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Receptive and expressive prosodic abilities in adults with Down syndrome

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RECEPTIVE AND EXPRESSIVE PROSODIC ABILITIES IN ADULTS WITH DOWN SYNDROME

by
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ABSTRACT

LOGAN ALEXANDRA KINGRY: Receptive and expressive prosodic abilities in adults with DS
(Under the direction of Toshikazu Ikuta and Susan Loveall)

Individuals with Down syndrome, a population that often struggles with communication, present a unique linguistic profile of strengths and weaknesses. Almost no research has examined prosody in adults with DS, despite the important role it plays in effective communication. The present study investigated the prosodic profile of seven adults with Down syndrome (18;07-34;11 years) using the Profiling Elements of Prosody for Speech and Communication (PEPS-C), and compared the group's expressive and receptive prosodic abilities to a group of seven adults with mixed-etiology intellectual and developmental disability (29;02-37;07 years) matched on nonverbal ability. Data analyses showed that the group with Down syndrome had a marginally significant lower score than the group with mixed-etiology intellectual and developmental disability on expressive contrastive stress. The group with Down syndrome also had relative weaknesses in expressive and receptive contrastive stress, expressive affect, and imitation but relative strengths in receptive affect, expressive and receptive turn-end, and expressive boundary. Although these observations mirrored aspects of the linguistic profile of Down syndrome, the results suggest a unique prosodic profile for Down syndrome that is not exclusively determined by their larger linguistic profile.

TABLE OF CONTENTS

SECTION I: INTRODUCTION

Despite the critical role it plays in effective communication, prosody has not been well studied in individuals with Down syndrome (DS), a population that often struggles with communication. Thus, there is a lack of information for researchers and practitioners who are developing intervention and therapy programs for individuals with DS. The purpose of the present study, therefore, was to examine the prosodic abilities of individuals with DS, including the expressive and receptive aspects of prosodic form and function.

Down Syndrome

Down syndrome (DS) is a neurogenetic disorder caused by an error during cell division that results in an extra portion or copy of chromosome 21, hence the frequently used interchangeable term for DS, Trisomy 21 (Jacobs, Baikie, Court Brown, & Strong, 1959; Lejeune, Gautier, & Turpin, 1959; Pangalos et al., 1994). DS can result in both cognitive and physical abnormalities (Stojanovik, 2011) and, notably, is the most common cause of intellectual disability in the United States with an occurrence of approximately 1 in every 691 live births (Parker et al., 2010).

Many individuals with DS present with a phenotypic profile that is unique to DS. Broadly speaking, this profile includes intellectual disability but with nonverbal abilities (i.e., visuo-spatial processing) that are typically stronger than verbal abilities (Abbeduto, Warren, & Conners, 2007; Fidler, Most, & Philofsky, 2009; Martin, Klusek, Estigarribia, & Roberts, 2009; Pettinato & Verhoeven, 2009; Silverman, 2007). Individuals with DS are also noted to have strengths in some aspects of adaptive and social functioning (Esbensen, Bishop, Seltzer, Greenberg, & Taylor, 2010; Fidler et al., 2009). Adaptive functioning strengths include lower rates of maladaptive behaviors (e.g. excessive noise-making, attention seeking, and aggression) and mental and behavioral disorders (e.g. anxiety, phobias, hyperactivity, and hypersensitivity) than peers with other neurodevelopmental disabilities (Abbeduto et al., 2007; Chapman & Hesketh, 2000). Strengths in social functioning include social interaction skills, such as empathy, cooperation, and sharing, that are commensurate with typically developing peers matched on mental-age (Abbeduto et al., 2007; Fidler et al., 2009; Laws & Bishop, 2004).

Linguistically, individuals with DS often have both speech and language delays that impact communication, with more difficulties in expressive versus receptive domains (Abbeduto et al., 2007; Grieco, Pulsifer, Seligsohn, Skotko, & Schwartz, 2015; Martin et al., 2009; Silverman, 2007; Stojanovik, 2011). In terms of speech, individuals with DS often have poor articulation and speech intelligibility, which have been attributed to structural abnormalities (e.g. small oral cavities, high arched palates, and large tongues), hearing loss, and low muscle tone (Barnes, Roberts, Mirrett, Sideris, & Misenheimer, 2006; Chapman, Seung, Schwartz, & Kay-Raining Bird, 2000).

Across the language domains of vocabulary, grammar, and pragmatics, individuals with DS demonstrate unique patterns of strengths and weaknesses. The emergence of early vocabulary milestones (e.g. the acquisition of first words) are often delayed in young children with DS when compared to typically developing children (Martin et al., 2009). However, in later childhood and early adolescence, receptive vocabulary appears consistent with nonverbal mental ability (Abbeduto et al., 2007). In fact, receptive vocabulary is often noted as a *relative* strength in DS, as it is not as severely impacted as other areas of language (Abbeduto et al., 2007; Grieco et al., 2015).

Grammar, clinically referred to as morphosyntax, in contrast to vocabulary, is often an area of significant difficulty for individuals with DS and even distinguishes DS from other neurodevelopmental disorders (Abbeduto et al., 2007). Relative to younger typically developing individuals of similar nonverbal mental age, individuals with DS often display poorer abilities in both syntactic comprehension and production (Martin et al., 2009; Silverman, 2007). This includes difficulties with verbs, nouns, grammatical morphemes, and sentence structure (Grieco et al., 2015). These delays in syntax have been identified via language samples, parent reports, and standardized assessments (Caselli et al., 1998; Cardoso-Martins, Mervis, & Mervis, 1985; Miller, 1995).

