Examination of Perceptual Subgroups of Ataxic Dysarthria Through Auditory Free Classification

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Abstract

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Ataxic dysarthria is highly heterogeneous in its presentation (Duffy, 2020). While the most salient and distinguishing perceptual characteristics center on articulatory and prosodic abnormalities, the manifestation can be quite variable. In fact, some characteristics are even contradictory in nature, such as prosodic excess and prosodic insufficiency. This perceptual heterogeneity has prompted research into possible subgroups within ataxic dysarthria. The current study expands upon the extant literature (e.g., Spencer, Amaral, and Lansford, 2022) which contends that subgroups are mediated by different patterns of motor control, namely an unstable pattern and an inflexible pattern (see Hartelius et al., 2000). In the present study, 25 speakers with ataxic dysarthria from a range of etiologies provided speech samples during two tasks: alternating motion rates (AMR) and connected speech (55 syllables from a reading sample). Seventeen experienced speech-language pathologists served as listeners and completed an auditory free-classification task (Clopper, 2008). Listeners grouped speakers who sounded similar based on their own judgment of the most salient perceptual characteristics. Listener ratings were compared with *a priori* expert determinations of speakers who fit the instability subgroup profile, the inflexibility subgroup profile, or a mixed subgroup of the two profiles. Results contributed to the existing evidence of perceptually distinct subgroups in ataxic dysarthria, and argue for continued investigation. The findings have implications for models of cerebellar disruption and can inform clinical diagnostic and treatment practices.

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Introduction

The cerebellum is a multi-faceted contributor to brain function, including sensorimotor and vestibular control, autonomic function, cognition, and emotion (Schmahmann, 2019). When damage or degeneration occurs in the cerebellar network, each of these distinctive functions can be negatively impacted. Most commonly associated with cerebellar disruption is motor impairment, specifically ataxia, which is linked to impaired gait, balance, and coordination of movement. Motor control challenges can also influence vision, such as nystagmus or other forms of oculomotor dysfunction (Kheradmand & Zee, 2011). Cognitive deficits from cerebellar disruption are often characterized by impairments in executive function, linguistic function, attention, memory, and visuospatial function (Tedesco, Chiricozzi, Clausi, Lupo, Molinari, & Leggo, 2011; Bodranghien et al., 2016), and may be accompanied by difficulty with emotional or affective regulation (Bodranghien et al., 2016). Most germane to this paper, cerebellar damage or degeneration can lead to the motor speech disorder of dysarthria.

Dysarthria is an umbrella term for a group of neurologic motor speech disorders that affect the execution and control of speech. Ataxic dysarthria is a specific type of dysarthria that involves damage or degeneration of the cerebellar control circuit, broadly causing insufficiency in the timing and coordination of speech (Ackermann, Mathiak, & Riecker, 2007). Etiologies of ataxic dysarthria include degenerative diseases such as hereditary spinocerebellar ataxias (SCAs), vascular disorders, traumatic injury, demyelinating disease such as multiple sclerosis, neoplastic disorders, and toxic or metabolic conditions. Due to the underlying incoordination, ataxic dysarthria can affect all subsystems of speech including respiration, phonation, resonance, articulation, and prosody. However, the most salient and distinguishing perceptual characteristics center on articulatory breakdowns and prosodic abnormalities (Duffy, 2020).

Articulation impairments are characterized by imprecise consonants and vowel distortions, and are often, but not always, irregular in presentation. Prosodic abnormalities are heterogeneous in nature and can be characterized by prosodic excess or prosodic insufficiency. For example, speakers with prosodic excess might have poor modulation of loudness and pitch, excess and equal stress, prolonged phonemes, slow rate, and prolonged intervals between speech transitions (Duffy, 2020). Conversely, speakers with prosodic insufficiency might present with monopitch and monoloudness (Spencer & Slocomb, 2007). These variable, and sometimes paradoxical, speech characteristics suggest there may be perceptually diagnosable subgroups of ataxic dysarthria.

Differential diagnosis of ataxic dysarthria subgroups, much like differential diagnosis of different types of dysarthria, would enhance specificity in clinical management and treatment, improve precision of participant selection and the focus of future research, and add to the growing body of research of the cerebellum's role in speech control and coordination. To that end, the intent of this study is to investigate perceptual subgroups of ataxic dysarthria.

Perceptual Subgroups

Perceptual subgroups of ataxic dysarthria were first explored by Boutsen, Bakker, and Duffy (1997). Twenty-seven speakers with ataxic dysarthria were asked to perform maximum syllable repetition diadochokinetic (DDK) tasks and oral reading tasks. A Q-factor analysis revealed three distinct groups of speakers based on repetition rate and intensity across DDK syllables. Group 1 presented with equalized rate and intensity across all DDK syllables (/pʌ/, /tʌ/, and /kʌ/); Group 2 and 3 demonstrated excess variability for the /pʌ/ syllable and /kʌ/ syllables. The authors concluded that ataxic dysarthria may not be a homogenous disorder in its presentation of speech characteristics stating that, "...the stability of articulatory timing, and to a small extent stability of intensity, can be differentially affected during fast syllable repetitions." (Duffy, 1997, p. 35).

A few years later, Hartelius, Runmarker, Andersen, and Nord (2000) introduced the concept of variability within ataxic dysarthria based on overall patterns related to the temporal speech characteristics of individuals with dysarthria from multiple sclerosis (MS). The investigation of the temporal characteristics was at two levels: duration and variability of syllable duration, and the duration of interstress intervals. The authors concluded that individuals with MS and ataxic dysarthria were demonstrating *inflexibility* of the temporal control of speech movements and/or *instability* of the temporal control of speech movements. Inflexibility was characterized as a reduction in variability in syllable length, rate of speech, and intonation, and was primarily perceived as insufficient in prosodic range. Instability, on the other hand, was characterized by increased variability in interstress intervals across repetitions as well as an excess in pitch and loudness variation which was primarily perceived as excess of prosodic range. Overall, the authors concluded that the results were consistent with inflexibility and instability in temporal control and served as a possible explanatory model for the contradictory perceptual characteristics.

The parallels between these two studies center on motor control and stability. That is, Group 1 from Boutsen and colleagues (1997) aligns with the inflexibility characterization from Hartelius and colleagues (2000). Conversely, Groups 2 and 3 from Boutsen and colleagues (1997) presented with excess syllable rate variation that more closely aligns with the instability description from Hartelius and colleagues (2000).

More recently, the concept of perceptual subgroups has been investigated by Spencer and colleagues. In their initial pilot study, Spencer and France (2016) used pre-recorded samples of ten individuals with ataxic dysarthria to examine the instability and inflexibility framework of Hartelius and colleagues (2000). The authors mapped perceptual characteristics onto notions of unstable and abnormally variable motor control (i.e., the instability group) and equalized, abnormally invariant motor control (i.e., the inflexibility group); see Table 1. Experienced speech-language pathologists rated perceptual speech characteristics using a categorical rating scale for the speech tasks of alternating and sequential motion rates (AMRs/SMRs) and approximately 30 sec of connected speech. For example, listeners would rate pitch during connected speech as variable, monopitch, or normal. Based on listener rating consistency and pre-established group membership criteria, the authors concluded that the most common pattern was instability, followed by a mixed profile, and inflexibility. This study provided initial evidence that speakers with ataxic dysarthria could be perceptually categorized by experienced, professional listeners into groups reflecting motor stability. However, the study was limited by the fair intra-rater reliability of some listeners, use of a categorical rating scale, use of prerecorded audio tasks, limited knowledge of the speaker's medical backgrounds, and the small sample size of ten speakers.

Table 1. Spencer and France (2016) summary of salient speech features consistent with patterns of instability versus inflexibility

	Instability	Inflexibility
Respiration	Incoordination of breathing with speaking Paradoxical breath patterning	Consistently reduced breath support
Articulation	Variable; possible undershoot and overshoot	Relatively consistent articulatory imprecision
Prosody	Inconsistent rate Uncontrolled loudness variation Variable pitch Variable stress patterns Variable rhythm (especially in AMRs/SMRs)	Decreased rate Monoloudness Monopitch Isochronous, metered stress patterns Steady rhythm (especially in AMRs/SMRs)

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

In a follow-up study, Spencer and Dawson (2018) contrasted the instability/inflexibility theory with the differential subsystem theory. That is, the authors wanted to see if the variation of perceptual speech patterns of ataxic dysarthria were due to the inflexibility/instability of motor control or to the differential impairment of subsystems of speech, such as respiration or articulation. The authors asked raters to listen to samples of AMRs/SMRs and conversational speech tasks to categorize and rate eight speakers with ataxic dysarthria. For this study, two rating scales were implemented. A visual analog scale was used, anchored by the putative perceptual speech characteristics of inflexibility and instability. Additionally, a 5-point rating scale was used to capture the presence/severity of speech characteristics across the subsystems of articulation, phonation, respiration, and prosody. This study further supported the findings of Spencer and France (2016) and aligned with the instability/inflexibility hypothesis introduced by Hartelius and colleagues (2000). Additionally, this study, while limited by a small sample size, found that there was no apparent connection between instability or inflexibility groupings and symptom duration, age, or type of spinocerebellar degeneration.

The instability/inflexibility theory was further investigated by Spencer and colleagues (Gore, 2020; Gore, Spencer, & Amaral, 2021) in 11 speakers with spinocerebellar ataxia. The authors were interested in examining the identification of subgroups based on perceptual ratings from diadochokinetic tasks, as well as a novel polysyllabic word repetition task chosen to highlight patterns of inflexibility. Listeners were 10 SLP graduate students who used online visual analog scales for their ratings. The authors reported additional evidence of the existence of subgroups, aligning with their previous work. 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 polysyllabic word repetition task was less successful at group differentiation given that it is designed to highlight patterns of inflexibility, but variability (instability) is harder to detect in single word productions.

Most recently, Spencer and colleagues (Amaral, 2021; Spencer, Amaral, & Lansford, 2022) examined subgroups of ataxic dysarthria across diadochokinetic tasks and connected speech tasks using an unconstrained auditory free-classification paradigm. This approach removed the bias associated with predetermined rating scales. The authors also added a guided classification task to compare with the auditory free classification task and to help contextualize the findings. Given the constraints of the COVID-19 pandemic, pre-recorded sound files drawn from two universities were used, and were selected based on *a priori* determinations of their fit with instability/inflexibility profiles. Listeners were SLP graduate students who were asked to organize speaker recordings into groups following the procedure of Lansford and colleagues (2014) for an auditory free classification. That is, listeners were able to create groups based on their own perceptual decisions. For the guided classification task, listeners were presented with pre-established descriptions of subtypes to then sort speaker files according to instability/variable characteristics, inflexible/ "robotic" characteristics, or indeterminate groupings.[1] Using a cluster analysis, the results of the free classification task mapped well onto *a priori* groupings of speakers with instability and inflexibility characteristics. The guided classification tasks further supported that the instability/inflexibility subgroups were significantly detectable perceptually. As with previous investigations, the AMR task was more sensitive to motor control differences and subgroup differentiation than the connected speech task.

Alternative Explanations for Variability within Ataxic Dysarthria

It is useful to consider other variables that could drive the disparate patterns of speech characteristics in ataxic dysarthria. For example, the severity of dysarthria or disease could alter the nature and presentation of speech impairment. Specifically, speakers with mild dysarthria

[1] The "variable"/instability group was described in the following manner: "These speakers have more unstable motor control. They are typically more variable with sound errors, stress, rhythm, and pitch. They tend to be faster or have inconsistent rate. They may have incoordination of breathing and speaking." The "robotic"/inflexibility group was described as having "more inflexible motor control; more equalized stress and rhythm, and more consistent errors; tend to be slower; may have monopitch/monoloudness."

may present with variability/instability of speech patterns and become more inflexible in their speech motor control with disease progression. Ackermann and colleagues (2007) postulated that dissolution and incoordination of verbal utterances in cerebellar disorders evolve across these two processes. That is, increased instability of temporal organization evolves into a slower and isochronous syllable pacing that is consistent with the pattern of inflexibility. However, this pattern may reflect the emergence of spastic dysarthria with disease progression, highlighting the importance of excluding speakers with mixed dysarthria to the extent possible. Additionally, Brendel and colleagues (2015) argued against severity influencing the distinct profiles of speech impairment in SCA speaker profiles based on group matching for disease duration and disease severity per the *Scale for the Assessment and Rating of Ataxia* (SARA; Schmitz-Hübsch et al., 2006). Amaral and colleagues (2022) also found a weak connection between dysarthria severity and perceptual subgroups, arguing against severity as the primary explanation for clusters of speech characteristics. Thus, at present, there is insufficient and conflicting evidence to attribute ataxic dysarthria speech patterns to severity and disease progression (Schalling et al., 2008; Brendel et al., 2015).

Another possible explanation for variability in perceptual characteristics is the *site of the lesion/disruption* to the cerebellar control circuit. For example, inferior portions of the cerebellum seem to be more active during speech execution while the anterior-superior areas are more active bilaterally during speech preparation (Duffy, 2020). There is considerable variability in the cerebellar regions which can lead to ataxic dysarthria such as superior cerebellar vermis, bilateral cerebellar hemispheres, and lateral and paravermal aspects of the hemispheres (Kent et al., 2000). Additionally, highly variable motor speech outcomes have been reported after removal of comparable cerebellar tumors (Cornwell, Murdoch, & Ward, 2005). Lesion location and its impact on speech characteristics are not well understood and even comparable lesion location or similar degenerative disease processes have not resulted in comparable speech outcomes (Spencer and Slocomb, 2007). Thus, profiles of speech characteristics due to damage to the cerebellar control circuit have yet to be clearly connected to lesion location.

Limitations in Previous Research

The emerging literature on subgroups of ataxic dysarthria has been somewhat constrained by methodological limitations. Sample sizes, often less than twenty, with a single medical diagnosis (e.g., Multiple Sclerosis, Friederichs Ataxia, etc.; Brendal et al., 2015, Hartelius et al., 2000, etc.) has limited generalizability. Additionally, some studies previously have used visual analog scales for listeners to rate perceptual speech characteristics. While informative, this method is constrained by researcher-imposed speech dimensions, and pre-conceived notions of perceptual groupings, which inherently biases listeners.

Auditory Free Classification

Bias associated with a researcher-defined rating system can be directly addressed with the implementation of a free classification paradigm. The free classification model developed by Imai and colleagues (1965; Imai and Garner 1966) was first used in cognitive psychology to explore the classification of visual stimuli in the absence of provided categorical labels. This paradigm allows the participants to create their own associations and an unlimited and self-determined number of groupings. As such, it is considered more flexible and allows researchers to explore dimensions of contrasting stimuli without the bias of researcher-imposed preconceived notions.

