<u>Vocal Patterns in Infants with Autism Spectrum Disorder: Canonical Babbling Status and Vocalization Frequency</u>

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Abstract:

Canonical babbling is a critical milestone for speech development and is usually well in place by 10 months. The possibility that infants with autism spectrum disorder (ASD) show late onset of canonical babbling has so far eluded evaluation. Rate of vocalization or "volubility" has also been suggested as possibly aberrant in infants with ASD. We conducted a retrospective video study examining vocalizations of 37 infants at 9–12 and 15–18 months. Twenty-three of the 37 infants were later diagnosed with ASD and indeed produced low rates of canonical babbling and low volubility by comparison with the 14 typically developing infants. The study thus supports suggestions that very early vocal patterns may prove to be a useful component of early screening and diagnosis of ASD.

Keywords: Canonical babbling | Volubility | Vocal patterns | Early detection

Article:

ASD and Early Vocal Development

Early intervention is critical for positive outcomes for children with autism spectrum disorder (ASD). Early identification of atypical behaviors that manifest during infancy could significantly impact age of diagnosis and subsequent initiation of intervention. Currently, the minimum age at which the majority of children with ASD can be reliably diagnosed with relative stability is 2 years (e.g., Chawarska et al. 2009; Lord 1995), but according to recent data from the Centers for Disease Control, many children are not diagnosed until preschool or kindergarten age (2012). Research targeting early detection has primarily focused on behaviors exhibited during toddlerhood (12–36 months) and preschool years (36–60 months) (e.g., Matson et al. 2009;

Volkmar and Chawarska 2008) after diagnosis has been made. Use of retrospective video analyses and studies of infant siblings of children diagnosed with ASD has allowed examination of possible indicators of ASD in the first year of life (e.g., Baranek 1999; Osterling et al. 2002; Zwaigenbaum et al. 2005). Still, the most widely used autism screening tool for young children, the Modified Checklist for Autism Toddlers (MCHAT: Robins et al. 2001) is recommended for ages 16–30 months.

We sought to identify potential communication markers of ASD that might be observed within the first year of life in a retrospective evaluation of data from infants recorded at home and later diagnosed with ASD. We focused on presumed precursors to language for two reasons: First, communication impairment is a core deficit in ASD, and second, evaluation of very early vocal behaviors in typically developing infants has already established markers that are critical to normal vocal communicative development. One robust pre-speech vocal milestone is the onset of canonical babbling. A canonical syllable (e.g., [ba]) is comprised of a consonant-like sound and a vowel-like sound, with a rapid transition between them (Oller 1980, 2000). A second potentially important vocal measure that we considered is volubility, the rate of infant vocalization independent of vocal type (Nathani et al. 2007; Obenchain et al. 1998).

Canonical Babbling as a Key Milestone

In typical development, infants from birth produce vegetative vocalizations (e.g., coughs, burps, etc.) and cry, as well as vowel-like sounds that become more elaborate with time, incorporating supraglottal articulations until canonical syllables emerge, usually by early in the second half-year of life. Robust onset of canonical babbling has been well documented in typically developing infants by not later than 10 months (Koopmans-van Beinum and van der Stelt 1986; Oller 1980; Stark 1980). The impression of robustness has been reinforced by the fact that no delay in onset of canonical babbling has been discerned in infants anticipated to be at-risk for communication deficits due to premature birth or low socioeconomic status (Eilers et al. 1993; Oller et al. 1995). Even infants with Down syndrome usually show normal ages of onset, although a group level delay of a month or more is detectable (Lynch et al. 1995b). Furthermore, infants tracheostomized at birth to provide an artificial airway that prevents or substantially inhibits vocalization for many months tend to produce age-appropriate canonical syllables within a short period after decannulation (Bleile et al. 1993; Locke and Pearson 1990; Ross 1983; Simon et al. 1983).

Only profound hearing impairment and Williams syndrome have been shown to produce consistent substantial delays in the onset of canonical babbling (Kent et al. 1987; Koopmans-van Beinum et al. 1998; Masataka 2001; Oller and Eilers 1988; Stoel-Gammon and Otomo 1986). Further supporting the idea that restricted hearing prevents experiences critical to onset of canonical babbling, age of onset in severely or profoundly hearing impaired infants has been reported to be positively correlated with age of amplification (Eilers and Oller 1994).

In infants without known disorders, onset of canonical babbling after 10 months has been shown to be a significant predictor of language delay or other developmental disabilities (Oller et al. 1999; Stark et al. 1988; Stoel-Gammon 1989). But late onset of canonical babbling is a rare occurrence in infants without easily diagnosed physical or mental limitations. The seeming resistance to derailment of this developmental milestone suggests that canonical babbling is of such importance in human development that it has been evolved to emerge within a relatively tightly constrained time period in spite of substantial variations in home environments and perinatal events. The importance of canonical babbling in predicting later language functioning is assumed to be due to the fact that words are overwhelmingly composed of canonical syllables, and thus lexical learning depends on control of canonical syllables.

To date, only two studies of which we are aware have targeted canonical babbling in ASD and neither specifically examined the onset of canonical babbling, But reasons for optimism that delays in onset of canonical babbling could constitute an early ASD marker can be found in research showing that various aspects of vocalization appear to be disrupted in young children with ASD (Paul et al. 2005; Peppe et al. 2007; Sheinkopf et al. 2000; Warren et al. 2010; Wetherby et al. 2004). Research using automated analysis of all-day recordings based on the automated LENA (Language Environment Analysis) system of classification has shown clear indications that young children with ASD (16–48 months) display low rates of canonical syllable production compared with typically developing infants, even after matching of subgroups for expressive language (Oller et al. 2010). Even more to the point, one recent study has assessed the usage of canonical syllables (though not the onset of canonical babbling) in infants at highrisk for ASD because they were siblings of children with ASD; seven of 24 participants in the study received a provisional diagnosis of ASD at 24 months (Paul et al. 2011). As a group, the at-risk infants (all 24) produced significantly lower mean canonical babbling ratios (canonical syllables divided by all "speech-like" vocalizations, i.e., those deemed "transcribable" by the researchers) compared to low-risk infants at 9-month of age, but there were no significant differences at 12 months. "Non-speech" vocalizations (those deemed "not transcribable" e.g., yells, squeals, growls) were not included in the evaluation of canonical babbling. Other vocal measures—especially number of consonant-like elements and number of speech-like and proportion of non-speech-like vocalizations—also appeared to be potentially useful indicators of emergent ASD.

