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Auditory-Perceptual Speech Features in Children With Down Syndrome

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Abstract

Speech disorders occur commonly in individuals with Down syndrome (DS), although data regarding the auditory-perceptual speech features are limited. This descriptive study assessed 47 perceptual speech features during connected speech samples in 26 children with DS. The most severely affected speech features were: naturalness, imprecise consonants, hyponasality, speech rate, inappropriate silences, irregular vowels, prolonged intervals, overall loudness level, pitch level, aberrant oropharyngeal resonance, hoarse voice, reduced stress, and prolonged phonemes. These findings suggest that speech disorders in DS are due to distributed impairments involving voice, speech sound production, fluency, resonance, and prosody. These data contribute to the development of a profile of impairments in speakers with DS to guide future research and inform clinical assessment and treatment.

Key Words: *Down syndrome; dysarthria; speech disorders; perceptual speech features*

Introduction

Down syndrome (DS) is the most common genetic cause of intellectual impairment, affecting approximately 1 in 700 births (Parker et al., 2010). Communication disorders are common in individuals with DS, including prominent language and speech deficits. Most data suggest that expressive language is more affected than receptive language (Cleland, Wood, Hardcastle, Wishart, & Timmins, 2010; Ferreira-Vasques & Lamonica, 2015; Ypsilanti & Grouios, 2008). In terms of speech, Kent and Vorperian (2013) provide perhaps the most comprehensive description of the nature of speech disorders in children and adults with DS to date. Based on review of the data from the last 6 decades, the authors describe distributed deficits in speech production involving voice, articulation, phonology, resonance, fluency, prosody, and intelligibility. Commonly exhibited speech features include dysphonia; speech sound production errors due to developmental delay, disordered articulation, abnormalities in craniofacial anatomy, and disorders of oral/nasal resonance; dysfluency (i.e., stuttering, cluttering, and/or other prosodic disturbance); and moderate to severe reductions in speech intelligibility (Kent & Vorperian, 2013).

In many instances, the auditory-perceptual speech features associated with specific neurological damage have been well-established (Duffy & Folger, 1996; Hanson, Yorkston, & Britton, 2011; Hartelius, Runmarker, & Andersen, 2000; Kluin, Gilman, Lohman, & Junck, 1996; Ludlow & Bassich, 1983; Scholderle, Staiger, Lampe, Strecker, & Ziegler, 2016; Spencer & France, 2016). Due to the nature of speech phenomena, such perceptual methods are particularly relevant for understanding speech disorders and remain the "gold standard" to establish diagnosis, severity, treatment plans, and functional change (Bunton, Kent, Duffy, Rosenbek, & Kent, 2007). Although instrumental measures such as acoustic and kinematic data are increasingly valuable in the clinic and laboratory, such measures are typically validated with perceptual data (Kent, 2009).

Darley, Aronson, and Brown (1969a, 1969b, 1975) pioneered the use of perceptual speech analysis in their study of 212 patients with dysarthria. These authors defined dysarthria as "a group of related speech disorders that are due to disturbances in muscular control of the speech mechanism resulting from impairment of any of the basic motor processes involved in the execution of speech" (Darley et al., 1975, p. 2). Darley and colleagues (1969a, 1969b, 1975) assessed the perceptual speech features in seven patient populations based on medical diagnosis and lesion site. Using a 7-point equal-appearing scale, three judges independently rated 38 perceptual speech features. This work generated perceptual descriptions of the speech features encountered in seven types of dysarthria and provided a conceptual framework for understanding motor speech disorders. Other researchers have continued to employ perceptual speech analysis to refine understanding of the speech features encountered in other conditions such as Parkinson's disease (Ludlow & Bassich, 1983) and unilateral cerebrovascular accident (Duffy & Folger, 1996). These and other perceptual studies have served as a foundation for subsequent research using instrumental approaches to study dysarthria (Bunton, Kent, et al., 2007; Duffy & Kent, 2001; Kent, 1996; Kent, Weismer, Kent, Vorperian, & Duffy, 1999). Furthermore, in the clinic, assessment of speech performance via perceptual methods remains the standard of care.

Despite the pervasive speech disorders that occur in DS, a comprehensive description of the perceptual speech features, including their frequency of occurrence and severity, is elusive. This does not appear to be due to lack of scientific interest, as research examining speech in DS has steadily increased since the 1970s (Kent & Vorperian, 2013). One obstacle is the complex nature of speech disorders in DS. That is, speech disorders in DS frequently co-exist with disorders in cognition, language, and/or other perceptual-motor functions. Additionally, structural abnormalities of the speech mechanism are common in DS. Substantial individual variability is likely another factor.

