Profiles of Vocalization Change in Children with Autism receiving Early Intervention

Running title: Vocalization change in children with autism

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Abstract

Children with Autism Spectrum Disorder (ASD) commonly present with co-morbid language impairment, negatively impacting their learning and participation across settings. Addressing these needs requires a detailed understanding of their communication trajectories. In this study, we used the Language ENvironment and Analysis (LENA) system to examine possible changes in children's (a) vocalizations and (b) ratio of speech to non-speech vocalizations over a 10-month period. Data for 23 children with ASD (17M, 6F; ages 32-67m) were analysed, including monthly 3-hour in-class recordings and standardized measures of language, cognition, and ASD characteristics. Using hierarchical generalized linear models, we found significant time-trends for child vocalizations ($p = < .001$) and the vocalization ratio ($p = .02$), reflecting a waxing and waning pattern. Children with higher expressive language scores (Mullen Scales of Early Learning, Vineland Adaptive Behavior Scales – 2nd Ed) and non-verbal cognition (Mullen Scales of Early Learning), and fewer ASD characteristics (Social Communication Questionnaire) demonstrated greater increases in the vocalization ratio over time (p values .04 - .01). Children with greater language and cognition difficulties were the most vocal, but produced a higher proportion of non-speech vocalizations. The results demonstrate that significant fluctuations, as opposed to linear increases, may be observed in children with ASD receiving intervention, highlighting the value of assessment at multiple time-points. In addition, the findings highlight the need to consider both the quantity (vocalization counts) and quality (ratio of speech to non-speech vocalizations) when interpreting LENA data, with the latter appearing to provide a more robust measure of communication.

Lay abstract

In this study, we examined possible changes in speech and non-speech vocalizations in 23 children with autism attending a comprehensive early intervention program over a 10-month period. Contrary to our expectation, we observed a waxing and waning pattern of change in children's vocalizations over time, rather than a steady increase. We also found evidence to suggest that looking at the quality of children's vocalizations (i.e., the ratio of speech to non-speech vocalizations) provides a more accurate picture of children's development than simply looking at the quantity (i.e., how frequently they vocalize).

Profiles of Vocalization Change in Children with Autism receiving Early Intervention

Social-communication impairment is a core diagnostic feature of Autism Spectrum Disorder (ASD), reflected not only in an individual's propensity to communicate, but also the modalities used, and the functions served. Commonly observed and diagnostically relevant features include reduced initiating and maintaining of social interactions, reduced use of gestures and facial expression as compensation for lack of speech, and a focus on communication to express needs and wants as opposed to sharing information (American Psychiatric Association, 2013). Approximately 60% of children with ASD have a co-occurring language disorder, characterised by persistent difficulties in the acquisition and use of language across modalities arising from deficits in comprehension and/or production (Posserud, Hysing, Helland, Gillberg, & Lundervold, 2018). Taken together, the communication differences observed in children with ASD, when compared to their neuro-typical peers, place them at substantial disadvantage with respect to learning, participating in, and benefiting from educational settings with long-term implications for their independence, health, and wellbeing (Howlin, Savage, Moss, Tempier, & Rutter, 2014; Magiati, Tay, & Howlin, 2014). Accordingly, understanding and addressing the communication needs of children with ASD is a key concern and priority for individuals with ASD (Autistica, 2016) as well as clinicians, parents, and researchers alike (Tager-Flusberg & Kasari, 2013; Trembath, Westerveld, & Shellshear, 2016).

Communication encompasses expressive and receptive language which in turn comprise domains of phonology, morphology, syntax, semantics, and pragmatics. Within this framework, expressive (or spoken) language provides a readily observable and intuitive measure of children's development, with well-established developmental benchmarks (Tager-Flusberg et al., 2009). In fact, although differences in social and non-verbal communicative behaviours emerge as early as 9 months of age (e.g., lack of joint attention or proto-declarative pointing), there is evidence to suggest that the most common trigger for parents to seek professional advice regarding possible ASD diagnosis is delayed emergence of spoken language between 12 to 18 months (De Giacomo & Fombonne, 1998; Herlihy, Knoch, Vibert, & Fein, 2015). Spoken language also provides an ecologically valid measure of change for children accessing early intervention programs and is among the most common measures employed in clinical trials. Encouragingly, communication has been shown to be among the most modifiable aspects of adaptive behaviour in early intervention trials (e.g., Dawson et al., 2010; Kasari, Paparella, Freeman, & Jahromi, 2008; Wetherby et al., 2014). However, close examination of findings reveals substantial individual variability in spoken language outcomes in these trials, while community-based research has revealed that as many as 30% of children receiving early intervention commence school unable to speak in sentences (Norrelgen et al., 2015; Rose, Trembath, Keen, & Paynter, 2016; G Vivanti, Dissanayake, & Victorian ASELCC Team, 2016).

The need to understand trajectories

Insights into the development of spoken language in children with ASD can be drawn from controlled trials that evaluate communication development in response to treatment (e.g., Romski et al., 2010; Smith, Klorman, & Mruzek, 2015) as well community-based cohort studies of children with uncontrolled treatment histories (e.g., Magiati et al., 2014; Wodka, Mathy, & Kalb, 2013; Yoder, Watson, & Lambert, 2015). In each case, research to date has revealed substantial variability in spoken language development over time. Pickles, Anderson, and Lord (2014), for example, completed a longitudinal study of 192 children initially referred for ASD. Using latent class growth analysis (seven classes), they reported variability between the ages 2 and 6 years, followed by stability in classes from 6 to 19 years. Wodka, Mathy, and Kalb (2013) identified that 47% of a cohort of 535 children with ASD (mean age 11 years, 6 months), who were minimally verbal before the age of 4, attained fluent speech (i.e., spoke in sentences) at or after the age of 4.. These findings highlight the need to examine spoken language skills over multiple time points as opposed to only pre-post assessment, which in turn requires ecologically assessment tools that can be administered regularly in children's everyday learning environments. Such an approach can help to elucidate individual variability amongst children and ultimately tailor approaches to individual strengths and needs.

Assessing language development

Various methods are available for assessing children's communication skills including direct assessment (e.g., Mullen Scales of Early Learning; Mullen, 1995), language sampling and analysis, and parent and teacher report (e.g., Vineland Adaptive Behavior Scales - 3rd Edition; Sparrow, Cicchetti, & Saulnier, 2016). For longitudinal research, the options are constrained by resources (e.g., cost of administering assessments and skilled assessors at multiple timepoints or cost of transcribing and analyzing spontaneous language samples); minimum time intervals between standardized assessments; sensitivity of measures to possible change in short periods of time; and the burden on children, families, and staff. There have been multiple calls for improved methods for assessing change amongst children receiving early intervention (Kasari, Brady, Lord, & Tager-Flusberg, 2013; McConachie et al., 2015; Trembath et al., 2016). In order to understand trajectories, it is important to employ a regular, systematic, and ecologically valid way of measuring children's communication. One option is the Language Environment Analysis (LENA®) system, which involves the use of small wearable digital recorders that measure the quantity of vocalizations produced by children and adults in the environment.

