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Estimates of the prevalence of speech and motor speech disorders in adolescents with Down syndrome

Erin M. Wilson^a, Leonard Abbeduto^b, Stephen M. Camarata^c, and Lawrence D. Shriberg^d

^aSpeech and Feeding Disorders Laboratory, MGH Institute of Health Professions, Boston, MA, USA; ^bMIND Institute and Department of Psychiatry and Behavioral Sciences, School of Medicine, University of California, Davis, CA, USA; ^cDepartment of Hearing and Speech Sciences, Vanderbilt University, Nashville, TN, USA; ^dIntellectual and Developmental Disabilities Research Center, Waisman Center, University of Wisconsin-Madison, Madison, WI, USA

ABSTRACT

Although there is substantial rationale for a motor component in the speech of persons with Down syndrome (DS), there presently are no published estimates of the prevalence of subtypes of motor speech disorders in DS. The goal of this research is to provide initial estimates of the prevalence of types of speech disorders and motor speech disorders in adolescents with DS.

Conversational speech samples from a convenience sample of 45 adolescents with DS, ages 10 to 20 years old, were analysed using perceptual and acoustic methods and measures in the Speech Disorders Classification System (SDCS). The SDCS cross-classified participants into five mutually exclusive speech classifications and five mutually exclusive motor speech classifications. For participants meeting criteria for Childhood Dysarthria or for Childhood Dysarthria concurrent with Childhood Apraxia of Speech, the SDCS provided information on participants' percentile status on five subtypes of dysarthria.

A total of 97.8% of participants met SDCS criteria for Speech Disorders and 97.8% met criteria for Motor Speech Disorders, including Childhood Dysarthria (37.8%), Speech Motor Delay (26.7%), Childhood Dysarthria and Childhood Apraxia of Speech (22.2%), and Childhood Apraxia of Speech (11.1%). Ataxia was the most prevalent dysarthria subtype.

Nearly all participants with DS in the present sample had some type of speech and motor speech disorder, with implications for theory, assessment, prediction, and treatment. Specific to treatment, the present findings are interpreted as support for motor speech disorders as a primary explanatory construct to guide the selection and sequencing of treatment targets for persons with DS.

Abbreviations: CAS: Childhood Apraxia of Speech; CD: Childhood Dysarthria; DS: Down syndrome; NSA: Normal(ized) Speech Acquisition; PSD: Persistent Speech Delay; PSE: Persistent Speech Errors; SD: Speech Delay; SDCS: Speech Disorders Classification System; SE: Speech Errors; SMD: Speech Motor Delay

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Motor speech disorders in Down syndrome

The research literature in Down syndrome (DS) includes frequent reference to a 'motor component' in the speech of children, youth, and adults with DS. Examples of such

CONTACT Lawrence D. Shriberg  shriberg@waisman.wisc.edu  University of Wisconsin-Madison, Waisman Center, Room 439, 1500 Highland Ave, Madison, WI 53705

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findings over the past three decades include Hamilton (1993), who in an early palatography study reported support for motor programming deficits and impaired tongue motility and more recently, Rupela, Velleman, and Andrianopoulos (2016), who interpreted their behavioural findings in a study of seven children with DS as support for concurrent dysarthria and apraxia. Following is a brief sample of the neuroanatomical findings that provide rationale for an initial estimate of the prevalence of motor speech disorders in DS using recently available behavioural methods and measures.

Neuroanatomical studies of adolescents with DS using autopsy findings and more recently using neuroimaging methods have consistently reported smaller cerebellar volumes among other anatomical brain differences (Coppens-Hofman, Maassen, van Schroyen, Lantman-de Valk, & Snik, 2012; Pinter, Eliez, Schmitt, Capone, & Reiss, 2001; see Lott, 2012 for review). Additional overviews and discussions of the cerebellum in brain networks relevant to speech and prosody disorders in DS include Ackermann (2008), Bayat, Hosseini, Shamsi, and Nami (2016), Breska and Ivry (2016), Chu and Barlow (2016), and Fiez (2016).

The presence of generalized hypotonia in individuals with DS is well-documented in the literature (Lott, 2012) and reportedly plays a role in delayed motor skill acquisition; however, a conclusive causal association between hypotonia and delayed motor development in children with DS has not been documented (Vicari, 2006). Hypotonia specific to the articulatory system has also been reported (Bunton & Leddy, 2011; Coppens-Hofman et al., 2012; Limbrock, Castillo-Morales, Hoyer, Stöver, & Onufer, 1993; Stoel-Gammon, 2001) and may explain some speech production characteristics of individuals with DS. Features of ataxic or flaccid dysarthria secondary to the observed hypotonia in individuals with DS have been described (Kent & Vorperian, 2013). Physiologic findings suggest that individuals with DS have limited tongue movement during vowel production and as a result, a reduction in acoustic vowel space, articulatory working space, and articulatory speed (Bunton & Leddy, 2011). But some researchers suggest that the anomalous muscle tone in individuals with DS does not influence the speech musculature sufficiently to alter speech production (Connaghan & Moore, 2013).

