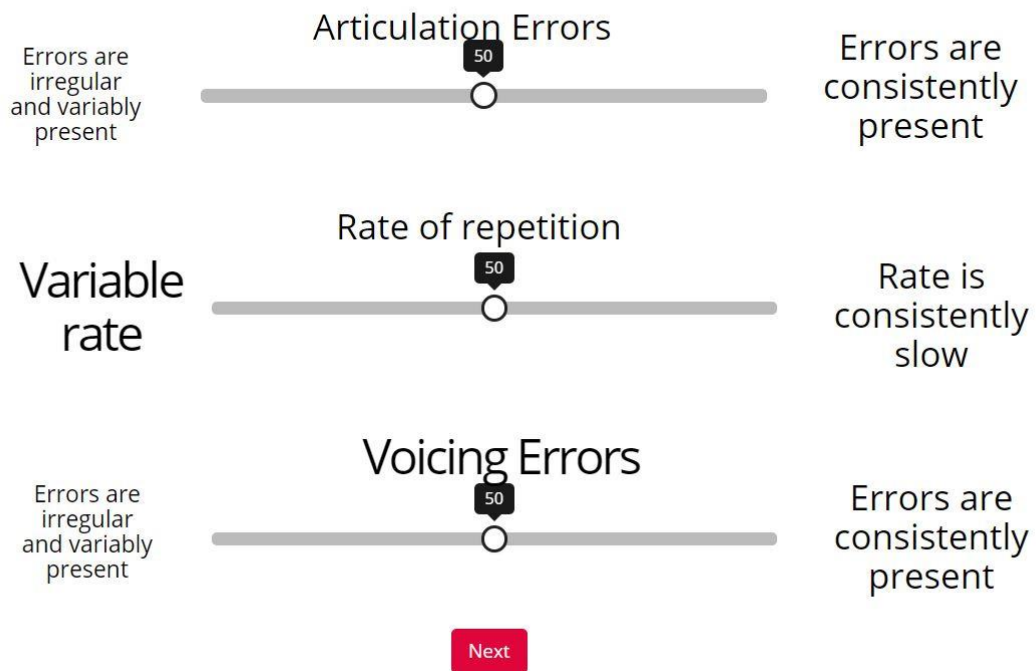


Figure 1b. Adapted visual analog scale (VAS) rating form for diadochokinetic task: Example for AMR /pa/.

Polysyllabic Word Repetition Task

Articulation Errors

Errors are irregular and variably present

No deficit detected

Errors are consistently present

.....

Stress Errors

Irregular and variable stress patterns

No deficit detected

Excess and equal stress

.....

Voicing Errors

Errors are irregular and variably present

No deficit detected

Errors are consistently present

.....

Figure 2a. Intended visual analog scale (VAS) rating form for polysyllabic word repetition task.