The free classification paradigm was applied to speech stimuli by Clopper (2008) in the classification of regional dialects of American English. This work supported the shift in free classification from purely visual to auditory stimuli. In more recent years, this auditory free-classification task has been applied to the study of dysarthria. Lansford, Liss, and Norton (2014) asked listeners to group the perceptual characteristics of dysarthric speakers into the established types of dysarthria (e.g., flaccid, spastic, ataxic, etc.). Twenty-three graduate students listened to the speech of 33 speakers with dysarthria and grouped speakers based on perceptual likeness on a visual grid (see Figure 1). In the grid, perceptually similar speakers are placed close to each other while perceptually divergent speakers are not. Thus, Lansford and colleagues (2014) demonstrated that free classification was feasible for categorization of perceptual characteristics and comparisons of dysarthria. Kim, Diehl, de Riesthal, Tjaden, Wilson, Claasen, and Mefferd (2022) also used auditory free classification for perceptual categorization of dysarthria from Huntington's disease (HD). They compared this approach to a statistical clustering approach using the guidelines of the Mayo Clinic dysarthria rating scale for hyperkinetic dysarthria. The study demonstrated substantial overlap between the subgroups of speakers using both systems but posited that the lack of complete overlap indicated that listeners may find certain perceptual speech characteristics in hyperkinetic dysarthria more salient than those suggested by the standardized Mayo Clinic dysarthria rating scale.

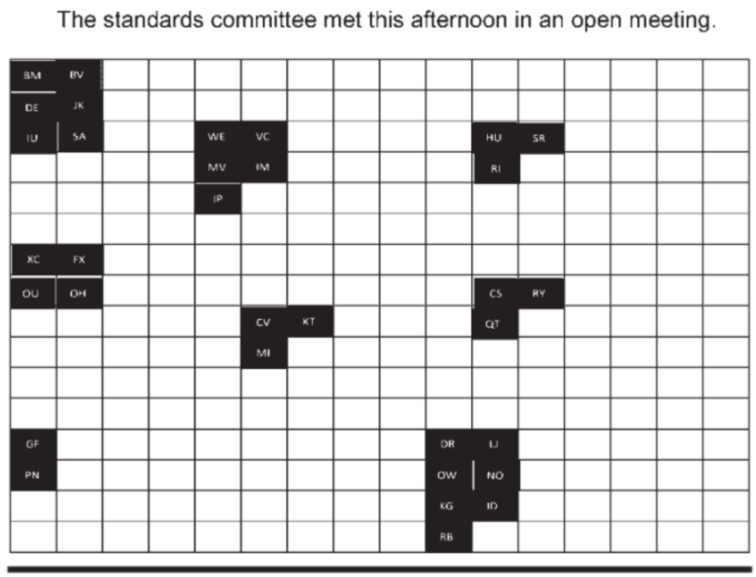

Figure 1. Lansford and colleagues (2014) example of one listener's free-classification task in the completed state. Icons that are touching were considered to sound similar to each other.

Thus, there are numerous benefits to the auditory free classification approach for examination of perceptual subgroups, including the identification of clusters based on the listener experience and not investigator-imposed classification guidelines. Additionally, free classification is less reliant on specialized programming skills or software, and therefore more feasible for researchers. To that end, this proposed study will examine the clusters generated by speech-language pathologists when using an auditory free classification paradigm to perceptually classify speakers with ataxic dysarthric. Specifically, the following research questions will be addressed:

(1) Will speakers with ataxic dysarthria be classified by speech-language pathologists into clusters in an unconstrained auditory free-classification task for AMR and connected speech tasks?

(2) Do the resulting clusters align with perceptual characteristics previously established for the instability and inflexibility subgroups (Spencer, Amaral, and Lansford, 2022)?

Based on previous literature and the studies by Spencer and colleagues (2016, 2018, in press), it is hypothesized that listener-derived clusters will map onto perceptual characteristics reflecting instability (e.g., variable stress and rhythm, faster rate of speech, etc.) and inflexibility (e.g., more equalized stress and rhythm, slower rate of speech, etc.). Additionally, stronger clusters will emerge for the AMR tasks than the connected speech task.

Methods

This study was conducted virtually to maximize sample size, and to acknowledge the ongoing public health concerns related to COVID-19. All procedures were approved by the Institutional Review Board of the University of Washington.

Participants

Speakers

Audio samples were collected from thirty-six speakers with ataxic dysarthria. Recruitment was through the Coordination of Rare Diseases at Sanford (CoRDS) registry, which facilitates the advancement of research for more than 10,000 rare diseases, including hereditary and acquired forms of ataxia. Recruitment efforts resulted in the immediate attainment of the target recruitment goal plus an extensive waitlist for participation.

Inclusion criteria for all speakers was a diagnosis of hereditary or acquired ataxia by a neurologist, age 18-85, and native American English speakers. Exclusion criteria included the presence of neurological compromise beyond ataxia or history of head trauma, disease duration less than one year, alcohol or drug dependency, and presence of a mixed dysarthria. Dysarthria determination was initially conducted during the screening process and confirmed later by two independent experts. The flow diagram (Appendix A) depicts the recruitment process, highlighting reasons for inclusion or exclusion. From the thirty-six original speakers, nine were excluded for a mixed dysarthria or negligible dysarthria based on judgment of the independent experts, one was excluded for perceived articulatory disorder, and one was excluded for poor recording quality, resulting in a final sample of 25 speakers.

Speaker demographic characteristics are summarized in Table 2. Speakers with ataxia included 12 men and 13 women ranging in age from 30 – 82 years with a mean of 53.72 years. Education ranged from 12 – 24 years (mean = 16.36, SD = 2.45). Disease etiology was diverse, and included spinocerebellar ataxia, cerebellar atrophy, Friedreich's Ataxia, idiopathic cerebellar degeneration, and ataxia caused by gluten inflammatory response. Average disease duration (time since diagnosis) was 9.56 years and ranged from 2 – 32 years. Participants were from a range of geographic areas including Western US, Northern/New England US, Southern US, and Midwestern US, and Canada.

Table 2. Demographics of the speakers with ataxic dysarthria

Speaker	Gender	Age (Yrs)	Education (Yrs)	Geographic Region	Disease Etiology	Disease Duration (Yrs)
S01	M	32	16	Midwestern US	SCA 2	7
S02	M	74	16	Northern/New England	Idiopathic Ataxia	6
S03	F	49	16	Midwestern US	SCA 1	7
S04	M	77	17	Southern US	SCA 5	22
S05	M	42	16	Southern US	SCA 2	3
S06	F	30	17	Western US	SCAr10	2
S07	F	48	18	British Columbia	POLR3A	20
S08	M	69	20	Western US	SCA 6	11
S09	F	32	16	British Columbia	ARCA1	6
S10	M	53	19	Midwestern US	SCA (Unknown)	8
S11	M	63	16	Western US	SCA 42	4
S12	F	32	17	Southern US	SCA 2	5
S13	M	75	16	Southern US	SCA 8	9
S14	M	63	18	Midwestern US	SCA 3	11
S15	F	60	14	Southern US	SCA 6	2
S16	F	40	18	Southern US	SCA 2	8
S17	M	51	14	Midwestern and Western US	Cerebellar Ataxia	5
S18	F	48	14	Southern US	Gluten Ataxia	3
S19	F	51	14	Midwestern US	Friedreich's Ataxia	15
S20	M	52	12	Midwestern US	Idiopathic Ataxia	4
S21	F	82	14	Western US	SCA 6	17
S22	F	66	13	Western US	SCA (Unknown)	32
S23	F	60	24	Southern US	SCA 8	25
S24	M	34	16	Northern/New England US	SCA 3	5
S25	F	60	18	Midwestern US	SCA 6	2
Mean (SD)	12 M **13 F**	**53.72 (15.07)**	**16.36 (2.45)**			**9.56 (7.80)**

Note. SCA= Spinocerebellar ataxia, SCAr10 = Autosomal Recessive spinocerebellar ataxia type 10, POLR3A: Pol III-related leukodystrophy, ARCA1: Autosomal recessive cerebellar ataxia type 1.

Listeners

Listeners were 17 speech-language pathologists recruited via posting on ASHA SIG 2 online forum, Facebook Medical SLP Forum, Washington State Speech-Language-Hearing Association Facebook page, and local clinics. See Appendix B for the recruitment posting. Inclusion criteria for listeners were experience working with individuals with dysarthria, no self-reported hearing difficulties, and native speakers of American English. The flow diagram (Appendix C) depicts the recruitment process.

Listener demographic characteristics are summarized in Table 3. SLP listeners included 3 men and 14 women ranging in age from 29-67 with a mean age of 40.94 years. Education ranged from 18-23 years (mean = 20.12). Years of clinical experience with adults with adults with dysarthria ranged from 4-34 (mean = 11.76). In the recent caseloads of listeners, the percentage of adult clients with dysarthria ranged considerably from “1-20%” to “81-100%”. The listeners had experience with adults with dysarthria in inpatient and outpatient settings, acute care, private practice, research/university settings, skilled nursing facilities, and home health.

Table 3. Demographics of SLP Listeners

Listener	Gender	Age (Yrs)	Education (Yrs)	Clinical Experience (Yrs)	% Recent Caseload Dysarthria	Work Settings
L1	F	36	22	8	61% -80%	Outpatient clinic, Acute care, Inpatient rehabilitation, Research/University settings
L2	F	67	18	34	41% - 60%	Outpatient clinic, Acute care, Inpatient rehabilitation
L3	F	48	23	6	41% - 60%	Outpatient clinic, Acute care, Skilled nursing facilities
L4	M	51	25	22	41% - 60%	Outpatient clinic, Home Health, Acute care, Inpatient rehabilitation, Skilled nursing facilities, Research/University settings
L5	F	41	18	15	1% - 20%	Outpatient clinic, Acute care, Research/University settings, Private practice
L6	F	30	19	4	41% - 60%	Outpatient clinic, Acute care
L7	M	29	18	5	1% - 20%	Acute care
L8	F	52	25	4	21% - 40%	Home Health, Inpatient rehabilitation, Skilled nursing facilities
L9	F	34	18	11	21% - 40%	Outpatient clinic, Acute care, Inpatient rehabilitation
L10	F	34	18	11	1% - 20%	Outpatient clinic, Acute care, Inpatient rehabilitation
L11	F	34	18	11	21% - 40%	Outpatient clinic, Acute care, inpatient rehab
L12	F	32	18	7	100%	Research/University Settings
L13	F	35	18	10	21% - 40%	Outpatient clinic, Home Health, Inpatient rehabilitation, Skilled nursing facilities
L14	F	42	19	15	21% - 40%	Outpatient clinic, Skilled nursing facilities
L15	F	43	23	10	1% - 20%	Outpatient clinic, Acute care, Inpatient rehabilitation, Research/University settings
L16	M	52	22	20	41% - 60%	Acute care, Inpatient rehabilitation, Skilled nursing facilities, Research/University settings
L17	F	33	22	8	81% - 100%	Acute care, Research/University settings
Mean (SD)	**3 M 14 F**	**40.94 (9.87)**	**20.12 (2.30)**	**11.76 (7.47)**	--	

Procedure

Speakers: Pre-session

Participants with ataxia were screened via preliminary telephone call to allow initial determination of the presence of dysarthria, and screening of inclusion and exclusion criteria (see Appendix D for phone screen talking points). If the participant met the criteria, a summary of protocol information discussed during the phone screen was emailed to the participant with the scheduling information for the experimental session, containing the time (in speaker's time zone) and date of the session, the speaker consent form, and the virtual assessment meeting link via HIPAA compliant Zoom.

Speakers: Experimental Session

The investigator opened the virtual meeting link ten minutes prior to the experimental session to check audio and visual quality, and functionality of the headset. Zoom settings were consistent across participants; for audio settings: (1) Suppress background noise—set to "Low", (2) Enable: "High-fidelity music mode", and for recording settings: (1) Set to store recordings to the lab server (L: drive), (2) Select "Record a separate audio file of each participant", (3) Deselect, "Display participants' names in the recording", and (4) Select, "Keep temporary recording files".

During the Zoom session, the functionality of the remote connection was determined, the ideal recording environment was discussed (i.e., reliability of Wi-fi, ability to see shared documents, adequate built-in or external webcam, functioning audio system, and recommendations that doors/windows are closed, all electronic devices silenced, etc.). Participants were asked questions pertaining to their diagnosis, primary symptoms, speech difficulties, history of speech therapy services, and medications, and confirmed information related to exclusion criteria (e.g., native language, history of neurologic compromise beyond ataxia or history of head trauma, drug and alcohol use, and vision) (see Appendix E).

The *Sentence Intelligibility Test (SIT)* (Yorkston, Beukelman, & Tice, 1996) was administered in a modified format for virtual administration for an estimate of intelligibility of the speakers. Speakers read one sentence each of 5-15 words in length, chosen randomly from the SIT sentences. Beverly and colleagues (2010) found that predictability of the SIT sentences had an influence on speech intelligibility scores. Thus, the selection of sentences was pseudo-random, drawing only from those sentences with adequate predictability metrics less than or equal to 50 percent predictability (Cannito et al., 2012) which applied to 568/660 sentences (44-55 per sentence length). Sentences were provided on a PowerPoint slide and visible to the speaker when the screen was shared.

The short version of the *Communicative Participation Item Bank* (CPIB; Baylor, C., Yorkston, K., Eadie, T., Kim, J., Chung, H., & Amtmann, D. (2013).; Yorkston et al., 2008; see Appendix F) was given for a measure of dysarthria impact on life participation. The ten questions were displayed visually, via shared screen, and verbally presented; after each item, participants were asked to verbally convey their choice (0-3 scale).

Speakers were then asked to perform speech alternating motion rates (AMRs /pʌ/, tʌ/, /kʌ/) given the following instruction: "I want you to say some sounds for me, and I'd like you to say them as quickly and as clearly as possible until you run out of breath. I'll give you an example." Finally, participants were asked to read *The Caterpillar* passage (see Appendix G) which was designed to inform the characterization of motor speech disorders (Patel et al., 2012). For this reading passage, the investigator shared their screen, making sure the entire passage was visible for the speaker. The investigator asked the participant to read the passage in its entirety using their "most natural and typical way of speaking." During all speech sample recordings, participants turned off their webcams to maintain confidentiality. A separate audio stream recording was also enabled to ensure only the participant's audio stream was used in data analysis.

Sessions took approximately 20-30 minutes. Speakers received a $15 compensation for participation[2] that was sent to them via Venmo or mailed check after the session.

Speakers: Data Processing

All sound files were edited using version 3.0.0 of Audacity ® recording and editing software. Participant audio streams were segmented into each task: AMRs, SIT sentences, and Caterpillar passage. The AMR file was then edited, as needed, to reduce silence between productions of /pʌ/, tʌ/, /kʌ/. *The Caterpillar* passage recording was clipped to only include the fourth and fifth sentences (indicated below), chosen for the relatively high number of multisyllabic words and prosodic variation.

