Research Article

Growth of Expressive Syntax in Children With Fragile X Syndrome

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Purpose: This research explored syntactic growth in children with fragile X syndrome (FXS) over a 5-year period, and variability in growth in relation to autism symptoms, nonverbal cognition, maternal responsivity, and gender.

Method: Language samples at 4 time points from 39 children with FXS, 31 boys and 8 girls, were analyzed using the Index of Productive Syntax (Scarborough, 1990) and mean length of utterance (Brown, 1973). The degree of autism symptoms was evaluated using the Childhood Autism Rating Scale (Schopler, Reichler, & Renner, 1988) at the first time point. Maternal responsivity estimates were averaged across time points.

Results: Children with FXS showed significant syntactic growth over time and a significant plateau (quadratic trend)

in the later observations. Children who exhibited more autism symptoms at Time 1 had significantly lower syntactic abilities over time than children who exhibited fewer autism symptoms. Nonverbal cognition significantly predicted mean length of utterance scores but not Index of Productive Syntax scores. Maternal responsivity was not a significant predictor of syntactic outcomes. Girls with FXS generally demonstrated better expressive syntax than boys with FXS with notable individual differences.

Conclusion: Despite significant growth over time, expressive syntax is a vulnerable domain for children with FXS, especially for those with severe autism symptoms. Clinical implications arising from the current findings are discussed.

Delays in expressive language are a common manifestation in children with fragile X syndrome (FXS) and affect domains such as vocabulary, morphology, and syntax (for a review see Abbeduto, Brady, & Kover, 2007; Finestack, Richmond, & Abbeduto, 2009). Particular challenges with expressive syntax have been reported by several studies (Estigarribia, Roberts, Sideris, & Price, 2011; Martin, Losh, Estigarribia, Sideris, & Roberts, 2013; Price et al., 2008; Roberts et al., 2007); yet, our understanding of change over time in expressive syntax and factors predicting change is limited by the lack of longitudinal studies. The present study is the first to our knowledge to examine growth and variability in expressive syntax over a 5-year period in children with FXS.

Factors Related to Language Development in FXS

FXS is the most common inherited cause of intellectual disability, resulting from a mutation in the Fragile X

Mental Retardation 1 gene, located on the X chromosome. According to the National Fragile X Foundation, the estimated prevalence of FXS is 1 in 3,600 to 4,000 for boys and 1 in 4,000 to 6,000 for girls (National Fragile X Foundation, 2015). Individuals with FXS are reported to show poor communication and language skills, with considerable variability in language profiles (see Abbeduto et al., 2007; Finestack et al., 2009).

Gender is one factor contributing to this variability. Due to the X-linked nature of the disorder, effects on girls tend to be moderated by the presence of a second X chromosome (Crawford, Acuña, & Sherman, 2001; Hagerman & Hagerman, 2002; Lachiewicz, 1995). In terms of language abilities, evidence indicates that girls are not as delayed as boys (Abbeduto et al., 2003; Finestack & Abbeduto, 2010; Fisch et al., 1999). Girls with FXS have generally demonstrated higher receptive (Abbeduto et al., 2003) and expressive (Finestack & Abbeduto, 2010; Finestack, Sterling, & Abbeduto, 2013; Sterling & Abbeduto, 2012) language skills, and tended to acquire specific language skills (i.e., vocabulary, syntax) faster over time than boys with FXS (Pierpont, Richmond, Abbeduto, Kover, & Brown, 2011), with the exception of a small proportion of girls who also have comorbid autism.

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Comorbid autism is another factor contributing to language variability in FXS. The prevalence of autism in FXS is estimated between 25% and 50% (see McDuffie et al., 2010), and is often an indicator of greater delays in communication and language (Abbeduto et al., 2007; Lewis et al., 2006; Philofsky, Hepburn, Hayes, Hagerman, & Rogers, 2004; Roberts, Mirrett, & Burchinal, 2001; Warren, Brady, Sterling, Fleming, & Marquis, 2010). However, several studies have reported no distinct language profiles between individuals with comorbid FXS and autism and individuals with FXS only, particularly in areas of expressive language, such as morphosyntax (Estigarribia et al., 2011; Kover & Abbeduto, 2010; Lewis et al., 2006; Price et al., 2008), lexical diversity, talkativeness, and dysfluency (Kover & Abbeduto, 2010). Inconsistencies between studies have been attributed to methodological differences, such as small sample sizes, different age ranges, different diagnostic criteria, and different measures (cf. discussion in Kover & Abbeduto, 2010; Price et al., 2008), suggesting the need for further investigations.

