



Vocalization patterns in young children with Down syndrome: Utilizing the language environment analysis (LENA) to inform behavioral phenotypes

Journal of Intellectual Disabilities

1–18

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DOI: 10.1177/1744629517708091

journals.sagepub.com/home/jid



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Date accepted: 11 April 2017

Abstract

Children with Down syndrome (DS) are at higher risk for both delayed expressive language and poor speech intelligibility. The current study utilized the quantitative automated language environment analysis (LENA) to depict mother and child vocalizations and conversational patterns in the home of 43 children with DS, chronologically aged 24–64 months. Children with DS displayed fewer utterances than typically developing children; however, there was wide variability. Furthermore, children with DS did not show increased vocalization counts across their chronological ages. In contrast to previous findings, this study found that the mothers of children with DS had a reduced number of vocalizations. However, the vocalizations increased with age in comparison to mothers of typically developing children. Implications for targeted interventions that facilitate learning opportunities in bidirectional contexts for children with DS and their parents are discussed, with particular attention to quantify behavioral phenotypes utilizing a novel expressive language assessment tool.

Keywords

behavioral phenotypes, Down syndrome, language development, LENA, vocalizations

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Introduction

Down syndrome (DS) is one of the most common genetic causes of intellectual disability (ID) (Abbeduto and Murphy, 2004; Chapman and Hesketh, 2000). The syndrome results from trisomy 21 and impacts many cognitive and developmental domains, in particular, expressive language development (Cicchetti and Beeghly, 1990; Roberts et al., 2007; Silverman, 2007). Despite considerable variability in the behavioral phenotype of DS, most empirical studies have reported consistent expressive language delays across the life span in comparison to other mentally age-matched individuals (Abbeduto et al., 2007; Chapman, 2006; Vicari, 2006; Yoder and Warren, 2004). This indicates that the group of children with DS share a similar linguistic profile with overall deficits in syntax, speech production, expressive language, and speech intelligibility (Abbeduto et al., 2007; Berglund et al., 2001; Chapman and Hesketh, 2000; Martin et al., 2009). However, many children with DS also show relative strengths, in comparison to their mental age (MA) matches, in the domains of gesture use, imitation, conversational style, and receptive language abilities (Chapman et al., 2012; Næss et al., 2011; Roberts et al., 2007).

While some aspects of language follow a trajectory similar to that of typically developing (TD) children (Abbeduto et al., 2007; Fidler, 2005; Roberts et al., 2007), syntax, meaningful speech, and expressive vocabulary appear to be significantly *less* well-developed in children with DS (Abbeduto and Murphy, 2004; Adamson et al., 2008; Martin et al., 2009). Specifically, research has shown convincing evidence that children with DS exhibit difficulties with syntactic or morphosyntactic processing due to deficits in phonological working memory, short-term auditory memory, and oral-motor issues (Adamson et al., 2015; Laws and Hall, 2014; Silverman, 2007). Given these early phonological impairments, children with DS often face challenges with subsequent and related language and literacy skills such as reading comprehension, vocabulary knowledge, and speech production in their academic trajectories (Chapman, 2006; Laws et al., 2016; Laws and Hall, 2014).

Early onset of language

Despite displaying initial trajectories similar to those of TD children, children with DS's expressive language skills remain significantly delayed (Cuskelly et al., 2016; Fidler, 2005; Polišenská and Kapalková, 2014; Thiemann-Bourque et al., 2014). More specifically, the onset of canonical babbling in infants with DS is only marginally delayed such that these skills develop in similar onset patterns as their same-aged peers (Chapman et al., 2012; Nathani et al., 2003; Thiemann-Bourque et al., 2014). In addition, studies have found that among infants with DS, the emergence of gestures and other nonverbal communication skills appear to follow a similar profile as TD children (Fidler, 2005; Iverson et al., 2003; Mundy et al., 1988; te Kaat-van den Os et al., 2014). While the overall expressive language skills of young children with DS lag when compared to their TD peers, there are periods in their developmental trajectories when their skills appear to follow typical patterns.

Notwithstanding these early communicative strengths, children with DS experience significant challenges in expressive language and auditory processing (Adamson et al., 2015; Laws and Hall, 2014; Mundy et al., 1995; Silverman, 2007; Zampini and D'Odorico, 2009). For instance, when compared to TD infants, infants with DS show more nonspeech-like sounds than speech-like sounds (Fidler, 2005; Lynch et al., 1995; Polišenská and Kapalková, 2014). In addition, young children with DS also show decreased vocal imitation that may alter later development of meaningful speech (Berger and Cunningham, 1983; Fidler, 2005). For instance, in a large-scale

study by Berglund and colleagues (2001), first word comparisons of 900 TD children and 330 children with DS between the ages of one and five were examined using the “Swedish early communicative development inventory words and sentences” (Berglund and Eriksson, 2000). Children with DS showed prominent delays in the development of first words in comparison to the TD group. In addition, children with DS lagged behind the TD group on pragmatics, grammar, and the maximum length of utterance scales (Berglund et al., 2001; Chapman et al., 2012). The study concluded that despite large individual variability in the expressive language skills of children with DS, there is a distinct developmental delay where spoken words appear at similar levels to that of TD children, but this rate of acquisition occurs at a much slower pace (Barrett, 2016; Berglund et al., 2001; Polišenská and Kapalková, 2014).