Clinical speech assessments typically comprise visual evaluation of oral-motor structures and functions, as well as elicitation of speech of varied complexity. Constrained, simple motor tasks such as vowel prolongation, diadochokinetic tasks, and word and phrase repetition, provide useful information about respiratory function, voice, resonance, articulation, and phonology. Connected speech samples are frequently elicited in more complex formulation tasks such as picture description or narrative retell. These dynamic tasks are extremely useful in observing integrated function of all the speech components such as prosody, intelligibility, and speech sound production across contexts (Duffy, 2005; Patel & Connaghan, 2014; Shipley & McAfee, 2009). The clinician must then synthesize the myriad of data collected from these assessment tasks to develop an individualized treatment plan. As many speakers with DS demonstrate deficits across speech domains, identifying and prioritizing appropriate targets for intervention can be challenging. Due to the "multidimensional" nature of speech disorders in DS, Kent and Vorperian (2013) suggest the development of a profile of impairments to allow the identification of general patterns of disorder to guide individual assessment. Such an approach may permit hierarchical analysis of areas of impairment and enhance assessment and treatment.

Therefore, we examined the auditory-perceptual speech features, their frequency of occurrence, and their severity in a sample of 26 children with DS as a first step toward the development of a profile of speech impairments in this population. Applying the system of perceptual ratings established by Darley, Aronson, and Brown (1969a, 1969b, 1975), three expert judges listened to speech performance during connected speech samples and made independent judgments regarding 47 perceptual speech features using a 5-point equal-appearing interval scale. We hypothesized that: 1) children with DS would present with auditory-perceptual speech features suggestive of distributed speech production impairments; and 2) trained, expert listeners would demonstrate at least moderate inter-rater agreement in their assessment of the presence and severity of these perceptual speech features.

Methods

Participants

Participants were recruited from the Down syndrome specialty clinic at Duke University Medical Center and 26 children enrolled and completed study tasks. Inclusion criteria included diagnosis of Down syndrome, age 8 or older, ability to follow instructions and/or imitate others, ability to hear speech at a conversational volume, and the primary use of speech for expressive communication. Initially, the exclusion criterion was an IQ score of ≤ 40 on the Kaufmann Brief Intelligence Test (KBIT-2; Kaufman & Kaufman, 2004), though this exclusion criterion was eliminated over the course of the study as it did not appear related to participation ability. Full IQ scores comprising both verbal and nonverbal portions of the KBIT-2 are reported for descriptive purposes.

The Duke Health Institutional Review Board approved this research and informed consent was obtained from each participant and/or parent/guardian.

Participant demographics. Individual participant demographic data are shown in Table 1. Sixteen participants were female and 10 were male. Twenty were Caucasian and four were African American. Race/ethnicity was unreported by two participants. Participant age ranged from 8 to 19 years with a mean age of 12.88 years (*SD* = 3.19; median = 12). Mean IQ was 52.6 (*SD* = 11.7; median = 53; range = 40–83). Mean Communicative Effectiveness Survey (CES) score by proxy was 20.69 (maximum score: 30; *SD* = 5.8; median = 21; range = 11–30).

Table 1
Participant Demographic Data

Subject Number	Age	Sex	Race/ Ethnicity	IQ (KBIT-2)	CES score
1	12	M	W	42	20
2	11	M	W	61	29
3	8	F	W	57	23
4	17	M	W	42	23
5	18	M	W	41	15
6	9	M	W	53	22
7	11	M	W	64	29
8	10	F	W	40	16
9	16	M	W	43	21
10	15	F	W	72	29
11	17	F	W	83	21
12	13	M	W	40	14
13	11	M	U	54	24
14	11	F	AA	61	11
15	9	F	AA	54	11
16	12	M	AA	45	20
17	13	M	AA	49	15
18	19	F	W	67	28
19	10	M	W	43	15
20	11	M	W	53	25
21	14	M	W	61	22
22	9	M	U	40	14
23	12	M	W	66	26
24	18	M	W	55	30
25	13	M	W	41	19
26	16	F	W	40	16

Note. KBIT-2 = Kaufmann Brief Intelligence Test; CES = Communicative Effectiveness Survey; M = male; F = female; W = white; AA = African American; U = Unknown.

Study Tasks

Speech recordings. Speech language pathologists (SLPs) obtained samples of connected speech by presenting participants with two pictured scenes (i.e., "Farm" and "Amusement Park") commonly used to elicit a spontaneous sample in motor speech evaluations (Shipley & McAfee, 2009). Participants were asked to provide a detailed description using complete sentences with the prompt, "Tell me about what you see in this picture." Additional attempts to elicit spontaneous connected speech were made by asking questions about the pictures and engaging the participants in discussion related to personally relevant topics of interest. Speech samples were recorded in a sound-attenuated booth using a high-quality digital recorder (Marantz PMD671) and head-mounted microphone (Shure SM10A) placed 2 centimeters from the left corner of the subject's mouth at a sampling rate of 44100 Hz. These recordings were later edited by an individual otherwise uninvolved in the study to present relatively long segments of speech production with minimal speech contributions from the SLP. The edited samples averaged 49.8 seconds in length (*SD* = 7.94; range = 33.20–68) with care to include multiword sequences to permit scoring of prosodic speech features.