Classification of motor speech disorders

A primary constraint on research questions about motor speech impairment in DS is the lack of a classification system for childhood motor speech disorders that includes operationalized and standardized perceptual and acoustic measures. The Method section and a Supplement for this research series [Supplementary Data] provide detailed information on methods and measures in the finalized version of a classification system termed the Speech Disorders Classification System (SDCS) developed for research in childhood speech sound disorders (Shriberg, Kwiatkowski, & Mabbie, 2019). The SDCS, which provides the research framework for the present study, has been used to classify participants' motor speech status in a number of studies of the genomic and behavioural substrates of persons with speech sound disorders (e.g., Baylis & Shriberg, 2018; Eising et al., 2018; Laffin et al., 2012; Raca et al., 2013; Redle et al., 2015; Rice et al., 2012; Shriberg, Jakielski, & El-Shanti, 2008; Shriberg, Paul, Black, & van Santen, 2011; Shriberg, Potter, & Strand, 2011; Truong et al., 2016; Worthey et al., 2013).

Statement of purpose

Notwithstanding substantial rationale for some type of motor component in the speech of persons with DS, there presently is no published source that estimates the prevalence of subtypes of motor speech disorders in persons with DS. The research in this report used the SDCS to address the following question:

What are the estimated prevalences of speech disorders and motor speech disorders in adolescents with DS?

Method

Participants

A database of originally 50 conversational speech samples was assembled from one retrospective study and two prospective studies. The assessment protocol in each of the three sites was similar, including a high-quality audio-recorded conversational speech sample. All participants in the three samples used speech as their primary means of communication and were native English speakers, according to parental report. Information about the data collection process at each site are detailed below.

Sample 1

The first of three samples of participants with DS included retrospective data from 29 participants, 10–18 years of age, who had been enrolled in a study of children with neurodevelopmental disorders at the University of Wisconsin-Madison (Abbeduto et al., 2003, 2008; Keller-Bell & Abbeduto, 2007). Participants were assessed in quiet rooms using recruitment procedures and an assessment protocol approved by an institutional review board. Participants had been recruited locally and nationally from support groups and professional associations, from specific urban areas using newspaper advertisements, and through a University participant research registry. The diagnosis of Trisomy 21 was confirmed through parental report, which was also confirmed in a majority of the cases by medical reports. The mother, father, and teacher of each participant were asked to complete the Autism Behavior Checklist (Krug, Arick, & Almond, 1980). If 2 of the 3 individuals scored the participant as ‘high’ (above 44), the participant was given a full autism evaluation. None of the participants in the present study met criteria for a diagnosis of autism spectrum disorder. The assessment protocol included a high-quality audio-recorded conversational speech sample in which participants were asked to talk about a series of daily life topics, including friends, families, and school. The analog recordings were digitized using conventional procedures (Flipsen, Tjaden, Weismer, & Karlsson, 1996).

Sample 2

A second sample of 17 participants with DS, 10–18 years of age, was prospectively recruited and assessed at the University of Wisconsin-Madison using procedures approved by an institutional review board. The catchment area included participants from local associations, schools, and clinics, and from facilities throughout Wisconsin and northern Illinois. All participants had a diagnosis of DS (Trisomy 21) confirmed by parental report and medical records where possible. Parents or caregivers completed a health and development inventory. Questions about participants’ medical, educational, and psychosocial

histories assessed the potential presence of autism or what was formerly termed Pervasive Developmental Disorder. The examiner, a paediatric speech-language pathologist, watched for autistic-like behaviour during the evaluation. The data for one potential participant was excluded from the present analyses due to a history and clinical presentation consistent with Pervasive Developmental Disorder. The assessment protocol, conducted in a quiet room, included a high quality digital audio-recorded conversational speech sample in which participants were asked to talk about a series of daily life topics, including friends, families, and school (Shriberg et al., 2010a).

Sample 3

A third cohort included four male participants with DS, ages 13–20 years, prospectively assessed by a paediatric speech-language pathologist in a quiet room at Vanderbilt University Medical Center. The four participants, who were currently participating in other clinical-research projects (Camarata, Yoder, & Camarata, 2006; Davis, Camarata, & Camarata, 2016), were assessed using the same protocol approved by the local institutional review board as the protocol used with the second cohort of adolescents with DS, including a high quality digital audio-recorded conversational speech sample in which participants were asked to talk about daily life topics.

