

Review

Speech Impairment in Down Syndrome: A Review

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Purpose: This review summarizes research on disorders of speech production in Down syndrome (DS) for the purposes of informing clinical services and guiding future research.

Method: Review of the literature was based on searches using MEDLINE, Google Scholar, PsycINFO, and HighWire Press, as well as consideration of reference lists in retrieved documents (including online sources). Search terms emphasized functions related to voice, articulation, phonology, prosody, fluency, and intelligibility.

Conclusions: The following conclusions pertain to four major areas of review: voice, speech sounds, fluency and prosody, and intelligibility. The first major area is voice. Although a number of studies have reported on vocal abnormalities in DS, major questions remain about the nature and frequency of the phonatory disorder. Results of perceptual and acoustic studies have been mixed, making it difficult to draw firm conclusions or even to identify sensitive measures for future study. The second major area

is speech sounds. Articulatory and phonological studies show that speech patterns in DS are a combination of delayed development and errors not seen in typical development. Delayed (i.e., developmental) and disordered (i.e., nondevelopmental) patterns are evident by the age of about 3 years, although DS-related abnormalities possibly appear earlier, even in infant babbling. The third major area is fluency and prosody. Stuttering and/or cluttering occur in DS at rates of 10%–45%, compared with about 1% in the general population. Research also points to significant disturbances in prosody. The fourth major area is intelligibility. Studies consistently show marked limitations in this area, but only recently has the research gone beyond simple rating scales.

Key Words: speech sound disorders, voice disorders, prosody, genetic disorders, fluency disorders

Speech production in Down syndrome (DS) is associated with significant impairments in spoken language (Fawcett & Perales, 2009; Leddy, 1999; Miller & Leddy, 1999; Rondal & Comblain, 1996; Timmins et al., 2009; Zink & Bialer, 1967). As shown in Figure 1, the number of studies on speech, voice, fluency and prosody, and intelligibility in DS has increased fairly steadily since the 1970s, with a substantial increase in the last decade. Studies focused on speech intelligibility have been reported only relatively recently and account for a major part of the increase in reports published since 1990. Figure 1 indicates that there is a reasonably sized literature on speech communication in DS.

Unlike earlier reviews, the present review covers articles published in the last 6 decades, offers systematic summaries of research participants (DS and comparison

groups) and research methods, and analyzes research progress in the four major aspects of speech production: (a) voice, (b) speech sounds (including articulation, phonology, and resonance), (c) fluency and prosody, and (d) intelligibility. The combination of fluency and prosody is based on the principle that both are most effectively expressed in units larger than the phone (e.g., as a syllable or multisyllabic strings). The last category, intelligibility, can be regarded as a joint product of the previous three and is the core of communication ability and disability. Although the relevant research in these areas overlaps, the categories are sufficiently distinct that they delineate the primary facets of speech difficulty in DS. The primary goal of this review is to inform clinical services and guide future research.

We used MEDLINE, Google Scholar, PsycINFO, and HighWire Press to search the literature published since 1950 and considered reference lists in retrieved documents (including online sources). The main search terms were *Down Syndrome*, *Down's Syndrome*, *Downs Syndrome*, *mongolism*, *mongoloid*, and *trisomy 21*, which were linked to additional terms including *articulation*, *babbling*, *cluttering*, *communication*, *consonants*, *conversation*, *cry*, *diadokinesis*, *disfluency* or *dysfluency*, *formants*, *infant vocalizations*, *intelligibility*, *nasality*,

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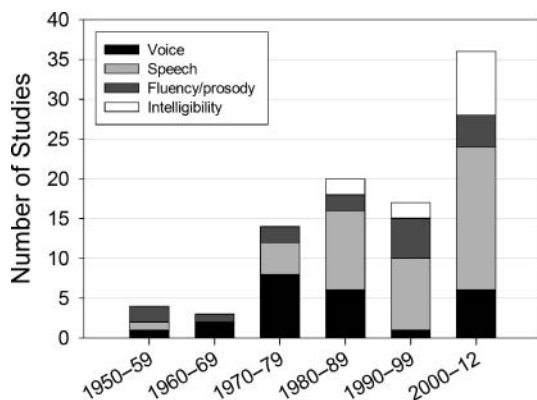
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Figure 1. The number of research articles on speech production in Down syndrome (DS) covered in this review, grouped in decade intervals since 1950. The number of articles is shown by category on the vertical axis, and decade intervals are shown on the horizontal axis. The four categories are voice, speech, fluency and prosody, and intelligibility.



phonation, phonology, phonological, prosody, speech, speech development, speech production, stuttering, voice, vocal quality, and vowels.

We compiled methods and results of studies in each of the four areas of speech production (voice, speech sound disorders, fluency and prosody, and intelligibility) for a given age group of participants (see Appendix A Tables A1 and A2 and Appendixes B, C, and E). When possible, we arranged the tables in a developmental perspective to show the results for adults and children (and, data permitting, children of different ages).

Given the phenotypic variation in DS (Reeves, Baxter, & Richtsmeier, 2001; Wiseman, Alford, Tybulewicz, & Fisher, 2009), it is important that sample size and participant characteristics be considered in generalizing the results of individual studies, so we have estimated the aggregate number of participants in each of the four areas of review. Both typically developing (TD) and atypically developing (AD) individuals have been used as controls in previous studies of DS, and the abbreviations *TD* and *AD* are used in both the text and tables to indicate these two general categories of participants. Case reports are not included in this review unless they provide methodological details relevant to group investigations. Treatment studies are excluded unless they present pre-treatment participant data on the categories listed earlier. Parental surveys are discussed and are summarized in Appendix D.

In the discussion, we highlight significant points of agreement and disagreement among the studies, relate the speech abnormalities to anatomic anomalies and other pathophysiology, and consider current perspectives

on the etiology and nature of speech disturbances in DS.

Voice

Review of Literature

We collected data on voice in DS from nearly 600 individuals, including children and adults (see Appendix A, Tables A1 and A2, respectively). The exact aggregate number is difficult to determine because some of the earlier studies may have reported on the same group of participants more than once. Research on vocal characteristics has focused mainly on vocal fundamental frequency (f_0) level and vocal quality, often with the hypothesis that DS is associated with a characteristic dysphonia. Low vocal pitch and hoarse, harsh, or raucous voice have frequently been ascribed to individuals with DS (Benda, 1949; Novak, 1972; Shprintzen, 1997; Strazzulla, 1953). These reports stimulated research on vocal characteristics of children and adults with DS.

Newborn and infant cry. Research in this area was published in the 1970s when there was a keen interest in the diagnostic significance of the newborn and infant cry (especially the pain cry, which could be elicited reliably). The cries of babies with DS were distinguished from those of healthy babies on the basis of spectrographic abnormalities such as stuttering, flat melody, and low pitch (Lind, Vuorenkoski, Rosberg, Partanen, & Wasz-Hockert, 1970). *Stuttering* was defined as “a special kind of tenseness which is periodically heightened during the cry, when attacks of glottal pressure are superimposed on the phonation” (Lind et al., 1970, p. 479). Vuorenkoski, Lind, Wasz-Hockert, and Partanen (1971) developed a cry score based on 13 acoustic characteristics that distinguished the pain cries of infants with DS from those of healthy infants. These studies indicate that the underlying disturbed infant cry in DS is most likely a result of abnormalities in respiratory and laryngeal function—this finding is not surprising given that the cry is formed largely by phonatory activity with relatively little participation of the vocal tract, except to maintain an open airway.

Vocal pitch and f_0 . The variable of f_0 is the primary acoustic correlate of perceptual judgments of vocal pitch. If vocal pitch is judged to be low in DS, then f_0 is expected to be lower in DS than in age-matched TD controls. Perceptual ratings of vocal pitch in DS are mixed (Montague, Hollien, Hollien, & Wold, 1978).

Quantitative studies based on acoustic methods, summarized in Appendixes A1 and A2, offer mixed results on vocal f_0 , with the majority of studies reporting no difference between individuals with DS and TD controls, although a difference may exist when age is taken into

account. One study demonstrated a low f_0 in the pain cry of infants with DS (Lind et al., 1970). One study of children with DS showed a higher f_0 (Weinberg & Zlatin, 1970), whereas another study of children with DS showed a lower f_0 compared with TD controls (Moran & Gilbert, 1978). Four studies showed a higher f_0 in adults with DS compared with TD controls (Albertini et al., 2010; Lee, Thorpe, & Verhoeven, 2009; Moura et al., 2008; Seifpanahi, Bahktiar, & Salmalian, 2011).

Rodger (2009) noted a discrepancy between perceptual judgments of pitch level and acoustic measures of f_0 , which may mean that perception of low vocal pitch is influenced by factors other than the actual frequency of vocal fold vibration. Researchers may help to resolve this discrepancy by examining a range of acoustic and perceptual factors associated with voice production in individuals with DS, taking into account a developmental perspective that covers the period from infancy to adulthood.

Vocal quality. Vocal quality has been studied with both perceptual and acoustic methods, as detailed in Tables A1 and A2 of Appendix A. In perceptual studies of voice in individuals with DS, researchers note, especially, breathiness and roughness. Published studies are by no means in complete agreement, but acoustic studies report increased frequency perturbations (e.g., higher values of jitter), amplitude perturbations (e.g., higher values of shimmer), and increased noise in phonation (e.g., reduced signal-to-noise ratio [SNR]). Discrepant results also have been reported for spectral tilt (Moura et al., 2008; Rodger, 2009). The variability in results among studies may be due, in part, to differences in participant samples, speaking task differences, language differences, or differences in the algorithms or equipment used to calculate the acoustic values. No single acoustic correlate of vocal quality in DS consistently emerges in the published literature, nor is it clear if a particular vocal quality persists in individuals with DS across various speaking tasks and if vocal quality in DS changes with development. Despite frequent comments in the clinical literature on vocal quality differences in DS, there has not been a satisfactory convergence on perceptual features or on acoustic correlates of vocal quality.

Anatomic anomalies and pathophysiology related to voice in DS. Some researchers have suggested that vocal features in DS are associated with anatomic and physiologic abnormalities such as hypothyroidism, absence of facial sinuses, or anomalies in laryngeal structures (Benda, 1949; Leddy, 1996; Novak, 1972). Endoscopic studies have shown that airway obstruction, which occurs in a significant proportion of individuals with DS, is often associated with laryngomalacia, tracheomalacia, or bronchomalacia (Bertrand, Navarro, Caussade, Holmgren, & Sanchez, 2003; Mitchell, Call & Kelly, 2003).

Laryngomalacia may affect the epiglottis and/or the arytenoid cartilages (Prescott, 1991; Roger, Denoyelle, Triglia, & Garabedian, 1995). Epiglottal involvement often appears as an elongation, with an inward folding of the walls that can obstruct the airway. The epiglottis is often omega shaped in cross section. This feature is by no means unique to DS, as it has been described in a significant percentage of TD children (Ferguson, 1970; Solomons & Prescott, 1987). With involvement of the arytenoid cartilages, enlargement is the most prominent feature. The cartilage is generally soft and pliable and is prone to dynamic prolapse over the larynx during inspiration, often resulting in inspiratory noise or stridor. Thompson (2009) presented evidence that laryngomalacia is associated with altered laryngeal tone and sensorimotor integration, a finding that may help to explain some of the cry abnormalities described earlier (see *Newborn and infant cry* subsection).

Discussion

It is likely that dysphonia of at least a mild degree is a common feature of speech in DS, although prevalence data have not been reported. (Prevalence is estimated to be about 6% for 8-year-old children in the general population [Carding, Roulstone, Northstone, & the ALSPAC Study Team, 2006]). Research on pain cry in neonates and infants with DS (reviewed in the *Newborn and infant cry* subsection) points strongly to the conclusion that vocal abnormalities are evident in the earliest stages of phonation.

Acoustic studies in adults indicate that vocal f_0 is generally higher in DS than in healthy controls, possibly because of the smaller body size of individuals with DS compared with that of TD controls (Myrelied, Gustafsson, Ollars, & Annerén, 2002; Rosenbloom, McGregor, Chen, An, Hsu, & Dupont, 2010). Because of the documented reduced body size in individuals with DS, growth curves specific to DS have been developed (Myrelied et al., 2002). If the size of the larynx is related to body size, individuals with DS may have a relatively small larynx compared with age- and sex-matched TD controls and, therefore, would have a higher vocal f_0 . This hypothesis would be supported if it could be established that laryngeal structures are smaller in individuals with DS than in healthy controls.

Perceptual studies of voice point to disturbances in vocal quality that are typically judged as breathiness and roughness. Acoustic studies often show increased perturbations and a reduced SNR, findings that are consistent with the results of perceptual studies. In general, vibratory aperiodicity, as measured by jitter and shimmer, has been attributed to four sources: (a) neurological, (b) biomechanical or structural, (c) aerodynamic, and

(d) Source \times Filter (Source \times Resonator) interaction (Titze, Horii, & Scherer, 1987). Any or all of these factors could account for vocal perturbation in individuals with DS, and different combinations of these factors could account for the variation in the results of studies on voice. A complicating factor in interpreting acoustic data for shimmer and SNR for children with DS is that the results for TD children would be considered as pathological values for adults (Glaze, Bless, Milenkovic, & Susser, 1988).

The larger picture of vocal quality includes oral/nasal resonance as well as characteristics derived from vocal fold function. As reviewed in the Speech Sound Disorders section below, resonance is altered in at least some individuals with DS; thus, the overall perception of vocal quality could be a combination of abnormalities in vocal fold vibration and atypical vocal tract resonances. Phonatory function may be affected by abnormal vocal fold behavior, loss of acoustic energy due to nasalization, and their interaction. Abnormalities of voice may have a significance that goes beyond a perceived difference in vocal quality, as they may signal inefficiencies in voice production that contribute to an overall difficulty in producing speech. The discordant results in published studies may be resolved by further study of age-related phenotypic variation in voice.

Another important question at the functional level is whether the vocal characteristics in DS are a result of laryngeal hyperadduction or hypoadduction. Pryce (1994) observed higher levels of electromyography (EMG) to initiate phonation in individuals with DS, a finding that is indicative of increased muscular activation of the larynx. If the laryngeal muscles are typically hypotonic, then it is possible that higher levels of muscular activation are needed to initiate and sustain phonation. Developmental factors may be relevant as well. Laryngeal hyperfunction in TD children has been described by Sapienza, Ruddy, and Baker (2004), who commented on the likelihood of false vocal fold adduction and the compression of the arytenoid cartilages to the petiole (the stalk of the epiglottis).