Language Environment Analysis (LENA)

The LENA system consists of a digital language processor (DLP) and speech recognition software. The device was specifically developed to monitor language environments, and has been reliably used for children between two and 48 months of age (Dykstra et al., 2013). The DLP records up to 16 hours of auditory input in the environment, and the software categorises the input into three variables/categories; (a) Adult Word Count (AWC); (b) Child Vocalizations (CV); and (c) Conversational Turns (CT), which includes responses to another within 5 seconds. The processor excludes from child vocalization counts overlapping adult-child speech, non-speech sounds such as crying, laughing, vegetative sounds, and other fixed signal sounds, as well as digitized sounds from television or music. The latter sounds are separately classified by the processor and can be identified manually using transcription software. The processor also discriminates between meaningful (≥ 35 dB HL) and distant sounds (less than 35 dB HL). LENA also provides an option for more detailed analyses of processed recording files via the LENA Advanced Data Extractor (ADEX; LENA Foundation, 2011). This includes the capacity to segment recordings and to differentiate between the duration of speech-like and non-speech vocalizations for each recording.

Language Environments and Vocalization Counts

To date, LENA has been used to examine language learning environments in a range of populations including children with language delay, hearing impairment, and Down syndrome, as well as those in bilingual learning environments (for a review see Wang et al., 2017). In the field of ASD, only a small number of studies have been conducted, focusing on children’s language learning environments and initial attempts to examine changes in communication over time. Warren et al. (2010), for example, compared child and adult vocalizations amongst 26 children with ASD and their parents, and a comparison group of 78 typically developing children and their parents. The authors reported no significant differences in adult word counts across the two groups, but reduced vocalizations and turn-taking for children with ASD, with parent and child vocalizations positively correlated. Dykstra et al. (2013), examined language learning environments for 40 children with ASD, age 3-5 years, attending self-contained classrooms for children with disabilities, at two time-points approximately 6 months apart. They reported significant increases in Child Vocalizations from Time 1 (M = 3.1, SD = 1.5) to Time 2 (M = 4.1, SD, 1.7), but no significant differences in Adult Word Counts or Conversational Turns. In addition, the authors reported a significant correlation between Child Vocalizations per minute and language age-equivalents obtained through direct testing (Preschool Language Scales 4th Edition) at Time 1 assessment, but not with visual reception age equivalent scores measured using the Mullen Scales of Early Learning nor ASD severity measured using the ADOS-G. The findings indicate that LENA may have good construct validity with another language measure (i.e., PLS 4th Ed), but that children's vocalization rates may not be associated with their cognitive development or ASD characteristics, at least cross-sectionally. Based on the findings, Dysktra et al., (2013 suggested that LENA may provide a sensitive measure of change in the communication of children with ASD. However, the authors noted potential limitations of the system including the fact that LENA only provides information regarding the quantity, not quality (e.g., intent, directness, vocabulary), of children's vocalizations, which may be particularly confounding in children with high levels of echolalia.

Speech versus Non-Speech Vocalizations

Rankine et al. (2017) examined the validity and reliability of LENA variables against transcribed samples and standardized measures, for a group of 18 minimally verbal children (i.e., not using phrase speech) with Phelan-McDermid Syndrome (commonly associated with ASD), aged 30 to 172 months. Each participant completed between one and three whole day recordings (maximum 16 hours) across home, education, and clinical settings. From these, 36 x 5-minute intervals were selected for transcription and analysis. The authors reported moderate overall reliability between human transcribers and LENA measures ($k = 0.455$, $p < 0.001$), 95% CI 0.437–0.473), with a tendency for LENA to underestimate the number of child vocalizations to avoid false positives (Rankine et al., 2017). In particular, LENA accuracy (against human transcribers) was negatively correlated with child chronological age, with the authors suggesting that acoustic changes in the voices of older children leading to increasing incidences of LENA misclassifying as adult vocalizations. Consistent with Dykstra et al. (2013), Rankine et al., (2017) suggested that high levels of echolalia and repetitive speech may confound interpretation of the data, with the quantity and quality of vocalizations not necessarily correlated. To illustrate, it is possible that two children – one with relatively mild ASD characteristics displaying frequent directed and flexible communication and another with severe intellectual disability and high rates of echolalia and repetitive speech may present with a similar rate of Child Vocalizations as measured using LENA (i.e., quantity of vocalizations), despite a fundamental difference in the quality of their vocalizations. Accordingly, caution appears warranted in interpreting the findings of studies focused solely on Child Vocalization counts in children with ASD.

Rankine et al., (2017) proposed that a ratio measure – calculated using the ADEX tool by dividing the total duration of the child's speech sounds by the duration of non-speech sounds–

may provide a more useful indication of children's communication development. Citing the work of Plumb and Wetherby (2013), they noted that a higher proportion of speech to non-speech vocalizations has been found to be positively correlated with other measures of expressive language (Plumb & Wetherby, 2013). Indeed, Paul, Fuerst, Ramsay, Chawarska, and Klin (2011) reported that high risk infants (older sibling with ASD) produced fewer speech-like, and more non-speech like vocalizations, compared to low risk peers based on analyses conducted between 6 and 12 months of age. Consistent with their hypothesis, Rankine et al. found the Vocalization Ratio to be a more reliable measure (against human transcribers) than child vocalization counts for their sample of minimally verbal children with ASD (i.e., not using phrase speech), who demonstrated higher levels of echolalia and repetitive speech. In addition, unlike the Child Vocalization count, the ratio was correlated with children's expressive language raw scores on the parent-report measure, the Vineland Adaptive Behavior Scales – 2nd Edition, although neither measure correlated with children's expressive language raw scores on the Mullen Scales of Early Learning. The authors suggested that the ratio may provide a novel expressive language measure child with ASD and proposed that its potential to provide a sensitive measure of change in expressive language over time be examined.

To date, only two studies have examined possible change in LENA measures over time amongst children with ASD. As noted, Dykstra et al. (2013) reported a significant increase in Child Vocalization counts for 40 preschool aged children with ASD, assessed approximately 6 months apart. More recently, Bak, Plavnick, and Byrne (2017) used LENA to examine language trajectories of nine minimally verbal children with ASD aged 6-10 years, attending two elementary schools, across one school year. They found no statistically significant changes based on group-level analyses of Child Vocalization counts or turn taking over the course of the year, with individual data indicating an increase for one of the nine children. The contrast in findings between the two studies may be due to differences in the populations (e.g., preschool versus school aged), methods of analyses (paired t-tests based on two time-points versus linear models with multiple time points), and apparent lack of statistical power in the case of Bak et al., (2017) due to a small sample size. It is also not clear if the use of a ratio measure, as proposed by Rankine et al., (2017) may have yielded additional insights, given the authors reiterated the need for caution in inferring communication quality based on a measure of vocalization quantity. Identifying, and ultimately understanding, the communication trajectories of children with ASD, would benefit from further research examining their vocal behaviour at multiple timepoints, and focusing on both the quantity and quality of the vocalizations.