In order to further our understanding regarding the varied factors involved in language development, Yoder and Warren (2004) studied predictors of later language in 39 toddlers with IDs, 17 of whom had DS. Their longitudinal correlational design utilized language measures, 15-min experimenter–child and parent–child sessions at time 1 and 6 months later at time 2. The results indicated that children with DS displayed less variable receptive and expressive language skills than children without DS. In addition, canonical vocal communication predicted later productive language in non-DS group compared to the DS group (Yoder and Warren, 2004). Taken together, these studies demonstrate the importance of considering multiple environmental factors such as parental or caregiver responses, child-initiated speech, as well as the etiology when mapping the trajectories of children with DS’s language skills.

Parent–child interactions among children with DS

Significant early communication skills develop within the context of bidirectional relationships between the caregiver and a child, resulting in repeated opportunities for meaningful interactions (Berger and Cunningham, 1983; Marder and Cholmáin, 2006). For those children who may face additional developmental and cognitive constraints, as do some children with DS, there may be fewer opportunities to fully engage in interpersonal exchanges (Marder and Cholmáin, 2006; Næss et al., 2011; Zimmerman et al., 2009). For instance, in one of the first parent–child interaction studies utilizing a DS sample (Cardoso-Martins and Mervis, 1985), mothers and their children were assessed during a 45-min free play session. Three groups of TD children were matched to the DS group based on their level of linguistic development, MA, and chronological age (CA). The study results indicated that mothers of children with DS, aged 21–33 months, had more directive interactions compared to the TD group. More specifically, the maternal speech of these pre-linguistic children with DS showed a higher proportion of imperatives sentences and a lower incidence of child-appropriate labels. Other studies that have examined parent–child interactions among children have also concluded that parental or caregiver language provides critical contextual factors for exploring language profiles of children with DS (Abbeduto et al., 2007; Fidler, 2005; Sterling and Warren, 2014).

Several empirical studies have documented the importance of contextual factors in mapping language phenotypes. For instance, Slonims and McConachie (2006) videotaped 23 mother–child interactions when children with DS were 8 and 20 weeks of age to compare how infants’ dyadic interactions affect mothers’ behaviors toward their babies. They also assessed social behaviors, family circumstances, maternal mental health, and infant temperament. Initially when infants with DS were 8 weeks of age, the mothers of DS and TD did not differ in their interactions. However, when infants with DS were 20 weeks of age, their mothers were rated as being more remote and

less sensitive in their interactions. Furthermore, the quality of the interaction between mothers and infants with DS was influenced primarily by the infant's behaviors at both 8 and 20 weeks. In contrast, the quality of the parent-child interaction in the control group was driven by infant behaviors at 8 weeks and external factors such as maternal depression at 20 weeks (Slonims and McConachie, 2006). These results are corroborated by previous studies (see Berger and Cunningham, 1983; Cardoso-Martins and Mervis, 1985; Crawley and Spiker, 1983) that highlight the need for assessing the linguistic feedback from parents and caregivers in order to disentangle both global and functional levels of expressive language delays in children with DS.

Only a few studies have analyzed the environmental content, feedback, and input within the parent-child conversational dyad. For instance, Thiemann-Bourque and colleagues (2014) explored the differences in adult vocalizations and child vocalizations (CVs) and conversational turn taking among nine infants and toddlers with DS, aged 9–54 months, and nine CA-matched TD children, utilizing automated vocal analysis software (e.g. language environment analysis (LENA) technology). The results indicated that children with DS heard approximately 22% fewer adult words per day and were engaged in fewer conversational turns (CTs) in comparison to TD peers. Further, parent vocalizations were reduced, but consistent, across age. Despite a small sample size, these studies highlight the need for assessing the verbal feedback, responsiveness, sensitivity, and reinforcement that parents or caregivers provide in the context of their children's vocalizations; thus, influencing the infant's subsequent acquisition of expressive language skills (e.g. Adamson et al., 2008, 2012; Sandbank and Yoder, 2016).