Other study tasks. The verbal and nonverbal portions of the KBIT-2 (Kaufman & Kaufman, 2004) were administered to all participants and standard scores were converted to a composite IQ. Initially used as a screening tool for inclusion/exclusion as described above in "Participants," IQ scores from the KBIT-2 were used for descriptive purposes and to determine associations between IQ and speech performance. The Communicative Effectiveness Survey (CES; Donovan, Kendall, Young, & Rosenbek, 2008) provided additional descriptive information. The CES is an 8-item tool that measures perceived speech effectiveness on a 1–4 scale (1 = not effective, 4 = very effective) in a variety of commonly encountered communication situations. The CES was administered via the participant's parent or guardian.

Listener training. Three speech-language pathologists (SLPs) made judgments regarding the presence or absence of 47 perceptual speech features and their severity in connected speech samples from 26 participants with DS. Each speech pathologist had between 10 and 13 years of clinical experience with perceptual assessment of speech in dysarthric individuals with diverse medical etiologies.

The judges participated in three listener training sessions prior to rating the speech recordings. Initially, the judges reviewed the list of perceptual speech features to be rated to ensure consensus among judges regarding their operational definitions (Table 2). The judges then listened to multiple randomly selected connected speech samples from study participants and openly discussed both the features they perceived and their severity ratings. Judges were encouraged to discuss any aspect of auditory-perceptual assessment with the group for calibration among listeners.

Rating sessions. Audio files were presented in a sound field environment at a comfortable fixed volume. The judges listened to the speech samples simultaneously, though each judge made independent ratings of the speech features. The audio files were played as many times as needed by any of the judges.

A 5-point equal-appearing interval scale was used (i.e., 0 = within normal limits, 1 = slight, 2 = mild, 3 = moderate, 4 = severe) to score each perceptual speech feature, similar to the scale used in previous perceptual rating systems (Duffy, 2005; Duffy & Folger, 1996). Agreement on the speech features and their severity was determined based on blind consensus agreement, defined as a difference of no more than one scale point across judges. In instances when consensus agreement was not achieved, the audio recording was replayed and rescored by each judge independently. If consensus agreement was still not obtained, the judges listened to the audio recording a third time and openly discussed their ratings until a final consensus score was obtained.

Auditory-perceptual speech features. A list of perceptual speech features of interest and their operational definitions was developed based on previous work (see Table 2; Anand & Stepp, 2015; Chenery, 1998; Darley et al., 1969a, 1969b, 1975; Duffy, 2005). During "Listener training" and "Rating sessions," it was necessary to add five speech features to the master list of auditory-perceptual speech features assessed in connected speech: excess pitch variation, aberrant oropharyngeal resonance, aberrant loudness pattern, aberrant pitch pattern, and snort. In such instances, these speech features were operationally defined by consensus. When these new speech features emerged during the rating sessions, the judges re-listened to the connected speech samples of all previous participants in order to provide independent ratings. A total of 47 perceptual speech features were identified as present or absent (with severity determined if present) in each of the 26 participants.

Statistical analysis. General descriptive analyses and distributions were examined for all variables. The frequency with which perceptual speech features occurred was tabulated. Severity of involvement across participants was determined based on mean scale values (MSVs). Spearman correlation coefficients were calculated to evaluate the correlation between the speech features and subject demographic data (e.g., gender, race, IQ score, CES score). Inter-rater agreement was assessed with Kendall's coefficient of concordance (*W*) for each perceptual speech feature to estimate blinded agreement among the three judges (Siegel, 1956). The guideline to interpretation of *W* provided by Schmidt (1997) was used in which values of 0.5 indicated moderate agreement, 0.7 designated strong agreement, and 0.9 signified unusually strong agreement. Logistic regression analyses were conducted to evaluate the association between dichotomized auditory-perceptual speech data across features and subject demographic data. Statistical significance was established at the 0.05 level for all analyses. Statistical analyses were conducted using SAS 9.4 (SAS Institute, Cary, NC).

Results

Perceptual Speech Features

Most frequently occurring auditory-perceptual speech features. The results of the listener ratings were dichotomized as present (scores = 1–4) or absent (score = 0) to determine the frequency with which each perceptual feature occurred in connected speech samples and tabulated across participants (see Table 3). A total of 35/47 speech features were identified as present in the participants' connected speech samples.