Given the heterogeneity in communication outcomes for children with ASD, both in treatment studies as well as in children with uncontrolled treatment histories, there is a well-established need to identify factors that may account for individual differences (G Vivanti, Prior, Williams, & Dissanayake, 2014). As noted, Dykstra et al (2013) found no significant correlations between the rate of Child Vocalizations and measures of cognition and ASD severity, taken at study intake, in 40 children with ASD. It is possible the lack of correlation may be due to equally high rates of vocalizations amongst children with the most significant (high rates of echolalia and non-speech vocalizations) and more subtle (high rates of spontaneous directed and flexible speech) communication needs. Here, the Vocalization Ratio proposed by Rankine et al., (2017) may offer greater insights. Furthermore, none of the studies to date have examined factors that may account for individual differences longitudinally: the question that is most pressing with respect to developing, evaluating, and implementing treatments to optimise children's communication.

The primary aim of this study was to examine the communication trajectories of children with ASD receiving comprehensive early intervention, in terms of possible changes in vocalizations assessed using LENA. Focusing on the age group examined by Dykstra et al. (2013) but with assessment at multiple time points as per Bak et al. (2017) and incorporating the ratio measure recommended by Rankine et al. (2017), the study was designed to provide a comprehensive interrogation of communication trajectories using LENA. Based on the findings of Dykstra et al., (2013) in children of similar age, we hypothesized that we would see a significant increase in Child Vocalizations, including improved Vocalization Ratios, over the course of the study. The second aim of this study was to examine factors that may be associated with individual differences in changes in vocalizations over time. We hypothesized that higher levels of baseline language skills and cognition (based on direct measures and parent report), and lower level autism symptoms, would be associated with greater increases in vocalizations overall, and improved ratios of speech to non-speech vocalizations. The third aim of this study was to examine possible sub-group differences in trajectories, related to child characteristics at baseline. We hypothesized that children with lower expressive language skills, lower cognition, and higher ASD symptoms at baseline (i.e., children with more complex needs) would, as a group, have equal or higher vocalization counts and lower vocalization ratios compared to their peers with the contrasting profiles.

Method

Design

We used a longitudinal cohort study involving children attending a comprehensive, group-based community early intervention program to address the research aims. The study examined communication change within a group of children receiving treatment, as opposed to change resulting from treatment due to the absence of a control group. The study was approved by the [name withheld for blind review] human research ethics committee (Ref: 2015/32).

Participants

The participants in the study were 23 children (17M, 6F) with mean age of 49.6 months (SD = 10.1, range = 32-67) with an existing diagnosis under DSM-5 to meet the service eligibility criteria which required diagnosis by a paediatrician, child psychiatrist and/or multidisciplinary team, verified by the research team using the ADOS-2. All families who attended the early intervention service were invited to have their children participate in this research, which formed part of a larger project examining communication development in children with ASD within the centre. LENA data were collected over the period 2015-2017, via monthly recordings. A total of 79 children participated in the study in the LENA component during the time period, of whom 23 children had sufficient data to be included in analyses. Specifically, children were required to have a recording at the beginning of the year (January-February) and 5 or more recordings in the ensuing 9-month period (total 10 months). The reasons for children not having sufficient data included (a) children commencing the program mid-year, (b) delays in parents returning consent forms, and (c) missing data due to child absence on the days of data collection or children electing not to wear the LENA device.

As indicated in Table 1, the participants presented with a broad range of social-communication skills and ASD characteristics. There were no exclusion criteria pertaining to co-morbidities including intellectual disability and language disorder for an ecologically valid sample representative of children seen in community practice. There were no significant differences between the children whose data were analysed, and the broader sample for whom some LENA data were available, with respect to chronological age (p = .61), gender (p = .54), expressive language (VABS-II, p = .37; MSEL, p = .07), ASD characteristics (SCQ, p=.*51*), the proportion of children for whom languages other than English were spoken at home (p = .54), the number of children in the child's home (p = .64), or birth order (p = .54). There was marginally significant difference on the MSEL non-verbal developmental quotient (p = .04) with the included children having on average higher scores (M = 64.1, SD = 21.9) than the excluded children (M = 53.4, SD = 19.2).

[Insert Table 1 about here]

Intervention Program

The study was conducted within a not for profit organisation in Queensland, Australia that provides early intervention to children with ASD from 2-6 years. The organisation has 10 early learning and child care centres around the country. The study was conducted in one of the centres, catering for approximately 48 children, and arranged into four classrooms of approximately 10-12 children with each led by an early years education teacher. Classes were grouped according to children's learning goals (reflecting developmental level) rather than chronological age with one staff member for a maximum 4 children. Staff included a team of speech pathologists, occupational therapists, behaviour therapists, early years teachers, and childcare professionals. Children in each class received the same level of input from these professionals. All staff completed the same induction and ongoing training program, participated in annual performance reviews (including review of clinical performance and application of the program), and were supervised across classrooms by the centre program manager.

The early intervention program has been previously described in Paynter, Scott, Beamish, Duhig, and Heussler (2012). It is comprehensive manualised program comprising both content areas (developmental domains) and teaching strategies derived from best practice guidelines

(Roberts & Williams, 2016). During the study, the program included children receiving a minimum 20 hours of intervention per week, as recommended by Roberts and Williams (2016). Each child had an individual plan based on her/his strengths and needs determined through multidisciplinary assessment and set in partnership with families. The program covered 11 domains: classroom attending, echoics, visual perception, social skills, play skills, self-help, intra-verbals, receptive and expressive language, imitation, and academic skills. Teaching occurred throughout the day in natural contexts such as free play, snack time, outside play, self-help activities, as well as circle and mat times using naturalistic strategies (e.g., pivotal response training, naturalistic teaching strategies) drawing from the National Autism Centre guidelines (2009).

Procedure

Children for whom consent was obtained were each assigned a digital language processor (DLP). The DLP, a lightweight audio recorder, was inserted into purpose designed vest, which children wore for one day per month. Children wore the vests from the beginning of the intensive program (09:00 daily), until their final meal break (approximately 14:00). Teachers were advised that children who showed visible distress from wearing the vest, or who communicated that they wanted to take the vest off, were to have the vest removed. As part of the research, children were also assessed using standard assessments and parent report measures, at intake, approximately 12-month intervals, and at exit from the program, as described below.