Polyword 1

▶ Play

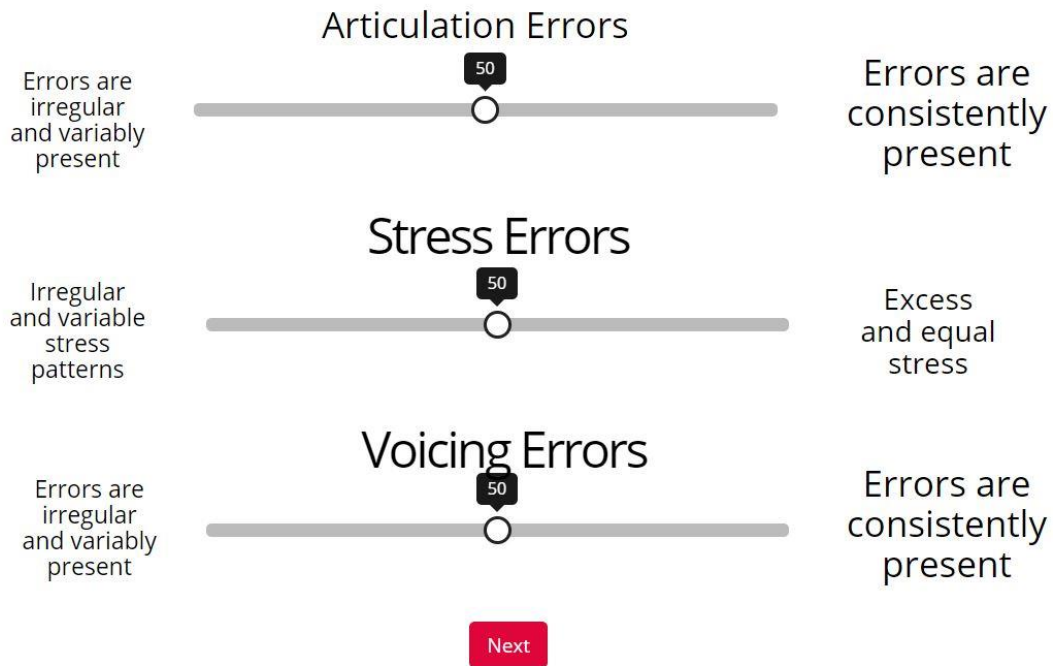


Figure 2b. Adapted visual analog scale (VAS) rating form for polysyllabic word repetition task.

Analysis

Subgroup alignment was based on visual/descriptive parameters. To determine intra-rater reliability, 20% of speaker samples (one speaker from each experiment group) were selected at random to be repeated during the listening session. For VAS ratings, intra-rater reliability of judgments was analyzed using intraclass correlation coefficients (ICC). Visual analysis of intra-rater reliability was also conducted using three levels of agreement. Full agreement was defined as ratings that are (1) within 20 points on the VAS and (2) on the same side of midline. Partial agreement was defined as ratings with greater than 20mm difference, but on the same side of midline, or ratings within 20 mm and on opposite sides of midline. No agreement was defined as ratings that are on the opposite side of midline and >20 mm apart.

Results

Reliability

Intra-rater reliability – DDK Task

Intra-rater reliability was variable across raters, with a range of intraclass correlation coefficients from -0.173 to 0.872; see Table 3. Though standard to use for intra-rater reliability, ICCs can misrepresent reliability as the coefficients reflect the sensitivity of the test to a shift in variability (that is, a rater can be highly consistently for the majority of ratings, but the shift to a few inconsistent ratings results in a marked reduction of the coefficient). Thus, visual analysis was also conducted using the criteria stated above. According to percent agreement, 64.6% of all ratings were in full agreement, with 85% of the Time 1 and Time 2 ratings having full or partial agreement (see Table 4).

Table 3. Intraclass correlation coefficients (ICC) by listener for the diadochokinetic rating task.

Listener	ICC	Significance (p)
L1	0.116	0.346
L2	0.872	0.000
L3	0.294	0.156
L4	0.433	0.072
L5	0.389	0.087
L6	-0.173	0.640
L7	0.167	0.285
L8	0.702	0.003
L9	0.081	0.390
L10	0.238	0.208

Table 4. Intra-rater agreement across listeners for the diadochokinetic rating task using visual analysis.

Level of Agreement	Definition	Percentage (across all 10 listeners)
Full agreement	<20 mm and same side of midline	64.6
Partial agreement	>20 mm and same side of midline or < 20 mm and opposite side of midline	20.4
No agreement	Opposite sides of midline and > 20 mm separation	15.0

Intra-rater reliability – Polysyllabic Word Repetition Task

Intra-rater reliability was variable across raters, with a range of ICC from -0.241 to 0.689; see Table 5. According to visual analysis, 76% of all ratings were in full agreement, with 87.8% of the Time 1 and Time 2 ratings having full or partial agreement (see Table 6).

Table 5. Intraclass correlation coefficients by listener for polysyllabic word repetition rating task.

Listener	ICC	Significance (p)
L1	0.109	0.300
L2	0.472	0.010
L3	0.689	0.000
L4	0.29	0.077
L5	-0.033	0.558
L6	0.235	0.245
L7	-0.241	0.862
L8	0.514	0.004
L9	0.014	0.476
L10	0.484	0.006

Table 6. Intra-rater agreement across listeners for the polysyllabic word rating task using visual analysis.

Level of Agreement	Definition	Percentage (across all 10 raters)
Full agreement	<20 mm and same side of midline	76.0
Partial agreement	>20 mm and same side of midline or < 20 mm and opposite side of midline	11.8
No agreement	Opposite sides of midline and > 20 mm separation	12.2

Testing Inflexibility/Instability Alignment of Speakers

VAS Ratings

Ratings on the 100-point VAS scale were analyzed according to side of midline. Ratings below 45 reflect items associated with instability, while ratings above 55 reflect items associated with inflexibility. Ratings between 45 and 55 were considered equal or uncertain attribution of instability and inflexibility. Criteria for the instability profile were that the majority of ratings for a given speaker are < 45, while criteria for the inflexibility profile were that the majority of ratings for a given speaker are > 55. The indeterminate/mixed profile would be that the majority of ratings fell between 45-55 (or if there was no majority pattern).