Within the pragmatic domain, individuals with DS often display both strengths and weaknesses. Individuals with DS are typically characterized as highly social and affectionate and as having the ability to develop interpersonal relationships with their peers (Abbeduto et al., 2007; Chapman & Hesketh, 2000; Martin et al., 2009). Language pragmatics (i.e. conversational regulation techniques such as conversational turn or code switching), conversational style (i.e. using personality assets such as kindness, humor, and forgiveness), and functional living skills (e.g. self-help skills and knowledge of appropriateness in a situation) are consistent with mental age and are seen as relative strengths compared to other etiologies of intellectual and developmental disabilities (IDD) (Fidler et al., 2009; Grieco et al., 2015; Martin et al., 2009). Individuals with DS also display more positive facial expressions than typically developing peers (Jahromi, Gulsrud, & Kasari, 2008; Kasari & Sigman, 1996). However, some research has found that while individuals with DS demonstrate strengths in pro-social behaviors, they may also use those behaviors as a form of social distraction to escape the responsibility of having to complete a requested task (Pitcairn & Wishart, 1994; Grieco et al., 2015). Individuals with DS have also been documented as displaying disruptive behaviors in social settings (e.g. impulsivity, tantrums, stubbornness) (Dykens & Kasari, 1997; Grieco et al., 2015).

Finally, individuals with DS often demonstrate significant weaknesses in verbal working memory (i.e. phonological memory) (Grieco et al., 2015; Martin et al., 2009; Roberts et al., 2005; Silverman, 2007). This is likely tied to difficulties with working memory more broadly (Conners, Moore, Loveall, & Merrill, 2011), which includes the simultaneous storage and successive processing of information. Poor phonological memory also impacts phonological awareness (e.g. phoneme and syllable segmentation, deletion, and rhyming) which, in turn, negatively affects reading development (e.g. sounding out words) (Grieco et al., 2015; Martin et al., 2009).

Prosody

Prosody is an umbrella term that refers to the rhythm and melody of speech and language, or, in other words, not *what* is said, but *how* it is said. Prosody includes both acoustic and perceptual qualities. Acoustically, prosody consists of changes in fundamental frequency, intensity, and duration during speech. Perceptually, those changes are perceived by listeners as alterations in pitch, loudness, and articulation rate (Peppé, Cleland, Gibbon, O'Hare, & Castilla, 2011). Prosody is used to express emotions and attitudes (affect), highlight the meaning of a word or purpose within an utterance (stress), discriminate grammatical units (boundary), and to indicate if a conversational utterance is meant as a question or a statement (turn-end) (Peppé, 2009; Stojanovik, 2011).

The ability to interpret the prosody of other people's speech and to apply prosody correctly in one's own speech is necessary for effective communication. Furthermore, prosodic deficits can cause breakdowns in communication (Stojanovik, 2011; Peppé, 2009; Zampini et al., 2016). For example, frequent juncture (i.e. pausing) in speech can create disfluency and impair a listener's comprehension of a speaker's message (Peppé, 2009). Although disordered prosody is typically expressive (Peppé, 2009), poor comprehension of another's prosody can also largely impact communication, as well as social interactions. For example, when a speaker uses sarcasm, his/her tone is used to indicate that the message is not to be taken seriously. If a listener does not or cannot perceive or understand this difference in tone, then a sarcastic comment may be misleading and at odds with the intended message (Peppé, 2009).

Prosodic Form and Function

The acoustic components of prosody (i.e. fundamental frequency, intensity, and duration) are often referred to as prosodic form. How high or low a sound is can be represented by fundamental frequency but is subjectively, and more commonly, described as pitch. Fundamental frequency can be calculated to display pitch heights at certain events within an utterance (Moura et al., 2008; Peppé, 2009; Zampini et al., 2016). The term intensity is used to describe how soft or loud a sound is and is subjectively described as loudness. The characteristic aspects of timing in speech (e.g. length, tempo, and rhythm) are referenced as duration.

Fundamental frequency, intensity, and duration combine and interact to establish different grammatical, emotional, and pragmatic functions. The grammatical functions of prosody include the use of stress placement to distinguish word classes (i.e. lexical stress; e.g. “CONtract” vs. “conTRACT”), pauses and the lengthening of final syllables to indicate where segments of utterances and phrases begin and end (i.e. syntactic chunking or boundary; e.g. “chocolate, cookies, and jam” vs. “chocolate cookies and jam”), and rising or falling pitch to indicate whether one is making a statement or asking a question (i.e. turn-end; e.g. “carrots?” vs. “carrots.”). In these cases, prosody is essentially acting as the spoken equivalent of punctuation (Peppé, 2009).

The emotional function of prosody involves distinguishing emotions, feelings, and attitudes (i.e. affect) (Peppé, 2009). This function of prosody is paralinguistic in that it adds circumstantial information to what a speaker says through the use of speech rate, pitch-height, loudness, and pitch-range (e.g. the greater range of pitch, the greater the emotional involvement) (Peppé, 2009). Affect provides the emotional indication of an utterance, revealing the feelings and attitudes of a speaker (e.g. whether an individual likes or dislikes something).

Finally, the pragmatic function of prosody concerns the intention of spoken utterances indicated by accented words or syllables (i.e. contrastive stress or focus). Contrastive stress is the use of prosodic form to emphasize specific information within an utterance to highlight or clarify the focus or purpose of something being said (e.g. “Bob asked for COFFEE” indicates Bob wanted coffee, not tea vs. “BOB asked for coffee” indicates Bob, not John, asked for coffee). This function is crucial to the practical delivery of information as well as to the structure of information given (i.e. new information vs. established information). The pragmatic functions of prosody are in place to create socially acceptable communication and are crucial to the communicative functioning of an individual.