[2] Funding for the study is through the American Speech-Language-Hearing Association's *Students Preparing for Academic-Research Careers* (SPARC) award to H.B.

"My most MEMORABLE moment was riding on the Caterpillar, which is a gigantic rollercoaster high above the ground. When I saw how high the Caterpillar rose into the bright blue sky, I knew it was for me."

Intelligibility ratings were independently conducted by two research assistants who were instructed to listen to each sentence only once, transcribe what they heard, indicate unintelligible words with an underscore with spaces between each perceived word boundary. Average intelligibility was calculated for each speaker. Rate of speech was calculated independently by two research assistants for each AMR syllable (/pʌ/, /tʌ/, and /kʌ/) and the two sentences of the Caterpillar passage. Research assistants were instructed to listen to each audio file and indicate duration and syllable count, including filler words in connected speech. Disagreements related to duration (beyond 0.25 sec) and syllable number were resolved by the investigator.

Dysarthria presence, type, severity, and characteristics were independently evaluated by two motor speech experts (KS and KL) based on audio recordings of AMRs, SIT sentences, and the full Caterpillar passage. The experts also made *a priori* determinations of the perceptual subgroups of inflexibility, instability, or mixed/indeterminate using previously established perceptual criteria (Spencer and France, 2016; Spencer & Dawson, 2019; Spencer, Amaral, & Lansford, 2022).

<u>Listeners: Pre-Session</u>

All potential listeners filled out an intake questionnaire (See Appendix H) to determine eligibility and provide background information regarding years of education, years of clinical experience with adults with dysarthria, percentage of recent caseload of adults with dysarthria, and settings in which they have worked with adults with dysarthria. Eligible listeners were emailed the IRB approved consent form to review. Once a statement of consent was received via email the study materials were sent. See Appendix I and Appendix J for email contact templates.

<u>Listeners: Experimental Session</u>

Given the size of file necessary to accommodate 50 speaker recordings, Google Drive was utilized for ease of sharing. Instructions for the task were outlined in the first three slides found in Appendix K. Aligning with the procedures and protocol of Lansford et al. (2014) and

Spencer et al. (2022), a single slide with a white background was used for each auditory free-classification task, one for connected speech (two sentences of the Caterpillar passage) and another for AMRs. Speaker audio recordings were embedded onto black rectangles with randomized two-letter identifiers to create a movable icon for each speaker sample. This allowed the listeners to click each icon to hear the audio file. Icons were placed in 5 columns to the left of a 14x10 cell gride (see Figure 2) and each icon was sized to fit exactly into each cell. The "notes" section was available for optional listener notations.

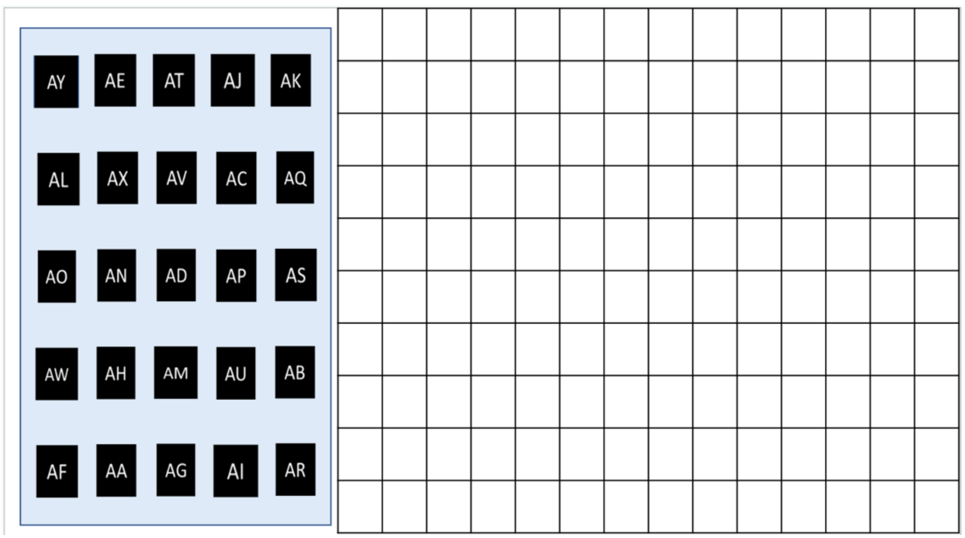

Figure 2. Screenshot of Free Classification task layout with speaker files and 14x10 grid

Listeners used their personal computers to complete the task. They were instructed to find a quiet workspace and were encouraged to use high-quality over-ear headphones when possible. Listeners were informed that all speakers had ataxic dysarthria and were from a broad range of geographic and dialectal backgrounds. They were instructed that perceived dialectal differences should not be used in grouping decisions. No other information was given about the speakers. Listeners were permitted, but not required, to use the notes section at the bottom of the slide to take notes throughout the tasks, to reduce working memory load. Notes were saved for descriptive purposes.

Listeners were asked to listen to the audio files and place them on the blank grid by dragging and dropping them into groups with speakers that sound similar— placed so the icons were touching each other on the grid. Figure 3 shows the parameters given regarding grouping.

They were not given further instructions about how to make their judgments and were not informed of the purpose of the study. Listeners were permitted to make any number of groups with any number of speakers in each group. They were also permitted to listen to each file and rearrange their groupings as many times as necessary. A 10-day time period was given to complete the task. Once the listeners completed the task, compensation was sent via Venmo or mailed check and editing permissions were removed from the Google Slides file.

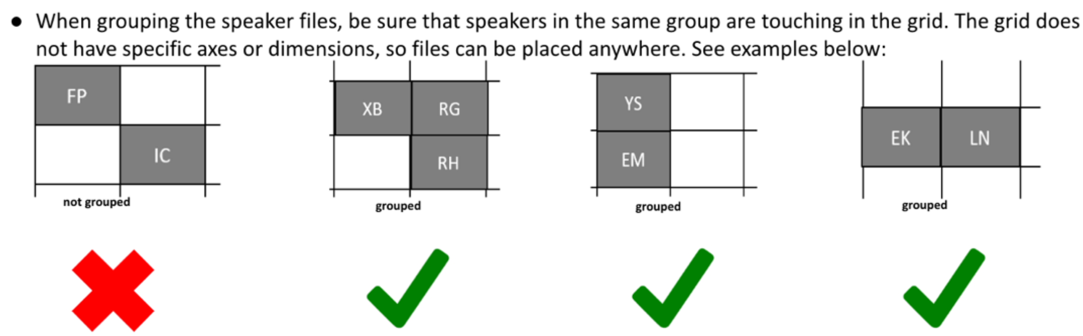

Figure 3. Screenshot of listener task grouping examples instructions.

<u>Listeners: Data Processing</u>

Google Slides were downloaded into Microsoft PowerPoint slides to follow the procedures outlined in Lansford et al. (2014) and Spencer et al. (2022). A Python script (Mazik, 2023) was used to gather positional information of icons for each task (AMR and connected speech) and create matrices that reflect neighboring icons in Microsoft Excel (see Appendix L). Icons that were parallel or perpendicular to another icon were marked as grouped, and all chaining icons (i.e., in the cluster but not next to each other) were marked as grouped. Figure 4 shows an example of chaining icons, where the BQ icon and the BU icon (for example) would be coded as part of the same group even though they are not parallel or perpendicular to one another. Additionally, icons that were not directly touching but were within 25% of the width or length of the icon away from their neighboring icon would still be marked as grouped to accommodate variability of icon placement within the grid.

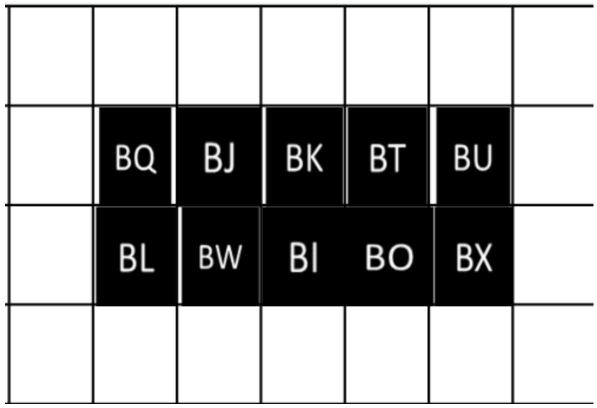

Figure 4: Screenshot Example of chaining aspects of Python script

The groupings obtained from each listener were arranged in a 25 x 17 speaker similarity matrix in Excel. A "1" was entered via python code into cells in which the two speakers were grouped together by a listener, while a "0" was entered if the speakers were not grouped together. Individual listener matrices were then combined into a pooled similarity matrix for each of the free classification tasks (AMR and connected speech). Point-to-point reliability between Python script calculations and hand calculations was conducted for 11.8% of matrices (4/34 [17 listeners x 2 speaking tasks] matrices).

Group comparisons were made using two-sample t-tests or single factor ANOVAs (SPSS v.29). The pooled similarity matrices were used to conduct cluster analysis (*hcultst() function*) and multidimensional scaling (*cmdscale() function*) which were conducted using R Statistical Soft Care (version 4.3.1).

Results

Reliability

Point-to-point reliability between the Python script and hand calculations of speakers who were grouped together (i.e., 1 or 0 entries in the similarity matrices) revealed 100% agreement.

Rate of speech calculations were completed by two independent research assistants and reliability was conducted on 100% of calculations. Point-to-point agreement for syllable counts was 59% and for duration measurements (within 0.25) was 96%. All inter-rater discrepancies were resolved by the PI.

A priori determination of dysarthria perceptual subgroups (instability, inflexibility, mixed/indeterminate) revealed 80% agreement (20/25) across the two expert raters (KS and KL), with full consensus following discussion. Of the 25 included speakers, 15 were characterized as aligning with the instability group, 2 were determined to align with the inflexibility group, and 8 were characterized as mixed[3]. Independent ratings of dysarthria severity (mild/moderate/severe) revealed 92% agreement (23/25) with full consensus following discussion.

Speaker Characteristics

Speaker characteristics and *a priori* subgroupings can be found in Table 4 below. Overall, dysarthria severity judgments ranged from mild to moderate, which is consistent with the intelligibility scores which averaged 92.3% and ranged from 81-99%. CPIB scores ranged from 3 – 25 (mean = 15.04, SD = 5.70; possible range 0-30); lower scores indicate more interference in participation in everyday communication participation situations. As anticipated, speakers with moderate dysarthria had lower CPIB scores than those with a mild dysarthria, as reflected in Figure 5.

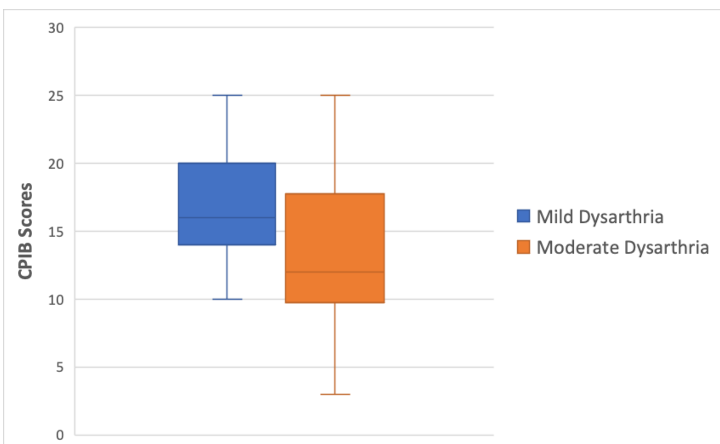

Figure 5: Boxplot of Communicative Participation Item Bank (CPIB) scores across speaker severity. Possible CPIB scores range from 0 (significant interference with participation) to 30 (minimal/no interference with participation).

[3] All speakers considered "mixed" had features of both instability and inflexibility speech characteristics (see Table 4); none were considered indeterminate.

Based on the judgments of two motor speech experts, 15 of the 25 speakers were characterized as aligning with the instability group, 2 were determined to fit with the inflexibility group, and 8 were judged to be mixed. Dysarthria characterizations of the two speakers in the inflexible group reflected shared features of slow rate, equalized stress, reduced pitch variation, and slow, regular AMRs, while speech patterns in the instability group were largely described as irregular articulatory breakdowns, irregular AMRs, and variable stress. The mixed group, as expected, reflected characteristics of both instability and inflexibility, e.g., variable stress and irregular AMRs paired with slow rate and equalized stress. The *a priori* inflexibility subgroup (n = 2) had the highest average intelligibility, with a mean of 94.77%. The instability subgroup (n = 15) had the lowest average intelligibility (mean = 91.64%), ranging from 81.42 - 98.64%. For the mixed subgroup (n=8), intelligibility averaged 93.78% and ranged from 89.09% - 95.45%.

Table 4: Characteristics of the speakers with ataxic dysarthria by *a priori* subgroup.