Most severely affected auditory-perceptual speech features. In order to determine the most

Table 2
Auditory-Perceptual Speech Features Assessed During Connected Speech Samples

Auditory-perceptual speech feature	Operational definition
Pitch level	Pitch of voice sounds consistently too low or too high for age and sex.
Pitch breaks	Pitch of voice shows sudden and uncontrolled variation (falsetto breaks).
Excess pitch variation	Alterations in pitch, too high, too low, or both, when pitch is expected to remain relatively unchanged.
Voice tremor	Voice shows shakiness or tremulousness.
Excess loudness variation	Alterations in loudness, either inadequate and/or excess loudness when loudness is expected to remain relatively unchanged.
Loudness decay	There is progressive diminution or decay of loudness.
Alternating loudness	There are alternating changes in loudness.
Loudness level (overall)	Voice is insufficiently or excessively loud.
Harsh voice	Synonymous with excessive vocal effort characterized by tongue retraction, and constriction of the pharyngeal constrictors, which is sometimes accompanied by nasality. The voice may be described by strident, metallic, or grating. There is often abrupt initiation of voice characterized by hard glottal attack.
Strained-strangled voice	Voice (phonation) sounds strained or strangled (an apparently effortful squeezing of voice through glottis).
Hoarse voice	Produced by interference with optimum vocal fold adduction. Characterized by a breathy escape on phonation, often accompanied by hard glottal attack, producing a low-pitched voice lacking an appropriate resonance.
Wet voice	Wet, liquid-sounding voice as heard when there is pooling of fluid around the vocal cords.
Glottal fry	Syncopated vocal fold vibration, which generally occurs over the lower part of the pitch range. There is a low-pitched, cracking type of phonation.
Breathy voice (continuous)	Voice is continuously breathy, weak, and thin.
Breathy voice (transient)	Breathiness is transient, periodic, and intermittent.
Voice stoppages	There are sudden stoppages of voice air stream (as if some obstacle along vocal tract momentarily impedes flow of air).
Imprecise consonants	Consonant sounds lack precision. They show slurring, inadequate sharpness, distortions, and lack of crispness. There is clumsiness in going from one consonant sound to another.
Irregular vowels	Vowel sounds are distorted throughout their total duration.
Hypernasality	Voice sounds excessively nasal. Excessive amount of air is resonated by nasal cavities.
Hyponasality	Voice is denasal.
Nasal emission	There is nasal emission of airstream.
Snort	Nasopharyngeal snorting sound.
Forced inspiration-expiration	Speech is interrupted by sudden, forced inspiration and expiration sighs.
Audible inspiration-expiration	There is audible, breathy inspiration and/or expiration.
Grunt at end of expiration	There is a grunt at the end of expiration.
Rate	Rate of actual speech is abnormally slow or rapid.

(Table 2 continued)

Table 2
Continued

Auditory-perceptual speech feature	Operational definition
Short phrases	Phrases are short (possibly because inspirations occur more often than normal). Speaker may sound as if he or she has run out of air. He may produce a gasp at the end of a phrase.
Increase of rate in segments	Rate increases progressively within given segments of connected speech.
Increase of rate overall	Rate increases progressively from beginning to end of sample.
Reduced stress	Speech shows reduction of proper stress or emphasis patterns.
Aberrant loudness pattern	Unusual or unexpected loudness variations.
Aberrant pitch pattern	Unusual or unexpected pitch variations.
Variable rate	Rate alternates from slow to fast.
Prolonged intervals	There is prolongation of interword or intersyllable intervals.
Inappropriate silences	There are inappropriate silent intervals.
Short rushes of speech	There are short rushes of speech separated by pauses.
Excess and equal stress	There is excess stress on usually unstressed parts of speech (e.g., monosyllabic words, unstressed syllables of polysyllabic words).
Prolonged phonemes	There are prolongations of phonemes.
Repeated phonemes	There are repetitions of phonemes.
Irregular articulatory breakdowns	There is intermittent, nonsystematic breakdown in accuracy of articulation.
Naturalness (overall)	How speech compares to that encountered in typical speech.
Inhalatory stridor	Similar to audible inspiration but characterized by actual rough phonation due to vocal fold approximation and oscillation during inhalation.
Weak pressure consonants	Pressure consonants lack acoustic distinctiveness or are weak because of excessive nasal airflow during their production.
Simple vocal tics	Repetitive, rapid, apparently involuntary noises or sounds (e.g., throat clearing, grunting) produced in isolation or during voluntary speech.
Palilalia	Compulsive repetition of words or phrases, usually in a context of accelerating rate and decreasing loudness.
Coprolalia	Involuntary, compulsive, repetitive obscene language or swearing, uttered loudly, softly, or incompletely.
Aberrant oropharyngeal resonance	Abnormal oral/pharyngeal resonance.