Indications for future research and clinical services. Despite a long history of research, the nature of the phonatory disorder in DS is not clearly established. Results of acoustic studies have been mixed, so it is difficult to draw firm conclusions or even to identify the most sensitive acoustic measures (e.g., jitter, shimmer, SNR) to be used in future research. The inconsistent results of efforts to identify acoustic correlates of perceived vocal abnormalities may mean that the vocal quality disorders are associated with a combination of acoustic characteristics that contribute, in varying degrees, to vocal quality among individuals with DS. Future research should be directed toward both structural (micro- and macro-anatomic features of the laryngeal tissues) and functional

objectives, taking into account developmental factors. New insights may be gained by pursuing methods of the kind described by Mehta and Hillman (2008). These include (a) perceptual assessment (use of the new Consensus Auditory–Perceptual Evaluation of Voice [CAPE–V] Inventory for the Auditory–Perceptual Assessment of Voice Quality; Kempster, Gerratt, Verdolini Abbott, Barkmeier-Kraemer, & Hillman, 2009); (b) acoustic assessment (use of new algorithms that are more robust across varieties of dysphonia and are capable of deriving vocal quality–related measures from conversational speech); (c) aerodynamic assessment (methods and devices for measuring phonation threshold air pressures and air flows); and (d) endoscopic imaging (high rates of image capture enhance the capabilities to examine the dynamics of vocal fold behavior). These research methods could be paired with a developmental perspective aimed toward the study of how laryngeal function changes with maturity and with the natural history of DS.

Speech Sound Disorders

Review of Literature

Studies in speech sound disorders in DS disclose a variety of problems affecting speech sound articulation, timing of syllable sequences, and phonological patterns. As shown in Appendix B, research in this category involved more than 700 participants, and the number of participants in individual studies generally ranged from fewer than 10 to as many as 66, with a mean of about 16.

Ontogeny of speech disorder. This subsection is concerned with the phonetic properties of speechlike vocalizations such as babbling, which involves supraglottal adjustments such as those of the jaw, lips, and tongue. Divergence in speech patterns between children with DS and TD children is clearly evident between the ages of 3 and 6 years (Bliele & Scharz, 1984; Moura et al., 2008; Smith & Stoel-Gammon, 1983). The stage of development at which differences in phonetic behavior emerge is less clear, but speech patterns may begin to diverge as early as the first year of life. Studies on early speech development in DS appear in the first section of Appendix B.

Although researchers in some studies did not find any remarkable differences in vocal development in infants with DS compared with TD infants (Dodd, 1972; Smith & Oller, 1981; Steffens et al., 1992), differences between infants with DS and TD infants have been observed. For example, studies have shown that infants with DS produced more nonspeech sounds and fewer speechlike sounds than did TD infants (Legerstee et al., 1992) and that the onset of canonical babbling was delayed by about 2 months in infants with DS and was less stable in infants with DS than in TD infants (Lynch et al., 1995). As discussed by Oller (2000), these conflicting

results may be attributable, in part, to different sampling intervals. Oller also noted that the delay in babbling onset in infants with DS is surprisingly small, especially when compared with the delays in gross motor skills such as sitting, crawling, standing, and walking (Palisano et al., 2001). In a similar way, Cobo-Lewis, Oller, and Lynch (1996) concluded that although attainment of canonical babbling was delayed in participants with DS, the delay was smaller than the delay for other milestones in motor and vocal development that the authors considered.

Smith and Stoel-Gammon (1996) reported no major differences in the development of specific types of babbling (e.g., reduplicated vs. variegated) in a comparison between infants with DS who were between 6 months and 2 years of age and TD age-matched infants. Research on phrasing in infant vocalizations showed that infants with DS have longer rhythmic units than do TD infants, but no differences emerged in overall vocal output or in the complexity of the rhythmic units (Lynch, Oller, Steffans, & Buder, 1995).

From these rather disparate findings, we can conclude that (a) the occurrence of babbling is typical but not universal in infants with DS (the same appears to be true of TD infants, but relevant data at the population level are surprisingly meager); (b) the age of onset of canonical babbling in infants with DS overlaps that in TD infants but may be somewhat delayed in infants with DS; (c) there may be differences in the features of babbling between infants with DS and TD infants; and (d) the delays in babbling are much less conspicuous than delays in gross motor skills such as crawling and walking.

Perceptual studies of vowel and consonant errors. An overall indication of vowel and consonant errors is expressed in the two measures of Percentage of Vowels Correct (PVC; Shriberg, Austin, Lewis, & McSweeney, 1997) and Percentage of Consonants Correct—Revised (PCC–R; Shriberg et al., 1997). (In the calculation of PCC–R, both clinical and nonclinical distortions are counted as correct; thus, only substitutions and omissions are counted as error sounds.) Van Bysterveldt (2009), reporting on 77 children with DS, obtained a mean PVC of 92.8 and a mean PCC–R of 78.2. In an intervention study of 10 children with DS in the age range of 4–5 years, van Bysterveldt, Gillion, and Foster-Cohen (2010) observed a mean PVC of 91.3 compared with a mean PCC–R of 50.6. These PCC–R values in DS exceed those for TD children compiled in the study by Bernthal, Bankson, and Flipsen (2009), except for one study of children with a mean age of 1;6 (years;months).

Several studies of speech in DS have noted vowel errors (Bunton, Leddy, & Miller, 2007; Van Borsel, 1996; van Bysterveldt et al., 2010). In their study of phonetic contrasts that are impaired in adults with DS, Bunton

et al. (2007) reported frequent errors with high versus low vowel and front versus back vowel. These errors indicate a limitation in the regulation of tongue height and advancement, which can occur because of anatomic factors and/or motor limitations. This issue is revisited in a subsequent discussion of acoustic studies of vowel articulation (see the *Acoustic and physiologic studies of speech in DS* subsection below).

Studies of both children and adults point to a higher than normal frequency of articulatory errors, with substantial involvement of consonants (Brown-Sweeney & Smith, 1997; Bunn, Simon, Welsh, Watson, & Elliott, 2002; Kumin, 1994; Roberts et al., 2005; Rosin, Swift, Bless, & Vetter, 1988; Schlanger & Gottsleben, 1957; Sommers, Patterson, & Wildgen, 1988; Timmins et al., 2009; van Bysterveldt, 2009; van Bysterveldt et al., 2010). Both the emergence and mastery of consonant phonemes in children with DS appear to be protracted processes, with substantial interindividual variability. The emergence of phonemes in the speech of children with DS does not seem to follow the order of published norms for TD children (Kumin, Councill, & Goodman, 1994). The most frequently misarticulated consonants may differ between children with DS and TD children. For example, Sommers et al. (1988) reported that for their group of 15- to 22-year-old participants, the 10 most frequently misarticulated sounds were (in descending order): /s/, /d/, /t/, /r/, /z/, /l/, /s/ blends, /r/ blends, /n/, and /v/. Errors on /d/, /t/, /n/, and /v/ are not common in TD children, and these sounds usually are mastered at an early age, with most children mastering /d/, /t/, /n/ by about 3 years of age (Bernthal et al., 2009). Of the 10 sounds listed by Sommers et al., seven involve the alveolar place of articulation, which is the most frequently used place of articulation in English and carries a significant intelligibility load (see the Intelligibility section). Bunton et al. (2007) identified phonetic contrasts that were most affected in DS. In addition to the vowel contrasts mentioned earlier, these included (a) simplification of clusters in both the word-initial and word-final position and (b) contrasts involving tongue posture, control, and timing (place of articulation for stops and fricatives).

General conclusions from perceptual studies of articulation. A condition is properly viewed as a *developmental delay* if the features of the condition follow the typical developmental course but with an overall delay in progress. The term *disorder* is applied if the features deviate from the pattern of typical development. Although developmental errors of articulation are prominent in DS, articulation errors of a nondevelopmental (“disordered”) nature also have been noted (Cleland, Wood, Hardcastle, Wishart, & Timmins, 2010; Dodd & Thompson, 2001; Kumin et al., 1994; Sommers, Reinhart, & Sistrunk, 1988).

Acoustic and physiologic studies of speech in DS. Studies involving acoustic and/or physiologic methods

are shaded in Appendix B to distinguish them from the more commonly used perceptual or transcription methods. The authors of several studies examined vowel production acoustically by examining formant frequencies. Novak (1972) commented that the overlap of first formant frequency–second formant frequency (F1–F2) areas for different vowels may explain listener difficulties in distinguishing vowels in DS, although Moran (1986) found no difference between DS and controls. Similarly, Saz, Simon, Rodriguez, Lleida, and Vaquero (2009) concluded from a study of Spanish speakers that errors in vowel identification were related to the confusability of vowel formant patterns as well as to poor control over the energy in stressed versus unstressed vowels and excessive variability in vowel duration.

Moura et al. (2008) reported that individuals with DS had a smaller ratio of the F2 frequencies for vowels /i/ and /u/ and called this ratio the *DS vocalic anatomical functional ratio*, implicating anatomy as the underlying basis of the formant frequency abnormality. However, this ratio may reflect either anatomic or motor factors (or both), as it is also a robust discriminator of dysarthric versus healthy speech (Sapir, Ramig, Spielman, & Fox, 2010). In a combined acoustic–articulatory study of two adults with DS, Bunton and Leddy (2011) reported a reduced range of F2 frequencies for the vowels /i/ and /u/, in agreement with Moura et al. (2008). Their data also show a smaller acoustic vowel area and a reduced articulatory working space compared with two age- and sex-matched healthy controls. The authors' most striking finding—markedly low F1 frequencies for the low vowels—could be explained by reduced mouth opening (and, probably, jaw lowering) in the participants with DS. In an acoustic study designed to identify the correlates of nasopharyngeal vocal quality (presumably a frequent characteristic of DS), F2 frequencies for the high vowel sounds were reduced in adolescent participants with DS, compared with TD children (Fourakis, Karlsson, Tilkens, & Shriberg, 2010). This feature was interpreted as evidence of backing of the tongue. The difference in F2 between /i/ and /u/ was virtually identical between the DS group and the TD group, which means that this dimension of the vowel space was not compressed in DS, contrary to the results of Moura et al. (2008).

Although it is reasonable to expect that vowel working space tends to be reduced in DS, studies on vowel formant frequencies in children and adults have been very limited and somewhat contradictory. More extensive data are needed from children and adults with DS. These data could be compared against normative data on acoustic vowel area that have been compiled for various age–sex groupings of speakers (Vorperian & Kent, 2007).

In a study of speech timing patterns, Brown-Sweeney and Smith (1997) did not find significant differences

between children with DS and TD children for durational measures, but the DS group was significantly more variable in two of seven segment measurements. Variability of word duration in children with DS also was reported by Hohoff, Seifert, Ehmer, and Lamprecht-Dinnesen (1998), whose results pertained to production of a single German word (*tasse*, meaning *cup*). These limited data point to increased variability in some temporal structures but not to abnormalities in the durations of segmental structure.

Physiologic methods are shedding new light on speech articulation in individuals with DS. Patterns of lingual contact have been studied with electropalatography (EPG; Gibbon, McNeill, Wood, & Watson, 2003; Hamilton, 1993; Timmins et al., 2009; Timmins, Hardcastle, Woods, & Cleland, 2011). Abnormalities observed in DS included both excessive and reduced areas of articulatory contact, moving contact, extended closure durations for occlusive consonants, and lengthened consonant transition times within clusters. Articulatory abnormalities were sometimes seen even when production of a speech sound was judged perceptually to be correct. Aerodynamic data on speech production in DS have seldom been reported, but Rosin et al. (1988) noted an increased intraoral air pressure for /p/ in speakers with DS. One interpretation of this result is that individuals with DS produce speech with greater respiratory pressures than do healthy controls. This possibility, together with the indication of increased muscular activation for phonation (Novak, 1972; Pryce, 1994; see the Discussion subsection of the Voice section), could mean that individuals with DS expend more energy in speech production than do TD speakers.

Phonological patterns. Articulation as a process is focused on physical production of sounds and the articulation data reviewed above answer questions such as “When are individual speech sounds mastered?” In contrast, phonology pertains to sound patterns such as those used to form words (e.g., the shapes of syllables within words), and phonological data are suited to questions such as “When are the phonological patterns of the language reliably produced to form words?” Studies of phonology in DS are summarized in Appendix B.

Phonological patterns in DS have been described for English speakers (Barnes et al., 2009; Cleland et al., 2010; Crosley & Dowling, 1989–1990; Dodd, 1976; Dodd & Thompson, 2001; Roberts et al., 2005; Sommers, Patterson, & Widgen, 1988; Stoel-Gammon, 1980; van Bysterveldt, 2009), Cantonese speakers (So & Dodd, 1994), Dutch speakers (Van Borsel, 1988), and Kannada speakers (Rupela & Manjula, 2007). As with studies of articulation (see the *Acoustic and physiologic studies of speech in DS* subsection of the Speech Sound Disorders section), phonological studies support a conclusion of combined developmental and disordered patterns in children with DS (Cleland et al., 2010; Dodd, 1976; Roberts et al., 2005; So & Dodd, 1994; Sommers et al., 1988). For

example, Sommers et al. (1988) observed the following nondevelopmental or disordered patterns: persistence of final consonant deletion processes, unusual difficulty with the acquisition of the liquids /r/ and /l/ and the nasals, and frequent errors with stop consonants. Unusual or atypical processes noted by van Bysterveldt (2009) included syllable reduction, glottal substitutions, epenthesis, metathesis, coalescence, and idiosyncratic substitutions. Nondevelopmental errors may be characteristic of a subtype of DS and may not necessarily occur in all individuals with DS.

Nasality and nasalance. *Nasality* is a perceived resonance quality that is related to velopharyngeal function. *Nasalance* is a physical measure of the ratio of nasally emitted acoustic energy to orally emitted energy. Nasality and nasalance are complementary measures, but they are not necessarily correlated in all speakers and speaking tasks.