Measures

LENA

The outcome measures were (a) changes in the participants' average number of vocalizations per minute (Child Vocalizations), calculated based on the first three hours of monthly whole-day recordings at the early intervention centre over the period of 12 months (school year) and (b) changes in the participants' Vocalization Ratio, again based on the first three hours of recording, calculated by dividing the duration of speech by the non-speech vocalizations using the ADEX tool. Our decision to analyze the first three hours of each whole day recording to ensure consistency across samples (e.g., avoiding inadvertently analysing sleep time for some children) was consistent to approaches taken in previous studies (e.g., Bak et al., 2017; Dykstra et al., 2013).

Mullen Scales of Early Learning (MSEL) (Mullen, 1995)

The MSEL is a standardized assessment which assesses cognitive ability and motor development in children up to 68 months, across five scales (Mullen, 1995). The Expressive Language (EL) scale age equivalent score was used in analysis. The EL scale consists of 28 items which examine language production. A nonverbal mental age composite was calculated by averaging age-equivalent scores on the Fine Motor and Visual Receptive scales. This score was used as an estimate of cognitive ability, as per previous ASD research (e.g., Paynter, Trembath, & Lane, 2018; Westerveld et al., 2016). Analyses were conducted using age equivalent scores to capture more fully the range of individual differences, as in previous studies (e.g., Paynter et al., 2018; Giacomo Vivanti, Dissanayake, Zierhut, & Rogers, 2012), as children with ASD often do not reach the basal raw score for meaningful MSEL standardized scores. Age equivalent scores were selected over raw scores (as in Bak et al., 2017) due to forming approximately ordinal evenly distributed scores, and thus consistent with assumptions for parametric analysis.

Vineland Adaptive Behaviour Scales – 2nd Edition (VABS-II) Parent Form (Sparrow, Cicchetti, & Balla, 2005)

The VABS-II is a parent or caregiver report form used to assist in understanding a child's adaptive behaviours across four domains. Items are rated on a 0 – 2 scale, indicating frequency of behaviour occurrence where 0 = never, 1 = sometimes or partially, and 2 = usually. The expressive language age equivalence score, a subset of the communication domain, was used in analysis as recommended by previous research with children with ASD (Yang et al., 2016)

Social Communication Questionnaire (SCQ) (Rutter, Bailey, & Lord, 2003)

The SCQ is a parent/caregiver report, ASD screening checklist, which assists in evaluating communication skills and social functioning in children with a suspected diagnosis of ASD. The SCQ consists of 40 yes/no answer questions which yield a total score between 0 and 39. Although the ADOS-2 was completed to verify diagnosis, this occurred at different stages during the children's intervention programs due to resourcing (rather than consistently at baseline), hence the SCQ was selected for use in analyses.

Analytical Plan

We used a hierarchical generalized linear methods analysis approach assuming an autoregressive covariance matrix, to examine LENA Child Vocalizations per minute and the Ratio of Speech to Non-Speech Vocalizations across the duration of time for the study, relevant to addressing all three study aims. The mean change in LENA measures over time was computed adjusting for chronological age, gender, year of intake and first month LENA recording. The models were assessed for convergence using existing non-missing data. Additionally, a regression-based imputation strategy (i.e. stochastic regression) using existing explanatory variables (chronological age, gender, intake cycle and first month recording) was performed to account for missing data (23.1%, based on each child having a maximum of 10 opportunities for recording in the year) within the cohort. Results were consistent for imputed and non-imputed samples, thus analyses for the non-imputed sample are reported. To address aims two and three, trajectories of predicted mean Child Vocalizations and Ratio of Speech to Non-Speech Vocalizations were plotted for the entire cohort and stratified by tertiles (to compare subgroups) for Vineland II Expressive Language, Mullen's Expressive Language, Mullen's DQ and SCQ scores. Contrasts were based on tertiles, as opposed to estimates of clinically relevant cut-offs for each measure, given that the sample was skewed towards children with more complex needs on all measures. To provide an indication of broader changes in children's expressive language, we used a paired samples t-test to examine changes in children's expressive language age equivalent scores on the MSEL that included the study period. Note that because the MSEL was administered approximately every 12 months to children in the study, the timing of LENA and MSEL assessments were not perfectly aligned. The MSEL was completed within 2 months of the first LENA recording for 18 children, and between 4-8 months for 5 children. All analyses were performed using SAS version 9.4 and SPSS Version 22.

Results

A total of 177 recordings were available for the 23 children included in analyses, representing 531 hours of recording. As presented in Table 2, mean child vocalizations per minute ranged from 2.54 – 3.60 at individual timepoints during the 10-month period. Vocalization ratios ranged from 1.29-2.22, indicating children produced approximately 2 times more vocalizations, compared to non-speech vocalizations. Results from our repeated measures analyses showed significant time trends for both child vocalization and ratio measures. A statistically significant increase in children's MSEL expressive language age equivalent scores was observed over the period of time that included LENA data collection, $t(21) = -5.454, p = < .001$, d = .772, from a mean of 23.68 (SD=10.77) at Time 1 to 32.00 (SD=13.15) at Time 2.

[Insert Table 2 about here]

To test our first hypothesis, that we would see a significant increase in children's Child Vocalizations and improved Vocalization Ratio measures over the course of the study, we used generalized linear mixed methods. Table 3 presents the mean change in Child Vocalizations and Vocalization Ratios relative to Month 1, adjusting for age, gender, intake cycle, and values for first month recordings. All point estimates for both measures exceed the values for the first month recordings, except Child Vocalizations in Month 9. Increases in Child Vocalizations were statistically significant for months 2 and 4, with increases in Vocalization Ratios statistically significant in months 2, 4, 5, 8 and 9.

Overall time trends were significant for both LENA measures (p-values <0.05), however with uniquely distinct patterns. Child Vocalizations peaked at month 4 and declined from then onwards until month 9 (Figure 1). In contrast, observations for the Vocalization Ratio revealed a cyclical waxing and waning pattern with higher measures in months 2 and 8 (Figure 2). Therefore, there was no uniform increase over time for either measure. Individual growth curves for each child are presented as supplementary material, revealing substantial within, and between child, variability.

[Insert Figures 1 and 2 about here]

Relationship between Baseline Skills and Vocalization Trajectories

Our second hypothesis was that higher levels of baseline language skills and cognition, and lower level autism symptoms, would be associated with greater increases in vocalizations. Using continuous measures of baseline language scores, we did not find significant associations between baseline language skills and the Child Vocalization measure. However, we found that higher VABS-2, MSEL age equivalent and MSEL DQ scores at baseline were associated with greater increases in LENA Vocalization Ratios over time (β=0.03 for VABS-2, p = < .001;

β=0.04 for MSEL, *p* = < .001; and β=0.01 for MSDQ; *p* = < .001). On the contrary, as expected SCQ scores were inversely associated with the Vocalization Ratio measure (β=-0.03; *p* = .03).