Measuring Prosody

Given its diversity, prosody has been measured in a variety of ways. Certain acoustic aspects of prosody, such as fundamental frequency and vowel duration, are typically measured through computer software (e.g. PRAAT) designed to analyze speech using recordings of imitative, elicited, or spontaneous utterances/phrases (Peppé, 2009; Pettinato & Verhoeven, 2009; Setter, Stojanovik, Ewijk, & Moreland, 2007; Zampini et al., 2016). Perceptually, prosody is often evaluated through subjective evaluations or ratings of individual scorers. One of the most commonly used assessments of prosody is the Profiling Elements of Prosody for Speech and Communication (PEPS-C; Peppé & McCann, 2003), a computerized semi-automated test battery which measures prosodic form and function abilities across both expressive and receptive tasks. The function tasks include turn-end, affect, boundary, and contrastive stress, and the form tasks include discrimination and imitation.

Prosody in Down Syndrome

While many studies have investigated language development in DS, almost no research has examined prosody in this population, despite the critical role it plays in effective communication. What research is available has reported conflicting results, likely impacted by the form or function of prosody assessed and the method of assessment. Additionally, the majority of existing studies focus on the prosodic abilities of children with DS, and almost no research has examined the prosodic abilities of adults with DS (Pettinato & Verhoeven, 2009; Stojanovik, 2011; Zampini et al., 2016). This is problematic, as previous research suggests that individuals with DS often experience changes in language, cognition, and behavior as they age (Chapman & Hesketh, 2000; Grieco et al., 2015; Martin et al., 2009).

Prosodic form. The aspect of prosodic form that has been most consistently evaluated in DS is fundamental frequency and when compared to typical development, both children and adults with DS have been documented as displaying differences in fundamental frequency. Using acoustic analysis, children with DS have been documented as producing a lower fundamental frequency, or overall pitch, than typically developing children matched on chronological age (Moura et al., 2008) and developmental age and vocabulary size (Zampini et al., 2016). However, adults with DS, particularly males, have been found to have a significantly higher average frequency than typically developing peers matched on chronological age during both a storybook activity (Lee, Thorpe, & Verhoeven, 2009) and an imitation task (Albertini et al., 2010). Albertini et al. (2010) also observed that the spectral mean frequency of speech did not change for individuals with DS relative to age and suggested that this could be attributed to difficulties of the upper respiratory function which is known to affect vocal productions.

Other aspects of prosodic form have not been investigated as much in individuals with DS. However, a small literature suggests that individuals with DS may have relative weaknesses in the production and comprehension of prosodic form and the ability to distinguish prosody when no meaning is involved (Albertini et al., 2010; Stojanovik, 2011; Stojanovik & Setter, 2011). Two studies have reported that the prosodic form of individuals with DS varies in frequency, intensity, and/or duration from typically developing individuals based on performance on imitation tasks (Albertini et al., 2010; Stojanovik, 2011). The study conducted by Stojanovik (2011) reported that children with DS scored significantly lower than two groups of typically developing children, matched on nonverbal ability or chronological age, on all prosodic form tasks on the PEPS-C (i.e. short-item discrimination, long-item discrimination, short-item imitation, and long-item imitation). Albertini et al. (2010) found that the intensity (i.e. loudness) of productions on an imitation task were significantly lower in adults with DS than in typically developing adults and the duration of imitations were significantly shorter for males with DS than typically developing males.

Prosodic function. The prosodic functions that have been most investigated in DS are the grammatical functions of turn-end and lexical stress. Individuals with DS demonstrate significant difficulties in expressing lexical stress and turn-end but slightly better skills in perceiving turn-end (Pettinato & Verhoeven, 2009; Stojanovik, 2011; Stojanovik & Setter, 2011; Zampini et al., 2016). For example, children with DS have been documented as performing significantly worse on the receptive and expressive turn-end tasks from the PEPS-C than two groups of typically developing children matched on either nonverbal ability or chronological age (Stojanovik, 2011), as well as a group of children with Williams syndrome (WS) matched on chronological age (Stojanovik & Setter, 2011). However, within the group with DS, children were better at comprehending the rise and fall of utterances than being able to express turn-end themselves. Additionally, Zampini et al. (2016) observed that children with DS had difficulty with ending interrogative utterances with the correct rising intonation. However, the comparison group in these studies included typically developing children matched on chronological age, making it difficult to evaluate the impact of intellectual disability on expressive and receptive grammatical prosodic ability more broadly.

As for lexical stress, children and adolescents with DS have been reported as having particular difficulty producing lexical stress patterns in multisyllabic words with weak-initial syllables (e.g. “maJORity”) when compared to typically developing mental-age matched children (Pettinato & Verhoeven, 2009). This difficulty producing multisyllabic words is indicative of a less developed phonological system and may be linked to poor phonological memory, which is often associated with the DS phenotype (Grieco et al., 2015; Martin et al., 2009; Pettinato & Verhoeven, 2009).

Affect has not been a large focus in the existing literature on prosody in DS, but within the limited research it appears that individuals with DS perform poorly on affective function output tasks (Stojanovik, 2011; Stojanovik & Setter, 2011). For example, Stojanovik and Setter (2011) noted that typically developing children matched on chronological age, typically developing children matched on nonverbal ability, and children with Williams syndrome matched on nonverbal ability all outperformed children with DS on the PEPS-C expressive affect task. However, the children with DS performed similarly to mental age-matched typically developing children and children with Williams syndrome on the receptive affect task, suggesting that receptive affect could be a relative strength for the DS phenotype when compared to nonverbal ability. This is also consistent with the broader linguistic profile of DS, in which receptive skills are typically stronger than expressive skills (Abbeduto et al., 2007; Grieco et al., 2015). These results suggest that individuals with DS may be better at comprehending the rise-fall/fall-rise dichotomy often used to indicate affect/emotion but are unable to express these accurately themselves.