Variability in language outcomes in FXS has been recently associated with maternal responsivity. Maternal responsivity refers to “a healthy, growth-producing relationship characterized by warmth, nurturance, and stability as well as specific behaviors, such as contingent positive responses to child initiations.” (Warren et al., 2010, p. 54). Two previous studies of children with FXS defined and measured responsivity according to the types of facilitative input and responses provided by mothers (Brady, Warren, Fleming, Keller, & Sterling, 2014; Warren et al., 2010). These studies specifically looked at how responsivity affected vocabulary outcomes. Warren et al. (2010) showed that early maternal responsivity predicted vocabulary outcomes at age 3 years. And Brady et al. (2014) showed that early and sustained responsivity over time were significant predictors of vocabulary outcomes through age 9 years. In both studies, children with more responsive mothers had significantly larger vocabularies, even after controlling for initial differences in nonverbal IQ and autism symptoms. However, the Warren et al. and Brady et al. studies did not measure responsivity effects on other aspects of language, such as expressive syntax.

Expressive Syntax in FXS

Many research studies have compared syntax in children with FXS to children who are typically developing (TD), or who have a different type of intellectual and developmental disability in order to determine if individuals with FXS have a specific profile of syntactic development. The age ranges and sample sizes for these studies are provided in Table 1.

Sudhalter, Scarborough, and Cohen (1991) found similar performance on mean length of utterance (MLU; Brown, 1973) and the Index of Productive Syntax (IPSyn; Scarborough, 1990) between their group of boys with FXS and Scarborough's (1990) TD children. Similar findings were reported in a subsequent study (Sudhalter, Maranion, & Brooks, 1992) with boys with FXS who did not differ in

the number of syntactic errors made during a sentence completion task, compared with a sample of developmentally matched TD children. It is possible that the smaller sample sizes (i.e., 19 and 11) in both studies by Sudhalter and colleagues (1991, 1992) did not allow for significant differences to emerge between FXS and TD groups.

On the other hand, Roberts et al. (2007) reported fewer words, shorter and less complex utterances in boys with FXS relative to TD boys, matched on nonverbal mental age, maternal education levels, and speech intelligibility. Likewise, Price et al. (2008) controlled for nonverbal mental age and maternal education levels, and found lower syntactic skills in boys with FXS with and without autism than in TD boys. Finestack and Abbeduto (2010) found overall lower performances by adolescents and young adults with FXS in the Developmental Sentence Scoring (Lee, 1974), compared with mental-age matched controls. In another study, Finestack et al. (2013) reported significant group differences between children and adolescents with FXS and TD children on conversational MLU. Estigarribia et al. (2011) matched their FXS groups with and without autism and TD group on nonverbal cognition, maternal education, and articulatory skill, and found that boys with FXS performed below the matched group on specific measures of noun and verb morphosyntax.

The studies reviewed above all compared expressive syntax in children with and without FXS cross-sectionally at a single point in time. Two recent studies provided data on syntactic development over time. Pierpont et al. (2011) examined the contributions of phonological and verbal working memory skills on growth of vocabulary and syntax over a 2-year period among adolescents with FXS. They reported significant relationships between the two memory domains and language growth in boys but not girls with FXS. In another study, Martin et al. (2013) reported growth over a 3-year period in vocabulary, syntactic, and pragmatic skills among boys with FXS, although their growth rate was significantly slower relative to TD boys.