Note. Anand & Stepp, 2015; Chenery, 1998; Darley et al., 1969a, 1969b, 1975; Duffy, 2005.

severely affected speech features across our participants, MSVs were calculated as seen in Table 4. A principal component analysis of the perceptual speech features during connected speech supported these findings in that the most heavily weighted speech features occurred commonly based on MSVs.

Inter-Rater Agreement

Based on initial independent rating by three judges, W was ≥ 0.5 indicating at least moderate agreement between judges for 32/35 (91%) perceptual speech features present in connected speech, as seen in Table 5. Consensus agreement across judges within one scale value was obtained in 94.5% of the time across speech signs and participants on the first attempt ($n = 1551$). When agreement was not achieved based on initial ratings, repeated listening and rating was sufficient to reach agreement an additional 5.4% of the time ($n = 88$). In 0.1% of instances ($n = 2$), it was necessary for the judges to have open discussion and further listening in order to obtain consensus ratings. Final consensus ratings were used to

Table 3
Dichotomized Auditory-Perceptual Speech Features Present in Connected Speech Samples in 26 Speakers With Down Syndrome in Rank Order

Perceptual speech feature (n = 35)	Rank order	Frequency of occurrence (n = 26)
Hyponasality	1	26
Imprecise Consonants	1	26
Inappropriate silences	1	26
Prolonged intervals	1	26
Hoarse Voice	5	25
Irregular vowels	5	25
Loudness level (overall)	5	25
Naturalness (overall)	5	25
Rate	5	25
Oropharyngeal resonance	10	24
Pitch level	10	24
Audible Inspiration-expiration	12	23
Prolonged phonemes	12	23
Reduced stress	14	21
Aberrant pitch pattern	15	20
Aberrant loudness pattern	16	18
Breathy voice (transient)	17	16
Excess loudness variation	17	16
Excess pitch variation	17	16
Repeated phonemes	20	8
Nasal emission	21	6
Breathy voice (continuous)	21	6
Variable rate	23	5
Harsh voice	23	5
Short rushes of speech	25	4
Snort	26	3
Alternating loudness	27	2
Glottal fry	27	2
Strained-strangled voice	27	2
Weak pressure consonants	27	2
Pitch breaks	31	1
Voice tremor	31	1
Wet voice	31	1
Hypernasality	31	1
Increase of rate in segment	31	1

calculate MSVs for each auditory-perceptual speech feature.

Inter-rater agreement for the 35 rated perceptual speech features can also be categorized by dividing W data into quartiles. At the 25th percentile, moderate inter-rater agreement was achieved as demonstrated by $W = 0.55$. At the 50th percentile and 75th percentile, strong inter-rater agreement was present based on $W = 0.67$ and $W = 0.78$, respectively.

Associations

Logistical regression revealed no associations between gender, race, age, IQ, or CES scores and dichotomized auditory-perceptual speech data. Significant negative correlation was found between overall scale of connected speech features and CES scores ($\rho = -0.49$, p-value = 0.0114).

Discussion

Perceptual Speech Features

These data contribute to our understanding of the nature of speech disorders in individuals with DS by comprehensively reporting the perceptual speech features present in 26 children. We hypothesized that connected speech samples in children with DS would demonstrate a pattern of perceptual speech features suggestive of distributed speech production impairments. Darley and colleagues (1975) suggested that MSVs of 1.5 be used as a cut off to determine the most critical speech features and we followed this convention. In rank order, the most severely affected speech features identified in our sample were naturalness, imprecise consonants, hyponasality, speech rate, inappropriate silences, irregular vowels, prolonged intervals, overall loudness level, pitch level, aberrant oropharyngeal resonance, hoarse voice, reduced stress, and prolonged phonemes. These speech features also occurred commonly in our sample, identified in 81 to 100% of our participants. These data suggest that speech disorders in DS reflect distributed speech production impairments involving voice, speech sound production, fluency, resonance, and prosody.