Preliminary analyses were completed to look for potentially relevant demographic and cognitive-linguistic characteristics of participants in the three samples. Although the descriptive findings for the four participants in the third cohort were consistent with those in the other two groups, only the two larger groups had a sufficient number of participants to test for significant between-group mean differences. The mean ages (in years) of participants in the three cohorts in the order described were 14.5 ($SD = 2.0$), 12.9 ($SD = 2.8$), and 15.8 ($SD = 3.1$), with a non-significant mean difference between the first two cohorts using 95% confidence intervals. The proportion of males in the first cohort (69%) was significantly larger than the proportion in the second cohort (29.4%; Fisher's Exact test p -value: 0.014).

The mean standardized IQ of participants in the first cohort (41.4, assessed with the Copying, Bead Memory, and Pattern Analysis subtests of the *Stanford-Binet Intelligence Scale, 4th edition*; Thorndike, Hagen, & Sattler, 1986), was significantly lower than the standardized IQ of participants in the second cohort (49.2; effect size: 1.09; confidence interval: 0.45–1.73) assessed with the *Kaufman Brief Intelligence Test – Second Edition* (KBIT-2; Kaufman & Kaufman, 2004). It is unclear whether the significant difference between the two samples is associated with participants or measures. The mean standardized IQ of the four participants in the third cohort assessed with the *Leiter International Performance Scale-Revised* (Roid & Miller, 1997) was 47.5.

Last, a language measure in conversational speech termed Average Words per Utterance, which is highly correlated with Mean Length of Utterance ($r > 0.90$ in pre-school-aged children; Shriberg & Kwiatkowski, 1994), was significantly greater for participants in the first cohort (5.0) than participants in the second cohort (3.5; effect size: 0.89; confidence interval: 0.21–1.58). The Average Words per Utterance for participants in the third cohort was 4.6.

To maximize the diversity and statistical power of participants with DS for the prevalence questions proposed, data for the three cohorts were combined to comprise a database of 50 participants. The prevalence analyses to be reported were completed on the 45 participants

whose conversational speech samples met requirements for classification of their status on the marker for childhood apraxia of speech (to be described). Specifically, childhood apraxia status could not be determined for five participants (10%) of the original sample due to one or both of two constraints. The first constraint was an insufficient number of utterances, due to low verbal output, limiting prosody-voice analyses and pause opportunities on the apraxia of speech marker. In associated research also using this marker with retrospective data, childhood apraxia status was classifiable for 415 of 448 (93.5%) children with idiopathic Speech Delay (SD; Shriberg, Kwiatkowski, & Mabbie, 2019) and averaged 95.7% classifiable samples for 312 participants with seven types of complex neurodevelopmental disorders (Shriberg, Strand, Jakielski, & Mabbie, 2019; the present group of adolescents with DS excluded). In prospective studies, children with this constraint are typically resampled on the same or another day to obtain a sufficient number of pause opportunities to compute a score on the apraxia marker. The second constraint was missing scores on the nonword repetition task used to resolve indeterminate scores on the apraxia marker because the participants had been assessed before the nonword repetition task, that provides part of this information, became available (cf. Shriberg et al., 2009). Table 2, discussed presently, includes a summary description of the 45 participants with DS.

Procedures

Data reduction and reliability estimates

Four research specialists completed narrow phonetic transcription, prosody-voice coding, and/or acoustic analyses on the original 50 conversational speech samples using methods and measures developed for research in speech sound disorders (Mabbie & Shriberg, 2017; Shriberg et al., 2010a, 2010b). A fifth person checked the phonetic transcriptions and prosody-voice coding for formatting errors and entered all data into a software suite titled *Programs to Examine Phonetic and Phonologic Evaluation Records* (PEPPER; 2019).

Table 1 includes estimates of the point-to-point interjudge and intrajudge reliabilities of the perceptual and acoustic data reduction methods obtained from the same randomly selected 10 (20%) participants. The first two sections of Table 1 include averaged interjudge and intrajudge transcriber agreement estimates for the transcribers who completed narrow phonetic transcription and prosody-voice coding of the 50 conversational samples. The average point-to-point interjudge and intrajudge agreement for the 10 transcription and prosody-voice variables ranged from 74.9% to 94.1%, which is consistent with the range of reliability estimates reported for these variables in research literatures in childhood speech sound disorders (cf. Shriberg et al., 2010b). The third section of Table 1 includes point-to-point interjudge and intrajudge agreement for 12 acoustic variables analysed in the PEPPER environment by the two acoustic analysts. With the exception of the intrajudge agreement estimate of 61% for Appropriate-Not Appropriate pauses used in the classification of childhood apraxia of speech, percentages of agreement for the remaining 11 variables ranged from 78% to 98%, also consistent with the range of reliability estimates reported for these variables in the research literature in paediatric speech sound disorders (cf. Shriberg et al., 2010b). The low estimate of intrajudge (but not interjudge) agreement on Appropriate-Not Appropriate pauses in Table 1 has not been observed in associated research. As reported elsewhere, interjudge and intrajudge agreement estimates for Appropriate-Not Appropriate pauses based on considerably larger sample sizes have averaged in the mid 70% to low 80% range (e.g., Shriberg et al., 2017).