Although nasality has been mentioned in some descriptions of speech in studies of individuals with DS, very few studies have directly assessed this aspect of speech production. In their study of pain cry in neonates and infants, Lind et al. (1970; see Appendix A, Table A1) remarked that hypernasality was a common feature in DS. Rolfe, Montague, Tirman, and Vandergrift (1979) noted that nasality was normal in most of their participants but that inconsistent hypernasality appeared in six children with DS. Hypernasality was not a prominent feature of speech reported in a parental report survey (Kumin, 2006), but lay individuals are not particularly discriminating when judgments of nasality are concerned. Kline and Hutchinson (1980) observed a marked increase in both perceptually judged nasality and acoustically determined nasalance in individuals with DS. Further study of oral/nasal resonance is needed, given that nasalization may contribute to abnormal vocal quality, reduced energy levels in speech (because of increased damping in sound transmission through the vocal tract), and reduced intelligibility (because nasalization can interfere with the production of phonetic contrasts). It is also possible that oral/nasal resonance balance is affected by abnormalities in the nasal cavities, the sinuses, and the tissue boundaries between the oral and nasal passages. As mentioned earlier, Fourakis et al. (2010) reported on the acoustic correlates of a vocal quality that they termed *nasopharyngeal resonance*. The origin of this quality is unclear, but it may be related to reports of hypernasality in DS.

Oral motor control in simplified speaking tasks. Clinicians commonly use *diadochokinesis* (DDK), also known as *maximum syllable repetition rate* or *alternating motion rate*, to assess oral movement skills in a task that makes modest demands on language ability and memory. Most studies of DDK in individuals with DS report a decreased rate (Brown-Sweeney & Smith, 1997; Hamilton, 1993;

Rosin et al., 1988), but McCann and Wrench (2007) observed a DDK rate similar to that seen in TD children, although they noted that the participants with DS were more inaccurate in performing the task.

The generally slow DDK rates reported for DS stand in contrast to some reports of an overall normal or even a rapid speaking rate. Fawcett and Peralego (2009) commented, "Probably one of the most striking characteristics of the speech of people with Down syndrome is a rapid rate" (p. 111). But rapid rate has not been uniformly confirmed in studies of DS, with at least one study reporting a slower speaking rate in words per minute for individuals with DS compared with TD controls (Chapman, Seung, Schwartz, & Kay-Raining Bird, 1998). Brown-Sweeney and Smith (1997) found that temporal segment durations in word production were not significantly different between speakers with DS and TD speakers even though the speakers with DS had slower DDK rates. Additional studies of speaking rate for both syllable repetition and meaningful speech are needed before firm conclusions can be drawn. The issue of speaking rate is revisited in the discussion of disfluency (see the Fluency and Prosody section), where rate is potentially related to the disorder of cluttering.

Anatomic anomalies and pathophysiology. Description of craniofacial anomalies is complicated by phenotypic variation and by developmental changes of specific features. Some characteristics of DS, including brachycephaly and the absence of nasal bone ossification, can be identified prenatally (Stempfle, Hutten, Fredouille, Brisse, & Nessmann, 1999). Craniofacial dysplasia is evident at birth and increases in severity with age until at least 14 years (Fischer-Brandies, 1988), although the rates and directions of growth appear to be similar to typical development (Frostad, Cleall, & Melosky, 1971).

Overall craniofacial anatomy. In an MRI study, Uong et al. (2001) noted that, compared with controls, participants with DS had reduced volumes of the airway, mandible, adenoid, and tonsil and a smaller mid- and lower-face skeleton and hard palate. The tongue, soft palate, pterygoid, and parapharyngeal fat pads seemed unaffected. Those authors concluded that the reduction in upper airway size is the result of soft tissue crowding within a smaller mid- and lower-face skeleton. An anthropometric study of craniofacial features showed a relatively small maxilla but a normal mandible (Allanson, O'Hara, Farkas, & Nair, 1993). A number of dental abnormalities have been reported (Cohen & Winer, 1965; Shapiro, Gorlin, Redman, & Bruhl, 1967). Anatomic studies have shown poorly differentiated mid-face muscles and the presence of muscles not seen in healthy individuals (Bersu, 1976, 1980).

Hypotonia. It is repeatedly asserted in the literature on DS that affected individuals have a hypotonic musculature (Desai, 1997). However, assessments of stiffness

do not necessarily support the contention that hypotonia is a pervasive characteristic (Connaghan, 2004). To the extent that hypotonia is present, it could explain some of the speech features that resemble the dysarthrias, with the expectation that these features would resemble those seen in flaccid or ataxic dysarthria, both of which are associated with hypotonia. Generalized hypotonia could help to explain altered function in the subsystems of speech production—especially the larynx, velopharynx, and the oral articulators.

The tongue. Macroglossia has historically been assumed to be a common feature of DS. This thinking led to surgical intervention by lingual resection, but it appears that an enlarged tongue in individuals with DS is more impressionistic than real. Ardran, Harker, and Kemp (1972) concluded from a radiographic study that none of the 16 children with DS had a generalized enlargement of the tongue, although regional enlargement was noted in five individuals. In a similar way, Guimaraes, Donnelly, Shott, Amin, and Kalra (2008) concluded that children with DS do not have true macroglossia but, rather, have relatively large tongues compared with the bony confines of the oral cavity. Evidence of abnormalities of the myofibers of the tongue also has been reported (Yarom, Sagher, Havivi, Peied, & Wexler, 1986).

The palate. Abnormalities in palatal anatomy have been recognized for decades (Benda, 1960; Oster, 1953). In one early study, it was concluded that the palates of individuals with DS were narrower but not higher than the palates of controls (Oster, 1953). More recently, however, Dellavia et al. (2007) reported no differences in the sagittal plane, but observations of the frontal plane showed a higher palate. In a similar way, Bhagyalakshmi, Renukarya, and Rajangam (2007) concluded that although individuals with DS had smaller values than age- and sex-matched controls for measures of palatal width, length, and volume they had greater values for the measure of average palatal height.

Škrinjarčić, Glavina, and Jukić (2004) found that shelflike or *stair palate* palatal shape was more than three times as likely to occur in participants with DS than in a control group. The authors also noted that the frequency of a shelflike palate diminished with age, which was attributed to the growth of craniofacial structures and increased tonus of the tongue and other orofacial muscles.

Beck (1997) suggested that the short, narrow palate with an essentially normal tongue would lead to fronted articulations of the tongue tip and blade, along with a fronting and raising of the tongue body setting. Brunner, Fuchs, and Perrier (2009) concluded that flat palates are associated with a greater acoustic sensitivity and, therefore, a smaller tolerance in articulatory positioning than arched palates. The acoustic effects of shelflike palatal shape apparently have not been studied.

Vocal tract and laryngeal configuration. Beck (1997) described significant differences in the “vocal setting” in DS compared with healthy controls, including protruded mandible; fronted tongue body; pharyngeal constriction; harshness; whisperiness; lax vocal tract; minimal range of lip, tongue, and jaw motion; nasality; and open jaw. Evidence of a relatively small oral cavity in the presence of seemingly normal pharyngeal length, pharyngeal volume, and vocal tract length was reported by Xue, Kaine, and Ng (2010), who used an acoustic reflection technique.

Auditory function. Reports on the prevalence of hearing loss in DS vary considerably, but some degree of hearing loss has been noted in audiometric studies of children (Balkany, Downs, Jafek, & Krajicek, 1979; Park, Wilson, Stevens, Harward, & Hohler, 2011; Roizen, Wolters, Nicol, & Blondis, 1993; Shott, Joseph, & Heithaus, 2001) and adults (Buchanan, 1990; Evenhuis, Van Zanten, Brocaar, & Roerdinkholder, 1992). Survey studies show moderate prevalence of hearing impairment (Kumin, 2006; Schieve et al., 2009). Hearing impairment certainly must be considered in explanations of delayed or disordered development of articulation, but, as Vicari (2006) observed, “There is no definitive evidence that language impairment in DS is merely a consequence of the hearing loss” (p. 356).

Summary

The craniofacial anatomy in individuals with DS is characterized by a compact mid- and lower-face skeleton, a tongue of average size, and a palate that is high and often shelflike. The developmental trajectory of orofacial characteristics is not well established. Developmental instabilities have been implicated in fluctuating dental asymmetry (Barden, 1980), which is an example of a more general pattern of developmental instability manifested as decreased developmental and physiologic buffering against genetic and environmental forces (Shapiro, 1975, 1983).

Discussion

Speech production in DS is compromised by several types of impairment. The relationship among these multiple impairments is not clear because the full range of impairments has rarely been examined in the same set of participants. There is reasonable agreement on the following general points:

1. Speech difficulties are not highly correlated with language or cognition, which may indicate that problems in speech are rooted in other factors, such as anatomy and motor control.
2. Reports are mixed on the extent to which infants with DS have atypical patterns of vocal development, but there appears to be some delay in the appearance

of canonical babbling. Any such delay is modest compared with delays in gross motor skills.

3. Articulatory and phonological studies show both delayed (i.e., developmental) and disordered (i.e., non-developmental) patterns in children with DS by about age 3 years, although other effects may appear at earlier ages.
4. Articulatory and phonological patterns in individuals with DS show inconsistent errors and possibly increased variability at the acoustic level—at least, for some segments. This fluidity of disordered patterns is an important clue to their etiology and a factor to be considered in assessment and treatment.
5. Although peripheral factors such as anatomic anomalies are not likely to explain all aspects of the speech disorder in individuals with DS, the deviations may impose some limitations on articulatory performance (Beck, 2010; Bunton & Leddy, 2011; Leddy, 1999). It is not well established how developmental changes in anatomy and physiology relate to articulatory and resonance features of speech.

Indications for Future Research and Clinical Services

Perceptual methods such as articulation testing and transcriptions of speech samples have provided a general description of speech sound disorders in DS. As indicated in Appendix B, the error patterns are complex and may be understood more fully from the use of instrumental methods, such as acoustic analysis, aerodynamic recordings, EPG, and movement transduction. It may be particularly informative to use combined methodologies to study speech production in DS (e.g., combining acoustic measures of speech with physiologic recordings). In addition, electromagnetic articulography (EMA) may be suitable to the study of speech movements in adults and children with DS. Researchers have successfully used this method to study speech articulation in children with dysarthria (Murdoch & Goozee, 2003). Reports of increased variability in speech production could be examined further with the *spatiotemporal index* (STI), a measure of variability in the production of several tokens of an utterance (Smith, Goffman, Zelaznik, Ying, & McGillem, 1995). It is also particularly important to study micro- and macro-anatomic development of the craniofacial system with respect to its motoric capabilities to determine structure–function relationships.

Fluency and Prosody Review of Literature

As noted in the introduction, fluency and prosody are grouped together in this review because they pertain

to speech behaviors that are best expressed in units larger than the phone (i.e., larger units such as the syllable or syllable string). In studies of fluency disorders, authors have used several different terms, including *dysfluency*, *disfluency*, *stuttering*, *cluttering*, and *stuttering/cluttering*. For the purposes of the present review, the word *disfluency* is a general term that includes all varieties of interruption in the flow of speech. Some of the reported disfluencies may be similar to those that occur in typical speech development.

Disfluency. Studies of speech disfluency in more than 300 participants have demonstrated that stuttering and/or cluttering occurs in individuals with DS at rates of 10%–45% (see Appendix C), compared with the incidence of about 1% in the general population (Guitar, 1998). It is generally not possible to distinguish normal developmental disfluencies from genuine stuttering or cluttering in this literature. Presumably, stuttering and cluttering were judged to be clinically significant. The published data do not permit conclusions on the persistence or developmental pattern of fluency disorders in individuals with DS.

In studies within the literature, stuttering has been demonstrated in 10%–45% of children with DS, with a mean of about 31%, or one in every three individuals with DS (Devenny & Silverman, 1990; Gottsleben, 1955; Keane, 1970; Preus, 1972; Rohovsky, 1965; Schlanger & Gottsleben, 1957). Rohovsky (1965) observed a rate of 36% in individuals with DS who were institutionalized, compared with 19% in those individuals with DS who were not institutionalized. Survey data confirm a rather high incidence of stuttering in individuals with DS: 17% in Kumin's (1994) parent report survey and 15.6% in Schieve et al.'s (2009) analysis of data from the National Health Interview Survey (Botman, Moore, Moriarity, & Parsons, 2000).

Other studies have provided information on the topography of stuttering. Otto and Yairi (1974) found statistically significant differences in disfluencies between 19 individuals with DS who were institutionalized, compared with an equivalent number of healthy controls. Analysis of the disfluencies with respect to seven categories of disfluency showed that the participants with DS had patterns similar to those observed in individuals with developmental stuttering. Willcox (1988) observed both similarities and differences in the types of disfluency in the speech of children with DS and in the speech of language-matched children without DS. She concluded that “it is clinically more appropriate to consider the speech non-fluencies of Down's syndrome individuals as part of a global language deficit rather than as a symptom of the syndrome” (p. 169).

The disfluencies in DS may take forms other than developmental stuttering. Cluttering may be even more frequent than stuttering. One of the first authors to note

the possibility of cluttering was Cabanas (1954), who asserted that the rhythm disorders in the individuals whom he studied should be called “cluttering” because of their restricted vocabularies, rapid speech patterns, and “lack of ideomotor equilibrium” (p. 36). Van Borsel and Vandermeulen (2008) classified a very large percentage of their 76 participants with DS as being either clutterers (about 80%) or clutterer–stutterers (about 17%). Preus (1972) noted that both stuttering and cluttering occur in individuals with DS and are not correlated.

Prosody. *Prosody* is a general term for the rhythmic and intonational aspects of language and includes rhythm, intonation, and lexical and emphatic stress. As shown in Appendix C, researchers in only a handful of studies—involving almost 50 participants—have examined prosodic features in the speech of individuals with DS, but all of these studies indicate that individuals with DS have limitations in the perception, imitation, and spontaneous production of prosodic features (Pettinato & Verhoeven, 2008; Reichle, Siegel, & Rettie, 1985; Shriberg & Widder, 1990; Stojanovik, 2010). Shriberg and Widder (1990) found that participants with a higher probability of being able to live independently also had better speech and prosodic capabilities. Prosodic features may have a bearing on intelligibility, insofar as increased intelligibility has been reported for prepausal rhythmic groups (Flipsen, 1999).

Discussion

Disfluency (either stuttering or cluttering) is highly likely to occur in DS, but it is by no means a universal characteristic of the syndrome. The types of disfluency are similar to those seen in developmental stuttering, which may be a sign of similarities in the origin of the disorder. The diagnosis of cluttering, as in the study by Van Borsel and Vandermeulen (2008), emphasizes the need to consider disfluency in relation to speaking rate, given that a rapid rate is frequently implicated in cluttering. Results on speaking rate in individuals with DS are mixed. The few studies reporting on prosody indicate that prosodic disturbance is a common feature of DS.