	Speaker	*A Priori*	CPIB Score	Rate of Speech AMR (Syll/sec)	Rate of Connected Speech (Syll/sec)	Mean Intelligibility (%)	Dysarthria Severity	Speech Characteristics
Inflexibility	S01	INF1	20	2.22	2.35	99	Moderate	Slow rate, equalized stress, reduced pitch and loudness variation, pitch breaks, mildly reduced articulatory precision; slow mostly regular AMRs with prominent voicing errors.
	S25	INF2	12	2.29	2.61	90	Moderate	Slow rate, imprecise articulation, equalized stress, some telescoping, mildly reduced pitch variation, slow/regular AMRs.
	Mean (SD)		**16 (4)**	**2.25 (0.04)**	**2.58 (0.13)**	**95 (4.32)**	**2 moderate**	

	Speaker	*A Priori*	CPIB Score	Rate of Speech AMR (Syll/sec)	Rate of Connected Speech (Syll/sec)	Mean Intelligibility (%)	Dysarthria Severity	Speech Characteristics
Instability	S02	INS1	16	3.79	3.02	99	Mild	Irregular articulatory breakdown, irregular AMRs, mildly inconsistent rate, hoarse, variable stress.
	S03	INS2	12	3.03	4.16	89	Moderate	Irregular articulatory breakdown, telescoping, variable stress (some equalized), irregular AMRs
	S05	INS3	24	3.97	4.15	97	Mild	Irregular articulatory breakdown, some telescoping, irregular AMRs.
	S06	INS4	16	3.31	3.43	98	Mild	Irregular articulatory breakdown, irregular AMRs, mildly inconsistent rate (and occasional equalization), occasional telescoping, mild harshness
	S07	INS5	20	4.07	3.71	98	Mild	Irregular articulatory breakdown, some telescoping, irregular AMRs, mild harshness
	S09	INS6	19	2.81	3.92	85	Mild	Irregular articulatory breakdown, some telescoping, variable stress (mild equalization), irregular AMRs.
	S10	INS7	14	3.13	3.82	84	Mild	Irregular articulatory breakdown, inconsistent rate, variable stress (occasional equalization), irregular AMRs, inconsistent hyponasality.
	S11	INS8	17	3.00	3.81	84	Moderate	Irregular articulatory breakdown, telescoping, inconsistent rate, variable stress, irregular AMRs.
	S13	INS9	16	4.61	3.69	93	Mild	Irregular articulatory breakdown, some telescoping, mild hoarseness, irregular AMRs.
	S14	IN10	20	3.79	3.45	81	Moderate	Irregular articulatory breakdown, telescoping, variable stress (some equalized), irregular AMRs (some mild blurring).
	S15	INS11	10	3.56	3.37	96	Mild	Irregular articulatory breakdowns, some telescoping, irregular AMRs.
	S16	INS12	25	2.84	3.35	93	Mild	Irregular articulatory breakdown, mildly slowed rate, mildly variable stress, irregular AMRs (stress/rhythm).
	S17	INS13	11	3.16	3.06	88	Moderate	Irregular articulatory breakdown, mildly slowed rate, variable stress (some equalized), irregular AMRs, some telescoping.
	S18	INS14	14	4.66	3.79	96	Mild	Irregular articulatory breakdown, faster rate, some telescoping, irregular AMRs.
	S21	INS15	17	3.40	3.65	94	Moderate	Irregular articulatory breakdown, variable stress (some equalized), variable rate, some telescoping, some distorted vowels, irregular AMRs.
	Mean (SD)		**16.73 (5.07)**	**3.54 (0.57)**	**3.63 (0.33)**	**92 (5.78)**	**10 mild 5 moderate**	

	Speaker	*A Priori*	CPIB Score	Rate of Speech AMR (Syll/sec)	Rate of Connected Speech (Syll/sec)	Mean Intelligibility (%)	Dysarthria Severity	Speech Characteristics
Mixed	S04	MIX1	4	4.66	3.79	89	Moderate	Articulatory imprecision, variable stress (some equalized), slower rate, prolonged phonemes, telescoping; slower AMRs with irregularities and difficulty coordinating breath and speech.
	S08	MIX2	11	2.69	3.47	90	Moderate	Imprecise articulation, variable stress (some equalized), incoordination of breathing and speaking, poor modulation of loudness, slower rate; irregular AMRs with slower rate, harshness, prolonged phonemes.
	S12	MIX3	19	3.44	4.01	95	Mild	Irregular articulatory breakdown, variable stress (some equalized), slower rate, irregular precision/voicing, rhythm of AMRs, possible speech/breathing coordination difficulty
	S19	MIX4	9	3.40	3.65	95	Moderate	Irregular articulatory breakdown, telescoping, AMRs slower and more rhythmic but irregular precision/voicing, variable stress (often equalized), variable (but mostly slower) rate.
	S20	MIX5	10	3.23	2.66	90	Moderate	More consistent imprecision, some shorter phrases, reduced variation of pitch, some reduced stress, irregular AMRs (short samples), some telescoping.
	S22	MIX6	12	2.43	2.69	95	Moderate	Irregular articulatory breakdown, irregular AMRs, variable rate, variable stress (often equalized), some harshness/strain (not consistently present).
	S23	MIX7	25	2.11	3.34	95	Moderate	Slow rate, equalization of stress but variable patterns, irregular articulatory breakdown, prolonged phonemes, reduced variation in pitch, some hoarseness; AMRs were slower, more consistent with rhythm and voicing errors, though some irregularities.
	S24	MIX8	3	2.29	2.61	91	Moderate	Imprecise articulation, slower rate, monopitch, telescoping, occasional difficulty coordinating breathing and speaking, reduced/equalized stress; AMRs slower, mostly rhythmic, imprecise, breathy (+ periodic vocal tremor).
	Mean (SD)		**11.63 (6.85)**	**2.87 (0.43)**	**3.12 (0.52)**	**93% (2.51)**	**1 mild 7 moderate**	

Note. CPIB =Communication Participation Item Bank. INF = Inflexibility, INS = Instability, MIX = Mixed subgroup.

Auditory Free-Classification Task and Cluster Analysis

Data from the individual similarity matrices of 17 listeners were summed to create a pooled similarity matrix for both the AMR task and the Connected speech task (see Tables 5 and 6). Entries in each matrix reflect the number of listeners who grouped two given speakers together (e.g., INS1 and INF1 were never grouped together, while INS1 and INS3 were grouped together by 7/17 listeners). For the AMR task, the number of clusters made by the listeners varied from 3 to 10 clusters, with the average number clusters being 5.65 (SD = 2.22). For the connected speech task, the number of clusters made by the listeners varied from 3 to 10 clusters, with the average number of clusters being 5.41 (SD = 2.09).

From the pooled similarity matrix, hierarchal clustering analyses were completed using an agglomerative (bottom-up) approach in accordance with the procedures in Spencer et al. (2022).

Table 5. Pooled similarity matrix for AMR speaker samples.

Pooled Matrix AMR

		INF1	INS1	INS2	MIX1	INS3	INS4	INS5	MIX2	INS6	INS7	INS8	MIX3	INS9	INS10	INS11	INS12	INS13	INS14	MIX4	MIX5	INS15	MIX6	MIX7	MIX8	INF2
		AY	AE	AT	AJ	AK	AL	AX	AV	AC	AQ	AO	AN	AD	AP	AS	AW	AH	AM	AU	AB	AF	AA	AG	AI	AR
INF1	AY	0	0	5	2	0	4	0	10	3	3	5	6	1	0	2	3	3	0	6	3	1	4	6	8	9
INS1	AE	0	0	1	2	7	2	5	0	2	1	0	1	9	7	2	2	2	9	0	1	3	4	1	0	1
INS2	AT	5	1	0	2	0	4	0	9	2	4	3	6	0	1	5	4	3	1	5	6	2	7	4	5	5
MIX1	AJ	2	2	2	0	2	3	1	2	6	7	2	1	3	3	1	3	6	4	5	3	3	2	3	3	0
INS3	AK	0	7	0	2	0	4	13	0	1	1	2	2	3	2	1	4	1	5	1	0	5	0	3	0	0
INS4	AL	4	2	4	3	4	0	2	3	6	4	7	7	2	2	1	10	3	4	4	4	4	2	7	3	5
INS5	AX	0	5	0	1	13	2	0	0	2	2	3	3	3	3	1	3	0	4	1	0	4	0	4	0	0
MIX2	AV	10	0	9	2	0	3	0	0	3	4	4	4	1	0	4	3	4	0	6	6	1	6	4	10	5
INS6	AC	3	2	2	6	1	6	2	3	0	5	4	4	3	4	3	4	6	3	4	3	0	4	6	3	1
INS7	AQ	3	1	4	7	1	4	2	4	5	0	5	5	2	1	2	4	5	1	4	4	3	6	5	5	2
INS8	AO	5	0	3	2	2	7	3	4	4	5	0	7	3	2	2	5	3	1	6	4	3	1	4	6	6
MIX3	AN	6	1	6	1	2	7	3	4	4	5	7	0	2	1	2	7	3	1	4	2	6	4	6	4	5
INS9	AD	1	9	0	3	3	2	3	1	3	2	3	2	0	10	5	2	3	10	1	3	4	4	0	1	0
INS10	AP	0	7	1	3	2	2	3	0	4	1	2	1	10	0	6	1	2	11	0	2	3	4	3	0	1
INS11	AS	2	2	5	1	1	1	1	4	3	2	2	2	5	6	0	1	5	2	2	8	3	5	0	2	1
INS12	AW	3	2	4	3	4	10	3	3	4	4	5	7	2	1	1	0	4	2	7	4	4	3	10	4	5
INS13	AH	3	2	3	6	1	3	0	4	6	5	3	3	3	2	5	4	0	2	7	5	3	5	2	5	3
INS14	AM	0	9	1	4	5	4	4	0	3	1	1	1	10	11	2	2	2	0	0	3	4	4	2	0	1
MIX4	AU	6	0	5	5	1	4	1	6	4	4	6	4	1	0	2	7	7	0	0	6	1	4	4	8	9
MIX5	AB	3	1	6	3	0	4	0	6	3	4	4	2	3	2	8	4	5	3	6	0	2	8	2	5	4
INS15	AF	1	3	2	3	5	4	4	1	0	3	3	6	4	3	3	4	3	4	1	2	0	3	4	1	2
MIX6	AA	4	4	7	2	0	2	0	6	4	6	1	4	4	4	5	3	5	4	4	8	3	0	4	3	3
MIX7	AG	6	1	4	3	3	7	4	4	6	5	4	6	0	3	0	10	2	2	4	2	4	4	0	4	5
MIX8	AI	8	0	5	3	0	3	0	10	3	5	6	4	1	0	2	4	5	0	8	5	1	3	4	0	9
INF2	AR	9	1	5	0	0	5	0	5	1	2	6	5	0	1	1	5	3	1	9	4	2	3	5	9	0

Table 6. Pooled similarity matrix for connected speech samples.

Pooled Matrix Connected Speech

		INF1	INS1	INS2	MIX1	INS3	INS4	INS5	MIX2	INS6	INS7	INS8	MIX3	INS9	INS10	INS11	INS12	INS13	INS14	MIX4	MIX5	INS15	MIX6	MIX7	MIX8	INF2
		AY	AE	AT	AJ	AK	AL	AX	AV	AC	AQ	AO	AN	AD	AP	AS	AW	AH	AM	AU	AB	AF	AA	AG	AI	AR
INF1	AY	0	0	0	3	2	0	0	6	0	1	1	0	2	1	0	0	5	0	2	2	5	8	5	7	5
INS1	AE	0	0	7	2	7	5	5	1	9	2	1	6	5	5	4	6	4	7	2	6	0	1	3	0	3
INS2	AT	0	7	0	1	9	4	7	0	11	2	5	7	6	6	2	5	4	4	3	2	1	1	2	0	1
MIX1	AJ	3	2	1	0	2	0	0	7	1	6	5	1	4	5	0	3	8	0	6	3	6	8	3	9	5
INS3	AK	2	7	9	2	0	8	9	1	10	4	4	5	4	4	6	4	4	7	3	6	1	2	1	1	2
INS4	AL	0	5	4	0	8	0	13	2	7	3	2	5	3	3	10	6	1	10	1	2	2	0	2	0	0
INS5	AX	0	5	7	0	9	13	0	2	9	3	2	7	6	4	10	6	1	9	1	2	1	1	3	0	0
MIX2	AV	6	1	0	7	1	2	2	0	1	3	4	3	2	4	2	4	6	2	5	3	5	7	9	3	9
INS6	AC	0	9	11	1	10	7	9	1	0	4	1	8	7	7	3	6	2	6	3	4	1	1	2	0	1
INS7	AQ	1	2	2	6	4	3	3	3	4	0	9	2	3	5	1	1	3	2	6	5	3	3	1	7	4
INS8	AO	1	1	5	5	4	2	2	4	1	9	0	3	4	6	3	2	5	4	5	3	4	2	3	5	4
MIX3	AN	0	6	7	1	5	5	7	3	8	2	3	0	6	8	7	8	2	6	4	3	2	1	5	0	2
INS9	AD	2	5	6	4	4	3	6	2	7	3	4	6	0	5	3	3	4	3	1	2	3	5	2	3	1
INS10	AP	1	5	6	5	4	3	4	4	7	5	6	8	5	0	2	7	3	3	6	4	4	3	3	2	1
INS11	AS	0	4	2	0	6	10	10	2	3	1	3	7	3	2	0	5	0	12	1	1	1	0	3	0	1
INS12	AW	0	6	5	3	4	6	6	4	6	1	2	8	3	7	5	0	3	5	4	4	3	2	5	1	3
INS13	AH	5	4	4	8	4	1	1	6	2	3	5	2	4	3	0	3	0	2	3	5	2	4	4	2	7
INS14	AM	0	7	4	0	7	10	9	2	6	2	4	6	3	3	12	5	2	0	1	1	1	0	3	0	1
MIX4	AU	2	2	3	6	3	1	1	5	3	6	5	4	1	6	1	4	3	1	0	4	7	5	4	5	6
MIX5	AB	2	6	2	3	6	2	2	3	4	5	3	3	2	4	1	4	5	1	4	0	1	2	2	2	6
INS15	AF	5	0	1	6	1	2	1	5	1	3	4	2	3	4	1	3	2	1	7	1	0	6	2	6	2
MIX6	AA	8	1	1	8	2	0	1	7	1	3	2	1	5	3	0	2	4	0	5	2	6	0	6	9	7
MIX7	AG	5	3	2	3	1	2	3	9	2	1	3	5	2	3	3	5	4	3	4	2	2	6	0	1	12
MIX8	AI	7	0	0	9	1	0	0	3	0	7	5	0	3	2	0	1	2	0	5	2	6	9	1	0	3
INF2	AR	5	3	1	5	2	0	0	9	1	4	4	2	1	1	1	3	7	1	6	6	2	7	12	3	0

The results of the cluster analysis are best visualized by dendrograms (see Figures 6 and 8) which illustrate how speakers with ataxic dysarthria were clustered together. Agglomerative clustering begins by assuming each speaker is in a cluster alone (as indicated by the individual lines for each speaker at the bottom of Figures 6, 8), then iteratively merges speakers together based on free classification similarity until there is a single cluster left (as indicated by the single line at the top of Figures 6, 8). The number of clusters resulting from the analysis were defined by the researchers based on gaps in the dendrogram (James, Witten, Hastie, & Tibshirani, 2017) together with findings from multidimensional scaling (see below) rather than by a pre-determined set number of clusters.

For the AMR task (Figure 6), three clusters emerged. Cluster 1 contained eight speakers total: five speakers that were determined by expert judgment, pre-experiment, to reflect the instability group and three speakers that were determined to be mixed inflexibility/instability. Cluster 2 contained both of the inflexibility speakers (INF1 and INF2) as well as the majority of mixed instability/inflexibility speakers, and a few speakers from the instability group (INS4, INS8, and INS12). Cluster 3 contained seven speakers that were determined by expert judgment, pre-experiment, to reflect the instability group.