The pragmatic functions of prosody have also been only minimally researched in DS. Research on contrastive stress (i.e. focus) suggests that individuals with DS have significant difficulty both comprehending and emphasizing the most prominent word in an utterance (Stojanovik, 2011; Stojanovik & Setter, 2011).

Current Study

Individuals with DS characteristically struggle with communication; however, they have a unique pattern of strengths and weaknesses within and across language domains. Prosody is essential for effective communication, yet almost no research has examined this skill in adults with DS. Establishing a profile of prosodic strengths and weaknesses for individuals with DS will allow for more effective speech-language intervention and therapy for this population. Prosodic weaknesses can be targeted to augment communicative abilities, while strengths can be honed and leveraged to improve communication. The purpose of this study, therefore, was to examine the prosodic profile of adults with DS. Specifically, we addressed the following research question: How do adults with DS perform on measures of receptive and expressive prosodic form and function when compared to other adults with mixed-etiologies of IDD matched on nonverbal ability? Based on previous research and aspects of the linguistic profile for DS, we hypothesized that adults with DS would perform better on word-level tasks versus phrase level tasks and receptive tasks versus expressive tasks. We also hypothesized participants with DS would demonstrate relative strengths in pragmatic functions (i.e. affect).

SECTION II: METHODS

Design

This study was conducted using a comparative between-subjects design. The between-subjects component was group: participants with Down syndrome versus mixed-etiology IDD matched on nonverbal ability. The dependent variables were expressive and receptive prosodic form and function abilities, measured by the PEPS-C.

Participants

Participants with DS. Seven adults with DS ages 18;07 to 34;11 years (*M* = 25;06; *SD* = 6.02) completed the study. They were recruited through the Down Syndrome Association of South Texas, the University of Alabama's Intellectual Disabilities Participant Registry, and by word of mouth. Inclusion criteria for the group with DS included the primary use of speech to communicate, English as their first language, and the comprehension and attention abilities to complete the tasks. These were confirmed by each participant's primary caregiver before testing.

Participants with IDD. Seven adults with mixed-etiology IDD ages 29;02 to 37;07 (*M* = 33;11; *SD* = 3.15) served as nonverbal ability-matched comparisons. These participants had diagnoses of autism spectrum disorder (*n* = 1), Prader-Willi syndrome (*n* = 1), Fetal Alcohol syndrome (*n* = 1), or an unspecified etiology or cause of mild to moderate intellectual disability (*n* = 4). These participants came from a larger study conducted in the Department of Communication Sciences and Disorders at the University of Mississippi examining the language abilities of individuals with IDD (*n* = 31). These participants were recruited through the North Mississippi Regional Center. As part of the larger study, participants completed the Kaufman Brief Intelligence Test (KBIT-2), Peabody Picture Vocabulary Test (PPVT-4), PEPS-C, and two experimental eye-tracking tasks. Data from the KBIT-2, PPVT-4, and PEPS-C were used for the current study. Inclusion criteria for the group with mixed-etiology IDD in the larger study included the use of speech as their primary method of communication and having the comprehension abilities to complete the tasks. Again, this was confirmed via caregiver report before testing.

Participant matching. Participants were matched on a 1:1 basis using nonverbal standard scores from the KBIT-2. First, nonverbal standard scores were calculated for each participant with DS. Next, a participant with IDD was selected from the larger study to serve as a match for each participant with DS. In order to be considered a "match", the participant with IDD had to be within four points of the target participant with DS on his/her nonverbal standard scores. When there was more than one participant with IDD that could be used for matching, we selected the participant with IDD who was also similar in chronological age, sex, and/or verbal ability.

An independent samples *t*-test was used to confirm that there were no significant differences between groups on nonverbal ability using the nonverbal standard scores from the KBIT-2, $t\ (12) = .24$, $p = 0.82$. This effect was small, Cohen's $d = .13$. Participant demographics and scores on the matching variable are detailed in Table 1.

Table 1

Participant Descriptive Statistics on Key Variables

	DS ***n*** = 7	IDD ***n*** = 7
Age		
Mean	25;06	33;11
SD	6.02	3.15
Min	18;07	29;02
Max	34;11	37;07
Sex		
Male	3	5
Female	4	2
IQ		
Mean	49.14	52.71
SD	8.30	12.69
Min	40	40
Max	63	77
V Ability		
Mean	51.86	58.86
SD	9.74	14.98
Min	40	40
Max	66	81
NV Ability		
Mean	54.43	52.86
SD	13.02	11.89
Min	40	40
Max	79	75
Vocabulary		
Mean	156.43	161.43
SD	14.22	30.03
Min	128	113
Max	168	200

Note: DS = Down syndrome. IDD = Mixed-etiology intellectual and developmental disability. Age = Chronological age. Sex = Sex of participant. IQ = KBIT-2 Intelligence Quotient. V Ability = KBIT-2 verbal standard score. NV Ability = KBIT-2 nonverbal standard score. Vocabulary = PPVT-4 Growth Score Value.

Measures

KBIT-2 (Kaufman & Kaufman, 2004; *30 min*.). The KBIT-2 is a standardized, norm-referenced assessment of both receptive and expressive cognitive abilities. It includes three subtests: *Matrices* (nonverbal), *Verbal Knowledge* (verbal), and *Riddles* (verbal). For the non-verbal subtest, *Matrices,* participants were asked to point to the picture/image that completed or answered a question or matrix. For the two verbal subtests, participants were asked to point to a picture that represented a word or question spoken by the tester *(Verbal Knowledge)* or to answer riddles with appropriate one-word answers *(Riddles)*.