Transactional model of development

Collectively, the universal linguistic profiles of TD children range from cooing (i.e. vowel-like noises) to babbling (i.e. repeated consonant-vowel combinations) to forming complete sentences (Barrett, 2016; Kuhl, 2004; Tsao et al., 2004). Given the robust literature on typical language development, it is evident that a large schism exists in the understanding of language trajectories of children with DS who often present varied behavioral phenotypes. Furthermore, even fewer theoretical models have been employed to enhance our understanding of the developmental trajectories of this group and, more specifically, environmental influences may contribute to the unique language patterns and outcomes among children with DS (O'Toole et al., 2016; Sandbank and Yoder, 2016; Yoder and Warren, 2004).

Studies conducted with different clinical populations have demonstrated the strengths of incorporating the transactional model of development (Cicchetti and Beeghly, 1990; Hodapp and Zigler, 1990; Sameroff, 2009; Sameroff and Chandler, 1975; Woynaroski et al., 2014; Yoder and Warren, 2004). The transactional model of influence is defined as the bidirectional and interdependent processes that affect the individual's social context over time. This model emphasizes the need to consider *both* parental- and child-level factors (Cicchetti and Beeghly, 1990; Sameroff, 2009; Sameroff and Chandler, 1975). For example, the transactions or interactions seen between a parent and a child occur within the context of ecological settings that are constantly changing and being changed by the participants (Sameroff, 2009; Sameroff and Chandler, 1975). This is best seen in intervention studies where developmental outcomes are a result of multiple sources of influence outside of the parent-child dyad such as siblings, peers, schools, and other settings. For instance, utilizing the transactional model, studies conducted by Yoder and Warren et al., 1998 have shown how intentional prelinguistic communication by children with DS elicits meaningful parental responses and influences later language in children with DS and other developmental

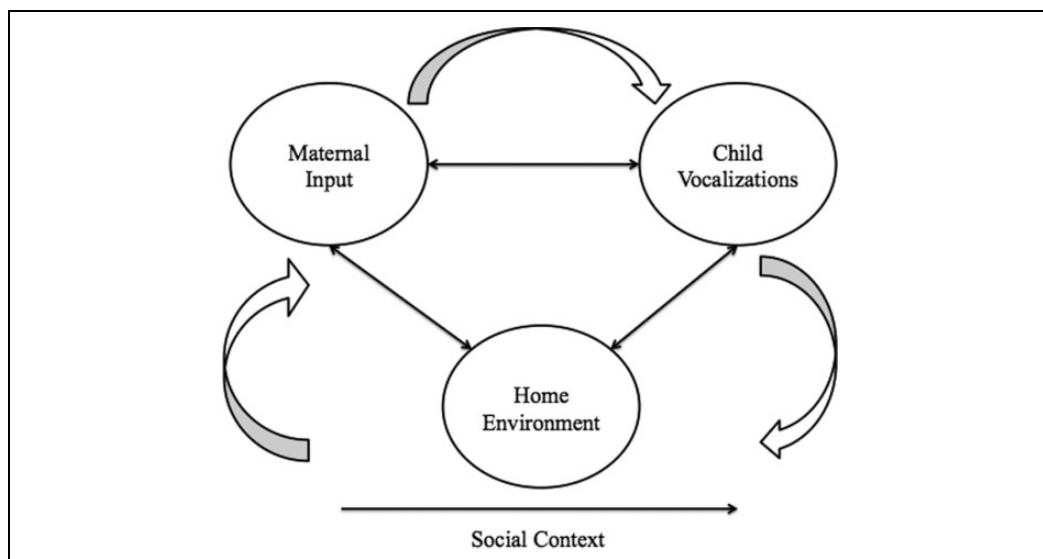


Figure 1. Transactional model of development in children with DS (Sameroff, 2009). DS: Down syndrome.

delays. More specifically, the capacity for language acquisition is influenced by the cumulative environmental factors such as linguistic mapping as well as the caregiving relationships that shape subsequent individual development in the domain of expressive and receptive language (Edgin et al., 2015; Shonkoff and Phillips, 2000; Warren et al., 1993; Yoder and Warren, 1999, 2004). In order to understand the developmental milestones and outcomes among children with DS, multiple sources of influence such as the parental or caregiver linguistic feedback as well as the social context needs to be considered (e.g. Hodapp and Zigler, 1990; O'Toole et al., 2016; Woynaroski et al., 2014). Yet, to date, few studies have reported on the bidirectional influences of the parent–child language interactions utilizing objective measures. The current study utilizes the transactional model of development in order to explicate the maternal language input and the influence on children's expressive language skills (see Figure 1).

The present study

Although there are a robust number of studies focusing on the parent–child communicative interactions, very few of these studies have investigated the relative contingency of parental and child concurrent language development among a young group of children with DS (Adamson et al., 2015; Thiemann-Bourque et al., 2014). Furthermore, even fewer studies have utilized objective tools to capture vocalizations and turn taking patterns outside of human coding and transcriptions. Given the transactional model of interdependent influences, the empirical literature needs more robust objective measures that describe the vocalizations and linguistic trends of children with DS (Sterling and Warren, 2014; Venuti et al., 2012; Yoder and Warren, 2004). The present study aims to highlight the developmental variability of vocalizations in young children with DS at a single time point by utilizing automated software that objectively captures audio input.