Many of our findings support studies previously reported in the literature, particularly related to voice, speech sound production, and fluency. Dysphonia of at least a mild severity was the most common voice feature identified in previous studies of individuals with DS–"low vocal pitch and hoarse, harsh, or raucous voice has been frequently been ascribed to individuals with DS" (Kent & Vorperian, 2013, p. 179). Our findings of abnormalities in overall pitch level and the presence of a hoarse voice are consistent with that

Table 4
Severity of 35 Auditory-Perceptual Speech Features Identified During Connected Speech Samples in 26 Individuals With Down Syndrome in Rank Order Based on Mean Scale Values

Perceptual speech feature	Rank order	Mean Scale Value	Median Scale Value
Naturalness	1	3.31	4.0
Imprecise consonants	2	3.15	3.0
Hyponasality	3	2.62	3.0
Rate	4	2.58	3.0
Inappropriate silences	4	2.58	3.0
Irregular vowels	6	2.50	2.5
Prolonged intervals	6	2.50	2.5
Overall loudness level	8	2.46	2.0
Pitch level	9	2.42	3.0
Aberrant oropharyngeal resonance	10	2.35	2.5
Hoarse voice	11	2.27	2.0
Reduced stress	12	2.15	2.0
Prolonged phonemes	13	2.00	2.0
Aberrant pitch pattern	14	1.81	2.0
Audible inspiration-expiration	15	1.62	2.0
Aberrant pattern of loudness	15	1.62	2.0
Excess pitch variation	17	1.46	2.0
Excess loudness variation	18	1.35	1.5
Breathy voice (transient)	19	1.12	1.0
Repeated phonemes	20	0.73	0
Breathy voice (continuous)	21	0.69	0
Harsh voice	22	0.46	0
Nasal emission	23	0.42	0
Variable rate	24	0.38	0
Short rushes of speech	25	0.27	0
Alternating loudness	26	0.23	0
Snort	26	0.23	0
Glottal fry	28	0.19	0
Strained-strangled voice	29	0.15	0
Pitch breaks	30	0.12	0
Weak pressure consonants	30	0.12	0
Voice Tremor	32	0.08	0
Wet voice	32	0.08	0
Hypernasality	32	0.08	0
Increase of rate in segments	32	0.08	0

research. The laryngeal anatomy changes dramatically during puberty, particularly in males who demonstrate laryngeal enlargement, vocal fold length, and vocal tract elongation in late puberty. This results in a corresponding drop in pitch, the perceptual correlate of fundamental frequency (*F*0; Fitch & Giedd, 1999). Females also demonstrate changes in laryngeal anatomy during puberty; however, these changes and the subsequent drop in *F*0 are less steep (Markova et al., 2016). Although age of puberty onset varies, 14 of the 26 of the participants in this study were between the ages of 12–18 and therefore likely to be entering or passing through puberty. Puberty-related changes to the voice, particularly pitch, may have had an effect on perceptual ratings.

Table 5
Agreement Among Three Expert Judges on Exact Scale Value Assigned to Each Auditory-Perceptual Speech Feature During Connected Speech

Connected Speech (35 of 47 features identified and scored)				
Perceptual speech feature	Rank Order	Coefficient of concordance (W)	Interpretation of agreement level	p-value
Excess loudness variation	1	0.8642	Strong	<.0001
Snort	2	0.8611	Strong	<.0001
Aberrant pattern of loudness	3	0.8528	Strong	<.0001
Loudness level (overall)	4	0.8449	Strong	<.0001
Wet voice	5	0.8299	Strong	<.0001
Repeated phonemes	6	0.8172	Strong	<.0001
Naturalness (overall)	7	0.8160	Strong	<.0001
Breathy voice (continuous)	8	0.8098	Strong	<.0001
Imprecise Consonants	9	0.7798	Strong	<.0001
Inappropriate silences	10	0.7679	Strong	<.0001
Aberrant pattern of pitch	11	0.7517	Strong	<.0001
Excess pitch variation	12	0.7515	Strong	<.0001
Irregular vowels	13	0.7470	Strong	<.0001
Nasal emission	14	0.7365	Strong	<.0001
Rate	15	0.7185	Strong	<.0001
Audible Inspiration-expiration	16	0.7091	Strong	<.0001
Pitch level	17	0.6982	Moderate	<.0001
Reduced stress	18	0.6685	Moderate	<.0001
Pitch breaks	19	0.6667	Moderate	<.0001
Alternating loudness	20	0.6531	Moderate	<.0001
Prolonged intervals	21	0.6391	Moderate	<.0001
Hoarse voice	22	0.6119	Moderate	0.0003
Prolonged phonemes	23	0.6021	Moderate	0.0005
Oropharyngeal resonance	24	0.5943	Moderate	0.0007
Breathy voice (transient)	25	0.5824	Moderate	0.0012
Weak pressure consonants	26	0.5791	Moderate	0.0013
Harsh voice	27	0.5520	Moderate	0.0037
Glottal fry	28	0.5349	Moderate	0.0067
Strained-strangled voice	29	0.5208	Moderate	0.0106
Short rushes of speech	30	0.5156	Moderate	0.0124
Increase of rate in segment	31	0.5102	Moderate	0.0147
Hypernasality	32	0.5031	Moderate	0.0181
Variable rate	33	0.4826	Weak	0.0321
Hyponasality	34	0.4762	Weak	0.0380
Voice tremor	35	0.4300	Weak	0.1103