Volubility in ASD

Volubility, or rate of vocalization, measured in terms of frequency of syllable or utterance production, may be limited in ASD, a possibility that is supported by automated analysis of data showing low volubility in ASD from all-day recordings on children from 16 to 48 months of age based on the LENA system (Warren et al.2010). Volubility in infants with severe or profound hearing loss and in infants with Down syndrome has not been found to be depressed compared with typically developing infants; however, infants from lower socio-economic status (SES) have been shown consistently to produce fewer utterances per minute than their middle or high SES

peers (Eilers et al. 1993; Oller et al. 1995). Research suggests that children in low SES experience less communication from caregivers (Hart and Risley 1995; Snow 1995). The lower volubility of these infants may be a product of decreased social-communication from adults, potentially resulting in lower levels of social motivation in the infants.

Variability in moment-to-moment parental interactivity clearly does affect infant volubility by the middle of the first year of life, as indicated by research on parent-infant interaction in the "still-face" paradigm. The work suggests a strong tendency in the particular case of parent still-face for infants to increase vocalization rate. Specifically, volubility during a baseline period of 1–3 min of face-to-face vocal interaction is substantially lower than during a following still-face period of 1–3 min where the parent withholds any facial or vocal reaction while continuing to look directly at the infant. This pattern is seen in infants after 5 months, but not at 3 months, where volubility does not change at the shift from face-to-face interaction to still-face (Delgado et al. 2002; Goldstein et al. 2009; Yale et al.1999). The results from the still-face paradigm are interpreted to mean that infants seek to re-engage the withdrawn parent during the still-face period, having learned by the middle of the first year that their vocalizations can have impact (Tronick 1982). This effect raises the question of whether infants with emergent ASD similarly increase their volubility to re-engage their caregivers after a period of withdrawn caregiver attention, or whether they decrease volubility, possibly due to diminished motivation to engage socially with others.

Frequency of vocalizations directed at others has been reported to be significantly lower in infants later diagnosed with ASD compared to typically developing infants at 12 months but not at 6 months (Ozonoff et al. 2010). It is also notable that frequency of vocalization based on parent report is predictive of language abilities in toddlers with ASD (Weismer et al. 2010). Paul et al. (2011) assessed frequency of vocalization in infants at high-risk and low-risk for developing ASD and found no difference between groups. However, the study did not actually test for volubility the way volubility is defined here and in much prior research. Frequency of vocalization was tallied in a special way in the Paul et al. study, by counting all speech-like (phonetically transcribable) and nonspeech-like (not phonetically transcribable) vocalizations that occurred within the first 50 speech-like vocalizations of each recorded sample. But not all participants produced 50 speech-like utterances, and in the ones who did, the length of recording required to reach the 50 speech-like utterance criterion was variable. Thus, rate of vocalizations per unit of time was not examined in this study; consequently, given the common usage of the term volubility, it is not possible to determine whether there was a difference in volubility between the groups. In addition, participants in this study were at high-risk for ASD—some were later diagnosed with ASD while some were not. This mixture may have attenuated group differences. It should also be noted that Weismer et al. included only child vocalizations directed at others while Paul et al. included all child vocalizations. Although ASD has roots in social impairments, vocalizations directed at others as well as independent vocal play might well be abnormal in ASD.

A New Study of Early Vocal Development in ASD

One reason the development of pre-speech vocal behaviors in ASD has not been well documented may be that ASD is not reliably diagnosed until long after canonical syllables are expected to emerge, thus making prospective analyses challenging. Retrospective interviews with parents whose children have been diagnosed with ASD regarding age at which canonical syllables emerged may be hindered by poor parent recall, given that parents are generally asked to remember the nature of child babbling that occurred one or more years prior to the time of the interview; also, parents' awareness of the diagnosis may bias their recall of the onset of canonical babbling. The effort by Paul et al. (2011) cited above represents a key advancement in methodology because they assessed infants known to be at-risk in a prospective fashion. Our approach seizes an additional opportunity afforded by the fortuitous existence of home video data from the first year of life that can be analyzed after diagnosis of ASD for comparison with similar video data from infants who did not receive the diagnosis.

As indicated in studies cited above, emergence of canonical syllables is a critical milestone in the development of spoken language, and delayed onset has been shown to be predictive of significant communication impairment. Canonical babbling and volubility have not been well characterized in infants with ASD. To arrive at a better understanding of these two variables as potential indicators of ASD risk in infants, we investigated vocalizations of infants later diagnosed with ASD and typically developing (TD) infants at two age ranges, 9–12 and 15–18 months, using retrospective video analysis methods. Previous research has suggested that nearly all TD infants reach the canonical babbling stage by 9–12 months (Eilers and Oller 1994), and on the assumption that a delay might be present in the children later diagnosed with ASD, we predicted such delay would be observed in this age range. We took the opportunity also to evaluate the available data at 15–18 months because any infant with a failure to show canonical babbling at that age would be greatly delayed in canonical babbling onset and would be considered at very high risk for a variety of disorders.

The coding scheme for this study is based on a widely applied method for laboratory-based evaluation of canonical babbling (Oller 2000). In accord with this method, infants are assumed to be in the canonical stage if they show a canonical babbling ratio (canonical syllables divided by all syllables) of at least .15, a value based on coding by trained listeners of a recording. A value of .15 or greater from such laboratory coding has been empirically determined in prior research as corresponding to parent judgments that infants are in the canonical stage (Lewedag 1995). It has been reasoned that parent judgments constitute the most appropriate standard for establishing this criterion value (Oller 2000). This reasoning is based on three points: (1) Parents respond to interview questions by providing very consistent and accurate information about canonical babbling in their infants (Papoušek 1994; Oller et al. 2001); (2) this parental capability is predictable, given that recognizing canonical babbling represents nothing more than being able to recognize syllables as being well-formed enough that they could form parts of words in real speech (and of course normal adults can easily recognize vocalizations of humans as speech or

non-speech); and (3) parents appear to intuitively understand that the onset of canonical babbling is an emergent foundation for speech, as evidenced by the fact that they initiate intuitive lexical teaching as soon as they begin to recognize canonical babbling in their infants (Papoušek 1994). Consistent parent recognition of the onset of canonical babbling runs in parallel with recognition of other developmental milestones (e.g., sitting unsupported, crawling, walking). In our study we could not use parents as informants about the age of onset of canonical babbling since that onset had occurred a very long time before our first contact with them. Consequently, the canonical babbling ratio, determined from recordings coded in our laboratory, provided the best available measure upon which to base inference about whether infants had reached the canonical stage.