Research questions

In order to understand the mother vocalizations and CVs as well as conversational turn taking among a larger sample, the current study assessed toddlers with DS and compared them with CA- and MA-matched TD children who were drawn from a larger sample (see Gilkerson and Richards, 2008). The following questions were examined:

1. What are the differences in mother vocalizations and CVs found within the home environment of CA- and MA-matched TD children and children with DS?
2. Do the mother vocalizations and CVs of children with DS differ across ages?
3. How do the LENA generated CV counts compare with parent-reported measures of expressive language?

We hypothesized that in comparison to CA- and MA-matched TD children, children with DS will have the overall lower rates of mother and CVs, with reduced amounts of conversational turns taking place within the home environment. Furthermore, we hypothesized that the maternal input would remain consistent across age.

Method

Participants

A total of 43 English-speaking families with children (29 males and 14 females) with DS between the CA ages of 24 to 64 months ($M = 39.69$, $SD = 11.17$) and MA ranging from 8 to 36 months ($M = 20.09$, $SD = 7.40$) participated in this study. Participants' MAs were obtained utilizing the LENA software's cognitive developmental profile (*Language Environment Analysis*; LENA Research Foundation, CO) as well as adaptive behavior via standardized developmental assessment of "Vineland Adaptive Behavior Scales-II" (Sparrow et al., 2005). The diagnosis of DS was confirmed by reviewing karyotype reports, and additional medical records were extracted for measures on cardiovascular health, surgeries, and body mass index scores. We compared the current study's dataset to the LENA Research Foundation's Natural Language Study (NLS; 329 TD children aged 2–48 months; Gilkerson and Richards, 2008) in order to provide a comparison for the children with DS as well as to describe the similarities and differences in parental input and CTs in the home environment. Table 1 provides participant information related to MA, CA, gender, race, and education for the families within the DS sample. Additionally, Table 1 includes the available data on the demographics of the TD sample, as obtained from the (Gilkerson and Richards, 2008) study. Chi-square analyses revealed no significant differences between the demographic characteristics and the age-groups of the participants in the current DS sample.

Recruitment

Families were recruited through flyers, advertisements in local support network groups, and newspapers. Exclusion criteria included diagnosis of DS other than trisomy 21 such as mosaicism or translocation, gestational age younger than 36 weeks, and a primary household with a non-English-speaking parent or other spoken languages besides English.

Table 1. Demographics of TD and children with DS and their mothers.

Child characteristic	DS group (N = 43) Mean (standard deviation)	TD group (N = 329) ^a Mean (standard deviation)
Mean CA in months	39.69 (11.17)	18.57 (93.82)
Mean MA in months	20.09 (7.43)	—
CA age range in months	24.05–64.24	2–48
24- to 36-month-olds	n = 20	—
37- to 64-month-olds	n = 23	—
MA age range in months	8–36	—
8- to 24-month-olds	n = 29	—
25- to 36-month-olds	n = 14	—
Gender (males–females)	29:14	167:162
Child's race	(%)	(%)
Hispanic or Latino	7 (16.3)	—
White	32 (74.4)	—
Biracial/multiracial	4 (9.3)	—
Mother's education	(%)	(%)
High school	1 (2.3)	108 (33)
Some college	9 (20.9)	92 (28)
Bachelor's degree	14 (32.6)	84 (25) ^b
Master's degree	11 (25.6)	—
PhD degree	8 (18.6)	—

TD: typically developing; DS: Down syndrome; CA: chronological age; MA: mental age.

^aLENA natural language study (Gilkerson and Richards, 2008).

^bCollege degree of at least a bachelor's degree.

Procedure

The institutional review board at the University approved all study procedures and informed written consent was obtained from parents. Participating families were mailed or hand delivered information including detailed written instructions for using the LENA's digital language processor (DLP), the surrounding hardware, and study questionnaires. Families were asked to complete additional background questionnaires and measures that are not included here due to the current study's goal of focusing on phenotypic behaviors as outputted by innovative technology such as the LENA. The DLP fits in a small pocket of a child-sized vest that is worn by the child during all recordings. The recorder stores 16 h of audio input that are later segmented into sounds from the environment (i.e. TV), an adult male, female, or the focal child. Mothers were instructed to begin recording in their child's natural home environment on a typical day as soon as their child was wakes from sleep and record the audio input for one full day continuously. Upon completion of the study, the families were compensated with gift cards for their time and efforts.