It is difficult to determine the degree to which stuttering or cluttering is comorbid with other speech and voice problems. It is also unclear if the nature and severity of the fluency disorder changes over the lifespan or if the “stuttering” in infant pain cry (Lind et al., 1970) is related to the later appearance of disfluencies in childhood. Disfluent speech in individuals with DS has been attributed to dysfunction in either motor control or language processes, such as utterance formulation or word finding (Leddy, 1999). Both kinds of dysfunction may need to be recognized in an integrated model, such as the *EXPLAN model*, which proposed to account for developmental stuttering (Howell, 2011; Howell & Au-Yeung,

2002). This model assumes that language planning (PLAN) as well as speech–motor programming and execution (EX) are independent processes, and it is the interface between these processes that determines the fluency of speech. An advantage of the *EXPLAN model* is that it can account for both language and motor influences on disfluent speech.

Limitations in prosody could be the result of motor difficulties, problems in coordinating speech motor control with phonological or other higher level representations, or even serious segmental (articulatory) errors that impede the effective production of speech across multisyllabic sequences. Prosodic abnormalities may have their origin in limitations of phonological processing (Pettinato & Verhoeven, 2008; Shriberg & Widder, 1990). It is also possible that prosodic difficulties contribute to problems in other domains. For example, Pettinato and Verhoeven (2008) concluded that “our findings are in accord with studies which suggest that underlying difficulties with the rhythmic and prosodic structure of speech are driving dysfluencies and reduced speech intelligibility in the speech of individuals with Down syndrome” (p. 58).

Indications for Future Research and Clinical Services

Disfluencies and dysprosody are fairly common in DS and constitute one part of a larger profile of communication disorder. A challenge for future research is determining the interactions between disfluencies and dysprosody with other aspects of communication—including syntactic, lexical, and phonological processes—in an effort to identify causal relations. In addition, research that combines methodologies (e.g., acoustics, EMA, and perceptual scaling) should be used in an effort to describe motor patterns associated with disturbances in fluency and prosody.

Intelligibility Review of Literature

Whereas the investigations in Appendix C address speech articulation or phonology, those in Appendix E specifically provide estimates of overall intelligibility. Definitions of *intelligibility* differ across published articles, as do the methods of assessing it. As Leddy (1999) pointed out, many reports assessed intelligibility incidental to other research goals, such as determining aspects of language formulation or vocabulary. We omitted parent surveys and intervention research; consequently, the total number of participants in studies that directly assessed intelligibility approaches 150 (see Appendix E), but the number is larger if related measures—such as

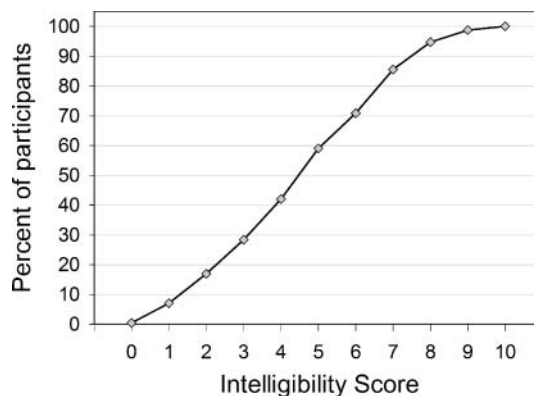
some reported in Appendix B—are included. When data from parental surveys are aggregated (Kumin, 1994, 2006), the number of participating units swells to more than 2,500. Several published intervention studies are not included in Appendix E because they reported only a change in intelligibility between pre-treatment and post-treatment rather than explicit pre- and post-treatment ratings. Appendix E includes a small number of studies in which intelligibility was assessed relative to an intervention.

Studies reporting intelligibility estimates. Reduced intelligibility results in difficult communication and can interfere with a variety of activities in everyday life (Barnes et al., 2009; Bray & Woolnough, 1988; Bunton et al., 2007; Kumin, 1994, 2006; Price & Kent, 2008; Rosin et al., 1988). Research that focuses on intelligibility, per se, is limited in the literature on DS. Diminished intelligibility is substantiated by parental report (see Appendix D) and clinical or laboratory testing (see Appendix E). The underlying causes of this problem can only be surmised from studies that examine aspects of speech production, as reviewed in the previous sections, along with studies of other domains of spoken language. It appears that intelligibility reduction is exacerbated by increased length of utterance (Kumin, 1994; Yoder, Hooshyar, Klee, & Schaffer, 1996) and nonfamiliarity of the listener (Kumin, 1994).

A variety of procedures are used to estimate intelligibility (Price & Kent, 2008), but the main methods that have been used in studies of individuals with DS are scaling procedures (e.g., percentage estimate of intelligibility; Kumin, 2006), word identification (Bunton et al., 2007), and scoring from transcriptions (Chapman et al., 1998; Chapman, Seung, Schwartz, & Kay-Raining Bird, 2000; Rosin et al., 1988). Regarding scoring from transcriptions, Chapman et al. (1998) wrote that “Intelligibility was scored as the proportion of complete and intelligible utterances over total utterances” (p. 864). Another approach is to measure correlates of intelligibility—for example, PCC (Barnes et al., 2009; Kennedy & Flynn, 2003; McCann & Wrench, 2007; Roberts et al., 2005; van Bysterveldt, 2009; van Bysterveldt et al., 2010). As was noted in the *Perceptual studies of vowel and consonant errors* subsection of the Speech Sound Disorders section, PCC values reported for children with DS are markedly reduced compared with the PCC values reported for TD children. We discovered only two studies of children with DS that reported PVC (van Bysterveldt, 2009; van Bysterveldt et al., 2010). The results indicate that production of vowels and diphthongs is more accurate than the production of consonants.

Figure 2 shows a cumulative plot of intelligibility scores derived from data of Kumin’s (1994) study. Note that 60% of the participants had an intelligibility rating of 5 or lower and that 89% of participants had a rating of

Figure 2. Plot of cumulative percentage of participants receiving a given intelligibility score in Kumin (1994). The horizontal axis is the intelligibility rating, and the vertical axis is the cumulative percentage. For example, about 60% of participants had scores of 5 or lower.



7 or lower. These results, which are based on parental ratings, agree with estimates of intelligibility reported by Chapman et al. (1988) and van Bysterveldt (2009), both of whom reported an average intelligibility score of about 80%.

Related measures. Measures of intelligibility are complemented by other measures including comprehensibility, listener comprehension, and communicative participation. *Comprehensibility* is defined as “contextual intelligibility,” or intelligibility when contextual information is present in different forms, such as semantic cues, syntactic cues, orthographic cues, and gestures (Yorkston, Strand, & Kennedy, 1996). Measures of listener comprehension evaluate listeners’ ability to interpret the meaning of messages without regard for accuracy of phonetic and lexical parsing (Hustad & Beukelman, 2002). *Communicative participation* is defined as communication in social contexts (Eadie et al., 2006). These latter three measures have been used only infrequently in the study of communication in individuals with DS, but Camarata, Yoder, and Camarata (2006) used a measure of speech comprehensibility defined as the *percentage of utterances that are comprehensible*. The advantage of this measure is that it is sensitive to communication success or failure, regardless of whether individual words are accurately identified by the listener.

Discussion

Several studies substantiate that intelligibility is a serious problem in individuals with DS, that it persists throughout life for many individuals, and that it may have negative effects on social and vocational pursuits. Very few of these studies have reported a detailed analysis of factors underlying reduced intelligibility, although

it can be assumed that disturbances in voice, articulation, resonance, fluency, and prosody all contribute to the problem. It is not known how difficulties in each of these areas contribute to an overall deficit in intelligibility. It is also not clear if the presence of unusual or atypical articulatory or phonological errors, as reviewed in the Speech Sound Disorders section, increases the risk of impaired intelligibility.

Indications for future research and clinical services. Reduced intelligibility in DS has been well documented, but the reasons for it have not been sufficiently explored. Impaired intelligibility is probably based, to some degree, on all of the other functions considered in this review (voice, speech sound production, fluency, and prosody), but a satisfactory study of their interrelationships would require many participants and several research methods. It is likely that progress could be made with less ambitious methods, such as acoustic studies of speech in DS. It may suffice to examine a set of acoustic features that appear to be related to speech intelligibility. One such set was described by Amano-Kusumoto and Hosom (2011) in a review of clear (highly intelligible) versus conversational (less intelligible) speech in healthy adults: formant transitions, temporal envelope, F1 and F2 ranges, formant bandwidth, and voice onset time. These features should be studied systematically in DS. As reviewed in the Speech Sound Disorders section, more data have been published on F1 and F2 ranges than on any other acoustic aspect of speech, but even these studies are not in agreement. In future studies, researchers could examine all the acoustic features mentioned, preferably in the same group of participants and with suitable TD controls. A better understanding of the bases of reduced intelligibility would help to guide clinical intervention. These bases may vary across individuals with DS, which is further reason to develop profiles of speech disorders that are linked to intervention strategies.

General Discussion

Given the evidence reviewed here, individuals with DS have difficulties in the domains of voice, speech sound production, fluency and prosody, and intelligibility. Children and adults with this syndrome face serious challenges in spoken communication, which may substantially interfere with their participation in social, educational, and vocational activities. The difficulties in communication are rooted in virtually all aspects of speech production, making it difficult to identify domains of strength that might be leveraged in the design of effective interventions. Although not every individual with DS will experience the full range of abnormalities noted in this review, multiple involvements are likely, and comprehensive assessments should be considered,

with due consideration of the results in treatment planning.

Population Sampling and Criteria for Selection of Control Groups

Shin et al. (2009) estimated that in 2002, there were 83,400 individuals with DS under the age of 20 years living in the United States. As noted in this review, the aggregate number of participants in each of the four areas of research related to speech communication in DS is in the low hundreds, which probably is not sufficient to assess phenotypic variation, especially because the majority of published studies focused on a small set of measures within any of the four research areas.

Control groups used in studies of speech in individuals with DS include mental age matches of TD individuals, chronological age matches of TD individuals, and participants with other types of disorders (e.g., children with fragile X syndrome and children with phonological disorders). Characteristics of control groups can strongly affect the validity of conclusions reached in studies of speech abilities. With mental age matching, there is no control for physical development and body size, both of which can substantially affect aspects of speech (in particular, acoustic measures of f0 and formant frequencies). Chronological age matching provides better control over physical development; however, it offers limited control over physical size and little or no control over language or cognitive capabilities or general experience (e.g., social interactions in different settings). Comparison with other types of developmental disorders can be revealing, but questions arise as to the need for matching body size, chronological age, and mental age. No single control group is satisfactory for all aspects of research on speech production, but a particular control group can be justified for studies of a highly specific nature.

Co-Occurrence and Impairment Profiles

Considering the broad spectrum of speech disturbances in individuals with DS, it is important to know patterns of co-occurrence. The unfortunate reality is that this information is not easily extracted from the literature. It has been established that many types of speech disorders in individuals with DS have high rates of co-occurrence or comorbidity in populations other than individuals with DS. For example, it has been estimated that developmental stuttering has a comorbidity of about 60% with speech, language, and other disorders (with articulation and phonological disorders being the most frequently co-occurring; Blood, Ridenour, Qualls, & Hammer, 2003). In a similar way, Arndt and Healey (2001) reported from a survey of 241 speech-language pathologists that

44% of 467 children who stuttered had a verified concomitant phonological and/or language disorder. In future studies of individuals with DS, researchers should examine the co-occurrence of voice disorders, articulatory–phonological disorders, fluency and prosodic disorders, and various aspects of language disorders. Identifying profiles of impairment may be an important step in selecting treatment strategies.

The multidimensional character of the speech disorder in individuals with DS is central to determinations of symptomatology and pathophysiology. A profile of impairments is one way to register the dimensions of the speech disorder in individuals with DS, and researchers can use this profile to identify general patterns of disorder in the population. Individual differences can be described relative to these general patterns. The classification of speech production difficulties into the four major classifications used in the present review cannot capture the interaction among these categories. Intelligibility—the most critical outcome with respect to communicative success or failure—is moderately to severely compromised in DS, yet it is one of the most poorly quantified aspects of speech production. Given the breadth of the difficulties in speech production, a hierarchy could be established to guide efficient assessment and treatment. Comprehensive testing allows the identification of co-occurring problems as well as the identification of areas of relative strength or competence.

Speech Disorders in Relation to Language, Cognitive, and Memory Functions

Speech cannot be isolated from other aspects of communication or cognition. Although this review focuses on speech production, problems with speech must be viewed in a larger context of perceptual, motor, and linguistic abilities. Speech problems in individuals with DS may be related to peripheral factors such as anatomic differences in the vocal tract, impaired hearing acuity during recurrent otitis media, and impaired motor function (dysarthria and/or apraxia) or to central factors such as language and cognitive dysfunctions. It is likely that several factors interact in the development and persistence of speech disorders in individuals with DS, each with a developmental trajectory that contributes to the overall interaction. Causal relationships among the various speech and language impairments are not easily determined. For example, it has been suggested that disfluencies are the result of language impairment (Willcox, 1988), underlying difficulties in the control of rhythm and prosody (Pettinato & Verhoeven, 2008), or a combination of language and motor limitations (Cabanais, 1954). Longitudinal studies may shed light on the relationships among the impairments noted in this review, but these studies are nearly nonexistent.

Short-term memory impairments have been noted in studies of individuals with DS (Bunn, Roy, & Elliott, 2007; Jarrold, Baddeley, & Phillips, 2002; Kanno & Ikeda, 2002; Laws, 1998; Vicari, 2006) and appear largely independent of speech articulation or speech perception abilities. Performance on certain speech and language tasks is likely affected by limitations in short-term memory.

Childhood Apraxia of Speech: A Component of DS?

More than 35 years ago, Dodd (1976) posited that the articulatory disorder in DS is rooted at least partly in “difficulties in programming the motor movements of speech” (p. 41). This implies that the motor disorder in speech is not only a *dysarthria* (typically defined as a disorder of execution) but perhaps also an *apraxia* (typically defined as a disorder of motor programming or sequencing). More recently, it has been proposed that children with DS have childhood apraxia of speech (CAS; Kumin, 2006; Rupela & Manjula, 2007). This proposal was based on similarities between speech behaviors in children with DS and those in children with CAS.