In the present study the following hypotheses were tested:

- 1. Infants later diagnosed with ASD will be less likely than TD infants to be in the canonical stage at each age (9–12 and 15–18 months), as determined by whether their canonical babbling ratios exceed the .15 criterion.
- 2. Infants later diagnosed with ASD will demonstrate significantly lower canonical babbling ratios (independent of the canonical stage criterion) compared to TD infants.
- **3.** Infants later diagnosed with ASD will demonstrate significantly fewer total vocalizations (lower volubility) at both age ranges compared to TD infants.
- **4.** A combined analysis using both volubility and canonical babbling status will significantly predict group membership.

Method

Participants

A total of 37 participants were included in the present study, 23 individuals later diagnosed with ASD and 14 individuals in the TD group (Table 1. There was one set of fraternal twins in the ASD group. Participants were drawn from a larger study conducted at the University of North Carolina-Chapel Hill based on availability of video recordings; participants must have had two 5-min edited video segments at 9–12 months and at least one edited video segment at 15–18 months. As part of the larger study, participants were recruited from the Midwest and Southeast over a 15-year time period. Recruitment criteria included: (1) child age between 2 and 7 years at the time of recruitment; (2) available home videotapes of the child between birth and

2 years of age that parents were willing to share; and (3) enough video footage for at least one 5—min codable segment (see video editing section below) of the child at either 9–12 or 15–18 months of age.

Table 1. Participant demographics

	ASD ; n = 23	TD ; n = 14
Age at 9–12 months; mean (SD)	10.89 (1.39)	10.63 (.53)
Age at 15–18 months; mean (SD)	16.33 (.83)	16.28 (.70)
Sex	19 Males, 4 females	11 Males, 3 females
Race	23 White, 1 Black	13 White, 1 Asian
Maternal education ^a	5.48	5.8 ^b
Childhood Autism Rating Scale; mean (SD)	34.17 (1.52)	16.15 (.39) ^c

^aMaternal education: 1 = 6th grade or lower; 2 = 7th to 9th grade; 3 = partial high school; 4 = high school graduate/GED; 5 = associate of arts/associate of science or technical training or partial college training; 6 = bachelor of arts/science; 7 = master of arts/science or doctorate or other professional degree completed ^bMissing information for two participants ^cMissing information for four participants

All participants included in the ASD group received a clinical diagnosis of ASD from a licensed psychologist and/or physician at a point after the recordings were made. Thus, our design is a retrospective analysis similar to others that have used home movies of children later diagnosed with ASD (Baranek 1999; Werner et al. 2000). A trained research staff member validated diagnoses for each participant using criteria from the Diagnostic and Statistical Manual IV (American Psychiatric Association 2000) and from one or more ASD screening and diagnostic tools, including: the Childhood Autism Rating Scale (CARS; Schopler et al. 1992), the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 1999), and/or the Autism Diagnostic Interview-Revised (ADI-R; Rutter et al. 2003). All participants had CARS scores and each participant in the ASD group had ADI/ADI-R scores and 13 of the 23 ASD participants had ADOS scores.

Typically developing group membership was based in part on scores within normal limits (i.e., not more than one standard deviation below the mean) on the Mullen Scales of Early Learning (Mullen 1995) and/or the Vineland Adaptive Behavior Scales (VABS; Sparrow et al. 1984). An additional exclusionary criterion for any participants in the TD group was any history of learning or developmental difficulties per parent report. Individuals with significant physical, visual or hearing impairments or known genetic conditions (e.g., Fragile X or Rett's Syndrome) associated with ASD were excluded. As indicated in Table 1, mean age (in months) was very similar across groups, gender was balanced, and the two groups were also similar with regard to SES based on maternal education. Our families were mostly middle SES with access to videotaping equipment.

The University of North Carolina-Chapel Hill Institutional Review Board approved the study, and all families signed informed consents. For more information regarding recruitment and inclusion criteria see Baranek (1999).

Video Editing Procedures

Families provided home videos of their child from birth to 2 years as available. The videotapes included footage from a variety of contexts including family play situations, vacations, outings, special events, and familiar routines (e.g., mealtimes), with individual variation in situational content of each family's videotapes as would be expected in home videotapes. All videotapes were copied, transformed to digital formats, and originals were returned to participating families.

Video editing guidelines first focused on the identification of video footage during which the child was consistently visible and for which the parents felt they could accurately identify the child's age. The two age ranges were originally selected for another study on early behavior in ASD (Baranek 1999). At the same time, the two age ranges are well-suited to our current purposes. The 9–12 months age range is the earliest age range in which parents had sufficient videotape footage for it to be useful in our research and represents the time period when a number of communicative behaviors emerge. Further, this is a time frame during which the vast majority of TD children would be expected to already be in the canonical babbling stage. The 15–18 months range provided follow-up on the same children with the expectation that monitored behaviors would be more consistent and would allow for confirmation or clarification of data from the earlier age. In TD children, canonical babbling is usually well consolidated by the 15–18 months age range (Vihman 1996; Oller 2000).

In editing tapes for the larger study, the aim was to compile two 5-min video segments for each child in the 9–12 age range, and two 5-min segments in the 15–18 months age range. On average, each 5-min segment consisted of 5 scenes. Research assistants who were blind to the research questions and not informed of the diagnostic status of the participants edited the videotapes and coded each scene for the following content variables: (a) number of people present; (b) amount of physical restriction on child's freedom to move, rated as low, medium, or high; (c) the amount of social intrusion another person was using to engage the child in interaction, rated as low, medium, or high; (d) and the types of events (e.g., meal time, bath time, active play, special events) (Baranek 1999). The assistants were instructed to quasi-randomly select a cross-section of scenes from the available footage in the designated age ranges, purposely including scenes from each 1-month age interval for which video footage was available within each age range, provided that the child was visible in each selected scene. All participants included in the current study had two 5-min compilations (i.e., 10 min total) for the 9–12 months age range, but at the 15–18 months age range, there were three TD infants and one infant with ASD for whom only a single 5-min segment was assembled due to insufficient video footage. As a result, the mean duration of samples at the 15–18 months age range was 9.5 min rather than 10.