In terms of speech sound production, our findings of imprecise consonants and distorted vowels are consistent with several studies that identified errors in consonant production (Brown-Sweeney & Smith, 1977; Bunn, Simon, Welsh, Watson, & Elliott, 2002; Kumin, 1994; Roberts et al., 2005; Rosin, Swift, Bless, & Vetter, 1988; Schlanger & Gottsleben, 1957; Sommers, Patterson, & Widgen, 1988; Timmins et al., 2009; van Bysterveldt, 2009; van Bysterveldt, Gillon, & Foster-Cohen, 2010) and vowel production (Bunton, Leddy, & Miller, 2007; Van Borsel, 1996; van

Bysterveldt et al., 2010). Disorders of speech fluency, in the form of stuttering and/or cluttering, have also been commonly reported in speakers with DS with the estimated prevalence ranging from 15% to 45% (Devenny & Silverman, 1990; Eggers & Van Eerdenbrugh, 2018; Gottsleben, 1955; Keane, 1970; Preus, 1972; Rohovsky, 1965; Schlanger & Gottsleben, 1957). In our sample, speech features consistent with disordered speech fluency include inappropriate silences, prolonged intervals, and prolonged phonemes.

More attention has been given to studying voice, speech sound production, and fluency in the literature than there has been examining resonance and prosody in speakers with DS (Kent & Vorperian, 2013). Resonance is the perceived quality of sound generated with phonation as it is modified by the pharynx, oral cavity, and nasal cavity, and is determined by the size and shape of these cavities, as well as velopharyngeal function. In terms of resonance, hyponasality and aberrant oropharyngeal resonance were both among the most commonly exhibited and severely affected speech features in our sample. Hyponasality (or denasality) refers to the perception of inadequate nasal resonance during the production of nasal sounds (/m/, /n/, /ng/) and vowels. To our knowledge, hyponasality has not previously been reported in speakers with DS. In contrast, although there are relatively few data, the presence of hypernasality has been reported in speakers with DS (Kline & Hutchinson, 1980; Lind, Vuorenkoski, Rosberg, Partanen, & Wasz-Hockert, 1970). However, we rarely perceived hypernasality in our sample.

During our rating sessions, we also noted what we termed aberrant oropharyngeal resonance due to what we perceived as abnormal resonance of the oral and pharyngeal cavities during speech production. Our term appears analogous to the resonance phenomenon reported in speakers with DS and other complex neurodevelopmental disorders described by Fourakis and colleagues (Fourakis, Karlsson, Tilkens, & Shriberg, 2010). These authors termed this finding nasopharyngeal resonance, defined as, "a 'muffled' 'back of the throat' quality consistent with the percept of 'sluggish or imprecise tongue movement'" (p. 9). Although not included in the original list of perceptual speech features from Darley and colleagues (1969a, 1969b, 1975), we felt that the addition of aberrant oropharyngeal resonance was necessary to capture our perceptual observations. Speakers with DS have known differences in craniofacial anatomy in speakers with DS that may influence resonance. For example, Kent and Vorperian (2013) note that speakers with DS may have disordered "oral/nasal resonance … (due to) abnormalities in the nasal cavities, the sinuses, and the tissue boundaries between the oral and nasal passages" (p. 184). Similarly, Uong and colleagues (2001) found that relative to controls, speakers with DS have reduced airway, mandible, adenoid, and tonsillar volumes, as well as a smaller mid- and lower-face skeleton. Theorized to be a consequence of more posterior movement of the tongue than appropriate during production of vowels, acoustic analysis found the perceptual construct of nasopharyngeal resonance to be associated with second formant (*F*2) values further back in the vowel space for speakers with DS and Fragile X syndrome than typically developing peers (Fourakis et al., 2010).

We also found it necessary to add snort as a perceptual speech feature, defined as the production of a nasopharyngeal snorting sound. Although snorting was not commonly encountered, it may be a distinctive speech feature in this population that is reflective of velopharyngeal dysfunction. Overall, our findings regarding resonance abnormalities and velopharyngeal dysfunction in speakers with DS suggest the need for future research, including replication of our perceptual findings and physiologic investigations to enhance understanding of the mechanisms of disordered resonance in DS.

Prosody refers to suprasegmental aspects of speech production including stress, intonation, and rate-rhythm (Yorkston, Beukelman, Strand, & Hakel, 1999). As previously mentioned, speech prosody in DS not related to fluency has received little study. However, Kent and Vorperian (2013) note that the handful of studies that have examined speech prosody in individuals with DS have all indicated "limitations in the perception, imitation, and spontaneous production of prosodic features" (p. 187). This observation is supported by findings in our sample, including speech features related to disordered prosody including speech rate and reduced stress.