AMR task

Nine of the 11 total speakers (A1, A2, A4, A5, A6, A7, A8, A9, A10) met the criterion to be classified as aligning with the instability subgroup. Five of these speakers (A1, A7, A8, A9, A10) had the strongest subgroup alignment, with 100% of ratings aligning with instability. One speaker (A3) demonstrated a mixed profile and one speaker (A11) met the criterion to align with the inflexibility subgroup. Table 7 and Figure 3 depict each speaker’s average ratings across articulation, rate, and voicing.

Table 7. Average listener ratings for AMR task: Instability (0-44 mm) reflected in blue; Indeterminate (45-55 mm) reflected in yellow; Inflexibility (56-100 mm) reflected in green.

		A1	A2	A3	A4	A5	A6	A7	A8	A9	A10	A11
Articulation	pa	43.2	33.2	42.5	28	40	53.2	11.4	32.2	28	38.6	53.2
	ta	18	38.8	45	40.6	35.2	40.6	23.4	26.2	30.8	31.8	59.8
	ka	40.8	49.25	46.6	40	36.4	36.6	33.4	30.8	35	36.6	56.4
Rate	pa	10.6	22.8	47	30.2	49	66.8	19.6	35.2	10.6	13	73.8
	ta	18.8	33	32.4	46	46	67.6	22.6	32.8	24	19.4	72
	ka	35.2	37.5	21.6	56.6	40	45.2	37.4	41	27	12.2	74.6
Voice	pa	24.8	19.6	47.25	42.6	33.6	32.6	11.4	21.8	18.4	33.4	48
	ta	38.2	36.6	41.6	54.4	35.6	28.4	27.8	19.6	29.2	31.6	43.4
	ka	43	49	47.4	41.8	20.8	20.2	22.8	21.8	28.8	29.4	79.2

Note. Scores reflects the average across five listeners for each AMR and each speech characteristic.

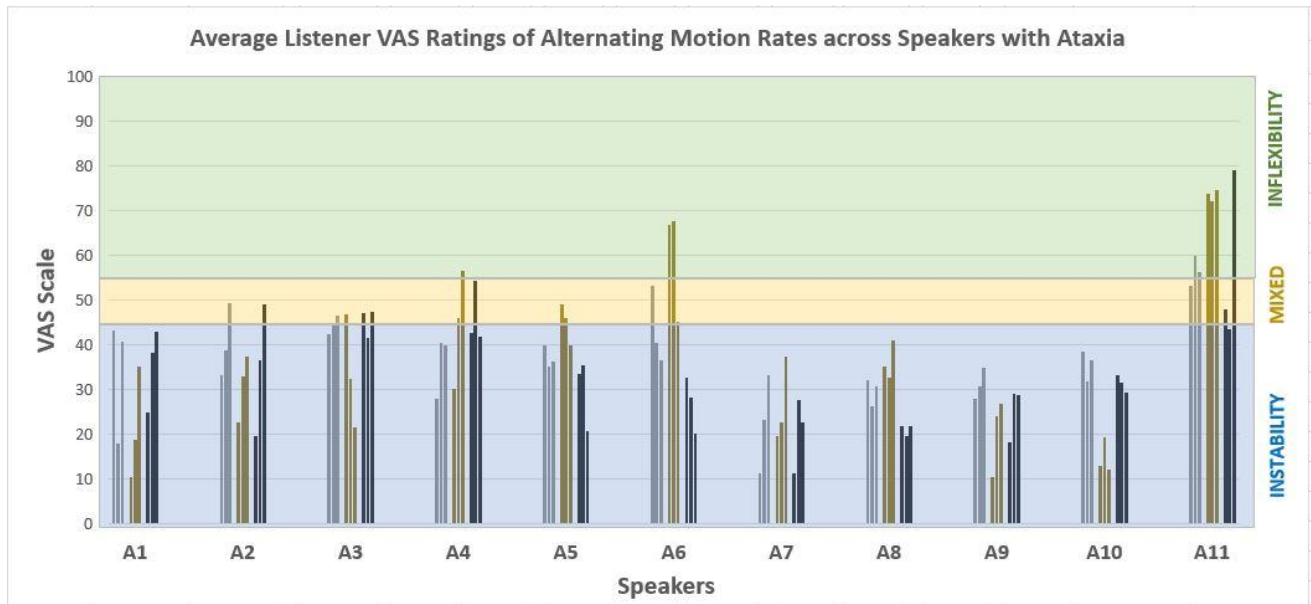


Figure 3. Average listener VAS ratings for the AMR task. Silver bars reflect articulation ratings (for /pa/, /ta/, /ka/, respectively), gold bars reflect ratings of rate, and black bars reflect ratings of voicing.