Data analysis: LENA

All vocalizations were evaluated and compiled using DLP that collects 16 h of continuous audio input in the child's home with the data stored for subsequent analysis of speech identification. Automated software analysis and validated processing algorithms separate speech from environmental sounds by identifying the frequency of words and babbles spoken near and by the focal

child. The variables of interest included adult word count (AWC), CVs, and CTs. More specifically, CV were calculated by the speech-related babbles or words initiated by the target child, separated by at least 300 ms of background sound, media sound, other individual's speech, or silence. AWC were the estimated number of utterances or adult words in adult speech segments and CT were identified as a sequence of speech-related sound segments with the first speaker classified as an adult and the second speaker as the focal child (Gilkerson and Richards, 2008; Oller et al., 2010). The reliability and validity of the LENA measures are described extensively in reports elsewhere (see Gilkerson et al., 2008; Xu et al., 2009).

Additionally, to code for meaningful utterances, the three 5-min segments within the 16-h LENA recordings with the highest CV counts were hand coded. Using guidelines suggested by Nathani and Oller (2001), the length of the longest meaningful utterance in the entire 15-min segment was coded, by excluding of repetitive speech, singing, and vegetative sounds. In order to validate the LENA-generated utterances, the hand scored utterances were correlated with the output from LENA and a correlation of $r = 0.62$ ($p < 0.05$) was found (Miller and Chapman, 1993).

Data analysis: Assessment of language

The *MacArthur-Bates Communicative Development Inventory* (MCDI; Fenson et al., 1993), one of the most widely used and validated parental report of language assessment in young children, was administered. This parent questionnaire assesses aspects of expressive language such as vocabulary and grammatical complexity. Part 1 of the scale words and sentences consists of a checklist of 680 words divided into categories typically produced by English-speaking children between the ages of 16 and 30 months. Part 2 of the scale assesses grammar acquisition, whereby parents indicate their child's ability to combine words, provide examples of the three longest sentences, and complete questions related to sentence structures and morphemes the child produces. The present study utilized the total scores reported by the parents on vocabulary production and the number of words used by the child.

Data analysis: Comparisons with TD children

The vocalization data from the current DS sample ($n = 43$) and the available data from the LENA Research Foundation's NLS with TD children ($n = 329$; Gilkerson and Richards, 2008) were used to examine the differences in mother vocalizations and CVs. The NLS with TD children provided full sample mean and standard deviation of the AWC, which was utilized to compare the differences in AWC between the TD and children with DS groups. For the variables of CTs and CVs (CV), the NLS data provided unique means and standard deviations for each month of age (i.e. individual means for 2–48 months). Additionally, using only the data from the current DS sample, AWC and CV were explored to examine the differences across ages. Finally, the LENA-generated CV were compared with parent-reported measures of expressive language.

Results

Differences in parent vocalizations and CVs found within the home environment of CA- and MA-matched TD children and children with DS were examined. Primary correlational analyses of the three LENA variables of AWC, CT, and CV in the DS sample showed significant associations between CV and CT ($p < 0.01$) as well as CT and AWC ($p < 0.01$). There were no statistically

Table 2. Correlations between total CVs, CTs, and AWC for children with DS.

Variables	AWC	CV	CT	DS group Mean	SD	TD group ^a Mean	SD
AWC	1	0.20	0.74 ^b	10061.23	7899.24	12297	6462
CV	0.20	1	0.71 ^b	1457.77	1105.91		
CT	0.74 ^b	0.71 ^b	1	301.33	260.48		

CV: child vocalization; CT: conversational turn; AWC: adult word count; DS: Down syndrome; TD: typically developing.

^aObtained from the LENA natural language study (Gilkerson and Richards, 2008).

^b $p < 0.01$

significant correlations between AWC and CV ($p > 0.05$) in the current DS group. These correlational analyses are provided in Table 2, which also displays the mean and standard deviation of the variables AWC, CT, and CV of the DS sample as well as the mean and standard deviation of the AWC from the NLS TD data.

Research question 1: Differences between mother vocalizations and CVs

In order to examine the differences in mother vocalizations and CVs found within the home environment of CA- and MA-matched TD children and children with DS, Z scores were calculated for the total CV and CT. In comparison to MA-matched TD children, overall children with DS had fewer utterances than the normative sample; however, this group also showed a wide range of variability ($M = -0.22$, range -1.88 to 3.88). Overall, children with DS also had less CT when compared to MA-matched TD children ($M = -0.44$, range -1.49 to 2.93 ; see Gilkerson and Richards, 2008 for unique mean and standard deviation values for CV for each month of age).