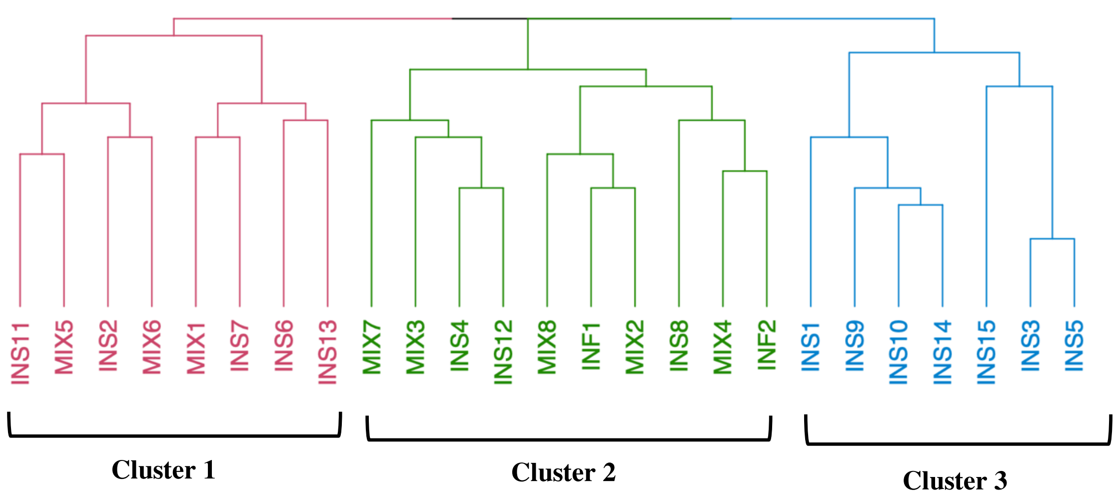

Figure 6. Dendrogram with three-cluster solution for the AMR task that shows the progressive linking of speakers based on listener groupings. The color-coding is used to demarcate the three clusters.

Multidimensional scaling (MDS) is helpful in visualizing the data by similarity; speakers closer together were rated more similarly by listeners than speakers farther apart. The three clusters identified by the hierarchical clustering are corroborated with findings from the MDS and are shown in the circles in Figure 7.

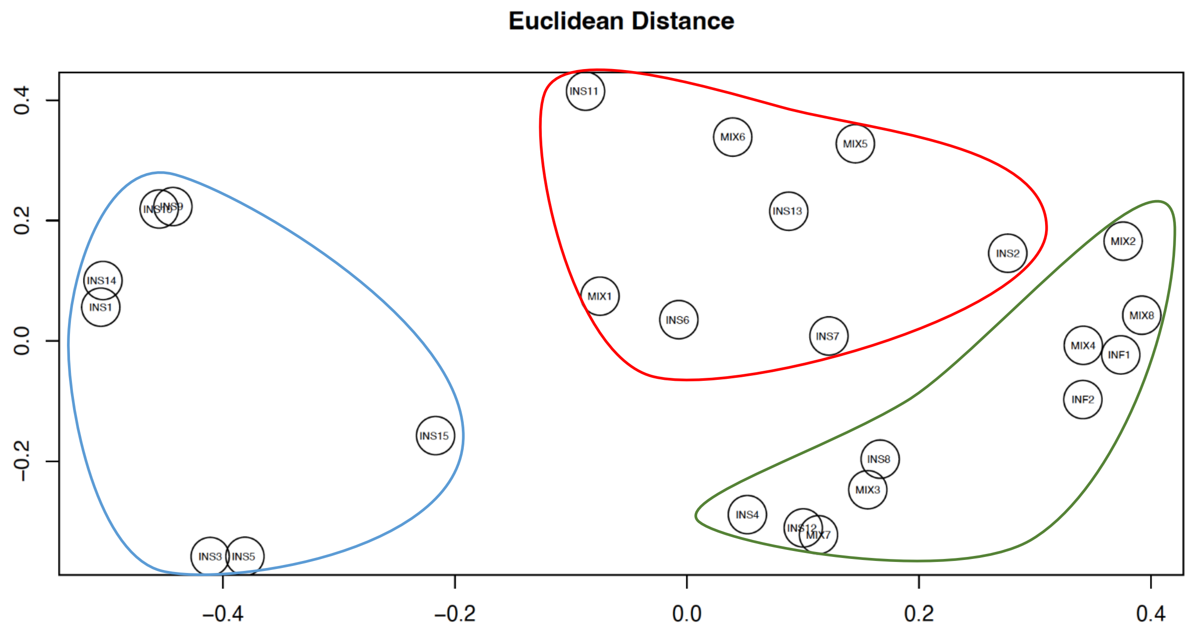

Figure 7: Multidimensional scaling plot reflecting the three-cluster solution for AMR task.

Across the three clusters for the AMR task, there did not appear to be an influence of gender with a mix of both men and women in each cluster, including 4 male speakers in each cluster (see Table 7). Additionally, there were no clear patterns regarding type of SCA or other disease etiology. With respect to speaker severity, all three clusters had a mix of mild and moderate speakers; however, Cluster 3 had the lowest percentage of moderate speakers (29.6%) compared to Cluster 1 (62.5%) and Cluster 2 (70%). There were no significant differences across clusters for age [F(2,22) = 2.07, $p = 0.15$], disease duration [F(2,22) = 0.19, $p = 0.83$] or average intelligibility [F(2,22) = 1.83, $p = 0.18$]. However, there was a significant difference for rate of speech (syllables/sec) for AMRs [F(2,22) = 23.18, $p < 0.001$]. Cluster 3 had the fastest rate of speech averaging 4.04 syll/sec, Cluster 1 had an average rate of speech of 3.14 syll/sec, and Cluster 2 had the slowest average rate of speech at 2.71 syll/sec.

The speech characteristics of Cluster 3, containing all *a priori* instability speakers, reflected predominant irregularities in the speech patterns across articulation and prosody, e.g., irregular AMRs, irregular articulatory breakdown, variable stress, etc. The speech characteristics of Cluster 1, with the majority of speakers judged to fit the instability profile, also reflected predominant irregularities in the speech patterns; while the 3 mixed speakers had mild indicators of inflexibility (e.g., perceived slower rate, more consistent articulatory imprecision), all three were noted to have irregular AMRs. The speech characteristics of Cluster 2 (both inflexibility speakers and the majority of mixed subgroup speakers) reflected some irregularities; however, this cluster had the most instances of slower rate, equalization of stress patterns, reduced variation in pitch, and reduced modulation of loudness. In particular, slowed rate was noted for 8/10 speakers in this cluster, including mildly slowed rate for a speaker classified as instability (INS12), consistent with the significant difference in syll/sec during the AMR task. Telescoping was observed in speakers across all clusters but was more frequently noted for Cluster 3 (71.4%) and Cluster 1 (75%) compared to Cluster 2 (50%). As a reminder, the overall speech characteristics (see Table 4) were derived from speech samples across multiple tasks and AMRs, whereas listeners were making judgments on only the AMRs in this task.

Table 7: Speaker characteristics grouped by listener-derived clusters for AMRs.

	Speaker	*A Priori*	Gender	Age	Etiology	Disease Duration (Yrs)	Dysarthria Severity	% Mean Intelligibility	AMR Rate of Speech (Syll/sec)
Cluster 1	S03	INS2	F	49	SCA 1	7	Moderate	89	3.03
	S09	INS6	F	32	ARCA1	6	Mild	85	2.81
	S10	INS7	M	53	SCA (Unknown)	8	Mild	84	3.13
	S15	INS11	F	60	SCA 6	2	Mild	96	3.56
	S17	INS13	M	51	Idiopathic Ataxia	5	Moderate	88	3.16
	S04	MIX1	M	77	SCA 5	22	Moderate	89	2.77
	S20	MIX5	M	52	Idiopathic Ataxia	4	Moderate	90	3.44
	S22	MIX6	F	66	SCA (Unknown)	32	Moderate	95	3.23
	Mean (SD)		**4M 4F**	**55.00 (12.37)**		**10.75 (9.86)**	**3 mild 5 moderate**	**90 (4.00)**	**3.14 (0.26)**
Cluster 2	S06	INS4	F	30	SCAr10	2	Mild	98	3.31
	S11	INS8	M	63	SCA 42	4	Moderate	84	3.00
	S16	INS12	F	40	SCA 2	8	Mild	93	2.84
	S08	MIX2	M	69	SCA 6	11	Moderate	90	3.26
	S12	MIX3	F	32	SCA 2	5	Mild	95	2.99
	S19	MIX4	F	51	Friedreich's Ataxia	15	Moderate	95	2.69
	S23	MIX7	F	60	SCA 8	25	Moderate	95	2.43
	S24	MIX8	M	34	SCA 3	5	Moderate	91	2.11
	S01	INF1	M	32	SCA 2	7	Moderate	99	2.22
	S25	INF2	F	60	SCA 6	2	Moderate	90	2.29
	Mean (SD)		**4M 6F**	**47.10 (14.32)**		**8.40 (6.73)**	**3 mild 7 moderate**	**93 (4.28)**	**2.71 (0.41)**
Cluster 3	S02	INS1	M	74	Idiopathic Ataxia	6	Mild	99	3.79
	S05	INS3	M	42	SCA 2	3	Mild	97	3.97
	S07	INS5	F	48	POLR3A	20	Mild	98	4.07
	S13	INS9	M	75	SCA 8	9	Mild	93	4.61
	S14	INS10	M	63	SCA 3	11	Moderate	81	3.79
	S18	INS14	F	48	Gluten Ataxia	3	Mild	96	4.66
	S21	INS15	F	82	SCA 6	17	Moderate	94	3.40
	Mean (SD)		**4M 3F**	**61.71 (14.66)**		**9.86 (6.15)**	**5 mild 2 moderate**	**94 (5.47)**	**4.04 (0.42)**

For the connected speech task, two clusters emerged (Figure 8). Cluster 1 contained 11 speakers that were determined by expert judgment, pre-experiment, to reflect the instability subgroup and one speaker that was determined to be mixed inflexibility/instability (MIX3). Cluster 2 contained both speakers determined by expert judgment to be in the inflexibility subgroup (INF1 and INF2), the majority of the mixed *a priori* subgroup (MIX1, MIX2, MIX4, MIX5, MIX6, MIX7, MIX8), as well as four speakers determined to be the instability subgroup. The two clusters identified based on hierarchical clustering are corroborated with finding from the MDS and are shown in the lower-dimensional projection by circles in Figure 9.

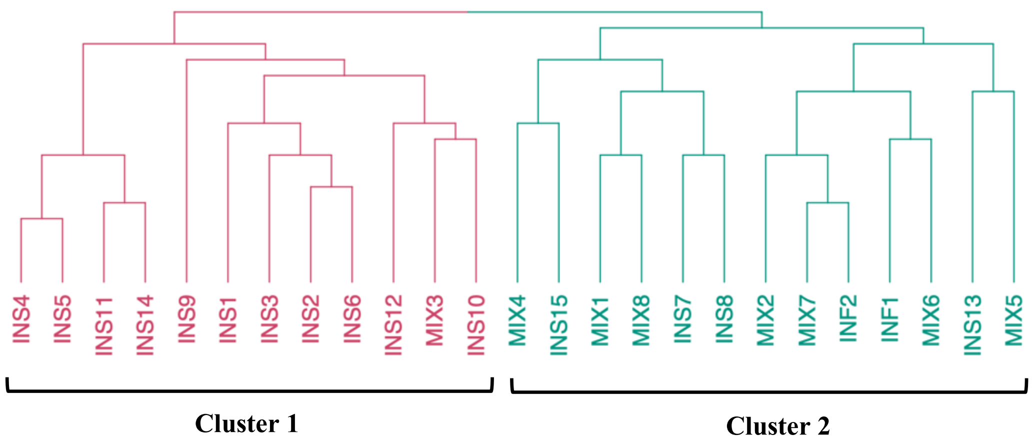

Figure 8: Dendrogram with a two-cluster solution for the connected speech task that shows the progressive linking of speakers based on listener groupings. The color coding is used to demarcate the two clusters.

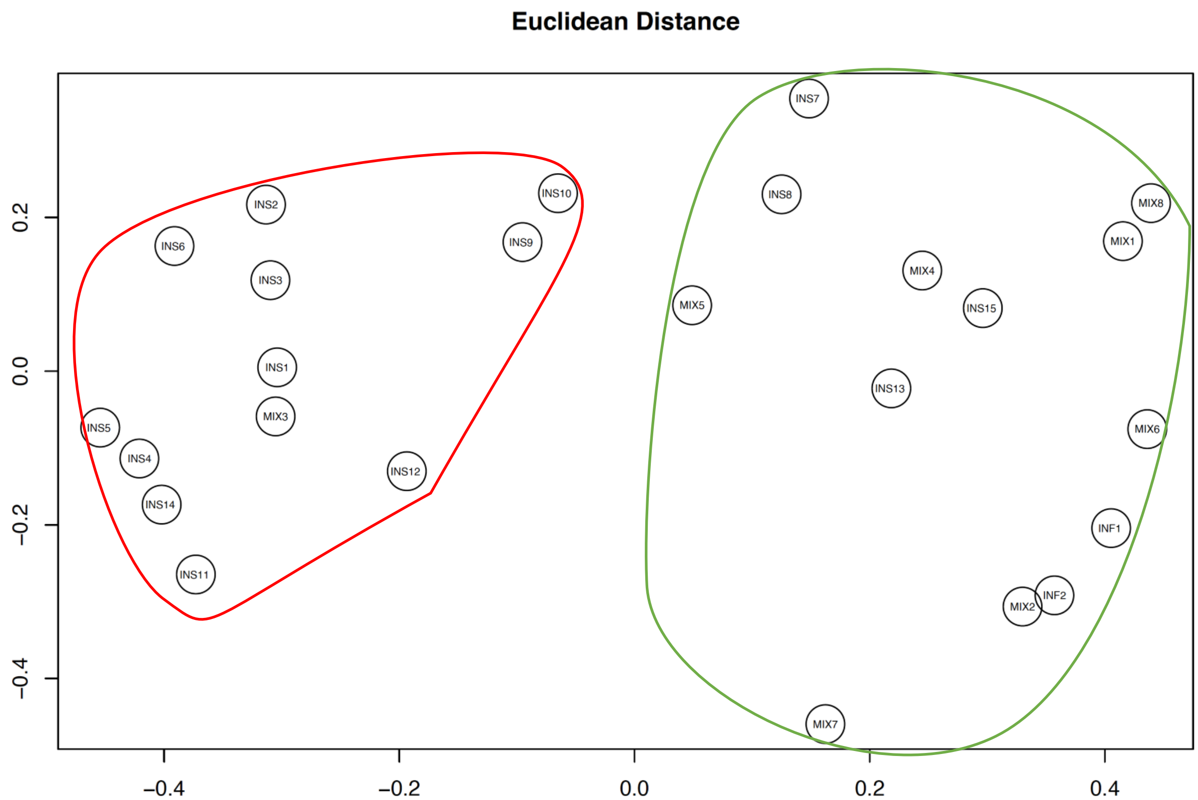

Figure 9: Multidimensional scaling plot reflecting the two-cluster solution for the connected speech task.