Vocalization Trajectory Subgroups

Our third hypothesis was that we would observe sub-group differences in Child Vocalizations and Vocalization Ratio trajectories. Vocalization Ratios increased across tertiles of baseline language skills (VABS-II EL: β=0.28, *p* = 0.04; MSEL EL: β=0.28, *p* =0.03 and MSEL DQ: β=0.24, *p* = 0.04). Results for Child Vocalizations however, showed an inverse association across MSEL DQ tertiles (β=-0.45, *p* = 0.01) and no significant association across other language groups. As presented in Figure 3, a near consistent pattern was observed in relation to Child Vocalizations, whereby children with lower expressive language skills (VABS-II and MSEL), lower cognition (MSEL DQ), and higher ASD characteristics (SCQ total score) at baseline demonstrated higher rates of vocalization at most time points compared to peers with contrasting profiles.

[Insert Figure 3 about here]

As presented in Figure 4, Vocalization Ratio data were also broadly consistent with our third hypothesis, with children with lower expressive language skills, lower cognition, and higher ASD severity at baseline having lower Vocalization Ratios (i.e., more non-speech) than children with contrasting profiles. However, overlap of subgroups is evident for some time point estimates. Based on visual inspection, a comparison of the 1st and 3rd tertiles provides the strongest contrast.

[Insert Figure 4 about here]

Discussion

The first objective of this study was to examine the communication trajectories of children with ASD receiving comprehensive early intervention, in terms of possible changes in vocalizations assessed using LENA. Consistent with Bak et al. (2017) we assessed children's communication at multiple time points over the course of the year to gain a fine grained understanding of the profile of any changes. We found a significant time trend for both Child Vocalization and Vocalization Ratio measures. However, we did not observe a uniform increase on either measure, but rather a waxing and waning pattern. It is possible that this pattern reflects individual and environmental factors that may be clinically meaningful, but not accounted for in our analyses. For instance, the rise in Vocalization Count from Month 1 to 4 may reflect children becoming increasingly confident (and thus vocal) in the new classroom environment, whereas the relative decline from Months 4-9 could be linked to children becoming more attuned to, and interested in, the communication of others, and thus listening more and vocalising less often. Clearly, these and other possible factors may be the focus of future research.

Nevertheless, with respect to our study hypothesis, our finding of trends, but not a significant uniform increase in Child Vocalizations is consistent with that of Bak et al. (2017), albeit in this study in a larger and younger sample of children. Similarly, the Vocalization Ratio measure, although presenting with a significant time trend (waxing and waning) did not show a uniform increase over time. We note that our finding regarding Vocalization Count contrasts with that of Dykstra et al., (2013), who reported a significant increase in Vocalization Count for 40 children with ASD receiving early intervention measured at two time points approximately 6 months apart. The difference may be accounted for by methodological differences. As illustrated in Figure 1, had we only examined data for the first and last month of recordings (as per Dykstra et al.,2013) we would have reported a mean increase of approximately one vocalization per minute, similar to that reported by Dykstra et al. In our view, the variability in our data, which is similar to that of Bak et al., (2017), suggests that caution should be exercised if basing judgements regarding change in children's vocalizations over time on just two LENA data time points. Indeed, as evidenced by the individual growth curves for each child (see supplementary material), considerable variability was observed within children over time, which may in part reflect natural within, and between days, variation in children's vocal behaviour (e.g., feeling more energetic, feeling unwell, engaged in more physical than social play during the recording period). Such variability should be considered when interpreting the findings of this and similar studies. The second objective of this study was to examine possible relationships between child characteristics at baseline (expressive language, cognitive skills, ASD symptoms) and changes in vocalizations over time. We found no significant relationships with respect to Child Vocalization, which may reflect the general lack of a consistent linear increase in vocalizations at the group level. In contrast, we found statistically significant relationships with the Vocalization Ratio, with children with higher expressive language scores on standardized measures (MSEL, VABS-II) and non-verbal cognition (MSEL DQ), and lower ASD symptoms (SCQ) demonstrating greater increases in the ratio over time. Thus, it appears that in considering LENA as an outcome measure in research or clinical practice, children's expressive language, cognition, and ASD severity at baseline may not yield sensitive information about how much change there will be in children's number of vocalizations. However, these factors may provide insight into change in the quality of vocalizations (i.e., percentage of speech to non-speech vocalizations; a finding that warrants further investigation.

The third objective was to examine possible sub-group differences in trajectories, based on child characteristics at baseline. We found significant differences in Child Vocalizations across tertiles, with the most vocal children being those with lower expressive language skills and lower cognition at baseline. The opposite effect was found with respect to the ratio measure, whereby these same groups of children had the lowest ratio of speech to non-speech vocalizations. Put simply, children with greater language and cognition needs were the most vocal, but produced a greater proportion of non-speech vocalizations. Previous authors (Bak et al., 2017; Dykstra et al., 2013; Rankine et al., 2017) have suggested that LENA data accuracy (against human transcribers), as well as interpretation of findings, may be confounded in cases where children present with high rates of echolalia or other repetitive and non-directed vocal behaviours. The Vocalization Ratio, proposed by Rankine et al., (2017), does not address the potential issue of non-directed echolalia inflating vocalization counts, given that any speech-like vocalization will still feature in the equation. However, the ratio does provide insight into the proportion of children's vocal behaviour that has a speech-like quality, with higher and improving ratios linked to more positive communication outcomes (Paul et al., 2011; Plumb & Wetherby, 2013). From a clinical perspective, the goal for children with ASD is not just to talk more, but to talk with increasing sophistication and flexibility; at the same time reducing non-speech related vocalizations such as crying. Our findings appear to support Rankine et al.,'s (2017) assertion that the Vocalization Ratio appears to offer different and valuable information regarding the quality, not just quantity, of vocalizations with relevance to clinical and research applications.

Limitations

The findings of this study must be considered with reference to several limitations. First, although analyses were based on a robust 531 hours of recordings, there was nevertheless approximately 23.1% missing data reflecting the clinical realities of embedding research in realworld clinical programs. In common with previous studies in this area, the small sample size limits the extent to which findings of the study can be interpreted for the broader population. However, with respect to statistical power, a post-hoc analysis using G*Power (Faul, Erdfeinder, Lang, & Buchner, 2007) indicated that we had 82.6% power to detect an effect size of 0.35, with our sample of 23 children and α at 0.05. Further, the use of tertiles maximized the available data for sub-typing and was theoretically driven; however, empirical subtyping (e.g., cluster analysis see Paynter et al., 2018) may have yielded differing subtypes and with a sufficient sample size may have been sensitive to detection of trajectories with more consistent patterns of growth, stability, or even regression. The fact that baseline MSEL non-verbal development quotients were significantly lower in children who had insufficient data to be included may indicate that LENA was less well tolerated by children with more complex needs, or that teachers took a more conservative approach in removing the devices if in doubt about children's comfort wearing them. In either case, future research should include more precise documentation for the reasons for missing data. To a similar end, our decision to not apply exclusion criteria based on comorbid diagnoses strengthens the external validity of the findings, but nevertheless introduced substantial heterogeneity into the sample. Finally, our decision to analyze the first three hours of each whole day recording to ensure consistency, although consistent with previous research (e.g., Bak et al., 2017; Dykstra et al., 2013), was nevertheless not consistent with the underlying LENA premise of the value of whole-day recordings and may have impacted the reliability of the measures.