SMR task

Six of the 11 total speakers (A3, A5, A7, A8, A9, A10) met the criterion to be classified as aligning with the instability subgroup. Three of these speakers (A8, A9, A10) had the strongest subgroup alignment, with 100% of ratings aligning with instability. Two speakers (A1, A11) demonstrated a mixed profile and one speaker (A2) met the criterion to align with the inflexibility subgroup. Table 8 and Figure 4 depict each speaker’s average ratings across articulation, rate, and voicing.

Table 8. Average listener ratings for SMR task: Instability (0-44 mm) reflected in blue; Indeterminate (45-55 mm) reflected in yellow; Inflexibility (56-100 mm) reflected in green.

	A1	A2	A3	A4	A5	A6	A7	A8	A9	A10	A11
Articulation	50.8	62.25	50	47.4	41	47.4	39.8	40.4	37.5	29.8	51.6
Rate	42	58.75	38.75	72.4	43.6	56.6	52	38.2	40.75	25.8	53.2
Voicing	46	52.25	42	36.2	40.6	38.4	30.6	35.2	35	22	45

Note. Scores reflects the average across five listeners for each speech characteristic.

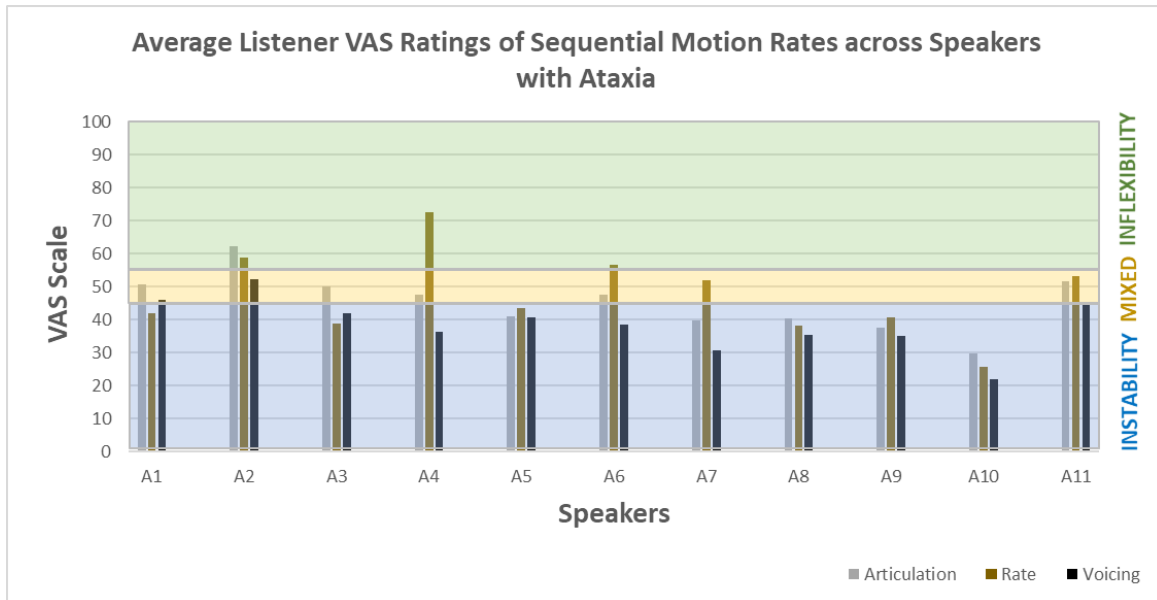


Figure 4. Average listener VAS ratings for the SMR task.

Polysyllabic Word Repetition Task

For 6 speakers (A1, A2, A3, A5, A9, & A11), the majority of their ratings aligned with a mixed profile. For 3 speakers (A4, A6, A7), the majority of their ratings aligned with the inflexibility profile, and for 2 speakers (A8 & A10), the majority of their ratings aligned with the instability profile. Figure 4 depicts each speaker’s average ratings for articulation, stress, and voicing.

Table 9. Average listener ratings for the polysyllabic word task: Instability (0-44 mm) reflected in blue; Indeterminate (45-55 mm) reflected in yellow; Inflexibility (56-100 mm) reflected in green.