Across the two clusters for the Connected speech task, there was a trend with respect to gender, with Cluster 1 predominantly female (8F, 4M) and Cluster 2 predominantly male (5F, 8M) (See Table 8). Additionally, there was a pattern with respect to severity, with Cluster 1 having predominantly mild severity rankings (10 mild, 2 moderate) and Cluster 2 having predominantly moderate severity rankings (1 mild, 12 moderate); however, intelligibility did not differ significantly between clusters [t(23) = 1.12, $p = 0.27$]. Rate of connected speech differed significantly between clusters [t(23) = 2.22, $p < .05$] with Cluster 1 averaging 3.60 syll/sec and Cluster 2 averaging 3.16 syll/sec. There were no obvious patterns with respect to type of SCA or other disease etiology, and the two clusters did not significantly differ with respect to age [t(23) = -1.37, $p = 0.18$] or disease duration [t(17) = -1.70, $p = 0.11$].

The speech characteristics of Cluster 1 had a predominant pattern of irregularity in terms of articulatory breakdown, inconsistent rate, variability of stress, and difficulty coordinating speaking and breathing. The one speaker in this cluster who received an *a priori* designation as mixed (MIX3) had characteristics that more strongly aligned with instability (irregular articulatory breakdowns, variable stress, irregular AMRs, possible speech/breathing coordination difficulty, etc.). The speech characteristics of Cluster 2 reflected some irregularity in articulatory breakdowns and variation in stress, but also contained a greater predominance of speakers with slower rates, equalization of stress patterns, reduced variation in pitch, and reduced modulation of loudness. Again, the speech characteristics reported in Table 4 were derived from complete speech samples across multiple tasks, whereas listeners were making connected speech judgments based on approximately 15 words (two sentences from the Caterpillar passage).

Table 8: Speaker characteristics grouped by listener-derived clusters for connected speech.

	Speaker	*A Priori*	Gender	Age	Etiology	Disease Duration (Yrs)	Dysarthria Severity	% Mean Intelligibility	Connected Rate of Speech (Syll/sec)
Cluster 1	S02	INS1	M	74	Idiopathic Ataxia	6	Mild	99	3.02
	S03	INS2	F	49	SCA 1	7	Moderate	89	4.16
	S05	INS3	M	42	SCA 2	3	Mild	97	4.15
	S06	INS4	F	30	SCAr10	2	Mild	98	3.43
	S07	INS5	F	48	POLR3A	20	Mild	98	3.71
	S09	INS6	F	32	ARCA1	6	Mild	85	3.92
	S13	INS9	M	75	SCA 8	9	Mild	93	3.69
	S14	INS10	M	63	SCA 3	11	Moderate	81	3.45
	S15	INS11	F	60	SCA 6	2	Mild	96	3.37
	S16	INS12	F	40	SCA 2	8	Mild	93	3.35
	S18	INS14	F	48	Gluten Ataxia	3	Mild	96	3.79
	S12	MIX3	F	32	SCA 2	5	Mild	95	3.17
	Mean (SD)		**4 M 8 F**	**49.42 (14.96)**		**6.83 (4.81)**	**10 mild 2 moderate**	**93 (5.34)**	**3.60 (0.35)**
Cluster 2	S10	INS7	M	53	SCA (Unknown)	8	Mild	84	3.82
	S11	INS8	M	63	SCA 42	4	Moderate	84	3.81
	S17	INS13	M	51	Idiopathic Ataxia	5	Moderate	88	3.06
	S21	INS15	F	82	SCA 6	17	Moderate	94	3.65
	S04	MIX1	M	77	SCA 5	22	Moderate	89	3.34
	S08	MIX2	M	69	SCA 6	11	Moderate	90	2.27
	S19	MIX4	F	51	Friedreich's Ataxia	15	Moderate	95	3.47
	S20	MIX5	M	52	Idiopathic Ataxia	4	Moderate	90	4.01
	S22	MIX6	F	66	SCA (Unknown)	32	Moderate	95	2.66
	S23	MIX7	F	60	SCA 8	25	Moderate	95	2.69
	S24	MIX8	M	34	SCA 3	5	Moderate	91	3.34
	S01	INF1	M	32	SCA 2	7	Moderate	99	2.35
	S25	INF2	F	60	SCA 6	2	Moderate	90	2.61
	Mean (SD)		**8 M 5 F**	**57.69 (14.05)**		**12.08 (9.08)**	**1 mild 12 moderate**	**91 (4.28)**	**3.16 (0.57)**

Discussion

The aim of this study was to examine if speakers with ataxic dysarthria could be classified by speech-language pathologists using a free classification approach in a way that aligns with pre-established notions of subgroups of instability and inflexibility. Based on studies to date, the "instability" group has been defined by unstable control and coordination of speech subsystems, with speech characteristics that reflect this instability, such as incoordination of breathing with speaking, variable articulation errors, uncontrolled loudness variation, and variable pitch, stress patterns, and rhythm (Spencer & Dawson, 2019; Gore & Spencer, 2020). Conversely, the "inflexible" group, which has more rigid motor control patterns and isochrony, has been characterized as having a slower rate of speech, reduced variability of pitch and loudness, more consistent articulatory imprecision, and more equalized stress patterns (Gore & Spencer, 2020).

Results from this study add to the growing empirical evidence (Spencer and France, 2016; Spencer and Dawson, 2019; Gore and Spencer, 2020; Spencer, Amaral, and Lansford, 2022) that perceptually salient subgroups exist within ataxic dysarthria and these subgroups align with an instability/inflexibility motor control framework (see Hartelius et al., 2000). This project also builds on the results of previous studies using auditory free classification as a means to better understand the heterogeneity within and across the dysarthria types (Lansford, Liss, and Norton, 2014; Kim, Diehl, de Riesthal, and Mefferd, 2020). Overall, this study found that listeners were able to broadly differentiate speaking patterns (Research question 1) in a manner largely consistent with instability/inflexibility characteristics (Research question 2), despite the inherent challenges of the perceptual task. The AMR and connected speech tasks were equally successful at clustering speakers, unlike Spencer et al. (2022) which reported clearer delineation of subgroups with the AMR task.

Listeners were asked to group 25 speakers with varying severity, speech characteristics, and etiologies across two speaking tasks: AMRs and two sentences from a reading passage. Given the unconstrained nature of the free classification task, they were given no guidance other than to group speakers who sounded similar (and to disregard gender and dialectical differences). Thus, listeners could approach this task by making nuanced distinctions between speakers, broad distinctions, or something in between. Adding to the complexity of the perceptual classification task were the *a priori* characterizations of the speakers. Unlike the previous free-classification study of ataxic dysarthria (Spencer et al., 2022), the speaker sample was markedly imbalanced, with 15 speakers fitting with the instability profile and only two speakers representing the inflexibility profile. Additionally, 8 speakers were included who were determined to have a mixed profile, with characteristics of both instability and inflexibility. Despite these challenges and sources of variability, listeners grouped the two inflexibility participants together across both speaking tasks, and those speakers were most frequently paired with "mixed" speakers who had features of both inflexibility and instability. Additionally, clusters containing all, or almost all, instability speakers emerged across both the AMR and connected speech tasks.

Free classification: AMRs

Three clusters emerged from the diadochokinetic task of alternating motion rates. AMRs were chosen over SMRs for their greater sensitivity to motor control differences in speakers with ataxia (Duffy, 2020; Spencer et al., 2022). Given the preponderance of speakers classified *a priori* as fitting with the instability group, it is not surprising that two clusters contained a majority of instability speakers. One cluster (Cluster 3) contained all instability speakers (n=7) and these participants appeared to be relatively milder in presentation overall. That is, the dysarthria severity of the majority of the speakers (5/7) was determined by the *a priori* judgments to be mild in nature with only two speakers rated as moderately dysarthria. Cluster 3 also had the highest mean intelligibility of all the clusters at 94%. Cluster 1, on the other hand, contained a majority of *a priori* instability speakers (5/8), but the participants were rated as more severe by the dysarthria experts, and had relatively slower diadochokinetic rates. Thus, listeners may have been noting overall irregularities pertaining to rate, rhythm, precision, etc. across those speakers, and distinguishing them based on perceived severity and/or speed of AMR production. Alternatively, listeners may have been differentiating these two instability-dominant clusters based on other speech features, such as dysphonia (present in three speakers in Cluster 3 but no speakers in Cluster 1); however, this was not a pervasive characteristic across speakers. The final cluster, Cluster 2, appeared to largely represent those speakers who depart from the typical presentation of motor incoordination and irregularity. That is, both of the inflexible speakers, as well as the majority of mixed speakers (5/8), had features of inflexibility, such as equalized stress and reduced pitch variation as well as a relatively slower rate of speech. Given the nature of diadochokinetic tasks, it is possible that listeners were largely using rate to differentiate groups, as rate is one of the most salient perceptual features during diadochokinetic tasks (Duffy, 2020; Kent, Kim & Chen, 2022). However, the variability of rate of AMR production within each group argues against this premise as there was rate overlap for individual speakers across clusters.

Free classification: Connected Speech

Two primary clusters emerged from the connected speech task. Listeners rated two sentences from the Caterpillar passage, chosen for the relatively high number of multisyllabic words and prosodic variation. The two-sentence sample expanded upon the previous use of one sentence samples in free classification studies of dysarthria (e.g., Lansford, Liss, and Norton, 2014) while remaining a reasonable task for the SLP listeners.

Despite the challenge of this perceptual task, and the sources of variability discussed earlier, Cluster 1 emerged with almost all *a priori* instability speakers (11/12) with one mixed instability/inflexibility speaker. The mixed subgroup speaker (MIX3) exhibited predominant instability characteristics (e.g., irregular articulatory breakdown, irregular AMRs, possible speech breathing coordination difficulty), but also had a slower rate and occasional equalized stress. Cluster 2 was more of a mix of *a priori* speaker groups, but contained both inflexibility speakers as well as the majority of mixed instability/inflexibility speakers (7/8). Four of the fifteen instability speakers (INS 7, 8, 13, 15) were also included in Cluster 2. These speakers, while largely displaying characteristics associated with instability/variability, all were perceived to have an unusual rate of speech ("inconsistent rate" for INS7, INS 8, INS15, and "mildly slowed rate" for INS13). Thus, listeners may have used abnormalities related to rate of speech in their grouping strategy. This notion fits with the significantly different rate of speech between Clusters 1 and 2. However, rate of speech did not appear to be the only grouping factor between the clusters as there was considerable overlap between individual speakers (e.g., 3.02 syll/sec for INS1 in the "faster" Cluster 1 and 4.01 syll/sec for MIX5 in the "slower" Cluster 2).

Across both clusters of connected speech task there appeared to be an influence of severity with Cluster 1 speakers having a predominately mild rating of severity, and Cluster 2 speakers having a predominately moderate severity rating. However, two other metrics of severity, intelligibility and disease duration, were not significantly different between the clusters. Because intelligibility ratings are primarily driven by articulatory precision (De Bodt et al., 2002;

Wei Xue et al., 2023; Plowman-Prine et al., 2009; Medoza et al., 2021), it is possible that the increased severity ratings of Cluster 2 were motivated more by the abnormal prosody associated with the inflexibility characteristics of the 9/13 speakers who had inflexibility features (INF1, INF2 and seven mixed speakers). The "robotic" characteristics associated with inflexibility, such as equalized stress patterns, monopitch, monoloudness, and slower rate, tend to be quite perceptually salient (Spencer et al., 2022) and may have driven the difference in group severity ratings.

A pattern of gender differences between clusters was also noted. That is, Cluster 1 was predominately female (8 female, 4 male) and Cluster 2 was predominately male (5 female, 8 male). However, it is unlikely that listeners were differentiating by gender, given the instructions to disregard gender and the lack of a pattern of gender-influenced speech features (e.g., lower pitch combined with faster rate and reduced pitch variation for the male dominant group) (Leung et al., 2018; Clopper et al., 2011).

Speaker and Listener Heterogeneity

A unique contribution of this study was the high variability of speaker diagnosis (15 unique diagnoses), and range of disease duration (2 – 32 years), age of the speaker (32 – 82 years), and dialectal backgrounds from across North America. For etiology in particular, patterns within or across clusters were plausible, especially given reports of varying speech characteristics across SCA types. For example, Sidtis and colleagues (2011) found differing speech patterns across speakers with SCA1, SCA5, and SCA6 diagnoses. As the present study had a maximum of four speakers per SCA type, delineation of patterns was challenging; future studies should conduct targeted recruitment for ample representation across SCA genotypes.

Dialectal speech patterns also varied considerably and represented each region of the United States and southwest Canada. While these varied dialects could have influenced perceptual classification, listeners were instructed to disregard dialect, and no notable trends were observed across dialectal backgrounds.

Another consideration of variability with this study was the broad range of experience for listeners. Clinical experience of listeners ranged from 4 years in the field to 34 years of clinical practice. Clinical practice settings also varied with some listeners only seeing patients in acute care settings, and other listeners working across all settings. Additionally, recent caseload for listeners with patients with dysarthria ranged from “1% - 20%” to “100%” of the caseload. Previous research has found that SLPs with regular interactions with individuals with dysarthria can generalize group-specific knowledge to aid in understanding other speakers with dysarthria; which suggests that those clinicians with more dysarthria experience would be better at initially understanding the speaker with dysarthria (Borrie, Lansford, & Barrett, 2021). It is expected that a listener group with such a broad range of clinical experience will have varying degrees of expertise in listening and making determinations in this high-level perceptual classification task. Some listeners reported spending 2-4 hours making their determinations, while other performed the task within 45 minutes.[4]

Methodology for Determining Dysarthria Subgroups

The present study implemented an unconstrained auditory free-classification approach. This method allows for freedom of decision making without researcher-imposed bias. Listeners were asked to place similar-sounding speakers in groups rather than using some pre-determined metrics.

The initial study of subgroups of ataxic dysarthria (Boutsen, Bakker, and Duffy, 1997) implemented a Q methodology that utilized correlations and factor analysis of acoustic variables such as shimmer, syllable repetition rates, and jitter values. While informative, this focus on discrete variables resulted in patterns of variability that were challenging to explain and not consistently correlated with overall severity. Later, in an investigation of speech impairment profiles specific to Friedreich’s ataxia (Folker, Murdoch, Rosen, Cahill, Delatycki, Corben, and Vogel, 2012), perceptual determinations were conducted using interval rating scales and supplemented with instrumental measurements across speech subsystems. The authors found that instrumental findings generally paralleled the perceptual measures of speech subsystem functioning, adding to validity of the use of perceptual classifications in subgroup determinations.