Z scores were also calculated for the CA-matched TD children for comparisons of CV and CT. A total of 10 participants were removed from the analysis since their CA was outside the bounds of the normative estimates of the NLS sample. When comparing children with DS to CA-matched TD children, 58% of children with DS had CT that were in the typical range. Additionally, despite a lower overall CV count, 52% (17/33) of children with DS displayed CV in the typical range when compared to CA-matched TD children. Table 3 provides the Z score values calculated for the CV and the CT for the current DS sample when compared to the NLS data of CA-matched TD children (see Gilkerson and Richards, 2008).

Research question 2: Differences in vocalizations in DS across age

The next analysis focused on whether or not the mother vocalizations and CVs of children with DS differed across age. Results showed that in contrast to the normative sample where CV counts increased significantly by age ($r = 0.61$, $p < 0.01$; Gilkerson and Richards, 2008), children with DS did not show increased CV counts across their CA ($r = 0.30$, $p = 0.06$). However, there were increased vocalization counts when examining across MAs ($r = 0.43$, $p < 0.05$). Despite the wide variability demonstrated among the group of children with DS in regard to CV, there was a lack of association observed in overall vocalizations and age.

For AWC, mothers of children with DS had a lower AWC ($M = 10,061$, $SD = 7899.24$) than mother of the TD group ($M = 12,297$, $SD = 6462$; Gilkerson and Richards, 2008). When we split the DS sample by mean CA ($M = 39.69$), each parent input variable differed with age. Specifically,

Table 3. Total CVs and CTs by CA for children with DS ($N = 33$).^a

Age	N	Mean CV	(CV) Z scores	CTC	(CT) Z scores
24	3	917	-1.28	208	-1.08
25		702	-1.44	359	-0.48
26		964	-1.28	65	-1.62
27	2	1807	-0.28	208	-0.98
28		1094	-0.96	148	-1.16
29	3	1835	-0.36	283	-0.78
30		1336	-0.83	118	-1.31
31		895	-1.10	154	-2.00
32	2	1265	-0.67	204	-0.84
35		1048	-0.97	291	-0.75
36		1094	-0.81	172	-1.00
37	4	883	-0.84	218	-0.71
38		713	-1.15	173	-1.21
39		645	-1.16	77	-1.29
41	3	544	-1.33	219	-0.79
42		1878	-0.38	473	0.04
43		2147	-0.26	272	-0.82
46	3	1582	-0.58	429	-0.09
47		1380	-0.86	476	-0.24
48		858	-1.06	207	-0.77

CV: child vocalization; CT: conversational turn; CA: chronological age; DS: Down syndrome.

^aPresent study data compared to TD data from the LENA natural language study (Gilkerson and Richards, 2008).

analysis of variance ANOVA analyses showed a lower AWC in the younger DS group (i.e. CA < 40 months) compared to in the older DS group, $F(1, 41) = 6.46, p = 0.02$. Age-group differences were also found for CT, $F(1, 41) = 13.59, p = 0.00$, with increasing turns across age as indicated by the lower z scores mean for the younger DS group in comparison to the overall mean (CT: $M = -1.02$ vs. $M = -0.61$, respectively).

Research question 3: Differences between LENA and MCDI language scores

Finally, we examined associations between the use of the LENA-generated CVs and the parent-reported measures of CV as captured by the *MacArthur-Bates Communicative Development Inventory* assessments. The analysis presented in this study utilized the total scores reported by the parents on total vocabulary production and how children with DS use words. Table 4 provides the mean and standard deviations for these variables. These results indicated that the LENA-generated CV were significantly associated with parental reports of total vocabulary production in children with DS ($r = 0.62, p < 0.01$). Furthermore, reports on words used by children with DS were also significantly associated with LENA outputs of CV ($r = 0.50, p < 0.01$) as well as CT ($r = 0.55, p < 0.01$). Furthermore, the assessment of vocabulary production and word use were also significantly associated with the CA of children with DS ($r = 0.70, p < 0.01$ and $r = 0.74, p < 0.01$, respectively).

Table 4. Total vocabulary production and word use in children with DS.

Measure	Mean	SD
MCDI total vocabulary production	147.16	178.60
MCDI total word use	2.12	1.89
MCDI % combining words	40.0	—
MCDI average number of words across utterances	2.57	2.77

DS: Down syndrome; MCDI: MacArthur-Bates child development inventory scale.

When the MAs of children with DS were assessed with the total vocabulary production and total word use based on parent-reported MCDI scores, the results showed positively significant associations ($r = 0.85$, $p < 0.01$ and $r = 0.83$, $p < 0.01$, respectively).

In summary, the results indicated that children with DS produced fewer utterances than the CA- and MA-matched TD sample. Additionally, CV counts of children with DS did not increase across their CAs. Children with DS also had less overall CT than their MA-matched TD counterparts. Finally, mothers of children with DS had lower AWC than the TD group; however, there were increasing CT across age in the sample with DS.