Although vocalization from the infants was common in these scenes, the segments were not specifically selected to capture vocal behavior. Therefore, volubility estimated from the present study may be lower than in prior works where infants have been observed in settings designed to maximize vocal interaction. Similarly, the video segment selection procedure may yield differences in canonical babbling from prior studies. In most studies, 20–30 min of vocal interaction have been recorded, whereas here we had less than half that amount of data per sample. Our procedure can be predicted to produce greater variability in canonical babbling ratios than in studies with longer sampling periods (Molemans 2011; Molemans et al.2011). Additionally, the audio–video quality of these home movies was not as good as would be expected in laboratory studies, another factor that could reduce perceived canonical babbling and volubility.

To ensure that the contexts in which children were recorded were comparable, specific content parameters were identified and compared (Tables 2, 3). No differences were found between the groups on any content parameter including: number of people present, level of physical restriction (i.e., amount of physical confinement such as a highchair versus free play; rated as low, medium or high), amount of social intrusion (rated as low, medium or high), and the total number of event types (e.g., meal time, active play). The number of times each event type (e.g., bath time, playtime) was represented in the ASD group versus the TD group for each age was compared using Chi square analyses. Results for the omnibus Chi square test failed to reach significance in the 9–12 months age group (p > 0.05), but did reach significance in the 15–18 months age group (p = 0.046). Typically developing children were more likely to be engaged in passive activities at the 15–18 months age range (p = 0.046; TD = 16.6 %, ASD = 4.6 %) according to follow-up analysis of the six event categories. See Tables 4 and 5 for the percentage in each category. For a comprehensive description of the coding procedures that yielded the data on situational context see Watson et al. (2013).

Table 2. Content variables for videos, 9–12 months

	ASD; mean (SD)	TD; mean (SD)
Number of people present	3.22 (1.53)	3.28 (1.24)
Amount of physical restriction ^a	1.58 (.35)	1.51 (.32)
Amount of social intrusion ^a	2.02 (.38)	2.04 (.32)
Total number of different event types	5.32 (1.05)	5.07 (1.02)

^aRated by coders on a 1–3 scale

Table 3. Content variables for videos, 15–18 months

	ASD; mean (SD)	TD; mean (SD)
Number of people present	2.84 (1.20)	2.82 (1.24)
Amount of physical restriction ^a	1.37 (.29)	1.28 (.33)
Amount of social intrusion ^a	2.06 (.40)	1.95 (.34)
Total number of different event types	5.34 (1.17)	5.23 (1.11)

^aRated by coders on a 1–3 scale

Table 4. Percentage of each activity type, 9–12 months videos

	ASD; $n = 23 (\%)$	TD ; $n = 14 (\%)$
Mealtime	10	11
Active	53.9	60.6
Bathtime	4.5	5.5
Other	2.5	4.1
Special activity	20.3	16.5
Passive activity	8.7	3.4

Table 5. Percentage of each activity type, 15–18 months videos

	ASD; $n = 23 (\%)$	TD ; $n = 14 (\%)$
Mealtime	7.5	2.6
Active	64	72.8
Bathtime	2.5	1.8
Other	8.3	11.4
Special activity	12.9	4.4
Passive activity	4.6	16.6

Coding Procedure and Observer Agreement

The videotapes analyzed in this study were coded for infant production of all syllables in speech-like vocalizations by two certified speech-language pathologists who were not informed of the diagnostic group of the infants. The intent was, of course, for the coders to be blind to diagnostic category, and with the exception of one infant to be discussed below, the coders reported they saw no reason to suspect any infant of having ASD.

We defined speech-like vocalizations (as in the primary literature on canonical babbling) to include both canonical and precanonical infant vocalizations (regardless of whether they would be deemed "transcribable"). Training of the two coders was provided by the last author, who originated the definition of "canonical syllable" used in this study, and who has conducted and collaborated on numerous studies on onset of canonical babbling, rate of canonical babbling, and volubility in infants (Cobo-Lewis et al. 1996; Lynch et al. 1995b; Oller and Eilers 1982, 1988; Oller et al. 2001, 1995, 1998). The two observers were trained in identifying canonical syllables and in counting all syllables independent of their canonical status. The video samples used during training were separate (although drawn from similar materials based on the home recordings) and not included in the analyses for this investigation.

Syllables were defined as rhythmic units of speech-like vocalization, excluding raspberries, effort "grunt" sounds (i.e., a schwa-like sounds produced as an artifact of physical exertion), ingressive sounds, sneezes, hiccups, crying and laughing. Within an "utterance", which was

defined as a vocal breath group (Lynch et al.1995a), it was possible to identify syllables as corresponding to sonority peaks (high points of pitch and/or amplitude) that are intuitively recognized by mature listeners. These rhythmic events occur in time frames typical of syllables in real speech (usually with durations of 200–400 ms). A canonical syllable is defined as including a vowel-like nucleus, at least one margin (or consonant-like sound) and a transition between margin and nucleus that is rapid and uninterrupted. In general, transitions that are too fast to be tracked auditorily (too fast to be heard "as transitions") are instead heard as gestalt syllables. Auditory tracking of these transitions focuses on formant (acoustic energy) transitions that can be measured on spectrograms as typically <120 ms (Oller 2000). Formants are audible bands of energy corresponding to resonant frequencies of the vocal tract that change as the tract changes shape or size. Audible formant transitions occur, then, when the vocal tract moves during opening from a consonantal closure into a vowel or vice versa.

Examples of canonical utterances (which must include at least one canonical syllable) are syllables that a listener might perceive as ba, taka, or gaga. Vocalizations produced while mouthing objects (e.g., toys or fingers) or eating were excluded from our analyses on the grounds that we could not be sure what role movement of the hands may have played in the apparent syllabification.

Videos were randomized and randomly distributed across the two coders with regard to diagnostic group. The 37 participants' videos were randomly split between the coders by participant and included both age ranges. The coders independently watched the videos, counting both syllables and canonical syllables in real time. This procedure is utilized regularly in the laboratories of the last author in accord with reasoning presented in recent papers, especially Ramsdell et al. (2012). This naturalistic listening approach mimics how a mother would hear her child, listening to each utterance only once.

The measure of canonical babbling ratio used here (number of canonical syllables divided by number of all syllables) is the measure utilized in the bulk of research on onset of canonical babbling to date. However, some studies have used a different ratio (number of canonical syllables divided by number of utterances). The former procedure is generally preferred nowadays because the resulting value can be interpreted as a proportion with values varying from 0 to 1, whereas the latter procedure yields a ratio with no effective upper limit (Oller 2000).