Investigations of subgroups of ataxic dysarthria from Spencer and colleagues (Spencer & France, 2016; Spencer & Dawson, 2018; Gore & Spencer, 2020) examined if speakers would be classified into the instability/inflexibility framework based largely on visual analog scale (VAS)

[4] Listeners were able to stop and start the task as necessary so timestamps for the listener task did not necessarily reflect time needed to complete task.

ratings. VAS ratings provide a simple way to measure a subjective experience, but can be constrained in the variables used, as well as using researcher-imposed variables that can influence perceptual perspectives. The most recent investigation from Spencer and colleagues (Spencer, Amaral, & Lansford, 2022) moved to the utilization of an auditory free-classification approach with listeners grouping based on their perception of the most salient speech patterns rather than investigator-determined metrics of speech characteristics typically found in VAS. This study found consistent clustering of speakers, particularly for the AMR task, and indicated this perceptual paradigm could be used successfully for subgroup differentiation.

The relative consistency of clustering of speakers in the present investigation suggests that the auditory free-classification method is a feasible and useful tool for speaker characteristic subgroup differentiation. This method was also used successfully by Lansford, Liss, and Norton (2014) for the classification of general dysarthria types. Most recently, research from Kim et al. (2022) compared the use of auditory free classification to a statistical clustering approach using the guidelines of the Mayo Clinic dysarthria rating scales for perceptual categorization of hyperkinetic dysarthria from Huntington's disease (HD). The study demonstrated significant overlap between the subgroups of speakers using both systems, but posited that the lack of complete overlap indicated that listeners may find certain perceptual speech characteristics more salient than those suggested by the standardized rating scale.

Converging evidence suggests that auditory free classification for perceptual subgrouping is valid and informative. Future subgroup classification studies should continue to explore the relationship between auditory free-classification outcomes and acoustic and perceptual measures of speech characteristics.

Models of Cerebellar Functioning

The greater prevalence of the instability subgroup is consistent of that of previous studies (Spencer & France, 2016; Spencer & Dawson, 2018), and reinforces the potential contribution of 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 (Izzzini-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.

Clinical Implications

Establishing subgroups for ataxic dysarthria would have several important clinical implications. 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). At present, graduate students and practicing clinicians are left to reconcile the broadly heterogeneous, and at times contradictory, list of possible speech characteristics of ataxic dysarthria (see Duffy, 2020; Darley, Aronson, and Brown, 1969). Second, 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.

While listeners can successfully use a list of the key, salient features to group speakers into instability ("variable") and inflexibility ("robotic") subgroups during a connected speech task (Spencer et al., 2022), AMRs may be the most clinically feasible task to support subgroup differentiation. First, AMRs are quite sensitive to differences in cerebellar motor control because they are maximum performance motor tasks (Duffy, 2020; Spencer et al., 2022) that require fast repetition of motor sequences. While the relationship to connected speech, intelligibility, and severity of dysarthria is not always clear (Wang, Kent, Duffy, & Thomas, 2009), AMRs are easy to administer clinically and impervious to language and cognitive impairment. Future efforts to determine the predictability of connected speech from AMR performance, in subgroups of speakers with ataxic dysarthria, would be informative.

While not the direct focus of this investigation, the findings regarding the Communication Participation Item Bank (CPIB) measure deserve mention. As discussed, speakers rated as having more severe dysarthria also endorsed more interference in day-to-day life on the CPIB, which is a finding consistent with other studies of dysarthria (see Baylor, Yorkston, Bamer, Britton, & Antmann, 2010; Page & Yorkston, 2022). Interestingly, the speakers in this study were relatively mild overall, with intelligibility scores between 84-99%, yet many still reported substantial impact to their daily communication participation via the CPIB. This finding serves as an important reminder to speech-language pathologists who may assume that higher intelligibility levels translate to ease of daily interactions. It also highlights the possible contribution that prosodic changes, and perceived naturalness, may have on communication participation, even in the context of preserved intelligibility (Anand and Stepp, 2015).

Limitations and Future Directions

Future studies would benefit from larger sample of speakers (particularly those representing the inflexibility subtype) and listeners. Despite the virtual nature of data collection, recruitment of listeners was particularly challenging, presumably because of the required experience/comfort with dysarthria. The virtual data collection format also resulted in reduced control over speaker recordings and listener tasks. More specifically, headphone quality, environmental distractors, etc. may have impacted the quality of the listener's participation. Similarly, limitation on speaker's audio quality may have influenced subtle differences in speech characteristics. However, virtual platforms have been shown to be effective and valid in other perceptual investigations of dysarthria (Lansford, Borrie, and Bystricky, 2016).

Looking forward, a consideration for future investigations includes the addition of more varied speaking tasks (such as a monologue or conversation speech sample) instead of, or in addition to, the reading task. Previous research suggests a task effect with speakers with dysarthria, including ataxic dysarthria (Kim, Sidtis, & Sidtis, 2023, Weir-Mayta, Spencer, Eadie, Yorkston, et al., 2017), so integration of findings across multiple speaking tasks would be illuminating. Additionally, the implementation of acoustic analysis software for automated calculation or rate/regularity of AMRs would improve accuracy, reliability, and efficiency.

The results of this study provide promising evidence of the existence of perceptually salient subgroups within ataxic dysarthria and that these subgroups align with an instability/inflexibility motor control framework. Continued investigation in this area is important for clinical treatment and practice; it would lead to more refined selection of participants in research on ataxic dysarthria and could inform models of cerebellar functioning and disease.

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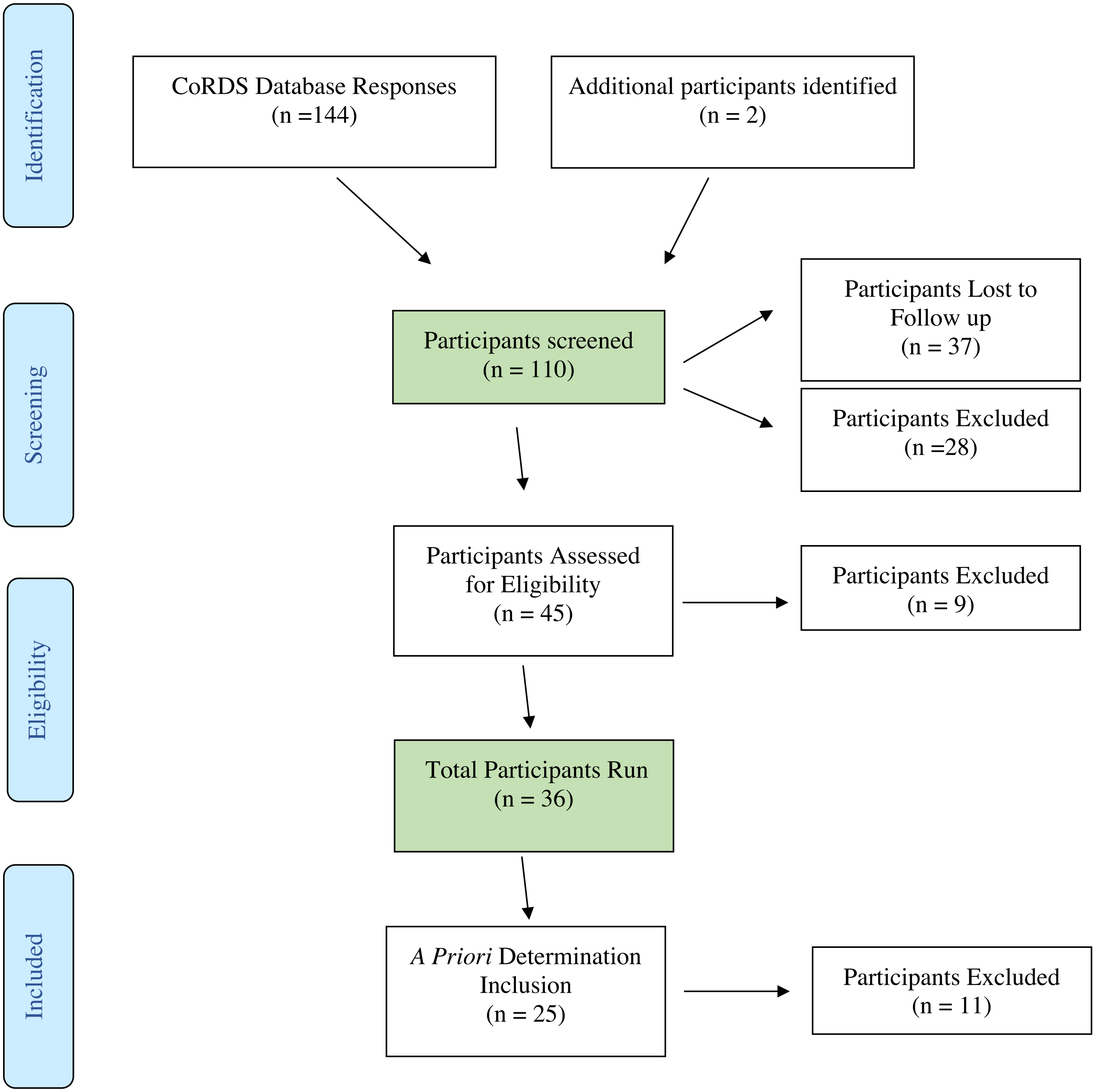
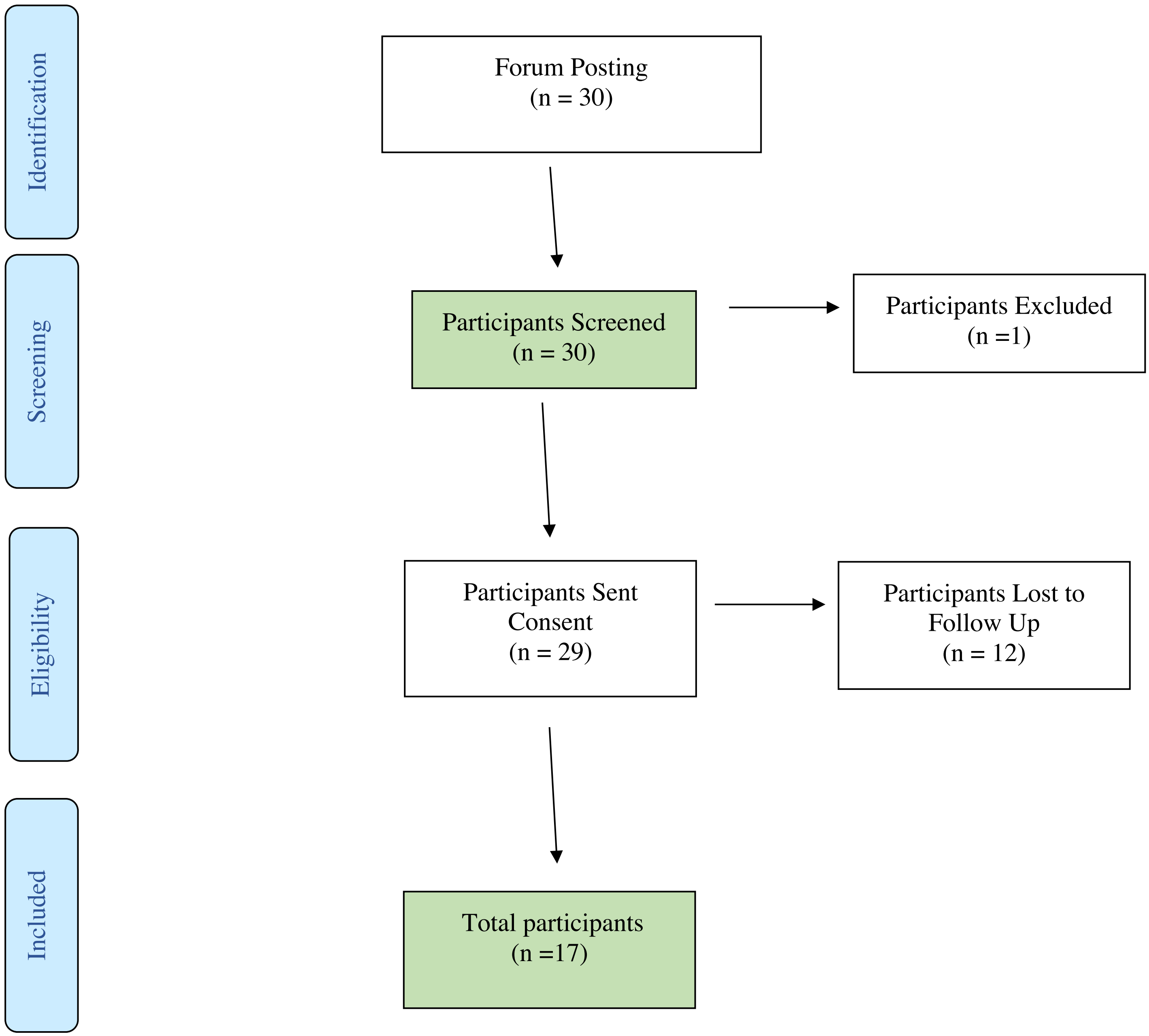
Appendix A: Flow Diagram: Speakers



Appendix B: Listener Recruitment Posting

My name is Haley Bouchard and I am conducting my master's thesis at the University of Washington with Dr. Kristie Spencer. We are in need of practicing SLPs to serve as listeners for our study about ataxic dysarthria. Listeners need to be native speakers of American English, have a minimum of 3 years of recent experience working with adults with dysarthria, and have no hearing difficulties. The experiment is conducted remotely and will take about 45-60 minutes. If you consent to the study, you will be given a 10-day timeframe to put your personal headphones on, listen to speech samples accessed on your personal computer, and make some perceptual judgments. Listeners will be compensated $20 upon completion. Please click here if you are interested in completing the screening questions. Participation is completely voluntary. If you have any questions, please feel free to contact us at motorspeechlab@gmail.com. Thank you so much for your consideration! *UW IRB: STUDY00008780, approved 12/2/22*

Appendix C: Flow Diagram: Listeners



Appendix D: Telephone Talking Points for Speakers with Ataxia

Hi, my name is Haley and I'm from a research lab at the University of Washington. We are a part of the Speech and Hearing Sciences department. We received your message about our study. If you'd like, I can tell you a little bit about it and you can decide if you'd like to hear more, okay?

We are currently conducting a study with people who have ataxia, and who have experienced changes to their speech from the ataxia. We want to learn about the different speech patterns that are caused by ataxia. Are you interested in hearing more about this study?

The study itself would take about 30 minutes and would be conducted via zoom. So you would be able to participate from your home or wherever you connect to the internet. During the session, we will audio record you doing some short talking tasks and give you a questionnaire about whether your speech interferes with your day to day life. Does this seem like something you might be interested in?

May I ask you a few more questions to see if you meet the criteria for this particular study?