Measures of Expressive Syntax

The current study focused on two measures of expressive syntax: MLU and the IPSyn. Both indices have been widely used in analyses of language samples to measure morphological and syntactic complexity in TD and clinical populations (Brown, 1973; Hewitt, Hammer, Yont, & Tomblin, 2005; Price et al., 2008; Rescorla & Turner, 2015; Rice et al., 2010; Roberts et al., 2007; Sudhalter et al., 1991).

MLU is widely used as a benchmark of early morpho-syntactic productions and as a supportive diagnostic tool for language impairment (see Eisenberg, Fersko, & Lundgren, 2001). However, concerns have been raised regarding the reliability of MLU to estimate syntactic complexity beyond the early stages of language acquisition due to the reported weak relationship between utterance length and grammatical complexity beyond an MLU of 3.0 (Klee & Fitzgerald, 1985; Rescorla, Dahlsgaard, & Roberts, 2000; Scarborough, Rescorla, Tager-Flusberg, Fowler, & Sudhalter, 1991).

Table 1. Participant characteristics from previous studies that examined expressive syntax in FXS.

Study	FXS without autism		FXS with autism		TD	
	<i>N</i>	Chronological age	<i>N</i>	Chronological age	<i>N</i>	Chronological age
Sudhalter et al. (1991)	19	5–36	n/a	n/a	15	24–48 ^a
Sudhalter et al. (1992)	11	6–41	n/a	n/a	11	4
Roberts et al. (2007)	35	34–167 ^a	n/a	n/a	27	25–79 ^a
Price et al. (2008)	36	2.9–14.4	35	3.5–14.0	46	2.1–6.6
Finestack & Abbeduto (2010)	17	11.38–21.54	n/a	n/a	21	3.61–6.66
Finestack et al. (2013)	22	10.18–16.01	n/a	n/a	27	3.11–6.77
Estigarribia et al. (2011)	35	2.10–14.4	33	3.6–13.11	46	2.1–6.7
Pierpont et al. (2011) ^b	44	10.17–16.01 ^c	n/a	n/a	n/a	n/a
Martin et al. (2013)	29	3.9–14.5 ^c	40	4.4–15.5 ^c	48	3.1–8.2 ^c

Note. FXS = fragile X syndrome; TD = typically developing; n/a = not available.

^aChronological age in months. ^bThe authors do not provide specific details about their participants who met the criteria for autism. ^cAt first visit.

The IPSyn (Scarborough, 1990) is a measure of morphological and syntactic structures from children's spontaneous language samples. The index was originally developed for children between the ages of 24 and 48 months, although it has been used beyond that age range (Hewitt et al., 2005; Oetting et al., 2010; Rescorla & Turner, 2015). The usefulness of the IPSyn as a measure of expressive syntactic abilities in TD children and various clinical populations has been shown in several studies (Price et al., 2008; Rescorla et al., 2000; Rescorla & Turner, 2015; Roberts et al., 2007; Scarborough et al., 1991). However, the index also has some limitations. The IPSyn measures the emergence of syntactic structures, rather than their probability of use in obligatory contexts (Scarborough, 1990). In addition, the development of the index was based on a sample of 15 TD children, which is too small to provide reliable normative data (Scarborough, 1990).

Current Study

Given the importance of expressive syntax in language use and communication, the disruptions that are observed among children with FXS will most likely limit their linguistic competency and affect their educational outcomes. Therefore, it is critical to further study the syntactic development of children with FXS, to identify variables associated with relatively better and worse syntactic development, and use this information to develop and investigate effective language interventions. An important limitation of many earlier studies is that they focused on static age periods, with little emphasis on the developmental progression of expressive language in FXS. In order to understand development in individuals with neurodevelopmental disorders, it is vital to conduct longitudinal studies and trace developmental trajectories across time (Karmiloff-Smith, 2012). In the present study we focus on syntactic growth, as measured by the IPSyn and MLU, in children with FXS over a 5-year period between 32 and 121 months of age, an important period in terms of complex language development. It is important to note that we also examine four potential predictors of variability in growth that were selected on the basis of past

research on language development in FXS: severity of autism symptoms, nonverbal cognition, maternal responsiveness, and gender. Identifying sources of variability can enhance our understanding of how syntax develops differently in individuals with different characteristics, and help lead to future intervention studies. This study addressed the following questions:

1. What is the nature of syntactic growth over time in children with FXS, as reflected by their performance on the IPSyn and MLU?
2. What type of strengths and weaknesses in expressive syntax do children with FXS demonstrate over time?
3. How do autism symptoms, nonverbal cognition, maternal responsiveness, and gender influence growth in expressive syntax in children with FXS?
4. What is the relationship between two measures of syntax, IPSyn and MLU, at both early and later stages of syntactic development in children with FXS?