In a coder agreement test, both observers independently coded twenty samples consisting of two 5-min segments of ten participants' video footage. A research assistant unaware of the study goals selected these test samples, and they represented both diagnostic groups and both ages. Reliability was gauged in accord with the degree to which coders agreed upon canonical syllables, total syllables, and whether the child was in the canonical babbling stage (i.e., had a canonical babbling ratio >0.15, the standard criterion). Inter-rater agreement ranged from good to excellent for canonical syllables (ICC = .98, CI 95 .96–.99) and for total syllables (ICC = .87, CI 95.61–.95). Reliability for canonical babbling ratios was also good (ICC = .89, CI 95.69–.96),

with agreement on the canonical stage criterion at 95 % for the twenty samples. Additionally, the coders differed by an average of only 10 % of the total range of canonical babbling ratios obtained, and the correlation across the ratios for the twenty samples for the two coders was .89. For volubility, the coders differed by an average of 13 % of the total range for volubility values, and the correlation across the twenty sample videos for the two coders was .91.

Results

Analyses were performed to confirm that the groups were matched on demographic variables. These analyses did not reveal significant differences between groups on any variable (see Table 1).

Initial descriptive statistics for within- and between-group variables revealed two outliers in the ASD group. Both cases produced very high canonical babbling ratios in the 9–12 months range (.93 and .64) relative to the mean for both groups (ASD = .12 for the 23 cases, TD = .17) (see Fig. 1). Based on prior research, the canonical babbling ratios observed for these two ASD cases were substantially higher than would be expected in TD infants in the 9–12 months age range—infants grouped as having English or Spanish at home, as high or low SES, and as born at term or prematurely all showed mean canonical babbling ratios under .4 from 8 to 12 months of age (Oller et al. 1997, Oller 1994). Analysis of z-scores revealed that infant 22 was 3.96 standard deviations above the mean for the present sample, and infant 23 was 2.73 standard deviations above the mean, further suggesting outlier status. On this basis we decided to eliminate these two cases in the primary analyses on canonical babbling; the remaining 35 cases (21 ASD, 14 TD) were analyzed to address our research questions regarding canonical babbling (see Figs. 1, 2 for canonical babbling ratios by participant at both ages, with the two outliers indicated). However, there were no significant outliers with regard to volubility, and thus we included data from all 37 cases for that analysis (see Figs. 3, 4 for syllable volubility by participant at both ages).

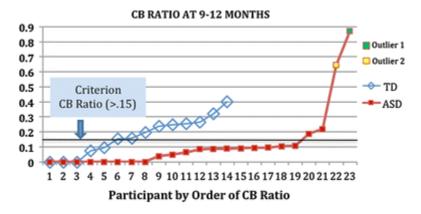


Fig. 1 Canonical babbling ratios by participant at 9–12 months

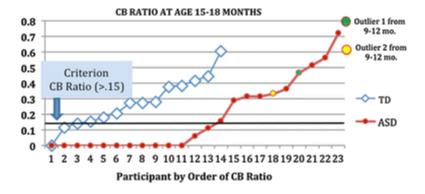


Fig. 2 Canonical babbling ratios by participant at 15–18 months

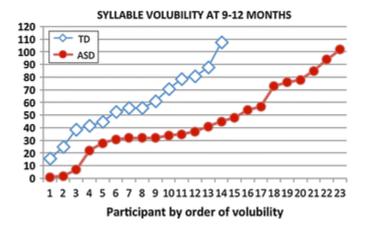


Fig. 3 Syllable volubility by participant at 9–12 months

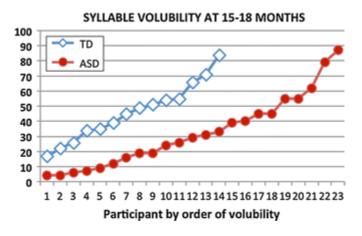


Fig. 4 Syllable volubility by participant at 15–18 months

Hypothesis 1

Infants later diagnosed with ASD will be less likely than typically developing infants to be in the canonical stage at each age (9–12 and 15–18 months).

Log odds ratios (log OR) were calculated to compare the classifications of both ASD and typically developing children with regard to their canonical babbling. The criterion for canonical babbling stage was set at 15 % or greater canonical syllables compared to all syllables; this is a common criterion in studies of canonical babbling, and is based on data reviewed in Oller (2000). TD infants were significantly more likely to have reached the canonical babbling stage based on the criterion than were infants later diagnosed with ASD at the 9–12 months age range (N = 35, log OR 2.84, $CI_{95} = 1.02-4.66$, p = 0.002), and remained more likely at the 15–18 months age range (N = 35, log OR 1.78, $CI_{95} = -0.04$ to 3.61, p = 0.054). As an easily interpretable effect size measure, the simple odds ratios (as opposed to the log odds ratio, which is statistically preferable for significance testing with small N's) can be considered; the simple ORs indicated TD infants were 17 times more likely (OR 17.1) to be categorized as in the canonical stage than ASD infants at 9–12 months and 6 times more likely (OR 5.96) at 15–18 months.

Hypothesis 2

Infants later diagnosed with ASD will demonstrate significantly lower canonical babbling ratios (independent of the canonical stage criterion) compared to typically developing infants.

Canonical babbling ratios of infants later diagnosed with ASD and TD infants were contrasted using a Mixed ANOVA. The between-subjects variable was diagnostic category (ASD vs. TD) and the within-subjects variable was age range (9–12 and 15–18 months). The mean canonical babbling ratios at 9–12 months were .06 (SD = .06) for the 21 infants later diagnosed with ASD and .17 (SD = .13) for the 14 TD infants; at 15–18 months the values were .16 (SD = .22) and .28 (SD = .16) respectively (Fig. 5). Analyses revealed a significant main effect for diagnostic category (F (1,1) = 6.79, p = .01, \mathfrak{n}_p^2 = 0.17), with infants later diagnosed with ASD producing significantly lower canonical babbling ratios, and a significant main effect for age (F (1,1) = 7.86, p < .01, \mathfrak{n}_p^2 = 0.19), with higher canonical babbling ratios at the older age. The effect size between groups for 9–12 months was d = 1.09 (a large effect) and for 15–18 months was .62 (a moderate effect; Cohen 1988). The age by diagnosis interaction was not significant (p > 0.66).

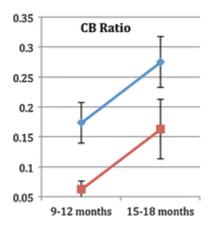


Fig. 5 Canonical babbling ratios by age and diagnosis

Hypothesis 3

Infants later diagnosed with ASD will demonstrate significantly fewer total vocalizations (lower volubility) at both age ranges compared to typically developing infants.