A diagnosis of CAS can be difficult, especially when this disorder is comorbid with other speech and language abnormalities associated with DS. CAS has been defined as “a neurological childhood (pediatric) speech sound disorder in which the precision and consistency of movements underlying speech are impaired in the absence of neuromuscular deficits (e.g., abnormal reflexes, abnormal tone)” (American Speech-Language-Hearing Association, 2007). There are only three features of CAS with widely acknowledged diagnostic validity: (a) inconsistent error production on both vowels and consonants across repeated productions of syllables and words, (b) lengthened and impaired coarticulatory transitions between sounds and syllables, and (c) inappropriate prosody. The diagnosis of CAS is usually made on the assumption that there is no evidence of craniofacial anomalies or of neurological abnormality in the speech musculature. It is obvious that this assumption cannot be made in individuals with DS, who are considered to have—at minimum—a hypotonic musculature and fairly distinctive craniofacial features, some of which affect the oropharyngeal structures involved in speech. The high prevalence of cluttering or cluttering–stuttering further complicates a confident diagnosis of CAS. This is not to say that CAS is unlikely to occur but, rather, that a confident diagnosis of this condition must take into account the combination of articulatory errors, abnormal muscle tone, and fluency disorders that appear to be common in individuals with DS. To some degree, CAS is a diagnosis of exclusion that is obviated in DS. The challenge, then, is to distinguish features of CAS from co-occurring

abnormalities related to neurological, structural, and, perhaps, other domains.

There is evidence of a general difficulty in praxis skills in individuals with DS (Bunn et al., 2007; Fidler, Hepburn, Mankin, & Rogers, 2005). Bunn et al. (2007) proposed that movement organization deficiencies in individuals with DS could reflect a difficulty in generating actions from memory. If this limitation is general across motor systems, then some aspects of speech production disorders would be based on deficiencies in central processes. Vulnerability of praxis skills is evident throughout life, as older individuals with DS appear to exhibit increased praxis disturbances (Daunhauer & Fidler, 2011).

Neural Abnormalities

Neural dysfunctions likely underlie many of the disorders considered in this review. Abnormalities of neuroanatomy and neural function have been described in several recent articles (Fidler et al., 2005; Nadel, 2003; Pinter, Eliez, Schmitt, Capone, & Reiss, 2001; Vicari, 2006), and these abnormalities could well be the basis for apractic and dysarthric characteristics of speech in individuals with DS. An important step in this effort is the systematic description of speech disorders in DS, including their natural history, comorbidity, and response to intervention.

Cross-Linguistic Research

The great majority of studies in this review pertain to speakers of English. Cross-linguistic studies are important because they establish features that are universal versus those that are specific to individual languages or families of languages. It is very difficult to determine from the published studies if there are strong cross-language correspondences in problems with voice, speech sound production, fluency and prosody, or intelligibility. The conclusions of this review may be used to form hypotheses for DS research in other languages.

Future Research

Perceptual methods—such as ratings of vocal quality and articulation tests—have provided basic information on characteristics of spoken language in DS, but the use of the instrumental techniques of acoustic and physiologic methods has been limited. EPG is one of the most frequently used of these techniques and has contributed especially to an improved understanding of lingual articulation. Acoustic methods have the potential for refined analyses of articulation and prosody. Aerodynamic recordings may reveal important aspects of voice and speech dysfunctions. A major direction for future research is the

application of instrumental techniques in a lifespan perspective to answer questions such as the following:

1. How do the air pressures and air flows for speech production in individuals with DS compare with those of TD controls at various times of development? For example, if intraoral air pressures are higher in individuals with DS than in TD controls (Rosin et al., 1988), then do individuals with DS drive the speech production system with unusually high pressures? If so, how does this feature relate to disturbances in voice, articulation, fluency, and prosody?
2. Assuming that vowel articulation is often impaired in individuals with DS (as perceptual studies indicate), what is the characteristic acoustic (F1–F2) space for vowels produced by children and adults with DS, and how does this result relate to reduced intelligibility? If atypical results are found in individuals with DS, are they the consequence of anatomic anomalies, motor control deficiencies, or both?
3. Despite longstanding comments on vocal quality abnormalities in DS, no consistent acoustic correlates have emerged. What are the acoustic patterns of phonation in individuals with DS in different phonation tasks, including sustained phonation, single-word production, and sentence recitation? How does phonation change during development and maturation? Do abnormalities in voice contribute to dysprosody?
4. Given the considerable evidence to date that DS is associated with prosodic abnormalities, what are the acoustic correlates of prosody in individuals with DS, and how do these differ from the correlates in TD controls?
5. Different conclusions have been reached on how speaking rate in individuals with DS varies across tasks. Acoustic and physiologic methods are well suited to the quantitative study of speaking rate. What is the effect of rate changes on segmental durations? Is rate a potent variable in intervention for speech?

One of the most productive approaches that researchers can use to address the foregoing and other questions listed in the conclusion of each major section in this review would be to use combined methodologies (a combination of perceptual, acoustic, and physiologic methodologies) to obtain detailed information on how voice, speech sound articulation, and fluency and prosody interact. Doing so would help to determine the intelligibility of speech in individuals with DS. It may be particularly fruitful to use such methods to determine speech production capabilities as a function of development. Such an approach to understand speech functions in DS may benefit the study of other complex disorders, such as childhood dysarthria, which involve a constellation of atypical patterns that interact to reduce speech intelligibility.

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Appendix A (p. 1 of 4). Summary of studies of voice in individuals with Down syndrome (DS).

Table A1. Summary of studies of voice in infants, children, and adolescents with DS.

Source	Participants	Method	Summary of results
Vuorenkoski, Lind, Wasz-Hockert, & Partanen (1971)	n = 30 DS (infants and neonates) n = 120 TD (infants and neonates) n = 90 AD with various pathologies, excluding DS Age range = 0 days to 8 months	<i>Acoustic assessment:</i> Derivation of a cry score (ranging from 1 to 4) calculated from 13 spectrographic features	93% of DS had an abnormal cry score, compared with only 6% of TD; distinguishing abnormalities included stuttering and melody.
Vuorenkoski, Wasz-Hockert, Lind, Koivisto, & Partanen (1971)	n = 3 DS (newborns) n = 8 TD (newborns) n = 9 AD (not DS; newborns)	<i>Perceptual assessment:</i> Auditory judgments of pain cries by a group of pediatricians and a group of medical students	Acoustic information from spectrograms improved the ability to identify medical status of newborns, especially for DS.
Lind, Vuorenkoski, Rosberg, Partanen, & Wasz-Hockert (1970)	n = 30 DS (infants) n = 120 TD (infants)	<i>Acoustic assessment:</i> Spectrographic features of pain cry	Participants with DS had abnormal features of pain cry, including long duration, low pitch, monotonous with flat melody form, nasal, and stuttering.
Weinberg & Zlatin (1970)	n = 27 DS (5;01–6;11)[yrs;mos] n = 66 TD (5;00–6;10)	<i>Acoustic assessment:</i> Analyses of M, SD, and range of speaking f ₀ , as determined with a dedicated device	Participants with DS had a higher mean f ₀ compared with controls.
Moura et al. (2008)	n = 66 DS (36 M, 30 F) Range = 3–8 yrs M _{age} = 5.8 yrs n = 204 TD (104 M, 100 F; M _{age} = 5.7 yrs)	<i>Acoustic assessment:</i> Voice assessments using Praat software (Boersma & Weenink, 2010) <i>Perceptual rating:</i> Modified GRBAS rating scale (Hirano, 1981)	Participants with DS had a lower f ₀ with elevated dispersion, greater measures of perturbation and noise higher, and lower value of spectral tilt.
Pentz & Gilbert (1983)	n = 14 DS (6 M, 8 F) Range = 7–10 yrs M _{age} = 9.42 yrs n = 14 TD (6 M, 8 F; 7–10 yrs; M _{age} = 9.25 yrs)	<i>Acoustic assessment:</i> Voice assessments using a Kay Visi-pitch, a Kay spectrograph, and an oscillograph. <i>Perceptual rating:</i> Ratings with Wilson Voice Profile (Wilson, 1972)	Participants with DS were significantly different for all variables. DS group had increased frequency perturbation, amplitude perturbation, and noise-to-harmonic ratios.
Pentz (1987)	n = 14 DS (6 M, 8 F; 7–10 yrs) n = 14 TD (6M, 8F; 7–10 yrs)	<i>Acoustic assessment:</i> Measurement of formant amplitudes using a spectrum analyzer	DS group was different only on the Severity subscale. DS had significantly reduced formant amplitude intensity levels.

Appendix A (p. 2 of 4). Summary of studies of voice in individuals with Down syndrome (DS).

Table A1 (cont'd). Summary of studies of voice in infants, children, and adolescents with DS.

Source	Participants	Method	Summary of results
Michel & Carney (1964)	n = 8 DS (all M; 8.5–10.5 yrs) n = 42 TD (all M; 7, 8, and 10 yrs)	<i>Acoustic assessment:</i> Determination of speaking f_0 using a phonellograph	DS group did not differ from TD group. DS pitch was normal with respect to age.
Albertini et al. (2010)	n = 48 DS* 27 M ($M_{age} = 9.6$ yrs) 21 F ($M_{age} = 9.8$ yrs) n = 46 TD 28 M ($M_{age} = 9.2$ yrs) 18 F ($M_{age} = 9.4$ yrs)	<i>Acoustic assessment:</i> Analyses with the KayPENTAX Real Time Pitch Model 5121 and Praat software (Boersma & Weenink, 2010)	DS group differed from TD only in the coefficient of variation.
Also in Table A2.	*Adults in Table A2.		See Table A2.
Hollien & Copeland (1965)	n = 9 DS (all F; 10 yrs) n = 36 TD (all F; 7, 8, and 11 yrs)	<i>Acoustic assessment:</i> Determination of speaking f_0 using a phonellograph	DS girls and TD girls had comparable mean speaking f_0 . No significant differences.
Montague & Hollien (1973)	n = 20 DS (10 M, 10 F; 7.8–13.5 yrs) n = 20 TD (10 M, 10 F; 8.0–13.2 yrs)	<i>Perceptual rating:</i> Judgments of presence of voice quality disorders by 16 listeners (8 native listeners and 8 SLP listeners)	DS had significantly higher ratings of breathiness and roughness. Also, DS had higher ratings of nasality, but these varied across participants.
Montague, Hollien, Hollien, & Wold (1978)	n = 20 DS (10 M, 10 F; 7.8–13.5 yrs) n = 20 TD (10 M, 10 F; 8.0–13.2 yrs) NOTE: Participants were the same as those selected in Montague & Hollien (1973).	<i>Perceptual rating:</i> Judgments of vocal pitch by 16 paid undergraduate college listeners	DS had lower pitch ratings as a group (60.2%), but a minority had higher pitch ratings (24.8%); differences in perceived pitch were not explained by f_0 , which was not different between groups.
Moody, Montague, & Bradley (1979)	n = 20 DS n = 20 TD NOTE: Participants were the same as those in the reliability study by Montague and Hollien (1973).	<i>Perceptual rating:</i> Ratings of voice using the Wilson Voice Profile System (Wilson, 1972) by 11 graduate students in communicative disorders	DS had higher ratings of deviations in severity, pitch, tension, and air loss.
Rodger (2009)	n = 22 DS (13 M, 9 F) Range = 10.01–20.33 yrs $M_{age} = 14.36$ yrs n = 52 TD (34 M, 18 F) Range = 1.01–18.67 yrs $M_{age} = 13.97$ yrs n = 8 TD (7 M, 1 F) Range = 10.0–15.0 yrs $Mdn = 12.17$ yrs	<i>Acoustic assessment:</i> Analyses of voice using Praat software (Boersma & Weenink, 2010) <i>Perceptual rating:</i> Ratings of voice using the Vocal Profile Analysis Scheme (Laver, Wirz, Mackenzie, & Hiller, 1991) <i>Other assessment:</i> Questionnaire-based analysis of judgments of voice	DS did not differ from controls in f_0 , jitter, shimmer, or SNR, but DS had higher values of spectral tilt. DS had lower pitch ratings.
Novak (1972)	n = 32 DS 19 M ($M_{age} = 13.3$ yrs) 13 F ($M_{age} = 12.8$ yrs) Range = 7–19 yrs n = 20 AD* (11 M, 9 F; range = 7–20 yrs)	<i>Acoustic assessment:</i> Measures of vocal f_0 using a spectrograph	DS did not differ in f_0 but had reduced voice range and “increased rustle” of voice attributed to squeezing of the larynx and irregularity of vocal fold vibration.
Also in Appendix B.	*AD participants with cognitive delay but not with DS.	See Appendix B for speech sounds findings.	

Note. Studies are arranged in order corresponding to approximate age of participants (youngest first). DS = participants with Down syndrome; F = female; M = male; TD = typically developing participants; AD = atypically developing participants; yrs = years; mos = months; GRBAS = Grade–Roughness–Breathiness–Asthenia–Strain voice rating scale; SLP = speech-language pathologist; f_0 = fundamental frequency.

Appendix A (p. 3 of 4). Summary of studies of voice in individuals with Down syndrome (DS).

Table A2. Summary of studies of voice in infants, children, and adolescents with DS.