Method

Participants

The participants for this study were 39 children, 31 boys and 8 girls, with FXS. All of the participants were part of a larger longitudinal study looking at family adaptation to FXS and the effects of maternal responsiveness on language development (see Brady et al., 2014; Warren et al., 2010). Data were collected between the years of 2004 and 2013. Participants represented a sample of convenience and were recruited from across the United States through advertisements at national conventions, use of a national research registry, and networking with the community of families who have a child with FXS. Children's chronological age ranged between 32 and 58 months ($M = 49.33$; $SD = 6.567$) at Time 1, and observations were repeated three more times for a total of four observations per child. Due to the broad age range, we did not conduct any analysis within time points, instead, we examined growth curves

across the four observations. The average maternal education level was at 15.5 years with a range from 9 to 20 years. Sixty-one percent of mothers had graduated from college, and 88% of the sample identified themselves as White.

Measures

Mullen Scales of Early Learning

Children's cognitive ability was evaluated using the Mullen Scales of Early Learning (MSEL; Mullen, 1995) at the first observation, when they were between 32 and 58 months of age. The MSEL is a standardized assessment for children between the ages of birth to 68 months. It consists of five subscales, but for our purpose we averaged age equivalent scores from the Fine Motor and the Visual Reception subscales to obtain a nonverbal age equivalent for each participant ($M = 30.27$, $SD = 9.292$; see Table 2).

Predictor Measures

The Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Renner, 1988). CARS scores used in this analysis were obtained from each child's first visit at 36 months or older and were used to evaluate the degree of autism symptoms. The CARS is a 15-item measure, and each item is rated on a scale from 1 (*within normal limits for age or developmental level*) to 4 (*severely abnormal for age or developmental level*). Scores range from 15 to 60, indicating no autistic symptoms (15–29.5), mild or moderate autistic symptoms (30–36.5), or severe autistic symptoms (37–60). For our sample, scores ranged between 16 and 36 ($M = 24.58$; $SD = 5.301$) (see Table 1), with 7 out of the 39 children having CARS scores between 30 and 36. CARS scores were arrived at by consensus between two trained graduate students who had observed the child over 2 days of assessments. The strength of this measure, and the reason we used it, was that it gave us a direct measure of autism symptomology concurrent with the collection of the other data, and thus served as more proximal measure than *autism diagnosis*.

Maternal responsivity. Maternal responsivity was measured as the occurrence of predetermined behaviors during mother–child interactions at each home visit, and was averaged across time points for the current study (see Table 2). Maternal interaction behaviors were coded on a behavior-by-behavior basis. Two graduate research assistants independently coded each videotaped observation file from the home visits using the Noldus Observer XT

software (Noldus Information Technology, 2008). Reliability between coders was assessed with Interclass Correlation Coefficients (ICCs) and ranged between .73 and .99. The coding system consisted of two levels. First, maternal behaviors were coded according to whether the mother (a) maintained, (b) redirected, or (c) introduced a topic. Then, within these categories the mother's behavior was coded as either (a) requested a verbal comply, (b) requested behavioral compliance, or (c) commented. Maternal use of gestures, recodes, communication breakdowns, and admonishments were also coded. Results from a previously completed principle components analysis (see Brady et al., 2014) indicated that the following three maternal behaviors reflected responsivity: (a) Maintain requests for verbal compliance, referring to questions or statements intended to elicit a verbal response (e.g., “Do you want peanut butter and jelly or just peanut butter?”), (b) Maintain comments (e.g., Mom says “Well done!” or “That’s the blue block.”), and (c) Maintain recodes, referring to verbal interpretation of child's communication act (e.g., Child says “ba” while pointing to a ball and mom says “You want your ball.”). Z scores were computed to indicate the frequency of each of these three behaviors relative to all participants' productions—then averages of the three z scores were computed for each dyad to use in our analyses.