The KBIT-2 provides raw scores for each individual subtest and age-equivalent and standard scores for verbal (via the Verbal Knowledge and Riddles subtests) and nonverbal (via the Matrices subtest) abilities. Standard scores are then combined to produce an overall IQ composite. For the purposes of this study, nonverbal standard scores were used to match participants on nonverbal ability, and verbal standard scores and IQ scores were used to describe the samples. Verbal age-equivalent scores were used to estimate the most appropriate start-point on the PPVT-4 for participants with DS and IDD.

Across all ages on the KBIT-2, internal-consistency ranges from 0.86 to 0.96 and test-retest reliability ranges from 0.88 to 0.93 (Kaufman & Kaufman, 2004). The KBIT-2 correlates with the Wechsler Intelligence Scale for Children, Fourth Edition (WISC-IV; Wechsler, 2003) 0.77 for the full IQ test.

PPVT-4 (Dunn & Dunn, 2007; *20 min*.) The PPVT-4 is a standardized and norm-referenced assessment of receptive vocabulary. The PPVT-4 displays four images on an easel page and asks a participant to point to the image that best represents the proctor's spoken word. The words become increasingly difficult as the test progresses. The PPVT-4 provides raw, standard, and growth score values (GSV; raw scores weighted for item difficulty). For the purposes of this study, GSV were used to describe the sample.

Across all ages, split-half reliability on the PPVT-4 ranges from 0.89 to 0.97 and test-retest reliability ranges from 0.92 to 0.96. The PPVT-4 correlates with the Expressive Vocabulary Test, Second Edition (EVT-2; Williams, 2007) from 0.80 to 0.84 for ages 2-61+ years and with the Clinical Evaluation of Language Fundamentals, Fourth Edition (CELF-4; Semel, Wiig, & Secord, 2003) from 0.67 to 0.79 for ages 5-12 years (Dunn & Dunn, 2007).

PEPS-C (Peppé & McCann, 2003; *1 hour*). The PEPS-C is a computerized, semi-automated test battery used to assess prosody. The PEPS-C is comprised of 14 tasks, examining parallel receptive and expressive abilities at two levels: prosodic function and prosodic form. For the current study, we used 10 of the 14 tasks. Eight of those tasks examined prosodic function. These included the functions of turn-end, affect, focus/contrastive stress, and boundary/syntactic chunking. Each function has both an expressive and receptive task. The expressive and receptive lexical stress or phrase stress tasks were not included in the current study as they were determined to be too difficult for participants. In addition, these tasks were not included in the previous versions of the PEPS-C, so there are no comparable data in the published literature. Two tasks were used to examine prosodic form. These included an expressive imitation task with both long and short items (i.e. phrases and individual words) and a receptive discrimination task with both long and short items. See Table 2 for a detailed description of each task.

Only a small amount of normative data is available on the PEPS-C. However, the PEPS-C diminishes the issues of confounding and subjective transcription found in other prosodic measurement tools as the tasks consist of perceiving or expressing meaning through prosody alone (i.e. responses to each task are single words or short phrases that only contain a target prosodic form or function instead of spontaneous utterances containing other language targets) (Catterall, Howard, Stojanovik, Szczerbinski, & Wells, 2006; Peppé, 2009; Peppé et al., 2011; Setter et al., 2007; Stojanovik, 2011). The PEPS-C is appropriate for ages 4 and older; therefore, the stimulus items are culturally appropriate, basic in nature, and easy to pronounce.

Receptive tasks. For all receptive tasks, including the discrimination task, an auditory stimulus is presented by the computer and is accompanied by two pictures. The participant is asked to choose the image (by pointing) that best corresponds to the auditory stimulus. For example, on the receptive turn-end task two pictures of a woman appear on the screen, one in which she is offering a food item on a plate and one in which she is reading about the food item in a book. The computer presents a word, (e.g. "carrots"), as either an offering with rising intonation (e.g. "carrots?") or a declarative with falling intonation (e.g. "carrots."). The participant then points to the answer they think is correct. The computer automatically records each response as correct or incorrect once the tester clicks the answer the participant chose. Each task included 16 items, and scores are reported as percent correct out of those items.

Expressive tasks. For expressive tasks, participants are asked to produce a word or phrase using the appropriate prosody. For example, on the expressive turn-end task a participant should say "honey?" with rising intonation when the picture displayed on the computer shows a woman offering honey. However, they would say "honey." with falling intonation if the picture displayed a woman reading about honey in a book.

Expressive task scoring. The expressive tasks were automatically recorded by the testing laptop. This allowed for scoring to be completed at a later date and on a separate computer and ensured that the judgement of the scorer was not biased by seeing the stimuli. Each expressive task also consisted of 16 items, and scores reflect the percentage correct out of scorable items. This is due to the possibility of answers being unscorable for expressive tasks. Items were considered unscorable (i.e. scored as a null item) for a number of reasons, including: a critical word was missing, participant gave an unintelligible response, participant used vocabulary words rather than prosody to indicate the image seen on the computer, no response was given, or a prompt interrupted the prosody of the answer. If unscorable answers were included, the percentage correct scores would not reflect the true prosodic abilities of the participants. For the current study, across all participants and expressive tasks, 20 imitation items, 20 affect items, 37 turn-end items, 50 boundary items, and 28 contrastive stress items were scored as null.