For this analysis, all 37 infants were included because there were no significant outliers. Volubility of infants later diagnosed with ASD and TD infants were contrasted using a Mixed ANOVA. The between-subjects variable was diagnostic category (ASD vs. TD), and the within-subjects variable was age range (9–12 and 15–18 months). Infants later diagnosed with ASD produced a mean of 4.55 (SD = .59) syllables per minute while TD infants produced a mean of 5.86 (SD = .67) syllables per minute at 9–12 months. At 15–18 months, infants later diagnosed with ASD produced a mean of 3.24 (SD = .49) syllables per minute while TD infants produced a mean of 4.63 (SD = .51) syllables per minute (see Fig. 6). Analyses revealed a significant main effect for diagnostic category (F (1,1) = 4.85, p = .034, η_p^2 = 0.12), and for age (F (1,1) = 4.96, P = .032, η_p^2 = 0.12). Thus, infants later diagnosed with ASD displayed significantly lower volubility than TD infants. The effect size for group at 9–12 months was d = 2.07 (large) and at 15–18 months was 2.77 (large).

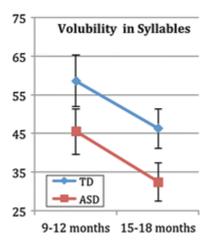


Fig. 6 Volubility by age and diagnosis

Hypothesis 4

A combined analysis using both volubility and canonical babbling status will significantly predict group membership.

Logistic regression analysis was conducted to test whether canonical babbling status (whether each participant met the .15 criterion) and volubility at age ranges of 9–12 and 15–18 months could reliably predict later diagnosis status (group membership). This test was conducted with all 37 cases included, partly in order to match the number of cases for the two predictor variables and partly because the goal of the analysis was to determine the potential practical utility of

identification of these children without any information other than volubility and canonical babbling ratio. This test may thus be the one of primary clinical interest, since it evaluates the circumstance that screening implies, where there would be no basis for knowing whether an infant might be an outlier on any variable. Without this evaluation there would be no direct indication in our results of the degree of group discriminability.

Statistical significance was reached in a test of the full model against a constant-only model, which indicated that, as a set, canonical babbling status and volubility reliably predicted later diagnosis ($\chi^2 = 9.82, p = 0.044, df = 4$). A small-to-moderate relationship between prediction and grouping was observed (*Nagelkerke's R* $^2 = 0.317$), with an overall prediction success of 75 % (64 % for TD and 82 % for ASD). However, further examination of the predictors using the Wald criterion revealed that when all four predictor variables were included in the model, none significantly contributed to prediction of group membership at an individual level (p > 0.05). The status of infants with regard to canonical babbling stage at the 9–12 months age range provided the largest observed predictive contribution, Wald = 3.06, p = 0.08, EXP(B) = 0.198. The contribution to group discriminability by volubility at 9–12 and 15–18 months age ranges approached nil, EXP(B) = 0.992 and 0.985 respectively.

Examination of the correlations among the predictor variables showed that all but volubility at 9–12 months were significantly correlated with all other predictors (Table 6), with volubility at 9–12 months significantly correlated with only canonical babbling at 9–12 months. This interrelation among the predictor variables suggests that, to some degree, they account for some of the same variance in diagnosis. However, the observed *EXP(B)* values (odds ratios of the outcomes given the value of an individual predictor) more strongly suggest that canonical babbling at 9–12 months accounted for the bulk of the variability in diagnosis.

Table 6 Intercorrelations between canonical babbling ratios and volubility

	1	2	3	4
1. Canonical babbling 9–12 months	ı	.352*	.528**	.354*
2. Volubility 9–12 months		_	0.21	0.14
3. Canonical babbling 15–18 months		.510**	_	
4. Volubility 15–18 months				ı

^{*} *p* < .05, ** *p* < .01

It seems clear that significance of the individual predictors in the logistic regression may have been hampered by the high level of relation among them. Individually, the volubility variables did not appear to have much influence given small Betas and high *p* values. When predictors were entered into the model in a hierarchical fashion, no matter how predictor entry was ordered (9–12 month variables at step 1 and 15–18 months variables at step 2, or CB variables at step 1 and volubility at step 2), only the 9–12 months CB variable was a significant independent predictor. R² changes and diagnostic ability for all of the regression and regression step iterations suggested little was added to the R² by adding variables in the second step (even with two more

predictor variables, only $\sim 2-3$ % was added to R^2), nor did these additions substantially alter the ability of the model to predict later diagnosis. The most efficient model appeared to be a logistic regression with 9–12 months CB as the only predictor.

Discussion

The importance of early intervention for children with ASD has resulted in attempts to quantify behaviors in infancy that may lead to early detection. Substantial effort has addressed gestural and social development and their potential roles in detection within the first year of life (e.g., Watson et al. 2013). The present results offer parallel findings in the domain of vocal development by demonstrating significant group differences in canonical babbling status, canonical babbling ratio, and total syllables produced (volubility) during the first year of life.

In our study, infants later diagnosed with ASD were significantly less likely to be classified as being in the canonical babbling stage, and demonstrated significantly reduced canonical babbling ratios compared to TD peers. Although significant group differences were apparent in both age ranges (9–12 and 15–18 months), the effect sizes for canonical babbling were larger at 9–12 months. Paul et al. (2011) demonstrated similar results in infants at high-risk for developing ASD who produced significantly lower canonical babbling ratios compared to low risk infants at 9 months, though at 12 months the differences were not statistically significant. Combined with the finding from Oller et al. (2010) that children with ASD up to 48 months of age show low canonical syllable production, the data here suggest that low production of canonical syllables may be a helpful marker for ASD from infancy into early childhood.

Since canonical babbling is well established in the vast majority of TD infants by 10 months (Eilers and Oller 1994), it might seem odd that several of the TD infants (5 at 9–12 months and 3 at 15–18 months) in the present study provided samples that did not meet the .15 canonical babbling ratio criterion for assignment to the canonical stage of vocal development. However, it is important to consider the fact that even infants who are clearly in the canonical stage based on parent report often fail to reach the criterion in a single laboratory sample of 20–30 min (Lewedag 1995). In addition, unlike the samples in prior research on canonical babbling, the samples here were not designed to elicit vocalizations, and consequently they may have been less rich in quantity and variety of vocalization than the samples that were used to develop the criterion. Further, our samples at 9–12 months were only 10 min in duration, and at 15– 18 months an average of slightly less than 10 min; it has been shown that variability in obtained canonical babbling ratios increases as the length of samples decreases (Molemans 2011; Molemans et al. 2011). Finally, our samples were based on home recordings with considerable noise and variable camera management that may have impeded our ability to recognize vocalizations in the samples. Consequently, we are not surprised that some of the TD infants failed to reach the criterion used to determine canonical status based on laboratory samples.