Future Directions

The findings of this study contribute to a growing understanding of the benefits and limitations of using LENA to explore and evaluate changes in the expressive communication of children with ASD. Importantly, our work, in building on that of others, is beginning to demonstrate the importance of considering not just how much children vocalize, and how this may change over time, but also the quality of those vocalizations with respect to the proportion of speech-like behaviour. Research in this field would benefit from the development of a set of guidelines for consistency in data collection (e.g., number of hours, measures used) which will be essential for replication and aggregation of data across studies, including the development of online repositories to cater for secondary analyses of single and combined study samples.

Our finding of time trends, and in particular, the waxing and waning pattern of the Vocalization Ratio, suggests potential value in further research examining LENA as a measure of change over time. We recommend such studies include data collection at multiple time-points, and the pairing of LENA data with observations of behavioural changes that may be occurring to potentially understand the fluctuations over time that we observed. For instance, reductions in vocalizations may be positive for some children, if they reflect increased listening, looking to, learning from, and generally engaging with others. We also note that LENA is not intended to provide a language sample, and our findings add weight to the argument that anything short of detailed language sampling and analysis (e.g., sole reliance on vocalization counts) is likely to yield an incomplete picture of children's communication skills (Trembath et al., 2016).

Conclusion

In this study, we presented three hypotheses. Our first, that we would see a significant linear increase in children's vocalizations, and ratio of speech to non-speech vocalizations, over time was not supported. Instead, we observed a waxing and waning pattern over a 10-month period amongst the children with ASD receiving early intervention. Our second hypothesis was partially supported, in that we found significant associations between the Vocalization Ratio and children's expressive language, non-verbal cognition, and ASD characteristics at Time 1, but not with respect to Child Vocalizations. Our third hypothesis was supported, in that children with lower language and cognition at Time 1 were found to be more vocal, but present with a lower ratio of speech to non-speech vocalizations, than children with the opposite profile over the course of the study. LENA was originally designed to capture the language learning environment, and while it has potential to provide more specific insights into children's expressive language development, it is important to consider the limitations of such an approach. The efficiency with which vocalization samples can be collected in natural environments has substantial potential value in clinical and research settings. However, our findings suggest the need for care when interpreting LENA data in children with ASD, particularly with respect to the potential for fluctuating patterns in children's vocalizations - indicating the value of assessment at multiple timepoints - and the need to distinguish between quantity (the amount) and quality (e.g., flexible and directed) of vocalizations.

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Table 1. Characteristics of the Study Sample‡

Characteristics	Study Sample, N=23
Chronological Age, months	49.6 ± 10.1
Gender	
Males	17 (73.9%)
Females	6 (26.1%)
p-value	0.02*
Intake Cycle Year	
2015	12 (52.2%)
2016	3 (13.0%)
2017	8 (34.8%)
p-value	0.07
Baseline VABS EL, continuous	20.5 ± 11.8
Baseline Mullens EL, continuous	22.9 ± 11.1
Baseline Mullens DQ, continuous	64.1 ± 21.9
Baseline SCQ, continuous	17.2 ± 6.0

‡ Data presented are mean values ± SD or frequencies (%) where appropriate.

*statistically significant

Table 2. Distribution of Total Vocalisations and Ratio of Vocalisations in Study Sample‡

Time	Sample Size (N)	Total Vocalisations (Mean ± SD)	Ratio of Vocalisations (Mean ± SD)
Month 1	23	2.74 ± 1.09	1.29 ± 0.59
Month 2	19	3.21 ± 1.17	1.78 ± 1.11
Month 3	18	3.25 ± 1.45	1.49 ± 0.92
Month 4	18	3.60 ± 1.42	1.95 ± 0.63
Month 5	22	2.99 ± 1.45	1.78 ± 0.83
Month 6	19	3.09 ± 1.28	1.57 ± 0.74
Month 7	18	3.14 ± 1.83	1.54 ± 1.05
Month 8	7	3.11 ± 1.75	2.22 ± 1.24
Month 9	19	2.54 ± 1.23	1.75 ± 1.02
Month 10	14	2.81 ± 1.17	1.40 ± 0.50
p-trend		0.01*	0.01*

‡ Data presented are mean values ± SD.

*statistically significant

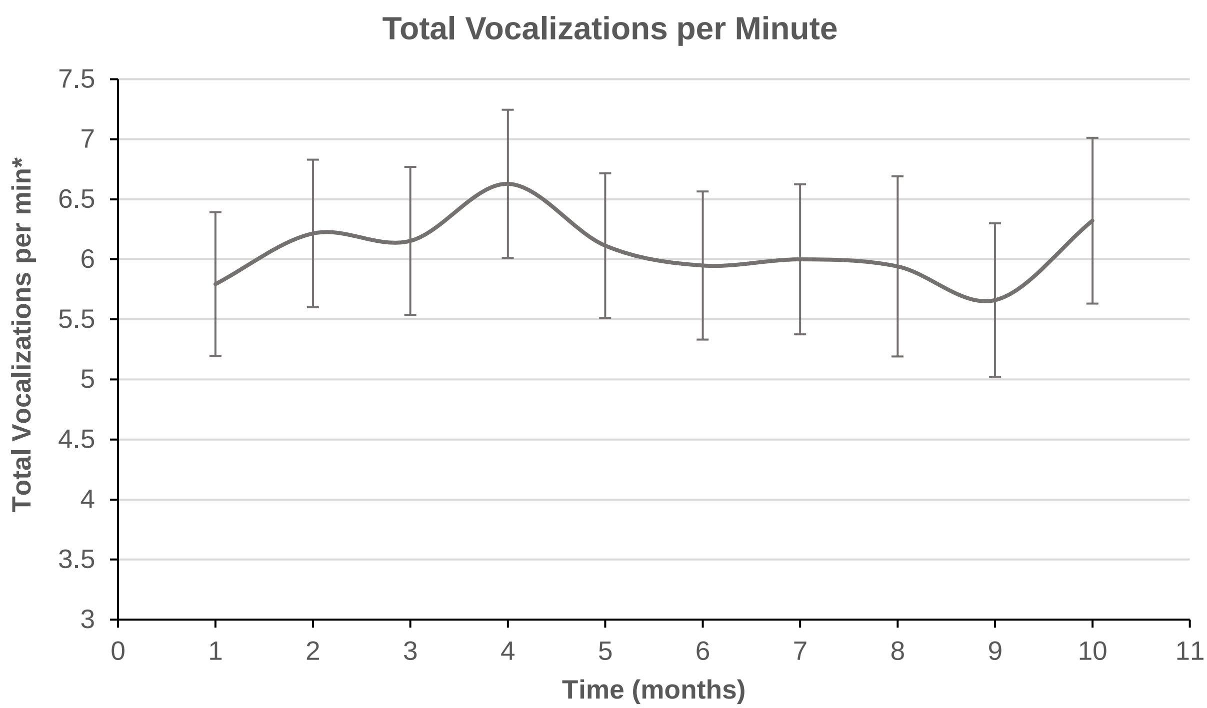
Table 3. Change in Total Vocalisations per min (TVPM) and Ratio of Speech to Non-Speech Vocalisations (RSNS VPM) over Time