Discussion

The current study is among the first few studies to utilize automated naturalistic recordings within the home to describe the quantitative nature of mother vocalizations—CVs as compared to CA- and MA-matched TD children. Given the varied data available on early language trajectories for children with DS, the study aimed to highlight specific group and individual variability of vocalizations of children with DS and the synchronicity of their mothers' vocalizations in a continuous 16-h audio sampling outputted by LENA technology.

CVs across age

Our findings indicate that children with DS decreased CV as a group in comparison to MA-matched TD children, replicating past studies (Berglund et al., 2001; Polišenská and Kapalková, 2014; Sterling and Warren, 2014). More specifically, this study provides further evidence of expressive language deficits that are seen *earlier* in development than previously reported, despite significant individual variability. In addition, the current study also found that the CV counts in children with DS did not increase with age, as expected in TD children, further replicating studies indicating a delayed onset of meaningful speech (Abbeduto et al., 2007; Roberts et al., 2007). A potential explanation for the lack of increase in CV counts across age could be that children with DS may be acquiring language later than TD (Abbeduto et al., 2007; Fidler, 2005; Sterling et al., 2008). More specifically, evidence from the exploration of early developmental precursors provides an understanding of the behavioral phenotypes that emerge early on and tend to remain plateaued in children with DS. For instance, studies such as Harris and colleagues (1997) have found that 39 children with DS between the ages of 12 and 76 months showed overall large variability but significant delays in the acquisition of words. Due to the variability in large word production seen in the children with DS and Williams syndrome, the authors split the sample into children with less than 50 words or greater than 50 words subsamples. Overall, both groups showed equal delays in the onset of language according to normative standards. However, the authors

noted understanding language acquisition in the early stages in children with DS, who initially show an advantage, but as vocabularies increase, this advantage disappears (Harris et al., 1997). Results, such as these, further highlight the importance of teasing apart the variability depicted in expressive language abilities among children with DS in order to provide appropriate qualifying and quantifying treatment outcomes that match the phenotypic profiles of different children with DS.

Presently, it is of interest that more than half of the children with DS in the current study demonstrated vocalizations similar to the number of CVs of the normative group (Gilkerson and Richards, 2008). This is a finding that is not often reported, and demonstrates the relative strengths in some areas of language development in young children with DS. Since children with DS are known to have global delays in expressive language development (Abbeduto et al., 2007; Chapman and Hesketh, 2000; Fidler, 2005), the variability in CV in this study highlights an important factor. More specifically, studies need to control for variability, especially during the early developmental years instead of portraying all children with DS as a homogenous group (Adamson et al., 2008; Roberts et al., 2007).

Adult word counts and CTs

The results in the current study indicated that the group with DS had a lower AWC and fewer CTs in comparison to MA-matched TD children. For AWC, results showed nonsignificant associations between AWC and CV for children with DS in this sample, a finding that suggests the absence of a relation in parent input and CV that is often found in TD children (Marder and Cholmáin, 2006; Mundy et al., 1995). In line with the transactional model of development (Sameroff and Chandler, 1975), the consideration of bidirectional and transactional processes that influence an individual's social context continues to be crucial. This finding may highlight the influence of other child-related mechanisms (medical or neurological) that may be directly influencing CT above and beyond AWC within this sample of DS (Fidler, 2005; Næss et al., 2011; Silverman, 2007). For instance, recent findings have depicted targeted relations between health factors and language in early development in DS (Edgin et al., 2015). Further, issues related to language development and higher-order health factors such as significant hearing loss, small oral cavity, poor facial musculature, hypotonia, respiratory issues, and apraxia among children with DS appear to contribute to the phenotypic profile of expressive language deficits (e.g. Abbeduto and Murphy, 2004; Roberts et al., 2007). Additionally, the impact of ID, short-term memory, and oral-motor issues also significantly contribute to language delays seen among this population (Abbeduto et al., 2007; Fidler, 2005; Martin et al., 2009). Therefore, despite consistent maternal input across the early developmental period, particular factors such as health and motor problems in children with DS may contribute to and influence the reduced amount of CT found in these children in comparison to MA-matched TD children (Haebig et al., 2013; Smith et al., 2014).

The current study also demonstrated significant associations between CTs and CVs, which are important to consider for understanding interaction strategies adopted by parents of children with DS. This contribution of the parental or caregiver language within the home environment can be especially important in understanding the language acquisition and developmental profiles of children with DS (Haebig et al., 2013; Mundy et al., 1995). Studies similar to Adamson and colleagues (2012) involving with children with DS, aged approximately 30.3 months have found that for parents with children with DS, they tended to provide more supportive scaffolding and increased symbol use in order to facilitate language learning. The authors noted that in comparison

to TD children, children with DS initiated less communication with overall lower quality of behavior patterns, resulting in subtle parental contributions, yet in line with TD parental communicative input. Although it is known that parent and caregiver input influences children in terms of acquiring language and learning (Adamson et al., 2012, 2015; Sterling et al., 2008), the lack of significant associations found between AWC and CV in this study highlights the need for going above and beyond observing just maternal input and focusing instead on explicating the child-related factors within the bidirectional context of parent–child interactions. Future studies should examine the extent of how both parent- and child-level factors may moderate the relation between parent input such as the pragmatics of the parental or caregiver language and a child’s vocal production.