Table 1. Estimates of the interjudge and intrajudge agreement for phonetic transcription, prosody-voice coding, and acoustic analyses.

Data	Agreement Types		No. of Tokens Analyzed	Variable	Average Percentage of Agreement		
	Interjudge	Intrajudge					
Phonetic Transcription	X		901 Utterances; 1977 Words	Broad	93.3		
				Narrow	74.0		
				Vowels			
				Broad	85.8		
	X		901 Utterances; 1996 Words	Narrow	73.0		
				Consonants			
				Broad	94.1		
				Narrow	78.0		
Prosody-Voice Coding	X	X	234 Utterances	Appropriate-Inappropriate	87.2		
				Appropriate-Inappropriate	91.1		
Acoustic Analyses	X		152 Consonants 784 Vowels	Phoneme			
				Duration			
				Consonants	84.5		
				Vowels	85.8		
			X		150 Consonants 783 Vowels	Phoneme	
						Duration	
						Consonants	89.0
						Vowels	87.1
	X		793 Vowels 142 Vowels 144 Vowels	Vowel Frequency			
				F0	97.9		
				F1	91.2		
				F2	95.2		
			X		792 Vowels 157 Vowels 156 Vowels	Vowel Frequency	
						F0	98.1
						F1	91.1
						F2	95.0
X		170 Pauses 160 Pauses 28 Pauses	Pause Variables				
			Pause – Non-Pause	94.1			
			Appropriate – Not Appropriate	77.5			
			Type 1 – Type 2	78.6			
		X		170 Pauses 155 Pauses 36 Pauses	Pause Variability		
					Pause – Non-Pause	91.2	
					Appropriate – Not Appropriate	61.3	
					Type 1 – Type 2	80.6	

Speech and motor speech classifications and measures

The perceptual and acoustic data from the conversational speech samples were used to classify participants into the five SDCS speech classifications and five motor speech classifications (Shriberg, Kwiatkowski, & Mabile, 2019). The Appendix, and a Supplement for this research series [Supplementary Data], provide detailed information on classification methods and measures. These documents include descriptions of the speech and motor speech classifications, and for participants meeting classification criteria for dysarthria, the five dysarthria subtypes.

Appendix

The speech classifications described in the Appendix are consistent with contemporary nosology in childhood speech sound disorders (e.g., Bernthal, Bankson, & Flipsen, 2009; Bowen, 2015; McLeod & Baker, 2017; Rvachew & Brosseau-Lapr e, 2016). Speech sound disorders are divided into Speech Errors (SE) and SD. To accommodate studies that include children with early disorders (e.g., SD) and those with persistent disorders (e.g., PSD), a slash convention (e.g., SD/PSD) is used to aggregate participants younger and older than 9 years of age (Shriberg, Gruber, & Kwiatkowski, 1994). Classifications are accomplished by software (PEPPER, 2019; see Shriberg et al., 2010a for an overview of PEPPER) that standardizes segmental information by age and sex using two reference databases totaling 200 typical speakers 3 to 80 years of age (Potter et al., 2012; Scheer-Cohen et al., 2013).

Two conventions are used to minimize the number of speech and motor speech classifications in the SDCS. First, in addition to including speakers with no speech errors, the Normal or Normalized Speech Acquisition (NSA) classification includes speakers with age-appropriate distortions, deletions, and substitutions (speakers with such behaviours in conversational speech are coded NSA- in the PEPPER software). Thus, an NSA classification does not necessarily indicate that a speaker makes no speech errors, but rather that his/her speech does not meet criteria for any of the four speech disorder classifications (i.e., NSA is the default classification).

A second convention is that speakers who meet criteria for both SE and SD are assigned the more severe classification, SD. Most speakers with SD also have common clinical distortion errors. Similarly, speakers who meet criteria for Speech Motor Delay (SMD) and one of the other three motor speech disorders described in the Appendix are assigned that classification, rather than SMD.

Notice in the Appendix that the motor speech classification of Childhood Dysarthria (CD) concurrent with Childhood Apraxia of Speech (CAS), abbreviated CD & CAS, is not just a coding procedure to make the motor speech classifications mutually exclusive. Concurrent rather than single deficits in neuromotor execution (dysarthria) and neuromotor planning/programming (apraxia) have significant genomic, neuropathological, and clinical correlates (see, for example, the implications of concurrent dysarthria and apraxia compared to apraxia alone in the *FOXP2* and other speech-genetics literatures; e.g., Carrigg, Parry, Baker, Shriberg, & Ballard, 2016; Morgan, Fisher, Scheffer, & Hildebrand, 2016; Rice et al., 2012; Shriberg et al., 2008; Worthey et al., 2013).