Given the strong links between the onset of canonical babbling and language development (Oller et al. 1997; Stoel-Gammon 1989), delayed onset of canonical babbling in infants with ASD may reflect latent communication impairment. It also may be that delayed canonical babbling directly contributes to communication symptoms in ASD. Canonical babbling requires motor ability as well as motivation to produce syllables, and practice in babbling may lay critical foundations for speech.

Prospective research on motor development in infants later diagnosed with ASD is sparse and often limited to high-risk groups, but available research does iSndicate that early motor impairment may be present (e.g., Matson et al. 2010; Manjiviona and Prior 1995; Page and Boucher 1998; Teitelbaum et al. 1998). Thus delayed canonical babbling may reflect an immature or disordered motor system with specific implications for speech.

If language develops as a consequence of social reinforcement of speech-like sounds that eventually evolve into true words (consider behavioral models of language development as in Hulit and Howard 2002; Goldstein et al. 2003; Goldstein and Schwade 2008; Goldstein and West 1999), then social reinforcement may encourage the production of canonical babbling. Children with ASD may be less motivated by social reinforcement, yielding less frequent vocal exploration and production of canonical syllables than in TD infants. To add to the problem, a delay in canonical babbling may result in reduction in caregiver social-communication directed toward the infant. On average, by 6-7 months and very rarely later than 10 months, canonical babbling emerges in TD infants (Eilers and Oller 1994). In response to recognition of canonical babbling, caregivers alter their communication pattern, sometimes attempting to direct the infant toward using canonical syllables meaningfully—for example, the parent who hears [baba] may reply, "Yes, that's a bubble" (Papoušek 1994; Stoel-Gammon 2011). Therefore, infants who are delayed in canonical babbling may also be delayed in their exposure to important linguistic input, and thus may be given less opportunity to learn words. A final point is that infants with ASD may simply have lower motivation to vocalize socially in the first place. This lower motivation could provide a further basis for slow vocabulary learning.

Our results on volubility included two statistically reliable findings. First, children in both groups had lower volubility at the second age than at the first. We attribute no particular theoretical importance to this finding but we take note of the fact that the lower level of volubility at 15–18 months compared to 9–12 months did correspond to greater physical movement of the children at the older age. In both groups combined, level of physical restriction during the selected recording samples was significantly less at the older age (p < .001). As reported earlier, level of physical restriction was not significantly different between diagnostic groups.

The second volubility finding is that infants later diagnosed with ASD produced significantly fewer vocalizations deemed to be relevant for the emergence of speech (both canonical and non-canonical sounds) at both age ranges (9–12 and 15–18 months) compared to TD peers. Other research has demonstrated that infants with ASD direct fewer vocalizations to others (Ozonoff et

al. 2010); our study extends this finding to a more general measure of volubility in terms of total vocalizations (syllables) rather than only ones directed to others. Our finding is also congruent with results from automated analysis of all-day recordings indicating low volubility in children with ASD at 16–48 months of age (Warren et al. 2010). The results may seem to run counter to Paul et al. (2011) whose sample of high-risk infants were reported to not produce significantly fewer vocalizations than low-risk infants. However, as described in the introduction above, the Paul et al. study did not report data in a way that can be directly compared with the volubility data reported here.

Some disability groups (e.g., hearing impaired infants and infants with cleft palate) have been reported to exhibit volubility similar to that of TD infants (Clement 2004; Chapman et al. 2001; Van den Dikkenberg-Pot Koopmans-van Beinum and Clement 1998; Nathani et al. 2007; Davis et al. 2005); however, infants from low SES households have been reported to have significantly decreased volubility in comparison to those from higher SES households (Oller et al. 1995). Children from low SES backgrounds are often presumed to be at-risk for language deficits. Although it would be impossible to identify and quantify all of the mechanisms through which poverty may affect language development, research has demonstrated that the amount of communication caregivers direct toward their children is decreased in low SES situations (Hart and Risley1995). This impoverished linguistic environment may result in decreased dyadic social and communicative interactions and thus in a decrease in overall volubility of infants.

It is important to note that the relatively well-matched SES between our two groups suggests that the differences in volubility were not attributable to differences in SES. In the case of low SES households, an impoverished linguistic environment due to lack of parent responsiveness might be expected to lead to decreased volubility of the infant and later language difficulty. For infants later diagnosed with ASD, reduced volubility may be affected by multiple factors, not related to inherent parental responsiveness, but related instead to the social impairments of ASD. One issue is that these children may experience less linguistic stimulation due to having disrupted sensory processing systems corresponding to sensory hyporesponsiveness; children with ASD are less likely to respond, or require substantially more stimulation to respond to environmental events (Baranek 1999; Baranek et al. 2013); Miller et al. 2001; Rogers and Ozonoff 2005). This characteristic of ASD is also reflected in the tendency for infants as young as 5 months who will later be diagnosed with ASD to be less likely than TD infants to respond to their name being called (Werner et al. 2000). This lack of responsiveness may indicate that infants with ASD are less affected by vocal communication from caregivers than TD infants. If so, the lack of responsiveness may reflect an effectively impoverished linguistic environment because of attenuated reception of caregiver input by infants with ASD and subsequent communication impairments. Indeed, sensory hyporesponsiveness has been shown to be associated with poorer language functioning in children with ASD (Watson et al. 2011).

An additional way that the environment for children with ASD may be impoverished could involve a social feedback loop (Warlaumont et al. 2010) that is under investigation using

automated analysis of vocalizations of parents and infants from all-day home recordings. Since infants with ASD produce fewer canonical syllables than TD infants, and since parents respond strongly with language stimulation to canonical syllables, an infant with ASD may actually hear less language from parents, because parents provide input that is tied to the infant's output. The infant's low volubility may then be aggravated by lower input levels resulting from the infant's own anomalous pattern of vocalization.

Finally, the logistic regression analysis with four independent variables (age 1 and age 2 canonical babbling classification and age 1 and age 2 volubility) demonstrated that classification of diagnostic category (ASD vs. TD) could be predicted with 75 % accuracy, even when the two outliers were included. The model more accurately classified infants later diagnosed with ASD (Sensitivity = 82.6 %) than TD infants (Specificity = 64.3 %). The strongest predictor of group membership was canonical babbling classification at 9–12 months as it alone correctly classified 90 % of infants later diagnosed with ASD and 63 % of TD infants. Thus, in the search for markers of ASD risk in infancy, canonical babbling status at 9–12 months appears to be the single best candidate among the variables considered in the current study. The utility of the measure as a group marker is age dependent, since a larger proportion of infants in the ASD group at 15–18 months had reached the canonical stage than at 9–12 months.