Table 2

Description of PEPS-C Tasks

Function tasks *Turn-end*	 In the receptive task, the participant is asked to indicate whether the utterance provided by the computer is a question or a statement (e.g. "honey?" vs. "honey."). In the expressive task, the participant is asked to produce an utterance as a question or a statement based on the image shown on the computer screen (i.e. a picture of a woman offering honey vs. a woman reading about honey in a book). Questions are conveyed with rising intonation and statements are conveyed with falling intonation.
Affect	In the receptive task, the participant is asked to indicate whether the person on the computer likes or dislikes a food by selecting a smiley face or a sad face (e.g. "eggs ☺" vs. "eggs ☹"). In the expressive task, the participant is asked to produce an utterance to indicate whether or not they like the food (e.g. if the participant likes eggs they will say "eggs" as if they really like eggs). Likes are signified by a rise-fall pitch contour and dislikes are signified by a fall-rise pitch contour.
Contrastive Stress	In the receptive task, the participant is asked to indicate which color socks the computer forgot to buy based on the word that was stressed in an utterance (e.g. "I wanted blue and BLACK socks"). In the expressive task, the participant is asked to use stress to correct the computer about what color/animal has the ball in a soccer game (e.g. "NO, the WHITE cow has the ball").
Boundary	In the receptive task, the participant is asked to select the picture on the computer that visually coordinates with how the computer prosodically chunks a phrase (e.g. a picture of a chicken, fingers, and some fruit vs. a picture of chicken fingers and some fruit). In the expressive task, the participant is asked to prosodically chunk a phrase to describe the picture shown on the computer screen (i.e. use boundary to correctly represent the image given).
Form tasks *Discrimination*	 In this receptive task, the participant is asked to indicate if two muffled utterances sound the same or different (e.g. "the bull's eye is red" vs. "the bullseye is red").
Imitation	In this expressive task, the participant is asked to repeat an utterance that the computer says, exactly how the computer says it (i.e. the participant should repeat the word or phrase using the same prosody that the computer did.)

Procedures

For participants with DS, testing was conducted individually in a quiet room at either the participant's home or another location convenient for the participant (e.g. a public library). For participants with IDD, testing was completed at the North Mississippi Regional Center or an associated community home or worksite. At the beginning of each session, the purpose of the study was explained to all participants, as well as the potential benefits and harms of participating. All parents and/or legal guardians of individuals with DS and IDD were required to give written informed consent and all participants were required to provide verbal assent (and written informed consent in the case of adults with DS or IDD who were able to legally sign for themselves) that they were willing to participate in the study.

The first measure administered was the KBIT-2. The tester then used the individual's verbal age-equivalent score from the KBIT-2 as the starting point for the PPVT-4. This was done to reduce testing time (i.e. to avoid having to drop back starting points multiple times to establish the basal), while also beginning at an appropriate level that was not too challenging (i.e. a starting point lower than the level for their chronological age).

Following the KBIT-2 and PPVT-4, the tester administered the PEPS-C. The subtests of the PEPS-C were administered in one of two predetermined, counterbalanced orders across participants. Alternating the order of the subtests also allowed us to control for variables such as possible fatigue, boredom, or hunger and to discern whether performance across PEPS-C tasks was impacted by these extraneous variables. Before beginning each paired task of the PEPS-C (i.e. turn-end, affect, boundary, contrastive stress, and discrimination/imitation), the tester used picture cards to introduce and name the images that were presented in that specific task to the participant to ensure that the participant was familiar with the pictures and terms used as testing stimuli. After reviewing the correct term for each image, the tester then checked for comprehension by asking questions about each card that gave the participant the opportunity to name the image themselves. For each consecutive task, additional vocabulary words were added to the existing terms (e.g. “In this next game, we are going to be talking about food again. Some are the same foods we were just talking about, but some of the foods are new. We’ll start by looking at the new foods.”). If a participant forgot the name of an image during a task, the tester could whisper the label to them without using any voicing so as not to give away any pitch information to the participant that could impact their expression of the task.

After all testing was completed, participants in this study (i.e. adults with DS) were debriefed and given a $10 Amazon gift card for contributing to the research. At the request of the North Mississippi Regional Center, participants with IDD were given a certificate of completion and new video games to share and play in their community recreation area.

Analytic Plan

A series of paired-samples *t*-tests were used to compare the groups on each of the ten PEPS-C tasks. Paired-samples *t*-tests were selected instead of independent samples *t*-tests because the participants were individually matched on nonverbal ability. A few of the dependent variables were not normally distributed (both the expressive and receptive contrastive stress tasks and the discrimination task for the group with DS), so we also examined these results using the nonparametric alternative Wilcoxon Signed Ranks test. The pattern of results did not change, and thus we chose to report the paired-samples *t*-test results below.

To control for family-wise error and the possibility of Type I errors, we utilized a Bonferroni correction (.05/10) and set our alpha at .005 to determine statistical significance. However, given our small sample, we also carefully examined eta squared effect sizes to help in guiding our interpretation of the results.

SECTION III: RESULTS

Preliminary Analyses

Correlations among variables for the two groups of adults can be found in Table 3. Nonverbal ability significantly correlated with the expressive affect and receptive turn end PEPS-C tasks for the group with IDD. The means and standard deviations of the performance of the two groups of adults on the PEPS-C function and form tasks are presented in Table 4.

Table 3

Correlations among key variables for DS above the diagonal and IDD below the diagonal

	1	2	3	4	5	6	7	8	9	10	11	12	13
1.Age	--	.69	.33	.26	-.51	.53	-.50	-.20	-.47	-.42	.26	-.42	.34
2.NV Ability	.46	--	.63	.26	-.12	.33	-.51	-.11	-.02	-.14	.34	-.43	.11
3.Vocabulary	.40	.90*	--	.60	.51	.76*	-.46	-.61	.57	.29	.66	.01	-.03
4.Imitation	-.76*	-.10	-.08	--	.11	.73	.17	-.95*	.44	-.06	.76*	.44	.10
5.E. TurnEnd	-.14	.44	.35	.46	--	.25	-.19	-.20	.73	.51	.28	.05	-.36
6.E. Affect	.25	.86*	.61	.05	.34	--	-.46	-.84*	.35	-.12	.56	-.01	.32
7.E. Boundary	.18	.58	.50	.01	.07	.46	--	-.02	-.08	.27	.20	.78*	-.47
8.E. ContStress	.06	-.41	-.29	-.64	-.54	-.43	-.51	--	-.55	.09	-.61	-.40	-.27
9.Discrimination	-.33	.48	.65	.59	.64	.24	.38	-.51	--	.25	.18	.27	.11
10.R. TurnEnd	-.02	.82*	.74	.44	.80*	.74	.38	-.63	.77*	--	.41	.56	-.78*
11.R. Affect	.30	.61	.79*	.14	.51	.32	-.04	-.28	.63	.68	--	.43	-.50
12.R. Boundary	.10	.64	.70	.13	.82*	.32	.34	-.33	.77*	.78*	.64	--	-.39
13.R. ContStress	-.27	.51	.48	.77*	.59	.57	.25	-.81*	.71	.83*	.57	.39	--