Supplement

The Supplement provides an overview of the methods and measures used to classify each participant's speech status and their motor speech status. The Supplement includes a graphic (figure S1) that illustrates the classification procedure and rules for classification. The Supplement also includes descriptions of the three measures used for motor speech classification, and copies of two of the three measures. Table S1 is the Precision-Stability Index, which is used to identify and quantify SMD and table S3 is the Dysarthria Index and Dysarthria Subtype Indices, used to identify CD, CD & CAS, and five subtypes of CD. Tables S2 and S4 include the calculations for each of the 32 Precision-Stability Index signs and 34 Dysarthria Index signs completed by the computer program. The Supplement also includes a brief description of the Pause Marker and Pause Marker Index, which, as

described elsewhere, are used respectively to identify and quantify the severity of CAS (Shriberg, Kwiatkowski, & Mabile, 2019; Shriberg et al., 2017; Tilkens et al., 2017).

Description of participants

Table 2 includes a brief summary of characteristics of the 45 participants (from the original group of 50 participants) who met SDCS requirements for cross-classification of their speech and motor speech status. The summary demographic, intelligence, language, and speech statistics for these participants are consistent with those reported in the literature on speech-language disorders in persons with DS. The standard deviations and/or ranges for the intelligence, language, and speech variables indicate a wide range of individual differences in cognitive, language, and speech status. Notably for the present context, some of the current participants had lower than typical speech scores on the Percentage of Consonants Correct (59.3%-93.3%), Percentage of Vowels Correct (77.7%-96.7%), and Percentage of Intelligible Words (50.1%-99%). The reduced intelligibility findings in Table 2 are the focus of research reported with the present participants in Wilson, Abbeduto, Camarata, and Shriberg (2019).

Results and discussion

Figure 1 is a summary of the cross-classification findings for the 45 participants with DS. The rows in Figure 1 include prevalence findings for NSA at the time of assessment and prevalence findings for the four speech disorder classifications. The speech classifications are arranged vertically in increasing order of clinical severity. The columns in Figure 1 include prevalence findings for the five SDCS motor speech classifications, arranged horizontally in putative increasing order of clinical severity.

Table 2. Description of 45 participants with Down syndrome.

Variable	<i>n</i>	%	<i>M</i>	<i>SD</i>	Minimum	Maximum
Demographic						
Chronological Age (yrs.)	45		14.2	2.3	10.0	20.0
% Male	45	55.6				
Intelligence						
IQ Composite ^a	15		47.4	5.3	40	57
Intelligence Quotient ^b	27		40.3	5.5	36	50
Language						
Average Words per Utterance	45		4.6	1.9	1.9	9.3
Oral Composite ^c	14		42.6	4.4	40	56
Speech						
Percentage of Consonants Correct	45		78.9	8.7	59.3	93.3
Percentage of Vowels Correct	45		89.9	4.3	77.7	96.7
Intelligibility Index (%) ^d	45		81.3	12.3	50.1	99.0

^a Standard scores for IQ Composite: *Kaufman Brief Intelligence Test – Second Edition*; Kaufman and Kaufman (2004).

^b IQ Scores: *Stanford-Binet-4th Edition*; Thorndike et al. (1986).

^c Standard scores for Oral Composite: *Oral and Written Language Scales (OWLS)*; Carrow-Woolfolk (1995).

^d Shriberg (1993).

Speech Disorders Classification System Summary (SDCSS): Group							Totals	
Speech Classification	Motor Speech Classification					n	%	
	No Motor Speech Disorder (No MSD)	Speech Motor Delay (SMD)	Childhood Dysarthria (CD)	Childhood Apraxia of Speech (CAS)	Childhood Dysarthria and Childhood Apraxia of Speech (CD & CAS)			
Normal(ized) Speech Acquisition (NSA) ^a	0	0	1	0	0	1	2.2	
Speech Errors (SE)	0	0	0	0	0	0	0.0	
Persistent Speech Errors (PSE)	0	1	0	1	0	2	4.4	
(SE/PSE)	0	1	0	1	0	2	4.4	
Speech Delay (SD)	0	0	0	0	0	0	0.0	
Persistent Speech Delay (PSD)	1	11	16	4	10	42	93.3	
(SD/PSD)	1	11	16	4	10	42	93.3	
Totals	n	1	12	17	5	10	45	
	%	2.2	26.7	37.8	11.1	22.2	100.0	

^a Includes children younger than 9 years old with age-appropriate distortions

Figure 1. Estimates of the prevalence of speech and motor speech disorders in adolescents with Down syndrome.

Speech classifications

As shown in the Totals columns in [Figure 1](#), 1 (2.2%) of the 45 participants with DS had normal or normalized speech at assessment. A total of 2 (4.4%) of the participants with DS had only the less severe speech disorder, Persistent Speech Errors (PSE). The remaining 42 (93.3%) of participants with DS met SDCS criteria for SD, with the chronological age of all but one participant meeting criteria for Persistent Speech Delay (PSD).