The results from the current study also emphasize the importance of considering age and developmental factors when quantifying the expressive language abilities and the behavioral phenotype in children with DS. As stated by Polišenská and Kapalková (2014), consideration should be given to provisions for not only *timely* interventions for children with DS, but also *targeted* interventions that support optimal development across the life span. Given that reduced differences in CV and CT were found in this study among children with DS in comparison to CA- and MA-matched TD children, it is important to recognize which age-groups require additional support. In order to minimize the adverse effects associated with poor speech production, targeted interventions for language development need to be appropriately introduced (Fidler, 2005; Polišenská and Kapalková, 2014). For instance, given that many children with DS displayed CV and CT in the normative range, it is important to tailor interventions and treatments that address other weaknesses and early red flags found in emerging language skills to further enhance communication patterns and interaction strategies in these young children with DS. In addition, by further validating the use of LENA with the parent-reported measure of MCDI, there is initial support for LENA as an innovative, objective assessment for capturing expressive vocalizations within a natural environment (e.g. Thiemann-Bourque et al., 2014; Zimmerman et al., 2009).

Limitations

The current study does have some limitations that might affect the generalization of the results. First, the LENA device is unable to capture the full range of communication, including discriminating between meaningful and nonmeaningful utterances or nonverbal communication (e.g. sign language). Second, no video observations were completed in conjunction with the LENA recordings, which limits the ability to capture facial expressions, eye contact, and other forms of nonverbal interactive styles. Third, 16 h of recordings were done at a single time point, whereas a longitudinal design would provide a more representative picture of the changes across age. Fourth, the study had a relatively small sample size from which it drew profiles of cross-sectional vocalizations in children with DS and their mothers. Fifth, the current sample had a predominantly White racial group as well as mothers with higher educational degrees (44%). The high-level of education may influence generalizability as related to the maternal investment and improvement strategies within the mother–child interactions. Finally, the recruitment strategy of using support groups could bias the sample to those families in access to these network groups and thus affecting the generalizability.

Implications and future directions

Despite these inherent limitations, there are also multiple strengths of the study. The LENA device may contribute to improved quantitative measures of expressive language profiles of children with

DS in an objective and efficient way that exceeds traditional methods of observation and analysis. While this study focused on the automatic coding of the language environment, providing one level of analyses, the LENA is also a very flexible tool for recording and manually coding language, including length of meaningful utterances, a technique used effectively elsewhere (e.g. Edgin et al., 2015). Furthermore, the clinical implications of the study for young children with DS and their language environments include tailoring parent-mediated interventions that use parents as coaches for facilitating early learning opportunities. Understanding both parent and child communication profiles will further our understanding of the nature and bidirectional influence of language development within this population. Additionally, researchers and clinicians can utilize LENA as a clinical tool for mapping treatment outcomes that can objectively classify cross-sectional age differences and life course language trajectories found in children with DS.

Overall, the language data collected from the three automated LENA measures revealed that early in the toddler years, mothers of children with DS may be interacting less often in terms of conversational turn taking with their children in comparison to MA- and CA-matched TD groups. Subsequently, these children may have fewer opportunities to participate in reciprocal social exchanges. It also contrasts normative data showing consistently higher maternal input across age. Therefore, very early in their child's development, mothers of children with DS should be encouraged to provide input that fosters bidirectional interactions and meaningful conversations in order to facilitate increased vocalizations similar to TD children (Roberts et al., 2007; Sterling and Warren, 2014). In addition, given that there are fewer studies that examine language delays in very young children with DS or studies that examine the influences of maternal responsiveness, careful consideration of how to tease apart the individual variability of children with DS is imperative in understanding both global and individual expressive language delays. Finally, the clinical implications for utilizing LENA for depicting behavioral phenotypes, describing developmental profiles, and targeting treatment outcomes early in development may aid in significantly altering and mitigating the course of subsequent speech-related delays as well as facilitate communication milestones in very young children with DS.

Acknowledgments

Thank you to the participants and their families. We would like to thank Dr Jamie Edgin for her invaluable support and collaboration with this project. We would also like to thank the Down syndrome research group for all their work on this project.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: grants from the LuMind Foundation and Research Down Syndrome (to JE) and the Sonoran University Centers for Excellence in Developmental Disabilities HHS #90DD0669, Administration on Intellectual and Developmental Disabilities.

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