Outcome Measures

Language samples. Language samples for this study were collected during home visits at four time points. The elapsed time was on average 16 to 18 months between the first and second time points, 30 to 31 months between the second and third time points, and approximately 18 months between the third and fourth time points (cf. description in Brady et al., 2014). Twenty-six of the participants had IPSyn scores at all four points, nine participants had scores only at three time points, and four participants had scores only at two time points. At the home visits, two graduate research assistants administered measures and filmed mother–child interactions. Language samples for the present study were combined from three contexts of mother–child interactions: 5 minutes of free play or craft, 10 minutes of naturalistic unstructured interaction, and 5 minutes of snack, for a total of 20 minutes. Free play was only used for observation points 1 and 2, and was replaced by an age appropriate craft activity (e.g., making

Table 2. Mean and standard deviations of child scores at first observation and of maternal responsivity across observations.

Measure	Boys ($n = 31$)			Girls ($n = 8$)		
	Mean	SD	Range	Mean	SD	Range
Chronological age in months	51.1	6.43	35–68	45.3	8.45	32–55
MSEL Nonverbal Age Equivalent	28.24	6.45	16.5–52.5	38.13	12.93	24.5–65.5
CARS total scores	25.2	4.77	16.5–36	20.2	4.08	17–29
Maternal responsivity	7.78	1.43	4.48–11.10	7.67	2.17	3.13–10.02

Note. MSEL = Mullen Scales of Early Learning; CARS = Childhood Autism Rating Scale.

paper puppets) for observation points 3 and 4. Note that we compared child MLU in free play to craft activity within the first two time points and there were no significant differences across these two contexts, $F(1, 159) = 1.05, p = .31$. Free play consisted of the mother and child playing with toys brought by the examiners or with their own toys. Naturalistic interaction occurred at each observation point and consisted of the mother and child performing routine activities together (e.g., chores, play). Snack also took place at each observation point and consisted of the dyad making and eating a snack together.

All interactions were transcribed from video recordings. Two research assistants completed transcription training and met an established criterion of at least 80% agreement for words transcribed, prior to transcribing samples used in this study. Transcripts were analyzed with the 2010 research version Systematic Analysis of Language Transcripts (Miller & Iglesias, 2012), to measure the number of different words and number of utterances produced by each child. The mean number of utterances analyzed at Time 1 was 95.33 ($SD = 10.327$, range = 63–100), at Time 2 was 98.33 ($SD = 7.414$; range = 61–100), at Time 3 was 98.88 ($SD = 4.643$; range = 77–100), and at Time 4 was 99.78 ($SD = 1.333$, range = 92–100).

IPSyn. The IPSyn was used to evaluate our participants' syntactic skills at each time point. The index contains 56 grammatical forms and four "other" items, divided in four subscales: Noun Phrases, Verb Phrases, Questions/Negations, and Sentence Structure. Scores per item range between 0 and 2; one point for each occurrence of a grammatical form, for a total of two points. For some "simple" items, automatic credit (labeled as Cr) is awarded in the presence of two instances of an "advanced" form. All item scores are summed into a maximum total of 120 points. IPSyn scores are derived on the basis of the child's first 100 intelligible utterances, with a minimum of 50 utterances required. In our sample, 31 children had at least 100 utterances at each time point, whereas eight children had between 50 and 100 utterances at a particular time point. For these eight children with corpora fewer than 100 utterances, we estimated IPSyn scores on the basis of the conversion table provided by Scarborough (1990; Table 2).