Motor speech classifications

Beginning with the left-most motor speech classification column in [Figure 1](#), 1 (2.2%) of the 45 participants with DS met SDCS criteria for No Motor Speech Disorder and 12 (26.7%) met SDCS criteria for SMD. The most prevalent motor speech classification was CD. As shown in [Figure 1](#), 17 (37.8%) of the participants met SDCS classification criteria for CD and an additional 10 participants (22.2%) met SDCS criteria for CD & CAS. Last, 5 (11.1%) of participants with DS met SDCS criteria for CAS.

Dysarthria subtypes

[Figure 2](#) includes group-level findings estimating the prevalence of each of five subtypes of CD in participants with DS. Findings are based on a total of 27 of the 45 (62.2%) participants in the sample who met SDCS criteria for CD or CD & CAS (see [Figure 1](#)). It is important to underscore that the percentage scores in [Figure 2a](#) do not scale severity of involvement. Rather, lower percentage scores on an index indicates more support for that subtype of dysarthria (i.e., fewer signs with typical age-sex performance on the index). The percentile scores in [Figure 2b](#) indicate a speaker's rank-ordered performance on each index relative to a reference database of 442 speakers at increased risk for motor speech disorders described in [Mabie and Shriberg \(2017, pp. 203–204\)](#). The percentile scores standardize potential age and sex differences in the magnitudes of the percentage scores across the five dysarthria indices. As described in the Supplement, subtype scores at or

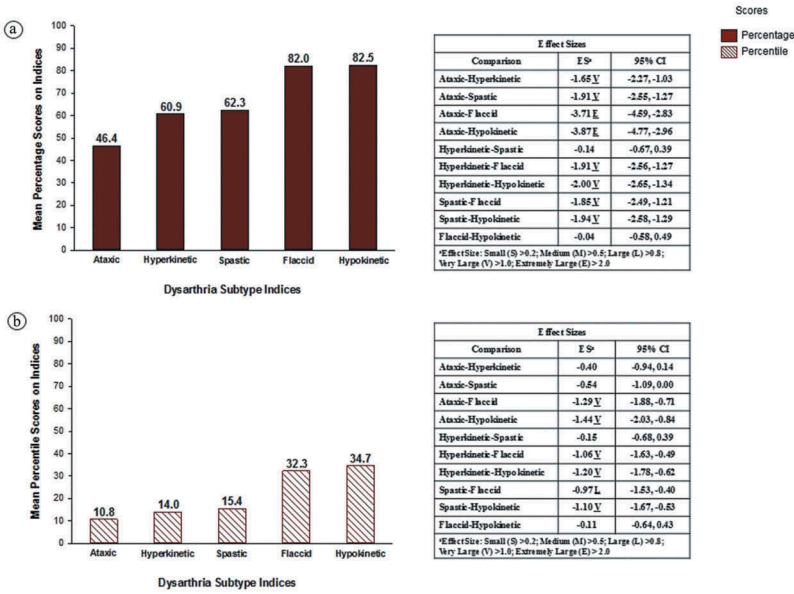


Figure 2. Estimates of the prevalence of five subtypes of dysarthria in adolescents with Down syndrome. The solid filled bars in Figure 2a are the mean percentage score findings for the 27 participants on the five dysarthria subtype indices described in the Supplement. The diagonal filled bars in Figure 2b are the mean percentile score findings for the same speakers on the five indices.

below the 10th percentile are considered positive for that subtype, and the five dysarthria subtypes are not mutually exclusive.

The shapes of the distributions of the averaged percentage scores and the averaged percentile scores in Figure 2 were similar across the five dysarthria subtypes. The descriptive statistics (means) provided the strongest support for the ataxic index, followed by scores on the hyperkinetic and spastic index, with the highest scores (i.e., least support for) flaccid and hypokinetic dysarthria.

These descriptive trends in Figure 2 are supported by the inferential statistics in the tabular data in each panel in Figure 2. To minimize Type II errors in this relatively low-powered study, inferential statistics are not adjusted for the few family-wise multiple comparisons (see Feise, 2002; Nakagawa, 2004; Perneger, 1998). For the percentage scores in Figure 2a, 8 of the 10 pairwise comparisons yielded statistically significant effect sizes (the confidence intervals did not cross 0.00), with nonsignificant effect-size findings only for the Hyperkinetic-Spastic and Flaccid-Hypokinetic comparisons. For the percentile comparisons in Figure 2b, 6 of the 10 pairwise effect size comparisons were statistically significant with nonsignificant effect-size findings for the Ataxic-Hyperkinetic, Ataxic-Spastic, Hyperkinetic-Spastic, and Flaccid-Hypokinetic comparisons. As shown in the two panels, the magnitudes of the effect sizes ranged from Large (L) to Extremely Large (E), with the largest effect sizes obtained for comparisons of ataxic scores with scores on two of the other four indices. As with the descriptive statistics findings described above, these inferential statistical findings are interpreted to indicate that participants with DS had the most speech, prosody, and voice signs consistent with ataxic dysarthria, the fewest signs

consistent with flaccid and hypokinetic dysarthria, with signs intermediate to these findings consistent with hyperkinetic and spastic dysarthria.