	A1	A2	A3	A4	A5	A6	A7	A8	A9	A10	A11
Articulation	58.2	43.08	42.37	63.66	48.34	57.33	64.97	40.41	50.09	40.73	51.59
Stress	47.54	54.33	46.66	70.28	49.89	60.63	50.85	33.82	52.91	43.62	56.35
Voicing	48.2	48.05	51.11	51.67	51.2	53.3	63.62	45.49	46.2	47.59	51.03

Note. Each of the 8 polysyllabic words were rated independently. Scores reflects the average across the 8 words and five listeners for each speech characteristic (i.e., 8 words x 5 listeners = 40 observations per speech characteristic of articulation, stress, and voice).

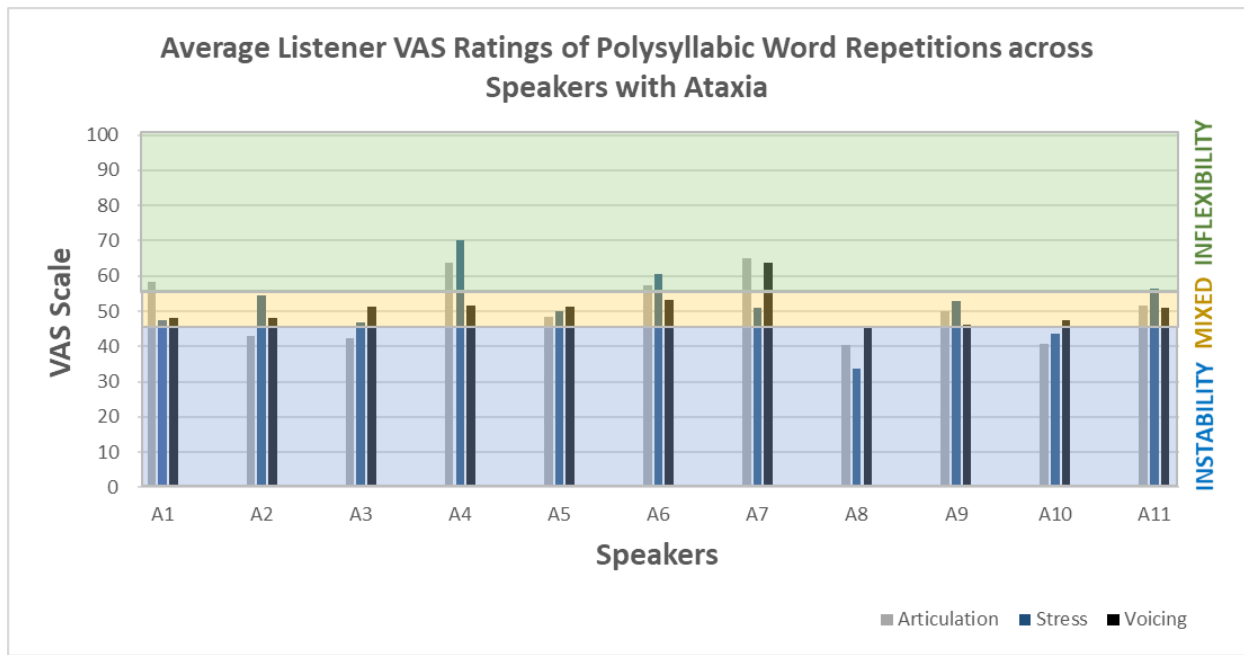


Figure 5. Average listener VAS ratings for the polysyllabic word task.

Discussion

The goal of this study was to further examine the alignment of speech profiles of individuals with ataxic dysarthria with patterns of instability, inflexibility, or a mixed presentation. Motivated by the pilot studies by Spencer & France (2016) and Spencer & Dawson (2018), the current study sought to determine whether subgroups would emerge based on perceptual ratings of speakers across two tasks: a maximum performance diadochokinetic task

and polysyllabic word repetition. Based on previous studies, it was hypothesized that listeners would identify perceptual patterns of instability/inflexibility across both speech tasks, with the subgroup of instability being most common. Our findings provide further confirmation of the existence of subgroups; these results are consistent with the findings of Spencer & France (2016) & Spencer & Dawson (2018) and support the instability/inflexibility hypothesis originally posited by Hartelius and colleagues (2000).