To help better understand the high canonical babbling ratios of the two outliers, the coders, both certified speech-language pathologists, viewed the videos from those infants again after their outlier status was identified. We speculated that the outlier status of these two infants may be related to the phenomenon of motor stereotypy that is common in ASD, that is, that the two infants were engaged, at least in the 9–12 months samples, in a motor stereotypy focused precisely on canonical babbling. In re-examining the videos of the two outliers, the coders looked for qualitative evidence that might speak to the credibility of this speculation. In the second viewing of the recordings, the coders noticed that the first outlier infant produced the majority of the canonical syllables during a single scene while walking outside. He repeatedly produced a [da] syllable during this brief episode, but did not direct his vocalizations to the caregiver. The sense that a prelinguistic vocal stereotypy may have been operating was enhanced by the fact that the same syllable was repeated throughout. The stereotypy of canonical babbling in this infant was reported by the coders as constituting the only evidence either had noticed as specifically suggesting the possibility of ASD while they were coding, and thus, this was the single case where the intended blinding of the coders to diagnostic group seems to have been foiled. The coders did not observe any other stereotypic behaviors vocal or otherwise in these samples. The second infant engaged in high canonical babble production while roughhousing with his father, but to our clinical eyes, that behavior did not seem particularly unusual. Further research on the possibility that babbling can be a focus of motor stereotypy in ASD seems in order. It may be worthy of note that the two outliers' CARS scores (25 and 31) fell within the range of the scores for the ASD group (23–50).

In addition to the findings suggesting possible clinically useful markers for ASD, the present results provide a new scientific view on the robustness of canonical babbling. There has been no prior empirical indication that canonical babbling onset is delayed in ASD, nor that volubility is low in infants later diagnosed with ASD. Our results thus suggest that the development of vocalization in infancy is affected by whatever the fundamental disorders of ASD may be. Assuming ASD to be a *social* disorder, it is not obvious that babbling would necessarily be disturbed in the disorder because the extent to which babbling is a social (as opposed to an endogenously generated) phenomenon is itself an empirical question. Our results can then be thought to provide a new empirical perspective on the possible social nature of babbling. The results also suggest that the vocal differentiation of the two groups is robust, given the relative clarity of the results indicating low canonical babbling and volubility in the infants in the ASD group, even though we had samples of low recording quality and very limited duration. The results seem especially significant in the context of a broad body of research cited above on the robustness of canonical babbling as a foundation for language and on the robust resistance of canonical babbling to delay as seen in prior studies cited in our paper—no delay has been found in cases of prematurity, low SES, or multilingual exposure.

Future Directions and Limitations

This study provides a proof of concept regarding the notion of atypical emergence of the canonical babbling stage in the developing infant who will later be diagnosed with ASD and the possibility that tracking canonical babbling in infancy may add to our repertoire of markers for ASD prior to 1 year of age. Future research to address some of the limitations of the current study and advance our understanding of the development of canonical babbling among infants with ASD is warranted by the findings of the current study. One limitation in the current study was the lack of a comparison group of infants with later diagnoses of non-ASD disabilities, which prevents us from definitively attributing the differences found in this study to ASD rather than general impairments in cognition or communication. Our working hypothesis to test in future studies will be that these differences in canonical babbling onset and in volubility are specific to ASD.

Another limitation was that our study used only short video segments from each time point, which surely impacted our ability to precisely assess important aspects of vocalization, because it has been shown that variability in obtained canonical babbling ratios increases as the length of samples decreases (Molemans 2011; Molemans et al. 2011). The low canonical babbling ratios obtained for a few of the TD infants presumably would not have occurred with larger sample sizes. In future studies we hope to obtain longer samples, and if possible to more precisely identify canonical babbling onset through longitudinal laboratory assessments paired with caregiver report of onset. But of course to make this possible, prospective studies may be necessary, with several years of follow-up, presumably taking advantage of the opportunity presented by sibling studies. Such studies would also afford the opportunity to obtain much better recordings than are available in retrospective studies such as the present one. Indeed,

sibling studies can now capitalize on all-day recording, yielding the opportunity to assess vocal development in ASD with much greater ecological validity and representativeness.

Onset of canonical babbling usually occurs between 5 and 9 months in TD infants. It appears from the present data that onset may occur within a much wider range in ASD. Quantification of onset in ASD may yield prognostic value regarding core communication symptoms. For example, if canonical stage onset is delayed beyond a certain threshold, the infant may be at especially high-risk for remaining nonverbal. Discovery of such a delay could allow specific interventions to be tailored based on prognosis earlier in development.

Future research should also focus on caregivers and their roles in canonical stage development and its identification. Prior work suggests that with TD infants, parents are extremely accurate in their reports of the onset of canonical babbling (Oller et al. 2001). If caregivers of infants with ASD are similarly capable of identifying onset of canonical babbling, it may be possible to use canonical babbling onset as part of a parent-report screening tool for early identification. In addition, alterations in communication directed to infants by caregivers as canonical babbling emerges may help to elicit and maintain social-communicative interaction, and subsequently impact language development.

Our findings on volubility represent another potential avenue for understanding early social-communication development processes in ASD. Perhaps the most intriguing aspect of this possibility is suggested by the proposal that there may be a feedback loop involving low canonical syllable production in ASD followed by low parental rate of vocalization to infants, aggravating the low volubility and low rate of canonical syllables in ASD (Warlaumont et al. 2010). We anticipate rapid growth of studies tracking this possibility, especially since there is a rapidly growing possibility of conducting some aspects of such analysis based on automated classification of vocalizations in all-day recordings as indicated by the growth of LENA system studies.

Clinical Implications

Our findings suggest that canonical babbling should be considered an important milestone in infancy that may be delayed in infants who are later diagnosed with ASD. If infants demonstrate delays in canonical babbling, a developmental assessment that includes evaluation of early warning signs for ASD should be administered. Although volubility appears less promising as a marker for ASD, it may be useful in combination with other items in the context of early identification screening tools. For infants demonstrating either low canonical babbling ratios or low volubility, interventions to draw infants' attention to social-communicative stimuli in that context of dyadic interactions may help stimulate growth of vocal communication.

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