Note: DS = Down syndrome. IDD = Mixed-etiology intellectual and developmental disability. Age = Chronological age. NV Ability = KBIT-2 nonverbal standard score. Vocabulary = PPVT-4 Growth Score Value. Imitation = PEPS-C imitation task. E. TurnEnd = PEPS-C expressive turn-end task. E. Affect = PEPS-C expressive affect task. E. Boundary = PEPS-C expressive boundary task. E. ContStress = PEPS-C expressive contrastive stress task. Discrimination = PEPS-C discrimination task. R. TurnEnd = PEPS-C receptive turn-end task. R. Affect = PEPS-C receptive affect task. R. Boundary = PEPS-C receptive boundary task. R. ContStress = PEPS-C receptive contrastive stress task.

* $p < .05$

Table 4

Scores (number correct/number scorable) on ten PEPS-C function and form tasks

Task	DS *M*	DS *SD*	DS *Range*	IDD *M*	IDD *SD*	IDD *Range*	***p***	***Eta squared***
Function Tasks								
E. TurnEnd	.65	.18	.43-1.0	.55	.32	0-.94	.36	.14
E. Affect	.50	.26	0-.80	.52	.21	.33-.94	.91	.002
E. Boundary	.83	.17	.62-1.0	.75	.19	.50-1.0	.43	.11
E. ContStress	.50	.17	.19-.75	.74	.15	.54-1.0	.04*	.54
R. TurnEnd	.73	.18	.56-1.0	.69	.24	.44-1.0	.67	.03
R. Affect	.81	.13	.63-.94	.72	.14	.44-.88	.15	.31
R. Boundary	.66	.11	.56-.81	.63	.12	.50-.81	.74	.02
R. ContStress	.53	.10	.38-.69	.62	.12	.50-.81	.28	.19
Form Tasks								
Imitation	.74	.13	.56-.97	.84	.07	.75-.97	.14	.32
Discrimination	.62	.15	.50-.81	.59	.18	.44-.94	.60	.05

Note: Imitation = PEPS-C imitation task. E. TurnEnd = PEPS-C expressive turn-end task. E. Affect = PEPS-C expressive affect task. E. Boundary = PEPS-C expressive boundary task. E. ContStress = PEPS-C expressive contrastive stress task. Discrimination = PEPS-C discrimination task. R. TurnEnd = PEPS-C receptive turn-end task. R. Affect = PEPS-C receptive affect task. R. Boundary = PEPS-C receptive boundary task. R. ContStress = PEPS-C receptive contrastive stress task
*$p < .05$

DS vs. IDD

To address our research question, we compared the performance of individuals with DS to individuals with mixed-etiology IDD on measures of expressive and receptive prosody. Examining group means, the group with DS appeared to score higher than the group with mixed-etiology IDD on expressive turn-end, expressive boundary, and receptive affect tasks. In contrast, the group with IDD appeared to score higher on both the expressive and receptive contrastive stress tasks and the imitation task, and groups were very similar on the expressive affect task, the receptive turn-end and boundary tasks, and the discrimination task.

However, paired samples *t*-tests indicated that the only difference to approach statistical significance was on the expressive contrastive stress task, t (6) = 2.64, p = .04, with a large effect, eta squared = .54. The group with IDD had significantly higher scores than the group with DS. However, after the Bonferroni correction to control for Type I errors, this result should be interpreted as marginally significant.

Although not statistically significant, there were several comparisons with large effects in which the group with DS had higher scores than the group with IDD, including expressive turn end (eta squared = .14), expressive boundary (eta squared = .11), and receptive affect (eta squared = .31). There were also comparisons with large effects in which the group with IDD had higher scores, including imitation (eta squared = .32), expressive contrastive stress (eta squared = .54), and receptive contrastive stress (eta squared = .19).

SECTION IV: DISCUSSION

The purpose of the present study was to examine the strengths and weaknesses of the prosodic profile of DS and investigate how the expressive and receptive prosodic abilities of adults with DS compared to those of adults with mixed-etiology IDD matched on nonverbal ability. We examined group performance and differences between the group with DS and the group with IDD on all PEPS-C tasks. Given our small sample size, we considered both the results of paired-samples *t*-tests and effect sizes in interpreting the pattern of results.

Prosodic Function

Across all prosodic functions, the only difference to approach statistical significance after the Bonferroni correction was expressive contrastive stress. Thus, contrastive stress appears to be the area of greatest weakness for the group with DS, most pronounced in the expressive domain, though also observed in the receptive domain. The group with IDD outperformed the group with DS on both contrastive stress tasks, with a marginally significant difference between groups after the Bonferroni correction on the expressive task and a large effect. Although the difference for the receptive task was not significant, there was also a large effect. This finding of a large effect size on both the expressive and receptive tasks coincides with previous research indicating that individuals with DS have significant difficulty perceiving and stressing the most important word in an utterance (Stojanovik, 2011; Stojanovik & Setter, 2011). This weakness observed in contrastive stress may be partially explained by aspects of the linguistic profile for DS. The PEPS-C contrastive stress tasks are phrase-level (vs. word-level) tasks, and, as such, include syntax. Syntax is an area of known difficulty for DS (Martin et al., 2009; Silverman, 2007) and therefore could have contributed to this pattern of results. The contrastive stress tasks may also tax working memory more so than other PEPS-C tasks as they require participants to remember more information for a single item at a time (e.g. the computer says “the blue sheep has the ball” but the participant sees an image of a white sheep and must correct the statement), and as working memory is typically poor in individuals with DS (Grieco et al., 2015; Martin et al., 2009; Roberts et al., 2005; Silverman, 2007), this weakness could be attributed to the complicated activities.