Time, months	Change in mean TVPM (95% CI) *	p-value	Change in mean RSNS VPM (95% CI) *	p-value
Month 1	0 (referent)		0 (referent)	
Month 2	0.42 (0.02, 0.81)	0.04*	0.58 (0.24, 0.92)	0.009*
Month 3	0.37 (-0.12, 0.85)	0.14	0.35 (-0.05, 0.75)	0.09
Month 4	0.83 (0.30, 1.35)	0.002*	0.54 (0.12, 0.97)	0.01*
Month 5	0.31 (-0.21, 0.83)	0.24	0.49 (0.08, 0.91)	0.02*
Month 6	0.18 (-0.37, 0.73)	0.51	0.35 (-0.08, 0.78)	0.11
Month 7	0.22 (-0.34, 0.79)	0.43	0.38 (-0.06, 0.82)	0.09
Month 8	0.12 (-0.60, 0.84)	0.74	0.84 (0.26, 1.42)	0.005*
Month 9	-0.15 (-0.71, 0.41)	0.60	0.57 (0.12, 1.00)	0.01*
Month 10	0.56 (-0.05, 1.18)	0.07	0.26 (-0.22, 0.74)	0.28
p-trend	0.007*		0.02*	

*Adjusted for age, (months), gender, intake cycle year and first month recording.