Kent and Vorperian (2013) emphasized the need to develop a profile of impairments to allow identification of general patterns of motor speech impairments encountered in individuals with DS to guide research efforts and clinical practice. The present data are an important first step toward the development of such a profile of impairments and suggest that speech assessment in DS should sample multiple aspects of speech production, minimally comprising the domains of voice, speech sound production, resonance, fluency, and prosody. This approach may allow for comprehensive yet efficient speech assessment while retaining the flexibility necessary for individual variability. Our findings also have implications for treatment of dysarthria in individuals with DS. Our data, for example, suggest there are multiple speech domains that may be appropriate to target in treatment with DS speakers. Additionally, treatments that target a relatively isolated aspect of speech production may be less impactful than those that are able to influence multiple domains of speech production.

Inter-Rater Agreement

We also hypothesized that trained, expert listeners would demonstrate at least moderate inter-rater agreement ($W \geq 0.5$) in their assessment of the perceptual speech features. Measures of inter-rater agreement are important to determine "if the rating scale is reliable and if the scale values are meaningful independent of the rater" (Bunton et al., 2007, p. 1482). Our findings showed at least moderate agreement within one scale value for 32/35 speech features identified by the judges in connected speech samples. Independent ratings of speech demonstrated exact agreement 60% of the time and agreement within one scale value was achieved 94.5% of the time across speech features. Additionally, across quartiles, moderate or better inter-rater agreement was achieved with W values ranging from 0.55 at the 25th percentile to 0.78 at the 75th percentile. The data provide support for the validity of using rating scales to identify salient perceptual speech features of speakers with DS independently of the rater.

Although comparisons between studies are challenging due to methodological variability (Bunton et al., 2007), our results are generally consistent with other reports of inter-rater agreement in the use of rating scales to assess perceptual speech features. For example, Darley, Aronson, and Brown (1969a, 1969b, 1975) calculated inter-rater agreement from a subsample of ratings by three listeners who used a 7-point rating scale to assess 37 perceptual speech features in samples from 150 patients with differing dysarthria types and etiologies. Agreement within one scale value was reported 84% of the time. Ozsancak and colleagues (2006) also reported on inter-rater agreement between two listeners who rated 33 speech features in 60 samples from participants with neurodegenerative dysarthria and healthy controls using a 5-point scale. Exact agreement was achieved 91% of the time and was higher in control participants (96.3%) than in speakers with dysarthria (86.1%; Ozsancak et al., 2006). Bunton and colleagues (2007) also studied listener agreement using a 7-point scale to assess 38 perceptual speech features. Ten inexperienced and 10 experienced clinicians rated connected speech samples in 47 dysarthric speakers with diverse medical etiologies. Overall, inter-rater agreement was found to be satisfactory, with exact agreement achieved 48% of the time and agreement within one scale value achieved 67% of the time in the current study. In contrast to prior studies (Zeplin & Kent, 1996; Zyski & Weisiger, 1987), Bunton and colleagues (2007) found no significant differences in inter-rater agreement between experienced and inexperienced listeners. Systematic listener training with well-defined task parameters and mutually accepted speech feature definitions may enhance inter-rater agreement, independent of experience with specific medical diagnoses. For example, despite varying frequency of clinical exposure to speakers with DS, the three experienced listeners in this study were able to achieve moderate or better inter-rater agreement 91% of the time following listener training.

Limitations of Research/Directions for Future Research

Limitations of the study include a small sample size and a reliance on perceptual data without supporting instrumental or acoustic data (e.g., EMG, kinematic, acoustic). Collection of intelligibility data would have enhanced description of the population and allowed post hoc analysis of which speech features made the largest contributions to decreases in intelligibility. As previously mentioned, we also introduced some novel perceptual speech features that require further validation and investigation including aberrant oropharyngeal resonance and snort.

Overall, it appears that resonance is an important aspect of speech in DS for future research. It is well established that resonance disorders have marked negative consequences for speech production. In this study, we found that resonance disorders occurred frequently, particularly hyponasality and aberrant oropharyngeal resonance. Considering the paucity of research that examines speech resonance in individuals with DS, as well as our poor agreement in the perceptual identification of hyponasality (0.4762), this appears to be a priority for future research including instrumental analyses.

Summary

Our findings regarding the perceptual speech features of speakers with DS have important implications for future research and clinical practice. Foremost among these is the notion that speech disorders in DS are complex in their nature and reflect distributed speech production impairments involving voice, speech sound production, fluency, resonance, and prosody. Cumulatively, these data suggest that children ages 8–19 present a distinctive pattern of auditory-perceptual speech features worthy of additional investigation and offer potential targets for improving speech. Our results also support the use of perceptual rating scales to identify speech features in speakers with DS.

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