Diadochokinetics

The majority of the speakers met the criterion for the instability profile based on their performance on the diadochokinetic tasks (alternating motion rates and sequential motion rates). That is, the majority of the speakers were judged to present with speech patterns that were highly variable across the characteristics of articulation, rate, and voicing. As anticipated, performance on the alternating motion rate (AMR) task most strongly reflected speakers' alignment to the instability subgroup. This is consistent with the idea that repetitive alternating movements are most sensitive to perturbations of motor control and thus the instability profile. AMRs require steady motor control and are useful for judging regularity of movement (Duffy, 2020), making them ideal to capture unusual variability in a speaker who aligns with this speech profile. The vast majority of our speakers met the criterion to be classified as aligning with the instability subgroup. In fact, five of these speakers had full agreement of the instability profile across ratings of all speech tasks (/pa/, /ta/, and /ka/) and characteristics (articulation, rate, and voice).

Performance across speakers on the sequential motion rate (SMR) task demonstrates a similar tendency towards unusual variability. However, as this task is comprised of a movement sequence versus movement repetition, it was not expected to be as sensitive to regularity of motor control, and the alignment to each speech profile subgroup was not as strong.

Polysyllabic Word Repetitions

The polysyllabic word repetition task was chosen in order to best identify and highlight patterns of inflexibility. Words were chosen to reflect varied stress placement, such that the equalized stress patterns should be easily detectable. Theoretically, a speaker who aligns with the inflexibility subgroup would receive ratings of "unusually consistent", especially on the stress ratings, as this characteristic would be sensitive to an excess/equal stress pattern characteristic of

some speakers with ataxic dysarthria (Duffy, 2020). Given the nature of the task, we anticipated that speakers with inflexibility would be readily identified with polysyllabic word repetition. However, speakers with instability were anticipated to fall in the middle/indeterminate range as it is challenging to identify unusual variability with this speaking task. Indeed, based on the performance on the word repetition task, the majority of speakers aligned with the indeterminate/mixed subgroup. Speakers who fall into this category received VAS ratings near the middle of the VAS scale.

Speech Subgroup Alignment

The greater prevalence of the instability subgroup with the DDK task is consistent of that of previous studies (Spencer & France, 2016 and Spencer & Dawson, 2018), and reinforces the potential contribution of the feedforward cerebellar control loops. During speech, speakers rely more heavily on the feedforward control loop due to the rapid nature of the movements (Parrell et al, 2017). That is, accurate speech production relies more on the transformation of a desired articulatory movement to the motor command needed to complete the action. Because speech is rapid, it is not as easily informed by the feedback control loop, using the error of one movement to inform the next (Tourville & Guenther, 2011). In their study of nineteen speakers with cerebellar degeneration, Parrell and colleagues suggested that on average, participants showed a reduced ability to adapt to motor perturbations when compared to the control group, which is consistent with feedforward control impairment. Therefore, the results of the current study provide a potential link between the instability/inflexibility theory of Hartelius and colleagues (2000) and the relative contribution of cerebellar feedforward/feedback mechanisms. The majority of our speakers aligned with a pattern of instability, which is suggested to be a manifestation of a compromised cerebellar feedforward control loop (Parrell et al., 2017). Thus, the instability profile, most commonly characterized and identified by irregular articulatory breakdowns, excess loudness variation, and variable pitch and rate may reflect impaired feedforward control. As summarized by Duffy (2020), feedforward control allows the cerebellum to receive advanced notice about the syllabic content of an utterance from the cortex, so that it can refine the temporal and prosodic properties prior to motor execution. These initial motor programming refinements allow speakers to have well-timed, durationally appropriate, controlled and coordinated speech.

The inflexibility profile, on the other hand, may be similarly associated with disrupted feedforward systems, but also a subsequent overreliance on feedback control loop mechanisms. The inflexibility profile is most commonly characterized by equalized stress, monopitch, monoloudness, and an overall unusual lack of variation in perceptual characteristics or “scanning speech”. As explained by Demopoulos et al. (2018), speakers who rely on immediate sensory feedback for moment-to-moment control of speech may have difficulty flexibly adapting their speech motor programs. This could result in overreliance on feedback which would lead to reduced speech adaptation and overcompensation, and the occurrence of equalized stress patterns (Izzini-Seigel et al., 2015). Parrell and colleagues (2017) similarly found that speakers with cerebellar lesions had compensatory responses that were larger compared to controls, showing an overreliance on the feedback system.

While statistical analysis of intra-rater reliability suggested dubious levels of reliability, these analyses notoriously misrepresent actual agreement when used with visual analog scales (VAS). Levels of VAS agreement when calculated with *a priori* criteria revealed fairly strong intra-rater reliability, with 85-88% full/partial agreement across both tasks. Rater reliability may be improved with increased experience of the listener. Because of protocol changes due to COVID-19, the listeners were first- and second-year speech-language pathology graduate students with varying exposure levels to motor speech disorders, instead of experienced speech-language pathologists.