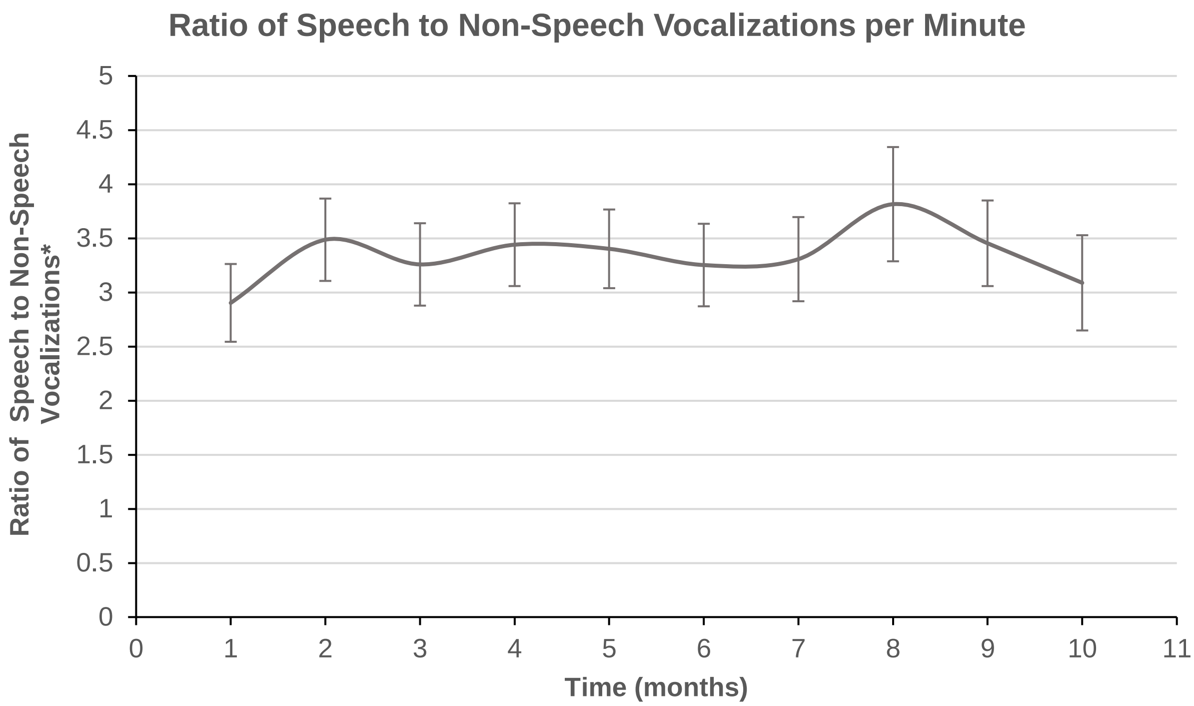
*statistically significant

Figure 1. Predicted mean total vocalizations per minute over time



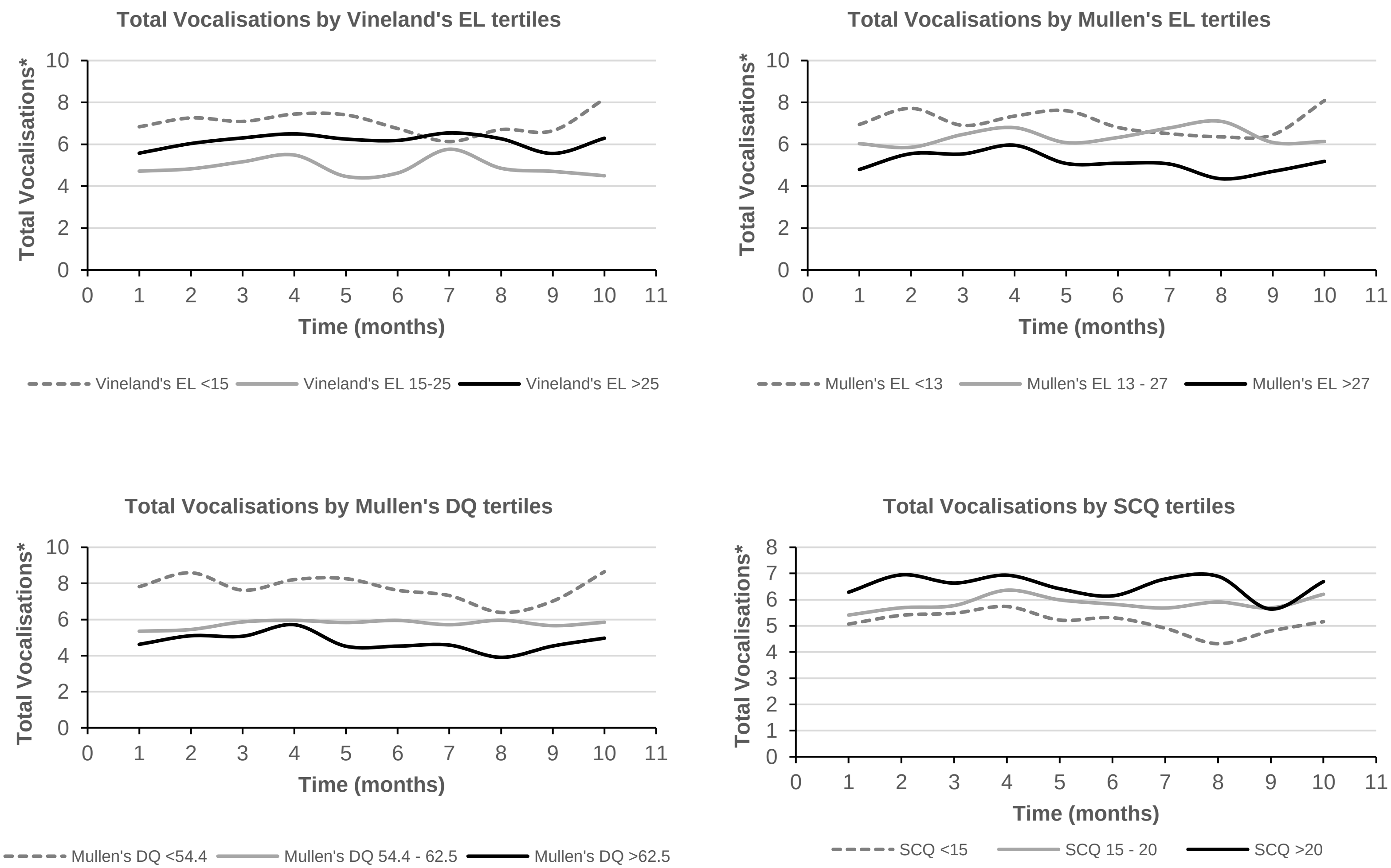
*Adjusted for chronological age, (months), gender and intake cycle year

Figure 2. Predicted mean ratio of speech to non-speech vocalizations per minute over time



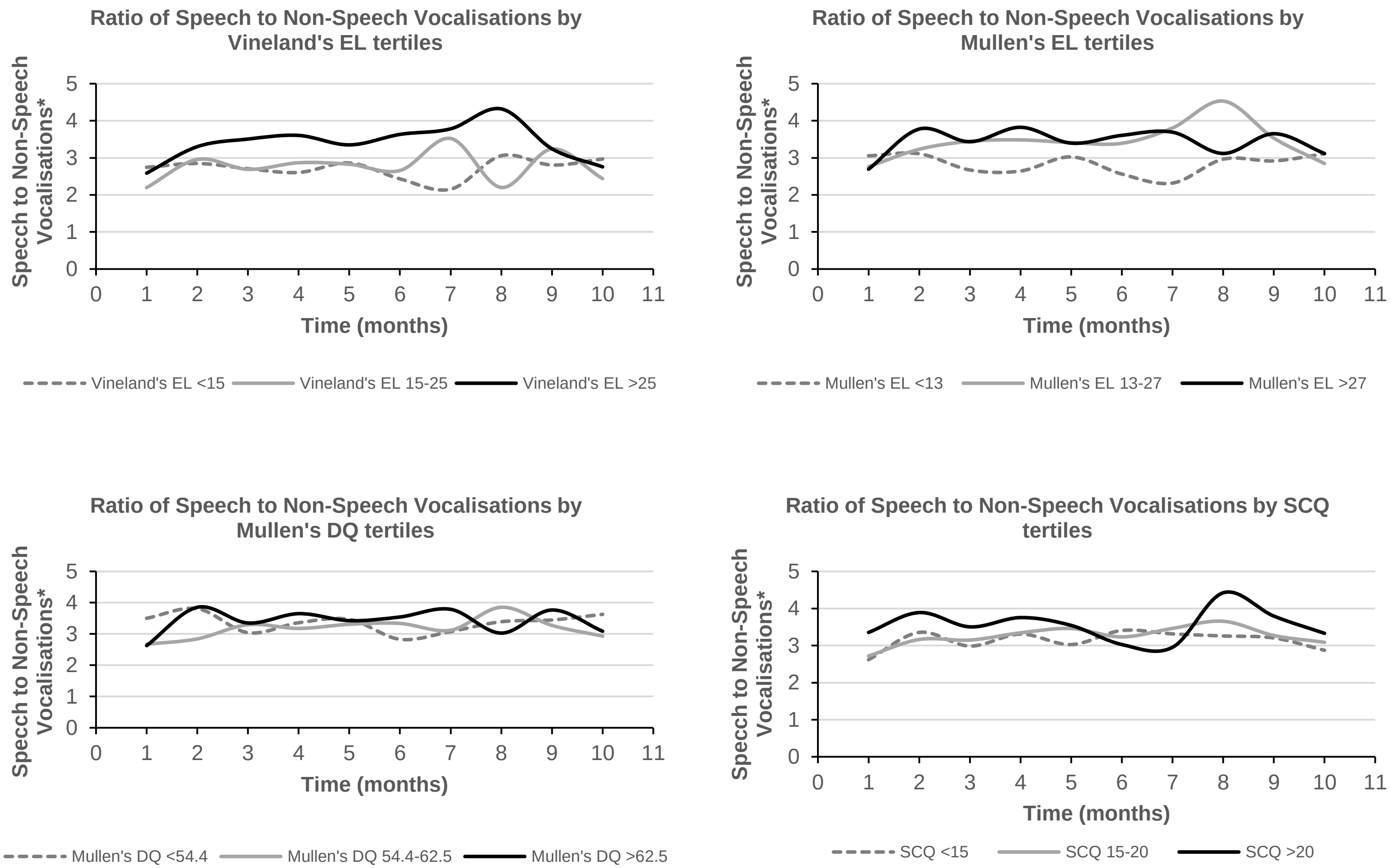
*Adjusted for chronological age, (months), gender and intake cycle year

Figure 3. Predicted mean total vocalizations per minute over time by measures of expressive language, cognition, and ASD characteristics at baseline



*Adjusted for chronological age, (months), gender and intake cycle year

Figure 4. Predicted mean ratio of speech to non-speech vocalizations per minute over time by measures of expressive language, cognition, and ASD characteristics at baseline



*Adjusted for chronological age, (months), gender and intake cycle year