1. Do you have a confirmed diagnosis of ataxia by a neurologist? Did you get that diagnosis more than a year ago?
2. Are you between the ages of 18 and 90?
3. Is English the first language you spoke?
4. Do you have a lot of problems with your hearing or vision even with glasses, hearing aids, or other corrective systems?
5. Do you have a prior learning disability?
6. Do you have a history of any neurological problems, such as a stroke, seizure disorder, head injury where you lost consciousness, etc.?

IF NOT ELIGIBLE: I'm sorry. At this time, we're only testing people who…. Thank you so much for your time and we will keep you in mind for future studies. Goodbye.

*IF ELIGIBLE***:** At this point, it sounds as though you are appropriate for the study. Would you like to get the (preliminary zoom / regular) session scheduled? We will send you a consent form to read ahead of time, then we can go over it together during the session.

Appendix E: Speakers Intake Questionnaire

Code # ___________

Demographics

Age:
Years of Education:
Location:

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 primary 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?

Appendix F: Communicative Participation Item Bank

The General Short Form of the Communicative Participation Item Bank (CPIB) and Scoring Guide

Instructions:
The following questions describe a variety of situations in which you might need to speak to others. For each question, please mark how much your condition interferes with your participation in that situation. By "condition," we mean ALL issues that may affect how you communicate in these situations, including speech conditions, any other health conditions, or features of the environment. If your speech varies, think about an AVERAGE day for your speech—not your best or your worst days.

Question	*Not at all* (3)	*A little* (2)	*Quite a bit* (1)	*Very much* (0)
1. Does your condition interfere with… **…talking with people you know?**	○	○	○	○
2. Does your condition interfere with… **…communicating when you need to say something quickly?**	○	○	○	○
3. Does your condition interfere with… **…talking with people you do NOT know?**	○	○	○	○
4. Does your condition interfere with… **…communicating when you are out in your community (e.g., errands, appointments)?**	○	○	○	○
5. Does your condition interfere with… **…asking questions in a conversation?**	○	○	○	○
6. Does your condition interfere with… **…communicating in a small group of people?**	○	○	○	○
7. Does your condition interfere with… **…having a long conversation with someone you know about a book, movie, show, or sports event?**	○	○	○	○
8. Does your condition interfere with… **…giving someone DETAILED information?**	○	○	○	○
9. Does your condition interfere with… **…getting your turn in a fast-moving conversation?**	○	○	○	○
10. Does your condition interfere with… **…trying to persuade a friend or family member to see a different point of view?**	○	○	○	○

Appendix G: “The Caterpillar” Passage

“The Caterpillar”

Do you like amusement parks? Well, I sure do. To amuse myself, I went twice last spring. My most MEMORABLE moment was riding on the Caterpillar, which is a gigantic rollercoaster high above the ground. When I saw how high the Caterpillar rose into the bright blue sky I knew it was for me. After waiting in line for thirty minutes, I made it to the front where the man measured my height to see if I was tall enough. I gave the man my coins, asked for change, and jumped on the cart. Tick, tick, tick, the Caterpillar climbed slowly up the tracks. It went SO high I could see the parking lot. Boy was I SCARED! I thought to myself, “There’s no turning back now.” People were so scared they screamed as we swiftly zoomed fast, fast, and faster along the tracks. As quickly as it started, the Caterpillar came to a stop. Unfortunately, it was time to pack the car and drive home. That night I dreamt of the wild ride on the Caterpillar. Taking a trip to the amusement park and riding on the Caterpillar was my MOST memorable moment ever!

Appendix H: Listener Questionnaire

1. Name: ______________________________

2. Age: _______

3. Education (years): _______

4. Native language (first language spoken): ____________________________

5. Do you speak any other languages fluently? _________________________

6. Do you have any history of speech or language difficulties? ______________

7. Do you have any difficulty with your hearing? ________________________

8. How many years of clinical experience do you have in adults with dysarthria?
____

7. What percentage of your current caseload is adults with dysarthria?
   a) 0
   b) 1% - 20%
   c) 21% - 40%
   d) 41% - 60%
   e) 61% -80%
   f) 81% - 100%

8. In what setting(s) have you worked with adults with dysarthria?
   - ❑ Outpatient clinic
   - ❑ Home Health
   - ❑ Acute care
   - ❑ Inpatient rehabilitation
   - ❑ Skilled nursing facilities
   - ❑ Research/University settings

Appendix I: Listener Email Communication First Contact

Hello!

My name is Haley Bouchard and I am reaching out regarding your interest in our ataxic dysarthria listener study. Thank you for completing the intake form! It appears you are eligible to participate in our study. To learn more about the study, please read the attached consent form. You do not need to sign or return this form to me. However, please let me know that you've read the form and agree to participate. We are happy to answer any questions about the study or consent form before you do so.

The study itself should take 45-60 minutes to complete on your computer. We ask that you do the study in a quiet environment with well-fitted headphones. Additionally, we ask that you complete the study within ten days. Upon completion, we will send you compensation via Venmo or mailed check. Please let me know your preference for compensation method and either your Venmo handle or address for mailing a check.

Thank you again for your interest. Please let me know if you have any questions. Once I receive your acknowledgement of consent (and compensation method), I will email you the study materials!

Best,
Haley

Appendix J: Listener Email Communication Study Materials

Hello,

Thank you for your response and for consenting to participate in our study of ataxic dysarthria. To proceed, you will need to click the link below to open the study materials in Google Slides and complete the tasks as described. The instructions for the listening task are on the first three slides.

When you are finished, please respond to this email to notify us. Please complete study within 10 days of this email **(Date+10**). Once the completed study is received, compensation will be sent via **mail or *venmo***. If you have any questions before beginning, please feel free to email me!
Study Materials

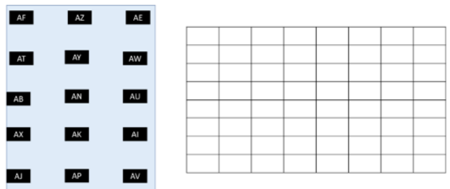
Thank you so much for your participation!

Haley

Appendix K: Listener Task Instructions

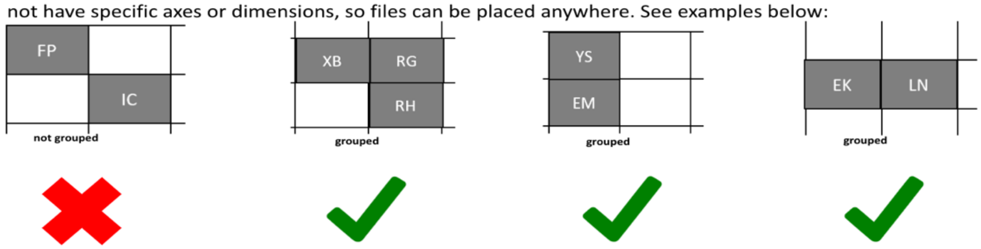
Thank you for participating in our experiment!

- To get started, please make sure you are in a quiet environment and using the highest quality headphones possible.
- We will be asking you to listen to speech samples and group them according to how similar they sound. This is a challenging perceptual task with no right answers, just do the best you can!
- On the following PowerPoint slides, you will find speaker files on the left and an empty grid on the right. Each speaker is given random initials. Here's an example:



- To listen to a file, click on the one you would like to hear and press "play" when the control bar appears.

- Each file represents an individual speaker with ataxic dysarthria.
- On one slide, the speakers are performing diadochokinetics (repetitions of /pə/, /tə/, and /kə/). On the other slide, they are saying 2 sentences. Please work through the slides in the order presented.
- Group the files by clicking and dragging them into the grid depending on how similar they sound to you. The PowerPoint must remain in “Edit” mode, rather than Slideshow, for you to make your groupings.
- You can listen to each file as many times as necessary.
- When grouping the speaker files, be sure that speakers in the same group are touching in the grid. The grid does not have specific axes or dimensions, so files can be placed anywhere. See examples below:



- Keep in mind that speakers are a mix of **males and females**, and perceived sex should **not** be used as a factor in your grouping decisions.
- Our speakers are from a broad range of geographic and **dialectal backgrounds**, so these perceived dialectal differences should **not** be used as a factor in your grouping decisions.
- You are permitted to make as many groups as needed with as many speakers in each group as needed.
- There is no time limit. You can listen to and re-arrange the files as many times as necessary.
- The notes section at the bottom of the slide is available for you to use however you see fit, but notes are not required.
- **The final product should be groups of speaker files that sounded similar to you.**
- When you are finished, email motorspeechlab@gmail.com to notify. If you have any questions before beginning, please contact Haley Bouchard at heb1218@uw.edu.

Thank you in advance for your help!

Appendix L: Matrix Coding Python Script

```python
#!/usr/bin/env python3
# -*- coding: utf-8 -*-
"""
Created on Thu Jun  8 17:49:25 2023
@author: haleybouchard
"""
from pptx import Presentation
import pandas as pd
import numpy as np
import os

#Folder that contains each particpants files
ppt_folder = r'/Users/haleybouchard/Dropbox/UW/Thesis/Matrix Coding/Powerpoints for Coding'

#File that containes the mapping between the letters and audio links
label_path = r'/Users/haleybouchard/Dropbox/UW/Thesis/Matrix Coding/Letters to Links.xlsx'

#Matrix Output Location
output_path = r'/Users/haleybouchard/Dropbox/UW/Thesis/Matrix Coding/Matrix_Output.xlsx'


#The slide numbers start at 0 i.e. slides 4 & 5 need to be entered as 3 & 4 respectivly
slide_no_1 = 3
slide_no_2 = 4

#The pair threshold is how close the boxes can be to each other to be considered pairs.
# A value of 1 indicated that two boxes are only pairs if their top left corners are
# within 1x of the height or width of each other (to the left/right or above/below).
#A value of 1.25 would be within 1.25x of the height or width so there can be a small gap.

pair_threshold = 0.25

labels = pd.read_excel(label_path)
letter_list = list(labels['Letters'])
speaker_list = list(labels['Speaker'])
link_list = list(labels['Audio Link'])

label_dict = dict(zip(link_list,tuple(zip(speaker_list,letter_list))))

def matrix_generator(slide,label_dict,pair_threshold):
# This function contains the logic to creat a matrix

    location_list = []
    name_list = []

    #Each shape that is in the "Picture" format has its location and dimension recorded. The audio link name is also recorded.
    for shape in slide.shapes:


        if shape.__class__.__name__ == 'Picture':

            address = shape.click_action.hyperlink.address

            name = label_dict[address]

            name_list.append(name)

            location_list.append([shape.left,shape.top,shape.width,shape.height])

        else:

            next

    # Converts name_list to multi-level
    name_list = pd.MultiIndex.from_tuples(name_list)

    count = len(location_list)

    pair_array = np.empty([count,count])


    #Pairs are generated for every rectangle that is next to another rectangle. This is determined by the x and y-axis locations.
    for column in range(0,count):

        for row in range(0,count):

            x_loc =   location_list[row][0]
            x_0_min = location_list[column][0]-(location_list[column][2]*pair_threshold)
            x_0_max = location_list[column][0]+(location_list[column][2]*pair_threshold)
            x_min =   location_list[column][0]-(location_list[column][2]*(1+pair_threshold))
            x_max = location_list[column][0]+(location_list[column][2]*(1+pair_threshold))

            y_loc =   location_list[row][1]
            y_0_min = location_list[column][1]-(location_list[column][3]*pair_threshold)
            y_0_max = location_list[column][1]+(location_list[column][3]*pair_threshold)
            y_min =   location_list[column][1]-(location_list[column][3]*(1+pair_threshold))
            y_max = location_list[column][1]+(location_list[column][3]*(1+pair_threshold))

            y_match = (x_loc < x_0_max) & (x_loc > x_0_min) & (y_loc < y_max) & (y_loc > y_min)

            x_match = (y_loc < y_0_max) & (y_loc > y_0_min) & (x_loc < x_max) & (x_loc > x_min)


            if x_match | y_match:

                pair_array[row,column] = 1

            else:

                pair_array[row,column] = 0

    set_list = []
    group_list = []

    #Each pair in the pair array is checked agains other pairs to capture common pairs and generate groups.
    for a in range(0,len(pair_array)):

        set_a = set(list(np.where(pair_array[a,:]==1)[0]))

        for b in range(0,len(pair_array)):

            set_b = set(list(np.where(pair_array[b,:]==1)[0]))

            if not(set_a.isdisjoint(set_b)):

                set_a = (set_a | set_b)

        set_list.append(set_a)
        group_list.append(list(set_a))

    group_list = np.unique(group_list)

    group_change = True
    change_count = 0

    #The groups are checked against each other until there are no more common pairs found. This code is likely redundant in most cases.
    while group_change:

        for a in range(0,len(group_list)):

            set_main = set(group_list[a])

            for b in range(0,len(group_list)):

                set_comp = set(group_list[b])

                if not(set_main.isdisjoint(set_comp)):

                    set_main = (set_main | set_comp)

                    change_count+1

            group_list[a] = list(set_main)

            if change_count == 0 and a == (len(group_list) -1):

                group_change = False

    group_list = np.unique(group_list)

    group_array = pair_array.copy()

    for group in group_list:

        for i in range(0,len(pair_array)):

            if i in group:

                for member in group:

                    if i == member:

                        group_array[i][member] = 0

                    else:

                        group_array[i][member] = 1


    group_matrix = pd.DataFrame(group_array, columns = name_list, index = name_list)

    return group_matrix


def matrix_to_excel(folder,output_path,label_dict,slide_no_1,slide_no_2,pair_threshold):
#This function formats the group matricies, creates an excel file, and pastes the results from each powerpoint.

    file_no = 0

    writer = pd.ExcelWriter(output_path,engine='xlsxwriter')
    workbook=writer.book
    worksheet=workbook.add_worksheet('Result')
    writer.sheets['Result'] = worksheet

    row = 0


    for file in os.listdir(folder):

        if file[-5:] != '.pptx':

            continue

        input_path = folder+'//'+file
        prs = Presentation(input_path)

        slide1 = prs.slides[slide_no_1]
        slide2 = prs.slides[slide_no_2]

        matrix1 = matrix_generator(slide1,label_dict,pair_threshold)
        matrix2 = matrix_generator(slide2,label_dict,pair_threshold)


        column = matrix1.shape[1]

        worksheet.write_string(row+2, 0, file[:-5])

        worksheet.write_string(row+3, 0, matrix1.columns[0][1][0])

matrix1.to_excel(writer,sheet_name='Result',startrow=(row+3) , startcol=0)

        worksheet.write_string(row+3, column + 3, matrix2.columns[0][1][0])

matrix2.to_excel(writer,sheet_name='Result',startrow=(row+3), startcol=column + 3)

        file_no = file_no + 1
        row = row+column+5

    writer.save()
    writer.close()
    return


matrix_to_excel(ppt_folder,output_path,label_dict,slide_no_1,slide_no_2,pair_threshold)

print('Done')
```