The possible existence of subgroups was originally investigated due to the variable and, at times, contradictory nature of the defining characteristics of ataxic dysarthria. The results of this study as well as those of previous studies point to the presence of subgroups within ataxic dysarthria, which matches the wide spectrum of speech characteristics perceived by clinicians (Duffy, 2020). Given the complexities of the cerebellar circuits, it reasonable to assume that neural disruption would not result in consistent, unified patterns of dysarthria across speakers. The same can be said of the basal ganglia circuits. To that end, subgroups of hypokinetic dysarthria have also been posited (Duffy, 2020). For example, a study by Sapir (2014) explored the idea that presentation of hypokinetic dysarthria in speakers with Parkinson’s disease may be based on a variety of factors other than basal ganglia pathology and lack of dopamine. Hyperkinetic dysarthria has also been noted to present differently in speakers with Huntington’s disease. Through a perceptual rating task, Diehl et al. (2019) found that four unique subgroups,

each characterized by unique speech characteristics, emerged among 48 speakers. Similar to the theories posited to explain the presence of subgroups in ataxic dysarthria, issues such as disease progression and divergent pathologic processes have been suggested to account for these subgroups and warrant further investigation.

Limitations and Future Directions

This study was limited by several factors. The group with ataxic dysarthria was smaller than anticipated because of the unexpected onset of COVID-19 public health restrictions during data collection. Ataxia is a relatively rare condition, and all speakers recruited were those with degenerative spinocerebellar ataxias (SCA1, SCA2, SCA3, SCA7, SCA14, and Freidreich's ataxia). Ataxia from acquired etiologies, such as cerebellar stroke or tumor, were not represented. Future studies should seek to include speakers of acquired etiologies in order to further investigate the relationship between mechanism of cerebellar damage and dysarthria presentation. As mentioned previously, the listeners in this study may also have affected the results. Because the listeners were students with limited experience rather than experienced speech-language pathologists as originally intended, strength of subgroup membership and intra-rater reliability may have been lower. Finally, the method of using a visual analogue scale may have limited the ratings by pre-determining perceptual characteristics. Future research should consider methodologies that avoid research-imposed constraints on the perceptual categories, such as auditory free classification (Clopper, 2008) which has recently been used to better understand dysarthric speech (Lansford et al., 2014, 2016).

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Appendix A: Perceptual rating form (Spencer and France 2016)

Listener #	Sample #			
DDKs				
Articulation	<input type="checkbox"/> <i>Variable errors</i>	<input type="checkbox"/> Consistent errors	<input type="checkbox"/> Normal	
Voicing	<input type="checkbox"/> <i>Variable errors</i>	<input type="checkbox"/> Consistent errors	<input type="checkbox"/> Normal	
Rate	<input type="checkbox"/> <i>Variable</i>	<input type="checkbox"/> Consistently slow	<input type="checkbox"/> Normal	
Rhythm	<input type="checkbox"/> <i>Variable</i>	<input type="checkbox"/> Normal		
Sustained Phonation				
Loudness	<input type="checkbox"/> <i>Variable</i>	<input type="checkbox"/> Consistently too loud	<input type="checkbox"/> Consistently too quiet	<input type="checkbox"/> Normal
Pitch	<input type="checkbox"/> <i>Variable</i>	<input type="checkbox"/> Normal		
Connected Speech				
Articulation	<input type="checkbox"/> <i>Variable errors</i>	<input type="checkbox"/> Consistent errors	<input type="checkbox"/> Normal	
Stress	<input type="checkbox"/> <i>Irregular placement</i>	<input type="checkbox"/> Equalized across syllables	<input type="checkbox"/> Normal	
Rate	<input type="checkbox"/> <i>Variable</i>	<input type="checkbox"/> Consistently slow	<input type="checkbox"/> Normal	
Pitch	<input type="checkbox"/> <i>Variable</i>	<input type="checkbox"/> Monopitch	<input type="checkbox"/> Normal	
Respiration	<input type="checkbox"/> <i>Incoordination of breath with speech</i>	<input type="checkbox"/> <i>Speech on residual air</i>	<input type="checkbox"/> Short phrases	<input type="checkbox"/> Normal
Loudness	<input type="checkbox"/> <i>Uncontrolled</i>	<input type="checkbox"/> Consistently too loud	<input type="checkbox"/> Consistently too quiet	<input type="checkbox"/> Normal

Note: Subgroup differentiation (italics, bolding) was not provided to listeners.