Source	Participants	Method	Summary of results
Schlanger & Gottsleben (1957)	$n = 44$ DS (ages not specified) $n = 472$ AD* M_{age} of total group (DS and AD) was 28.9 yrs	<i>Perceptual ratings:</i> Assessments of speech articulation, voice, and stuttering	72% (32 participants) of participants with DS were judged to have a voice disorder.
Also in Appendix B.	*AD individuals with other etiologies of mental retardation; all were residents of a training school.	See Appendix B for speech sounds assessment/findings.	
Moran & Gilbert (1978)	$n = 16$ DS 8 M ($M_{\text{age}} = 38;04$)[yrs;mos] 8 F ($M_{\text{age}} = 41;02$; institutionalized adults) $n = 16$ TD 8 M ($M_{\text{age}} = 37;08$) 8 F ($M_{\text{age}} = 41;02$; adults with no cognitive impairment)	<i>Acoustic assessment:</i> Analysis of speaking f_0 using an oscillograph	DS (both males and females) had higher mean f_0 than did controls.
Wold & Montague (1979)	$n = 51$ DS (16 yrs and older)	<i>Perceptual ratings:</i> Identification of voice qualities by 2 trained listeners	Most DS voices were rated as breathy; pitch was rated as either low or high.
Moran & Gilbert (1982)	$n = 16$ DS (8 M, 8 F; adults)	<i>Acoustic assessment:</i> Analysis of f_0 and other voice features (f_0 perturbation and noise-to-harmonic ratio) using oscillograph and spectrograph <i>Perceptual ratings:</i> Ratings with Wilson Voice Profile (Wilson, 1972) by 17 CSD graduate students	DS had variable patterns across individuals; authors concluded that abnormal voice quality reflected the interaction of several factors. DS had abnormal profiles, with breathiness being the most frequent voice quality disorder.
Moran (1986)	$n = 14$ DS (8 M, 6 F; 20–43 yrs; adults who were institutionalized) $n = 14$ TD (8 M, 6 F; 19–54 yrs; adults with hoarse voices but without cognitive impairment)	<i>Acoustic assessment:</i> Measures of SFF of three prolonged vowels using Kay Visi-pitch and measures of vowel formants using a Voice Identification Series 700 spectrograph <i>Perceptual ratings:</i> Judgments of whether a speaker had DS and ratings of nasality by 16 listeners (8 faculty in special education and 8 faculty in SLP)	DS did not differ in f_0 or vowel formants. DS identified at better than chance and received higher nasality ratings.
Pryce (1994)	$n = 30$ DS (16 M, 14 F) $n = 30$ AD with learning disabilities (15 M, 17 F) $n = 30$ AD with functional dysphonia (8 M, 11 F) $n = 30$ TD with normal voice (15 M, 14 F)	<i>Physiologic assessment:</i> Level of laryngeal EMG needed to initiate phonation.	DS had higher levels (almost 2 times greater) of EMG to initiate phonation.
Lee, Thorpe, & Verhoeven (2009)	$n = 9$ DS (4 M, 5 F) Range = 17.0–29.0 yrs $M_{\text{age}} = 24.7$ yrs $n = 9$ TD (matched for age and sex, speaking Standard British English) $M_{\text{age}} = 23.5$ yrs	<i>Acoustic assessment:</i> Analysis of organic and linguistic pitch ranges, voice compass, and declination; and acoustic analyses of phonation, including maximum phonation time, jitter, and shimmer	DS had (a) normal respiratory capacity, reduced organic pitch range, and reduced linguistic pitch range; (b) intonation patterns with a high f_0 and reduced dynamics; and (c) reduced jitter and normal shimmer.
Albertini et al. (2010)	$n = 30$ DS adults* 17 M ($M_{\text{age}} = 28.7$ yrs) 13 F ($M_{\text{age}} = 23.2$ yrs) $n = 60$ TD adults 30 M ($M_{\text{age}} = 48.7$ yrs) 30 F ($M_{\text{age}} = 44.7$ yrs)	<i>Acoustic assessment:</i> Analyses with the KayPENTAX Real Time Pitch Model 5121 and Praat software (Boersma & Weenink, 2010)	DS had higher mean vocal f_0 with reduced f_0 variation and lower energy.
Also in Table A1.	*Data for children are reported in Table A1.	Same as in Table A1.	See also Table A1.

Appendix A (p. 4 of 4). Summary of studies of voice in individuals with Down syndrome (DS).

Table A2 (cont'd). Summary of studies of voice in infants, children, and adolescents with DS.

Source	Participants	Method	Summary of results
Seifpanahi, Bakhtiar, & Salmalian (2011)	<i>n</i> = 22 DS (14 M, 8 F) Range = 20–28 yrs <i>M</i> _{age} = 25 yrs <i>n</i> = 22 TD adults (matched for age and sex)	<i>Acoustic assessment</i> : Analyses of voice using Dr. Speech 4.3U from Tiger Electronics	DS group had higher <i>f</i> ₀ and lower jitter; there was no difference in MPT and shimmer.

Note. Studies are arranged in order of date of publication (earliest first). SFF = speaking fundamental frequency; EMG = electromyography; MPT = mean phonation time; CSD = communication sciences and disorders.

Appendix B (p. 1 of 6). Summary of studies of speech sound disorders (articulation, phonology, and resonance) along with related oral-motor functions in individuals with DS.

Source	Participants	Method	Summary of results
Legerstee, Bowman, & Fels (1992)	<i>n</i> = 8 DS (4 M, 4 F; 56–66 days old when study began)	<i>Perceptual rating</i> : Longitudinal study of infant reactions to different situations, with vocalizations categorized as melodic (speechlike), vocalic (nonspeechlike), or emotional	DS produced more vocalic (nonspeechlike) sounds and fewer melodic sounds than did TD infants studied previously.
Dodd (1972)	<i>n</i> = 10 DS (5 M, 5 F; infants 9–13 months) <i>n</i> = 10 TD (5 M, 5 F; infants 9–13 months)	<i>Acoustic assessment and transcription</i> : Measures of utterance frequency and duration; counts of phonetic constituents of utterances	DS did not differ from control group on any measure.
Smith & Oller (1981)	<i>n</i> = 10 DS (infants) <i>n</i> = 9 TD (infants)	<i>Transcription</i> : Determination of age of reduplicated babbling, developmental trends for place of consonant articulation, and developmental aspects of vocalic productions	DS, similar to controls, began to produce canonical, reduplicated babble at 8.0–8.5 months. DS had patterns that were highly similar to those in the control group.
Steffens, Oller, Lynch, & Urbano (1992)	<i>n</i> = 13 DS (infants; 4–18 months)* <i>n</i> = 27 TD (infants; 4–18 months)* *Longitudinal study across this age period	<i>Perceptual assessment</i> : Categorization of vocalizations into 4 types: quasivowel, full vowel, marginal syllable, and canonical syllable	DS developmental patterns not significantly different from TD. Large variability noted in both groups.
Lynch, Oller, Steffens, & Buder (1995)	<i>n</i> = 8 DS (infants; 2–12 months)* <i>n</i> = 8 TD (infants) *Longitudinal study from 2 to 12 months of life	<i>Acoustic/perceptual assessment</i> : Judgments by nontrained adults of phrasing in infant vocalizations were made between nonvegetative utterances, temporal utterances, and utterance durations	DS rhythmic units longer in DS, but there were no differences between groups in overall vocal output or in the complexity of the rhythmic units.
Lynch, Oller, Steffens, Levine, Basinger, & Umbel (1995)	<i>n</i> = 13 DS (infants; 4 M, 9 F) <i>n</i> = 17 TD (infants; 17 M, 10 F)	<i>Perceptual judgment</i> : Categorization of vocalizations including syllable type (canonical, marginal, quasis resonant, fully resonant)	DS were delayed by about 2 months in onset of canonical babbling relative to reported onset for TD infants; DS infants also had less stable babbling patterns.
Smith & Stoel-Gammon (1983)	<i>n</i> = 5 DS (2 M, 3 F; longitudinal observations from 3 to 6 yrs of age) <i>n</i> = 4 TD (3 M, 1 F; longitudinal observations from 18 to 36 months of age)	<i>Transcription</i> : Longitudinal observations of singleton stops, consonants, and clusters	DS performance was similar to that of controls, but DS had a considerable delay in sound acquisition.
Bleile & Schwarz (1984)	<i>n</i> = 3 DS 1 M = age 4;06 (yrs;mos) 2 F = ages 3;04 and 3;06	<i>Transcription</i> : Analysis of free-play speech using 3 methods: phonological oppositions, phone acquisition, and phonological processes	DS had developmental delays; the 3 methods provided complementary information on phonological development.

Appendix B (p. 2 of 6). Summary of studies of speech sound disorders (articulation, phonology and resonance) along with related oral-motor functions in individuals with DS.

Source	Participants	Method	Summary of results
Stoel-Gammon (1980)	<i>n</i> = 4 DS* (ages 3;10–6;03) *Comparisons with typical development in literature	<i>Transcription:</i> Analyses of spontaneous speech to determine phonetic inventory, accuracy of target phonemes, and characterization of errors in terms of phonological processes	In DS, correct sound production tended to be limited to particular word positions; DS had phonological patterns similar to those reported for TD.
van Bysterveldt (2009)	<i>n</i> = 77 DS (ages 5;08–4;11)	<i>Assessment battery:</i> Articulation, phonological awareness, letter knowledge, real word decoding	In DS, PCC–R scores ranged from 55.2% to 93.5% (<i>M</i> = 78.2%), and PVC scores ranged from 69.9% to 100% (<i>M</i> = 92.8%). Evidence was seen for both developmental and nondevelopmental speech errors.
van Bysterveldt, Gillon, & Foster-Cohen (2010)	<i>n</i> = 10 DS (5 M, 5 F) Range = 4;04–5;05 <i>M</i> _{age} = 4;11	<i>Assessment battery:</i> A number of receptive/expressive language and phonological awareness tests were used to determine pre-treatment and post-treatment status	In DS, PCC–R scores ranged from 22.4% to 76.1%, and PVC scores ranged from 84.6% to 100%.
Moura et al. (2008)	<i>n</i> = 66 DS (36 M, 30 F) Range = 3–8 yrs <i>M</i> _{age} = 5.8 yrs <i>n</i> = 204 TD (104 M, 100 F; <i>M</i> _{age} = 5.7 yrs NOTE: All participants were speakers of Portuguese.	<i>Acoustic assessment:</i> F1–F2 frequencies for the 5 main Portuguese vowels <i>Perceptual rating:</i> Modified to Portuguese GRBAS rating scale by two expert SLPs.	DS had smaller value of the ratio between F2 for /i/ and F2 for /u/ (termed the <i>DS vocalic anatomical functional ratio</i>); DS also had smaller F1–F2 area. All parameters showed significant differences between the two groups.
Kumin, Council, & Goodman (1994)	<i>n</i> = 60 DS (31 M, 29 F; range = 9 mos–9 yrs)	<i>Transcription:</i> Emergence of phonemes in transcriptions obtained from structured therapy sessions in a play environment	DS had considerable variation in age of emergence of individual sounds; DS had a different order of emergence compared with published norms for TD.
Borghi (1990)	<i>n</i> = 50 DS (25 M, 25 F) Range = 5.0–19.1 yrs <i>M</i> _{age} = 9.2 yrs Participants divided into 3 age categories (5.0–7.11 yrs, 8.0–11.9 yrs, and 12.0–19.1 yrs)	<i>Articulation testing:</i> Fischer–Logemann Test of Articulation Competence (Fischer & Logemann, 1971)	DS had persistent articulation errors noted across the 3 age ranges; 7 phonemes were determined to be the most error prone.
Crosley & Dowling (1989)	<i>n</i> = 22 DS (10 M, 12 F) Range = 6;06–12;07 <i>M</i> _{age} = 9;08	<i>Transcription and coding:</i> Analysis of phonological processes	For DS, sentence length was a primary predictor of cluster reduction and liquid simplification; liquid /r/ was more difficult than liquid /l/.
Crosley & Dowling (1989–1990) Roberts et al. (2005)	<i>n</i> = 22 DS (10 M, 12 F; range = 6;06–12;07) <i>n</i> = 32 DS (all M; range = 4–13 yrs) <i>n</i> = 50 AD* (all M; range = 3–14 yrs) <i>n</i> = 33 TD (range = 2–6 yrs) NOTE: Participants were matched to DS and fragile X groups on developmental age. *Fragile X	<i>Transcription and coding:</i> Analysis of phonological processes <i>Articulation testing:</i> GFTA–2 (Goldman & Fristoe, 2000)	DS had phonological patterns similar to those of younger TD. DS had more consonant errors than either of the other 2 groups.
Dodd (1976)	<i>n</i> = 5 DS, home-reared (ages 6;06–8;05) <i>n</i> = 5 DS, residential (ages 12;04–14;09) <i>n</i> = 10 TD (5 home-reared and 5 residential) <i>n</i> = 10 severely subnormal (5 home-reared and 5 residential)	<i>Transcription:</i> Phonological analyses of oral responses to picture identification (spontaneous and imitative)	DS had more errors than did comparison groups; DS performed better on imitation than on spontaneous naming.

Appendix B (p. 3 of 6). Summary of studies of speech sound disorders (articulation, phonology and resonance) along with related oral–motor functions in individuals with DS.

Source	Participants	Method	Summary of results
Brown-Sweeney & Smith (1997)	<i>n</i> = 8 DS (<i>M</i> _{age} = 7.0 yrs) <i>n</i> = 8 DS (<i>M</i> _{age} = 12.0 yrs) NOTE: 2 TD groups age matched to DS groups.	<i>Acoustic assessment:</i> Measurements of voice onset time, vowel duration, and word duration from oscillographic tracings	DS had greater temporal variability, poorer articulatory accuracy, and slower syllable repetition rate; speech timing and maximum syllable repetition rates were good predictors of single-word accuracy.
Hohoff, Seifert, Ehmer, & Lamprecht-Dinnesen (1998)	<i>n</i> = 10 DS (8 M, 2 F; <i>M</i> _{age} = 7.0 yrs) <i>n</i> = 10 TD (5 M, 5 F; <i>M</i> _{age} = 7.1 yrs)	<i>Acoustic assessment:</i> Spectrographic analyses of the test word <i>tasse</i> (including temporal and spectral features); compared with peripheral factors including angle class, overbite, oral–motor ability, hearing disorder, and logopedics	DS had a longer and more variable duration of the test word and a less sharp production of the fricative /s/. (Acoustic features are not correlated to the peripheral factors under study.)
Dodd & Thompson (2001)	<i>n</i> = 15 DS (12 M, 3 F; range = 5.6–15.8 yrs) <i>n</i> = 15 AD* *Phonological disorder characterized by inconsistent errors	<i>Transcription:</i> 25-Word Inconsistency Test (Burt, Holm, & Dodd, 1999)	DS did not differ in the number of whole words produced inconsistently, but there were differences in the quality of the inconsistent errors.
Rupela & Manjula (2007)	<i>n</i> = 7 DS (3 M, 4 F; range = 11.5–14.5 yrs) <i>n</i> = 7 mental age–matched children with mental retardation but without DS <i>n</i> = 6 TD (3 M, 4 F; range = 4–5 yrs) NOTE: Participants were speakers of Kannada.	<i>Transcription:</i> Analysis of phonotactic patterns in conversational speech	DS had a higher percentage of the occurrence of simpler phonotactic patterns.
Schlanger & Gottsleben (1957)	<i>n</i> = 44 DS (ages not specified) <i>n</i> = 472 controls AD (<i>M</i> _{age} = 30 yrs)	<i>Perceptual assessment:</i> Assessments of speech articulation, voice, and stuttering	95% of DS were judged to have an articulatory disorder.
<i>Also in Table A2.</i>	<i>See Table A2 for details.</i>	<i>See Table A2 for voice assessment/ findings.</i>	
Van Borsel (1988)	<i>n</i> = 5 DS (all F; range = 16;05–19;09) NOTE: Participants were speakers of Dutch.	<i>Transcription:</i> Phonetic and phonological analyses of speech	DS speech errors were highly similar to those reported in young TD.
Timmins, Hardcastle, Wood, & Cleland (2011)	<i>n</i> = 26 DS (15 M, 11 F) Range = 8;3–18;9 <i>M</i> _{age} = 13.4 <i>n</i> = 10 TD matched for cognitive age Range = 3;8–7;1 <i>M</i> _{age} = 5.9	<i>EPG assessment:</i> Articulatory contact for the obstruent /t/ in the word <i>toe</i>	DS differed from TD in type of contact, with most frequent atypical patterns being forward movement, increasing contact, and minimal contact.
Cleland, Wood, Hardcastle, Wishart, & Timmins (2010)	<i>n</i> = 15 DS (12 M, 3 F) Range = 9.0–18.0 yrs <i>M</i> _{age} = 14.3 yrs	<i>Standardized testing:</i> Standardized speech, language, and cognitive assessments	DS had atypical and often unusual errors co-occurring with developmental errors; speech measures were not correlated with language or cognitive measures.
Rosin, Swift, Bless, & Vetter (1988)	<i>n</i> = 10 DS (all M; range = 10.6–17.5 yrs) <i>n</i> = 10 AD (all M; with mental retardation of unknown etiology; range = 12.5–18.7 yrs) <i>n</i> = 10 TD (all M; with chronological age matched to DS; range = 12.2–18.6 yrs) <i>n</i> = 10 TD (all M; with mental age matched to DS; range = 5.1–6.11 yrs)	<i>Transcription and standardized testing:</i> Speech assessments with intelligibility rating, GFTA–2, and oral–motor evaluation. <i>Aerodynamic assessment:</i> Intraoral air pressure for bilabial stop /p/	DS had more articulatory errors and more abnormalities of oral structure than did other groups. DS had higher intraoral air pressures for /p/ in different phonetic contexts than did other groups.