Inspection of participant-level data for the grouped data in [Figure 2](#) confirmed that for the participants with DS who met SDCS criteria for CD with or without concurrent CAS, ataxic dysarthria was the most frequent subtype. A total of 21 (77.8%) of the 27 participants who met SDCS criteria for CD or CD & CAS met the $\leq 10^{\text{th}}$ percentile criteria for ataxic dysarthria. However, additional analyses also indicated that 12 (57.1%) of these participants also had dysarthria subtype scores at the 10^{th} percentile or lower on 2 to 5 of the other subtype indices. Participants meeting the $\leq 10^{\text{th}}$ percentile criteria on more than one index might be classified as having mixed dysarthria. However, the significantly high inter-correlations among scores on several of the CD subtype indices (Shriberg & Mabbie, 2017), which use the signs and sign weighting described in Duffy (2013), likely contribute to the present CD subtype findings.

Summary

Findings indicate that nearly all of the 45 participants with DS who met criteria for classification using SDCS methods had some type of speech (97.8%) and motor speech (97.8%) disorder. CD was the most common motor speech disorder classification (37.8%), followed by SMD (26.7%), CD & CAS (22.2%) and CAS (11.1%). Among the five subtypes of dysarthria assessed by the Dysarthria Subtype Indices, ataxic dysarthria was significantly most prevalent, but there was support for mixed dysarthrias in over 50% of participants in the present study with CD or CD & CAS.

Conclusions

Methodological considerations that constrain generalizations from the present findings include limitations in sample size and in the sensitivity of the behavioural methods. We note that a primary challenge of translational science is to instrument valid, reliable, and time efficient measures to make them maximally accessible for research and clinical practice. The present SDCS perceptual and acoustic data reduction procedures will require such enhancement. The following conclusions include research and clinical perspectives on the three primary findings of this study.

Most adolescents with Down syndrome in the present study had persistent speech delay, rather than resolved speech delay or only persistent speech errors

As calculated using chronological age, nearly all of the classifiable participants in the present sample of adolescents with DS met SDCS criteria for PSD (93.3%), indicating that they continued to have speech sound deletions and substitutions past 9 years of age. This finding differs from findings for children with idiopathic SD. If speech sound disorder persists in children with the latter classification, it typically is limited to persistent common distortions (i.e., PSE; Shriberg et al., 1994). Longitudinal research to track the type and stability of persistent deletion and substitution errors in persons with DS during developmental epochs could be theoretically and clinically informative, using alternative

age and developmental status metrics as potential moderating variables (e.g., nonverbal age, language age, motor age). Clinical studies might observe how treatment directed at the deletions/substitutions of SD/PSD versus treatment targeting the common and uncommon speech sound distortions of SE/PSE might increase intelligibility. Greater sensitivity to spatiotemporal differences in distortions (i.e., differences in place, manner, voicing, duration, and force) using contemporary signal processing modalities could be informative for explication and increased clinical efficacy in treatment designs (e.g., Hitchcock, McAllister Byun, Swartz, & Lazarus, 2017; McAllister & Ballard, 2018; McAllister Byun et al., 2017; McAllister Byun & Hitchcock, 2012; Preston, Leece, & Maas, 2016).

Most adolescents with Down syndrome in the present study had some type of motor speech disorder

The central finding of this study was the high prevalence of motor speech disorders in adolescents with DS. The 97.8% prevalence rate for motor speech disorders in persons with DS in the present sample was the highest prevalence rate among 8 types of complex neurodevelopmental disorders assessed and classified using the same methods (Shriberg, Strand, et al., 2019). The high rate of CD & CAS in the present study, indicating that nearly one of every four participants with Trisomy 21 met criteria for CD & CAS, is consistent with CD & CAS in DS reported by Rupela, Velleman, and Andrianopoulos (2016). It is also consistent with studies reporting high rates of CD & CAS in children with certain types of complex neurodevelopmental disorders (Shriberg, Strand, et al., 2019), and in case studies of CAS associated with *FOXP2* disruptions (Morgan et al., 2016). Alternatively, the present findings implicating several motor speech phenotypes in DS could be associated with limitations in the ability of the present behavioural methods to discriminate among subtypes of motor speech disorders. More generally, there are continuing cautions about limitations in the categorical versus dimensional classification assumptions in the Mayo Clinic system on which the present indices of paediatric motor speech disorders were based (e.g., Morgan & Liégeois, 2010; Weismer, 2006), issues that go beyond the focus of the present paper.