Appendix B: Perceptual rating forms (Spencer and Dawson 2018)

1) 5-Point Rating Scale and Visual Analog Scale Rating Form for Diadochokinetic Task

Rater: _____ Participant Code: _____

Diadochokinetic Task (AMRs/SMRs)

	0	1	2	3	4
Articulation errors	Absent	Mild	Moderate	Marked	Severe
Errors are irregular and variably present	_____				Errors are consistently present
Rate of repetition (slow)	Normal	Mild	Moderate	Marked	Severe
Variable rate	_____				Rate is consistently slow
Voicing Errors	0	1	2	3	4
Errors	Absent	Mild	Moderate	Marked	Severe
Errors are irregular and variably present	_____				Errors are consistently present

2) 5-Point Rating Scale and Visual Analog Scale Rating Form for Connected Speech 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

Abnormal speech breathing	0 Absent	1 Mild	2 Moderate	3 Marked	4 Severe
Highly variable breath groups	_____				Consistently short phrases

Appendix C. Primary neurologic symptoms reported by speakers.

A1	Impaired vision, gait
A2	Incoordination, impaired balance, double vision, spinal degeneration
A3	Impaired balance, (fine motor) dexterity, impaired gait; minor vision loss
A4	Incoordination (walking and driving), impaired fine motor skills
A5	Impaired balance and gait, impaired fine motor skills (writing)
A6	Tremors, impaired gait, balance, and fine motor skills (writing)
A7	Impaired balance, blurry vision, arm cramps
A8	Impaired gait, incoordination, difficulty with writing, vision changes
A9	Incoordination, limb pain, impaired balance/gait, double vision, difficulty falling asleep, memory impairment
A10	Tremors, mild incoordination
A11	Impaired balance/gait, chronic exhaustion, sleep difficulties, cramps in calves, limb weakness

Appendix D: Experiment groups and order of presentation of speakers:

Speakers presented	
Group A	Group B
A1	A2
A4	A3
A7	A5
A8	A6
A9	A10
A1 (repeat)	A2 (repeat)
	A11

Appendix E. Familiarization Task and Instructions for Listeners.

Hello! Thank you for participating in this experiment. Please have your headphones on as you will soon be listening to audio samples from several different speakers. Some speakers will sound more robotic, lacking the typical variability and melody of speech. They will often have:

- Equalized stress patterns
- Monopitch
- Monoloudness
- Relatively consistent articulation errors.

Here is an example of this unusually consistent way of speaking:



Other speakers are more variable than what is typical. They will often have:

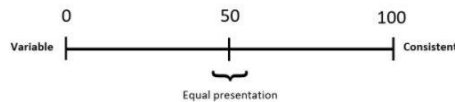
- Variable pitch
- Variable rate
- Variable loudness
- Irregular/inconsistent articulation errors.

Here is an example of this highly variable way of speaking:



Next

You will be asked to listen to audio samples and determine if the speakers sounds robotic and unusually consistent OR if they sound unusually variable. You will be asked to make these judgments across aspects of speech, such as rate, stress patterns, voicing, and articulation. For example, if someone says /pa-pa-ba-pa-ba-pa/ for the target "pa", then their voicing errors are variable and you would mark the "variable" end of the scale. A "0" would be extremely variable while a 45 would be variability that's almost not detectable. On the other hand, if the speaker says /ba-ba-ba-ba-ba-ba/ for the target "pa", then their voicing errors are consistent and you would mark on the other end of the scale (100 for extremely consistent, 55 for barely noticeable). A score in the neighborhood of 50 suggests equal presentation of variability/consistency or too difficult to differentiate. There may not be equal representation of the "subgroups" (highly variable vs robotic) within your sample, so it is fine if your ratings tend to cluster towards one side of the scale. If you don't notice any abnormality during the speaking task, please write "none" in the fill-in box in the top right corner of the screen. Please only do so if you find ALL aspects of speech to be intact.



Next

You will listen to 6 audio sets. In each set, you will hear productions of DDKs as well as 8 polysyllabic words (each word repeated 3 times). You will be able to listen to each audio 4 times. It is recommended that you first listen to the entire audio all the way through, and then listen to it once before each rating you make. You will not be able to go back to a previous screen after you continue to the next, so please make sure your ratings are final before proceeding to the next screen.

Click next to begin!

Next