Appendix B (p. 4 of 6). Summary of studies of speech sound disorders (articulation, phonology and resonance) along with related oral-motor functions in individuals with DS.

Source	Participants	Method	Summary of results
McCann & Wrench (2007)	n = 12 DS Range = 10.08–18.75 yrs M _{age} = 15.02 yrs n = 4 TD Range = 5.4–7.1 yrs M _{age} = 4.63 yrs	<i>Acoustic and EPG assessment:</i> Analysis of DDK rate and accuracy	DS and TD had similar DDK rates, but DS group was more inaccurate.
Timmins, Cleland, Rodger, Wishart, Wood, & Hardcastle (2009)	n = 20 DS (11 M, 9 F) Range = 8;0–19;0 M _{age} = 13;01 n = 8 TD (6 M, 2 F) Range = 4;0–8;0 M _{age} = 6;01	<i>Transcription and EPG:</i> Study of the production of the palatal fricative in the phrase “a sheep”	DS had inconsistent production, with more errors observed in EPG than in perceptual judgment.
Novak (1972)	n = 32 DS (19 M, 13 F; range = 7–19 yrs) n = 20 AD controls* (11 M, 9 F; range = 7–20 yrs) n = 10 TD controls** *With cognitive delay but no DS **For X-ray portion of study only	<i>Acoustic assessment:</i> Measures of vowel formant frequencies <i>Pneumographic assessment:</i> Breathing patterns <i>Imaging assessment:</i> X-rays of vocal tract <i>Other assessment:</i> Otolaryngologic exam	DS had overlapping F1/F2 areas for different vowels. DS had shallow breathing, frequently abdominal. DS had altered shape of resonating cavities. DS had rough, overlarge tongue; hypertrophy of tonsils; and small, narrow epipharynx.
Also in Table A1		See Table A1 for voice findings.	
Fourakis, Karlsson, Tilkens, & Shriberg (2010)	n = 8 DS (sex not specified; range = 15–17 yrs) n = 8 fragile X (all M; range = 15–19 yrs) n = 5 TD (sex not specified; age 14 yrs) n = 5 TD (all M; age 16 yrs)	<i>Acoustic assessment:</i> Measures of F1 and F2 in an effort to determine the acoustic correlates of nasopharyngeal resonance, which was judged to characterize the majority of the samples in DS, some of the samples in fragile X, and none of the samples in TD	DS had reduced F2 frequencies for the high vowels /i/ and /u/.
Rolfe, Montague, Tirman, & Vandergrift (1979)	n = 6 DS (5 M, 1 F; noninstitutionalized adults; range = 26–30 yrs) NOTE: Participants were perceived as having hypernasal speech by 2 SLPs	<i>Perceptual ratings:</i> Ratings by 2 groups of listeners who differed in clinical experience	DS had essentially normal ratings of nasality.
Kline & Hutchinson (1980)	n = 20 DS (10 M, 10 F) n = 20 AD controls* (10 M, 10 F) n = 20 TD (10 M, 10 F) NOTE: All groups were ages 15–35 yrs. *AD with idiopathic mental retardation	<i>Acoustic assessment:</i> Measures of nasalance using TONAR II (Fletcher, 1972) <i>Perceptual ratings:</i> Ratings of nasality	DS had larger nasalance values. DS had higher ratings of nasality.
Beckman, Wold, & Montague (1983)	n = 2 DS* (1 M, 1 F; adults) *Both participants had perceived voice disorders.	<i>Acoustic assessment:</i> Analysis of first three formants with computer-generated vocal tract shapes. Measures of sustained vowels, f ₀ , F1–F3, and jitter.	In DS, the pharynx cavity is lengthened, and the oral cavity is shortened.
Moran (1986)	See Tables A1 and A2.	<i>Acoustic assessment:</i> Measures of F1 and F1 for vowels /i/, /u/ and /a/	DS not different from TD in F1:F2 ratio.
Sommers, Reinhart, & Sistrunk (1988)	n = 22 DS* (range = 15;02–22;02) n = 24 DS* (range = 13;0–17;01) *Same participants as those in Sommers, Patterson, and Wildgen (1988)	<i>Coding of articulatory errors:</i> Articulation assessed in spontaneous picture-naming test, imitation test, and a sample of spontaneous, conversational speech	Both groups of DS had patterns of delayed and deviant productions.

Appendix B (p. 5 of 6). Summary of studies of speech sound disorders (articulation, phonology and resonance) along with related oral–motor functions in individuals with DS.

Source	Participants	Method	Summary of results
Sommers, Patterson, & Wildgen (1988)	$n = 24$ DS (10 M, 14 F; range = 15;02–22;02)	<i>Orthographic transcription</i> : Presence of natural phonological processes determined from connected speech, picture naming, and imitation	In DS, patterns of both delayed and disordered phonology were observed.
Hamilton (1993)	$n = 3$ DS (2 M, 1 F; ages 17, 17, and 20 yrs, respectively) $n = 1$ TD (F; adult)	<i>EPG assessment</i> : Analysis of DDK performance	DS had various irregularities in EPG patterns—including excessive contact areas and reduced contact areas, asymmetrical contacts, and prolonged contacts—and in slow DDK rates.
Van Borsel (1996)	$n = 20$ DS (10 M, 10 F) Range = 15;04–28;03 $M_{\text{age}} = 20;10$ $n = 20$ TD (10 M, 10 F) Range = 2;06–3;04 $M_{\text{age}} = 3;00$ NOTE: All participants were speakers of Dutch.	<i>Transcription</i> : Examination of consonant, vowel, and diphthong production to determine sounds in error, error rate, and nature of errors (error type)	DS had patterns similar to those of the TD group; this was interpreted as evidence of developmental delay to account for speech patterns in DS.
Bunn, Simon, Welsh, Watson, & Elliott (2002)	$n = 14$ DS (6 M, 8 F) Range = 22–36 yrs $M_{\text{age}} = 29.2$ yrs $n = 15$ AD* (5 M, 10 F) Range = 21–41 yrs $M_{\text{age}} = 29.1$ yrs *Developmental delays	<i>Transcription</i> : Reading, repeating, and formulating speech from a picture following presentation of word and picture sequences	DS had more memory errors and also had more speech production errors in the repetition and formulation tasks (but not in reading).
Carlstedt, Henningsson, & Dahllöf (2003)	$n = 9$ DS* (6 M, 3 F; $M_{\text{age}} = 5.6$ yrs) $n = 11$ DS** (6 M, 5 F; $M_{\text{age}} = 5.6$ yrs) *PPT treatment group **Control group	<i>Articulation testing and oral exam</i> : Consonants, nasals, and vowels perceptually assessed, questionnaire, and intraoral exam	DS participants round their lips more during spontaneous speech.
Barnes, Roberts Mirrett, Sideris, & Misenheimer (2006)	$n = 34$ DS (all M) Range = 4.3–15.9 yrs $M_{\text{age}} = 7.9$ yrs $n = 59$ AD (all M; fragile X) Range = 2.9–14.0 yrs $M_{\text{age}} = 9.1$ yrs $n = 36$ TD (all M) Range = 2.5–6.6 yrs $M_{\text{age}} = 4.6$ yrs NOTE: TD boys were developmentally matched to DS and FX.	<i>Oral–motor exam</i> : Assessment of structure and function using an adapted version of Robbins and Klee's (1987) Oral Motor Speech Protocol	<i>Structure</i> : Boys with DS had more atypical oral structures than did the 2 comparison groups. <i>Oral and speech function</i> : Boys with DS performed more poorly than did TD boys.
Barnes et al. (2009)	$n = 34$ DS (all M) Range = 4.5–16.0 yrs $M_{\text{age}} = 9.7$ yrs $n = 31$ AD (all M; fragile X and ASD) Range = 5.0–15.4 yrs $M_{\text{age}} = 10.1$ yrs $n = 32$ AD (all M; fragile X only) Range = 3.2–14.5 yrs $M_{\text{age}} = 10.9$ yrs $n = 45$ TD (all M; developmentally matched to other groups) Range = 2.8–7.8 yrs $M_{\text{age}} = 5.0$ yrs	<i>Phonological assessment</i> : Measures of phonological accuracy, phonological process occurrence, and intelligibility determined for connected speech samples	Boys with DS scored lower than did other groups on phonological accuracy and on occurrence of phonological processes. DS had greater delays in all phonological measures.

Appendix B (p. 6 of 6). Summary of studies of speech sound disorders (articulation, phonology and resonance) along with related oral–motor functions in individuals with DS.

Source	Participants	Method	Summary of results
Saz, Simon, Rodriguez, Lleida, & Vaquero (2009)	<i>n</i> = 3 DS (1 M, 2 F) 1 M = age 18 yrs 2 F = age 13 yrs <i>n</i> = 11 AD* (6 M, 5 F; range = 11–21 yrs) *Individuals with cognitive or physical impairments affecting speech NOTE: Compared with a reference corpus of 13- and 14-year-old females.	<i>Acoustic assessment:</i> Analysis of how vowel production varies using LPC formant frequencies, f_0 , tone, intensity, and duration. <i>Perceptual assessment:</i> Judgments of vowel production	No specific conclusions on DS. 2 of 3 DS had substantial vowel errors.
Bunton & Leddy (2011)	<i>n</i> = 2 DS (both M; ages 29 and 26 yrs) <i>n</i> = 2 TD (both M; ages 29 and 26 yrs)	<i>Acoustic and radiographic assessments:</i> Analysis of vowel formant frequencies using LPC; kinematic studies of tongue articulation using X-ray microbeam	DS had smaller acoustic vowel space (F1 and F2), reduced articulatory working space, and slower articulatory movements.

Note. Information on age and sex is included, whenever available. Studies involving children are listed according to approximate age (youngest first). Within age groups (e.g., infants and adults), studies are listed in chronological order of publication. Unless stated otherwise, the participants were speakers of English (or, in the case of infants, had English as the ambient language). PCC–R = Percentage of Consonants Correct—Revised (Shriberg & Kwiatkowski, 1982); PVC = Percentage of Vowels Correct (Shriberg, Austin, Lewis, & McSweeney, 1997); DDK = diadochokinesis; F1 = first formant; F2 = second formant; GFTA–2 = Goldman Frisloe Test of Articulation—Second Edition (Goldman & Fristoe, 2000); EPG = electropalatographic; PPT = palatal plate therapy; ASD = autism spectrum disorder; LPC = least percent correct. Gray shaded cells indicate an instrumental methodology.

Appendix C (p. 1 of 3). Summary of studies of fluency and prosody in individuals with DS.

Source	Participants	Method	Summary of results
Reichle, Siegel, & Rettie (1985)	<i>n</i> = 8 DS (4 M, 4 F) Range = 2.2–3.75 M_{age} = 2.73 yrs	<i>Perceptual ratings:</i> Imitations of adult vocalizations that were systematically varied in pitch, duration, and loudness	No relationship between imitative performance for prosodic features and speech sounds; no particular prosodic feature was more likely to be imitated than another.
Stojanovik (2010)	<i>n</i> = 9 DS Range = 8;03–12;05 (yrs;mos) M_{age} = 9;09 <i>n</i> = 8 TD (MA-matched controls) Range = 4;02–5;07 M_{age} = 5;05 <i>n</i> = 8 TD (age matched to DS; CA-matched controls) Range = 8;00–11;00 M_{age} = 9;08 NOTE: Sex not specified for any participants.	<i>Standardized testing:</i> Assessment of prosody with the computerized battery, PEPS–C	DS had significantly lower scores than did the CA-matched group on all aspects of prosody. DS had significantly lower scores than did the MA group on the production of affect, on the production of pre-final narrow focus, and on all four tasks assessing prosody. DS receptive language abilities were unrelated to prosodic abilities.
Nash & Snowling (2008)	<i>n</i> = 17 DS (7 M, 10 F) Range = 9;05–17;0 M_{age} = 14;0 <i>n</i> = 17 TD* (6 M, 11 F) Range = 5;06–9;05 M_{age} = 7;02 NOTE: Matched pairwise to DS participants for receptive vocabulary age.	<i>Verbal fluency task:</i> Semantic and phonological representations observed in a verbal fluency task.	DS had reduced productivity in both semantic and phonological tasks; this was interpreted to reflect less efficient retrieval strategies. DS produced fewer clusters in phonological task. Reduced productivity in semantic/phonological fluency is a result of impaired processing.

Appendix C (p. 2 of 3). Summary of studies of fluency and prosody in individuals with DS.