The speech, prosody, and voice signs of individuals with Down syndrome classified as CD were most consistent with ataxic dysarthria

Descriptions of the characteristics of ataxic dysarthria include a ‘scanning pattern of speech, disturbed articulation of both consonants and vowels, and abnormal voice quality’ (Kent et al., 2000, p. 1275). Spencer and Slocumb (2007) concluded that impairment of articulation and prosody is central to all descriptions of ataxic dysarthria, noting that ‘... a critical role of feedforward processing by the cerebellum has been established and linked to speech motor control and to aspects of ataxic dysarthria’ (p. 63). Similarly, using fMRI, Urban (2013) described cerebellar activation during speech production. Ogawa, Yoshihashi, Suzuki, Kamei, and Mizutani (2010) proposed that cerebellar dysarthria is characterized by ‘... ataxic speech, and is mainly caused by impairment of the upper cerebellar hemisphere in the distribution of the superior cerebellar artery (SCA)’ (p. 861). The link between the known cerebellar differences

in persons with DS paired with the motor speech disorder results and ataxic dysarthria findings further support the role of the cerebellum in speech production and the potential causative role of cerebellar dysfunction in motor speech disorder and intelligibility deficits in DS. The perspective that the cerebellum is involved both in speech motor programming and in articulatory execution is consistent with the high prevalence of CD & CAS in the current study.

Overall, the present findings underscore the contribution of motor speech disorders to the complex of communication challenges for speakers with Down syndrome and highlight the need to consider the role of speech motor control during assessment and management (Namasivayam et al., 2013). If characteristics of motor speech disorders are observed at assessment, personalized treatment may require targeted precision and stability goals across multiple subsystems, including respiration, phonation, prosody, resonance, and articulation (McDaniel & Yoder, 2016; Miller & Pennington, 2011). The present findings are interpreted as support for motor speech disorders as primary explanatory constructs to guide the selection and sequencing of treatment targets within these subsystems for persons with DS.

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Appendix: Speech, motor speech, and dysarthria subtype classifications in the Speech Disorders Classification System (SDCS).

Table A1. The five Speech classifications and five Motor Speech classifications in the SDCS are each mutually exclusive. The five dysarthria subtype classifications are not mutually exclusive. That is, a speaker can meet percentile criteria for more than one of the five listed dysarthria subtype classifications (i.e., mixed dysarthria). See Supplement for the procedures and measures used to classify each motor speech disorder.

SDCS Classifications and Dysarthria Subtypes	Abbreviation	Age (yrs;mos) at Assessment	Description	References ^a
Five Speech Classifications				
Normal(ized) Speech Acquisition	NSA	3–80	Does not meet criteria for any of the four Speech Disorder classifications	2, 3, 4
Speech Errors	SE	6–8;11	Age-inappropriate speech sound distortions	3, 4
Persistent Speech Errors	PSE	9–80	Age-inappropriate speech sound distortions that persist past 9 years of age	4, 5
Speech Delay	SD	3–8;11	Age-inappropriate speech sound deletions and/or substitutions	3, 4
Persistent Speech Delay	PSD	9–80	Age-inappropriate speech sound deletions and/or substitutions that persist past 9 years of age	3, 4, 5
Five Motor Speech Classifications				
No Motor Speech Disorder	No MSD	3–80	Does not meet criteria for any of the four Motor Speech Disorders classifications	2, 6, 8
Speech Motor Delay	SMD	3–80	Meets PSI criterion for SMD	2, 6, 8
Childhood Dysarthria	CD	3–80	Meets DI and DSI criteria for CD	2, 6, 8
Childhood Apraxia of Speech	CAS	3–80	Meets PM criterion for CAS	6, 7, 8
Childhood Dysarthria & Childhood Apraxia of Speech	CD & CAS	3–80	Meets SDCS criteria for CD & CAS	2, 6, 8
Five Dysarthria Subtypes				
Ataxic		3–80	Cerebellar disorder	1, 2
Spastic		3–80	Upper motor neuron disorder	1, 2
Hyperkinetic		3–80	Basal ganglia disorder; increased movement	1, 2
Hypokinetic		3–80	Basal ganglia disorder; decreased movement	1, 2
Flaccid		3–80	Lower motor neuron disorder	1, 2

Notes: PSI = Precision-Stability Index; DI = Dysarthria Index; DSI = Dysarthria Subtype Index; PM = Pause Marker.

^a 1. Duffy (2013); 2. Mabbie and Shriberg (2017); 3. Shriberg (1993); 4. Shriberg, Austin, Lewis, McSweeney, and Wilson (1997); 5. Shriberg et al. (2010a); 6. Shriberg and Mabbie (2017); 7. Shriberg et al. (2017); 8. Tilkins et al. (2017).