In contrast, receptive affect emerged as an area of relative strength for the group with DS, evident by the large effect size between groups. This is consistent with previous research (Stojanovik, 2011; Stojanovik & Setter, 2011), that has also documented receptive affect as an area of relative strength for individuals with DS and suggests they are able to comprehend the use of prosody to express emotional affect. However, both groups performed poorly on the expressive affect task. In fact, expressive affect was one of the lowest scoring tasks for both groups. This could suggest that individuals with IDD, including DS, struggle to express affect using prosody. Interestingly, both affect tasks are word-level tasks. Given that the use of syntax is limited in these tasks, combined with known social strengths in DS, we may have expected participants with DS to exceed on both affect tasks. However, it is possible that the expressive affect task was particularly challenging in some way. Although the images for the affect tasks were presented and reviewed for comprehension before beginning the tasks, participants may not have been familiar with certain items (e.g. "leeks") and therefore had difficulty appropriately expressing like or dislike for those items.

Turn-end was another area of relative strength, both receptively and expressively for the group with DS. On the expressive task, the group with DS outperformed the group with IDD with a large effect size between groups. On the receptive task, the two groups performed similarly, but the task was one of the highest scoring tasks observed within the group with DS. This finding is consistent with previous research (Stojanovik, 2011; Stojanovik & Setter, 2011) suggesting that individuals with DS are better at comprehending than expressing turn-end. Like affect, the turn-end tasks are word-level tasks, and this relative strength may also be linked to the lack of syntactic expectations within the tasks.

Finally, expressive boundary (i.e. syntactic chunking) was also observed as a relative strength in DS, with a large effect size between groups. In fact, across all form and function tasks, the group with DS had the highest average score on expressive boundary. Interestingly, the boundary tasks are phrase-level tasks and since syntax is an area of difficulty for DS (Grieco et al., 2015; Martin et al., 2009; Silverman, 2007), we would expect this to be a relative weakness. Despite the large effect size, expressive boundary was also one of the highest scoring tasks for the group with IDD. This may indicate that the expressive boundary task is relatively easy; the task includes images that may reduce working memory load and help participants place the boundary in the correct place (e.g. a picture of a chicken, human fingers, and fruit vs. a picture of chicken fingers and fruit). Thus, this relative strength could be partially due to the ease of this task compared to others. Receptive boundary does not appear to be a relative strength or weakness for DS, as the group with IDD scored similarly with only a small effect size between groups.

Prosodic Form

On the prosodic form tasks, imitation appeared to be an area of relative weakness for participants with DS, at least in comparison to other individuals with IDD matched on nonverbal ability. The imitation task includes phrase-level targets, therefore this relative weakness could be partially attributed to the syntactic difficulties within DS. However, despite the large effect between groups, within the group with DS individuals scored relatively high on the imitation task compared to other tasks. Discrimination does not appear to be a relative strength or weakness for DS, as the group with IDD scored similarly with only a small to medium effect size between groups.

Linguistic Profile Considerations

Because the linguistic profile of DS suggests expressive language is poorer than receptive language (Abbeduto et al., 2007; Grieco et al., 2015), we would expect the group with DS to score higher on receptive tasks than expressive tasks. However, while there was a slight trend of better receptive than expressive scores within the group with DS, there was no clear, overall distinction between the two domains. In fact, the group's highest scoring task was an expressive task (i.e. expressive boundary).

Another known linguistic difficulty in DS is poor syntax (Martin et al., 2009; Silverman, 2007). We observed that the group with DS tended to perform better on tasks that targeted word-level versus phrase-level items, but again (as with the receptive/expressive observation), there was no clear separation between word and phrase level tasks. Thus, while we did see some aspects of the linguistic profile of DS represented in the data, our results suggest a unique prosodic profile for DS that is not exclusively driven by their larger linguistic profile.

Limitations and Future Directions

The current study has several limitations that are important to consider. First and foremost is the small sample, which limits our confidence in the generalizability of the results. However, given the medium and large effect sizes observed, future research on the syndrome specific prosodic profile of DS is warranted. Future studies should also consider matching participants on variables other than and/or in addition to nonverbal ability (e.g. verbal ability or IQ) and to compare participants with DS to other types of peers (e.g. typically developing peers matched on mental age). The nature of comparative research in IDD is that by matching on one variable, you are often mismatching on other variables. Hence, more research with different comparison groups matched on additional variables is needed to fully capture the prosodic profile of DS. Lastly, the current study utilized the PEPS-C for measuring expressive and receptive prosody. However, the PEPS-C is not norm-referenced and the different tasks are not necessarily equal in difficulty, syntactic expectation, or demands on the working memory, therefore future research may utilize other measures of prosody that better represent prosodic ability.

Clinical Implications

The results of the current study indicate that clinicians should focus on explicit training in expressive and receptive contrastive stress skills in clients with DS. The prosodic function of contrastive stress reveals the intention behind an individual's utterances and therefore is crucial for effective communication. In addition, clinicians could perhaps leverage certain prosodic areas that are not as impaired (e.g. receptive affect, turn-end, boundary) in order to further improve communication.

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