Source	Participants	Method	Summary of results
Willcox (1988)	<i>n</i> = 5 DS* (3 M, 2 F; range = 10;10–15;01) <i>n</i> = 5 TD** (all M; range = 2;00–2;08) *Considered nonfluent. **Matched for language.	<i>Perceptual ratings:</i> Analysis of frequency and type of disfluencies	Similarities and differences observed in the disfluency types of the 2 groups. The mean number of nonfluencies for DS was 7.4 (per 100 words) and 3.6 for TD. Results were questionable because of individual differences. Repetitions were most common for both groups. Percentages of prolongations were much lower in the TD group than in the DS group.
Pettinato & Verhoeven (2008)	<i>n</i> = 16 DS (10 M, 6 F; range = 11.00–20.00) <i>n</i> = 12 TD* (range = 4.06–7.00 yrs) *Matched on receptive vocabulary level with sex balance similar to that for DS group.	<i>Perceptual ratings:</i> Examination of the production (using a nonword repetition task) and perception of word stress (using XAB discrimination task)	DS had processing difficulties in both the production and perception of more difficult and later acquired stress patterns as well as weak word-initial syllables.
Van Borsel & Vandermuelen (2008)	<i>n</i> = 76 DS (51 M, 24 F, 1 unknown) Range = 3.8–57.3 yrs <i>M</i> _{age} = 22.8 yrs	<i>Perceptual ratings:</i> Used the Predictive Cluttering Inventory (Daly, 2006), which was administered by 26 SLPs.	78.9% of DS had scores that classified them as clutterers, and 17.1% of DS had scores that classified them as clutterer–stutterers.
Gottleben (1955)	<i>n</i> = 36 DS (23 M, 13 F) Range = 8;11–51;07 <i>M</i> _{age} = 27;03 <i>n</i> = 36 AD* (23 M, 13 F) Range = 9;07–76;05 <i>M</i> _{age} = 28;03 *Individuals with mental retardation but not DS.	<i>Perceptual ratings:</i> Judgments of stuttering by 3 individuals	33% of DS were identified as stutterers.
Schlanger & Gottleben (1957)	<i>n</i> = 44 DS (ages not specified) <i>n</i> = 472 AD* <i>M</i> _{CA} = 28.9 yrs for all participants (DS and AD) *The AD group comprised AD individuals with other etiologies of mental retardation; all were residents of a training school.	<i>Perceptual ratings:</i> Assessments of speech articulation, voice, and stuttering	45% of DS were judged to stutter.
Also in Table A2.			
Rohovsky (1965)	<i>n</i> = 9 DS (2 M, 7 F) Range = 10;07–19;03 <i>M</i> _{age} = 15;10 (institutionalized) <i>n</i> = 18 DS (9 M, 9 F) Range = 9;04–19;07 <i>M</i> _{age} = 14;06 (noninstitutionalized)	<i>Perceptual ratings:</i> Fluency judgments (severity, incidence, and reactions of stuttering) by 10 graduate students enrolled in the study of speech and hearing	Stuttering identified in 35% of the institutionalized group and 19% of the noninstitutionalized group (both high and low verbal groups). A greater incidence of stuttering was found in females than in males.
Preus (1972)	<i>n</i> = 47 DS (21 M, 26 F; age 7+ yrs) NOTE: Participants were part of an Oslo-based day home for individuals with mental deficiencies.	<i>Perceptual ratings and transcription:</i> Analysis of stuttering and cluttering behaviors by 10 judges familiar with the individual with DS, based on a spontaneous speech sample <i>Articulation testing:</i> Articulation test used to screen for articulatory disorders	Stuttering on 5% of words was observed in 34% of individuals with DS, secondary symptoms were observed in 29.8% of individuals with DS, and cluttering was observed in 31.9% of individuals with DS. 52% of individuals with DS were judged to be stutterers. 46.8% showed no signs of cluttering. 10.6% had a pronounced tendency to stutter, and 31.9% were clutterers.

Appendix C (p. 3 of 3). Summary of studies of fluency and prosody in individuals with DS.

Source	Participants	Method	Summary of results
Otto & Yairi (1974)	<p>$n = 19$ DS* (9 M, 10 F) Range = 14;00–31;00 $M_{\text{age}} = 21;00$ $n = 19$ TD Range = 15;00–32;00 $M_{\text{age}} = 22;04$ *Institutionalized NOTE: TD group matched to DS group on sex, age, and race.</p>	<i>Perceptual ratings:</i> Analysis of 7 disfluency categories for samples of spontaneous speech	Individuals with DS were more disfluent on categories that are regarded as most typical of developmental stuttering.
Devenny & Silverman (1990)	<p>$n = 31$ DS (20 M, 11 F) Range = 30.0–57.5 yrs $M_{\text{age}} = 40.0$ yrs (M) $M_{\text{age}} = 41.5$ yrs (F)</p>	<i>Transcription and standardized testing:</i> Analysis of the relationship between speech disfluency and manual lateralization	42% of DS were judged to be stutterers; increased disfluency was associated with increased nonrighthandedness.
Devenny, Silverman, Balgley, Wall, & Sidtis (1990)	<p>$n = 8$ DS (8 M; stutterers) Range = 40.0–39.5 yrs $n = 8$ DS (8 M; fluent) Range = 40.0–39.5 yrs</p>	<p><i>EPG assessment:</i> Verbal and manual motor production tasks at two levels of complexity (simple and complex) Simple: DDK rate and finger tapping Complex: Imitating sentences and pegging a pegboard</p>	Compared with the fluent controls, the individuals who stuttered were faster on the simpler tasks but slower on the more complex tasks.
Ferrier, Bashir, Meryash, Johnston, & Wolff (1991)	<p>$n = 18$ DS ($M_{\text{age}} = 19.55$ yrs) $n = 18$ fragile X ($M_{\text{age}} = 21.63$ yrs) $n = 18$ AD autism ($M_{\text{age}} = 16.68$ yrs) NOTE: All groups had 10 adults and 8 children. Mean ages are for the group as a whole. For children, the mean ages were 9.31 yrs for DS, 9.2 yrs for fragile X, and 9.17 yrs for AD. Sex was not specified for any of the groups.</p>	<i>Transcription and coding:</i> Analysis of conversational roles, conversational skills, and articulatory fluency (among others)	DS had significantly more disfluencies (6.1%) than did the group with autism (1.6%) but was not significantly different from the group with fragile X (4.9%).
Flipsen (1999)	$n = 6$ DS (2 M, 4 F; range = 21;00–39;00)	<i>Perceptual ratings:</i> Determination of intelligibility and segmental accuracy	In DS, prepausal rhythm groups were more intelligible.
Shriberg & Widder (1990)	$n = 8$ DS (part of a larger group of forty 20- to 50-year-old noninstitutionalized adults with mental retardation)	<i>Transcription:</i> Narrow phonetic transcription of recorded speech samples to determine segmental and suprasegmental (prosodic) characteristics	DS had problems with most prosodic variables, including rate, phrasing, stress, and voice quality.
<p><i>Note.</i> Studies are listed in approximate order of age of participants. PEPS-C = Profiling Elements of Prosody for Speech and Communication (Peppé & McCann, 2003); MA = mean age; CA = chronological age.</p>			

Appendix D. Summary of parental surveys on communication problems in Down syndrome (DS).

Kumin (1994)

Method: Questionnaire survey of parents with a child with DS. There were 937 respondents. Included were questions relating to intelligibility, oromotor skills, specific speech skills, and speech skills in conversation.

Results: Among the findings were the following: frequent difficulty being understood (58%), tongue thrusting (28%), weak facial muscles (16%), difficulty with chewing (16%), difficulty with swallowing (14%), difficulty with articulation (80%), difficulty with rate of speech (49%), stuttering (17%), and difficulty with voice (13%). In addition, the following results pertained to questions about speech skills in conversation: difficulties with sequencing (56%), increasing length of words (62%), and sentences in conversation (69%).

Kumin (2006)

Method: Questionnaire survey of parents with a child with DS. There were 1,620 respondents. Included was an estimate of intelligibility based on a 10-point scale (1 = *completely unintelligible*, 10 = *completely intelligible*).

Results: Average intelligibility rating was 4.97; girls had significantly higher ratings than boys; speech intelligibility was inversely correlated with apraxia.

Schieve et al. (2009)

Method: Used data from the National Health Interview Survey, a household survey using in-person interviews. Data were reported for 146 individuals with DS (85M, 63F) divided into three age groups for the age range of 3–17 years.

Results: 15.6% of individuals with DS were reported to be stutterers.

Appendix E (p. 1 of 2). Speech intelligibility.

Source	Participants	Method	Summary of results
Barnes et al. (2009)	See Appendix B.	<i>Phonological assessment:</i> Perceptual and acoustic measures of phonological accuracy and processes	DS scored lower in accuracy and processes and used fewer intelligible words.
van Bysterveldt (2009)	See Appendix B.	<i>Transcription:</i> Determination of percentage of intelligible utterances in narratives and connected speech	DS had mean intelligibility scores of 83.1% for narratives and 80% for connected speech.
Parsons, Iacono, & Rozner (1987)	Children who had tongue-reduction surgery: $n = 18$ DS (9 M, 9 F) Range = 5.08–19.60 yrs $M_{\text{age}} = 11.50$ yrs Comparison group of children who did not have tongue-reduction surgery: $n = 9$ DS (7 M, 2 F) Range = 5.33–18.66 yrs $M_{\text{age}} = 9.50$ yrs	<i>Perceptual transcription and parental questionnaire:</i> Calculation of the ratio of total number of consonant substitutions and omissions, divided by the total number of consonants in words attempted by the participant	Ratio of consonant errors was about 0.40 for both groups, pre- and post-treatment. There was no significant difference in intelligibility across time (i.e., not attributed to surgery or maturity); there was no significant difference in intelligibility between surgery group and nonsurgery group, although parents in both groups rated their children as showing improvement.
Chapman, Sueng, Schwartz, & Kay-Raining Bird (1998)	$n = 47$ DS (29 M, 18 F; range = 5;06–20;06 [yrs;mos]) $n = 47$ TD (22 M, 25 F; range = 2;02–6;01)	<i>Transcription:</i> MLU and total number of words spoken, analyzed by SALT transcription program (Miller & Chapman, 1990)	DS had more utterance attempts and spoke with more word tokens, types, and longer MLU. Omissions were more common in older children with DS. Children with DS had poorer intelligibility.
Chapman, Sueng, Schwartz, & Kay-Raining Bird (2000)	See Chapman et al. (1998), above.	<i>Transcription:</i> MLU analyzed by SALT transcription program (Miller & Chapman, 1990); two models compared language comprehension with language production: Model I (without comprehension) Model II (with comprehension)	For DS, Model II explained 68% of the variability in number of different words, 80% of the variability in MLU, and 32% of the variability in intelligibility.

Appendix E (p. 2 of 2). Speech intelligibility.

Source	Participants	Method	Summary of results
Cleland, Timmins, Wood, Hardcastle, & Wishart (2009)	<p>$n = 6$ DS* (5 M, 1 F) Range = 10;01–18;09 $M_{\text{age}} = 12.74$ yrs *All DS were part of the EPG therapy group. In a larger study, 27 participants were randomly assigned to one of three groups: EPG therapy, speech therapy, or control. Data from only 6 participants with DS are reported. All of these were in the EPG therapy group.</p>	<p><i>Standardized testing, EPG, oral–motor exam:</i> Speech, language, and cognitive tests included EPG, oral–motor exam (Robbins & Klee, 1987), and DEAP phonology test (Dodd, Hua, Crosbie, Holm, & Ozanne, 2002).</p>	<p>Post-treatment, all participants showed qualitative and quantifiable differences in EPG patterns as well as improvements in DEAP PCC.</p>
Dodd & Thompson (2001)	<p>$n = 15$ DS (12 M, 3 F; range = 5;07–15;02) $n = 15$ AD* (12 M, 3 F; range = 3;07–5;05) *Children with phonological disorder</p>	<p><i>Phonological assessment:</i> Perceptual assessments of speaking characteristics and phonological errors, using the 25-Word Inconsistency Test (Burt et al., 1999).</p>	<p>DS were not significantly different from comparison group; trend for DS was to use fewer phonemes. AD group made more errors involving addition or deletion of consonants.</p>
Roberts et al. (2005)	<p>See Appendix B.</p>	<p><i>Transcription:</i> Determination of PCC phonological processes, and proportion of whole-word proximity (see Appendix B)</p>	<p>DS had phonological patterns that were delayed relative to TD controls, but DS also differed in some respects from TD patterns; word shapes in DS were reduced because of omitted syllables, reduced consonant clusters, and deletion of consonant singletons.</p>
Kennedy & Flynn (2003)	<p>$n = 3$ DS (ages 7;02, 8;04, and 8;10; sex not specified)</p>	<p><i>Mainly perceptual:</i> Perceptual assessments and comprehension detection using a phonological awareness–based intervention</p>	<p>DS improved phonological awareness targeted in intervention; overall PCC did not significantly improve following intervention.</p>
Rosin et al. (1988)	<p>See Appendix B.</p>	<p><i>Standardized tests and transcription:</i> SALT transcription program</p>	<p>DS were less intelligible, especially in areas such as PCC, MLU, developmental level, and amount of cueing needed.</p>
Wood, Wishart, Hardcastle, Cleland, & Timmins (2009)	<p>$n = 2$ DS (1 M, 1 F) $M = 14$ yrs $F = 11$ yrs NOTE: Both participants received EPG therapy.</p>	<p><i>Standardized tests:</i> Assessment of cognition (WPPSI-III; Wechsler, 2003), language (BPCS-II [Dunn, Whetton, & Burley, 1997] and CELF-P [Wiig, Secord, & Semel, 1992]), and speech (DEAP; Dodd, Hua, Crosbie, Holm, & Ozanne, 2002).</p>	<p>DS intelligibility increased from 72% to 76% and from 59% to 65% for the female and male participant, respectively; variability decreased for female participant but remains higher than that of TD.</p>
Yoder, Hooshyar, Klee, & Schaffer (1996)	<p>$n = 8$ DS ($M_{\text{age}} = 83$ months) $n = 8$ AD* ($M_{\text{age}} = 44$ months) *Matched to DS group on MLU. There was no DS, but there was a language delay.</p>	<p><i>Perceptual assessment:</i> Intelligibility and length determined with SALT transcription program (Miller & Chapman, 1990)</p>	<p>DS had over 3 times as many multiword, partially intelligible utterances. However, overall, there were no significant differences in intelligibility.</p>
Bunton, Leddy, & Miller (2007)	<p>$n = 5$ DS (5 M; range = 26–39 yrs)</p>	<p><i>Perceptual assessment:</i> Intelligibility test and perceptual scoring by listeners and transcribers</p>	<p>DS had a wide range of intelligibility scores (41%–75%) and made errors especially on cluster–singleton production (word initial and word final), vowels, and place of production for stops and fricatives.</p>

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