Coding reliability on the IPSyn was completed by two graduate students in speech-language pathology. An initial training established that the primary and reliability coders met a criterion of 80% agreement across three consecutively scored transcripts. Following the training, each transcript was individually coded, and 25% of the transcripts at each time point (approximately 8–9 transcripts) were randomly selected to be independently coded by both coders. Agreement for the overall score for the transcript was computed by comparing the two scores (i.e., lower score divided by higher score and multiplied by 100). Agreement ranged between 86.05% and 100%, with a mean of 93.47% ($SD = 4.20$).

MLU-Morphemes. MLU is calculated by dividing the total number of morphemes divided by the number of

utterances. Utterances counted toward MLU consisted of at least one intelligible word and potentially, some unintelligible words (marked with X); 13.18% of utterances in the entire sample were partially unintelligible. Utterances that were entirely unintelligible (marked with XXX) were not considered toward MLU. We also followed Systematic Analysis of Language Transcripts conventions to consider mazes (e.g., repetitions, reformulations) and exclude them from further analysis. Elliptical responses were counted toward MLU. Utterance boundaries were determined by intonation pattern, speaker turn, and completeness of thought.

Analysis Procedures

Modeling procedures. In order to address our question about growth over time in expressive syntax by children with FXS, overall trajectories for IPSyn scores and MLU across a 5-year period were analyzed within a multilevel modeling framework using SAS PROC MIXED. The observations for each individual in the current data can be viewed as repeated measurements (Level 1) within individuals (Level 2). Age was centered on 48 months so that all intercepts are interpreted as level of communication at 48 months. This age was chosen because it represented the earliest age where most of the participants had a data point. The multilevel modeling analysis provides fixed effects that reflect the average trajectory, and random effects that reflect the departure of individuals from the average effect. Of primary interest was whether growth in expressive syntax could be observed over time. An empty model was fit initially in order to partition the variance into between- and within-person variance and calculate the ICC. Next a growth model with linear and quadratic terms for fixed effects and random intercepts was fit to explore overall changes across time and individual differences in level of IPSyn scores and MLU.

For the IPSyn, we also modeled subscale scores over time. Models for Noun Phrases, Verb Phrases, Questions/Negations, and Sentence Structure subscales were compared with overall IPSyn growth models to determine if growth in any of these subscales significantly differed from any of the other subscales.

A secondary aim was to explore the possible influence of predictors on syntactic growth, including autism symptoms, nonverbal cognition, and maternal responsivity. In order to address this aim, CARS scores, nonverbal IQ, and maternal responsivity scores were centered such that 0 was the mean across participants to facilitate interpretation of results. A predictor model was then estimated including CARS, nonverbal IQ, and maternal responsivity as predictors of level of IPSyn scores and MLU.

In order to address how well each of our measures of expressive syntax compared with each other, we compared scores from the IPSyn and MLUs collected within each time point, using a simple scatterplot. Based on visual analysis and previous research findings (e.g., Rescorla et al., 2000; Scarborough et al., 1991), we derived two sets of correlations between the IPSyn and MLU, one for MLUs above 3.0 and one for MLUs below 3.0.

Results

Growth Over Time in Expressive Syntax

Growth Over Time in IPSyn

Figure 1 shows the IPSyn scores over time for all participating children, with boys denoted by solid lines and girls denoted by dashed lines. Initial rapid linear growth followed by a slowing of the growth rate over time with a slight decline for some children can be observed in the trajectories. There was significant linear growth over time with scores increasing on average by .55 with every month increase in age. Average IPSyn score at 48 months (the intercept age) was 39. Parameter estimates obtained from Model 1 (see Table 3) indicate that there was significant variability in intercepts (measures obtained at 48 months) across children. In addition, the ICC was .31 indicating that 31% of the variance in IPSyn scores was due to between-person differences and 69% was due to longitudinal changes over time within the person.

Fixed effects terms for intercept, linear growth, and quadratic growth were included in the initial growth model with random effects for intercept and slope. That is, the trajectory of growth with age was modeled in the fixed effects while allowing individual intercepts and slopes to vary (random effects). Restricted maximum likelihood estimation was used when evaluating random effects. Attempts to include random slopes in the model resulted in estimation problems that were most likely due to the small sample size. Therefore, only random intercepts were included in the final model.

Growth Over Time in MLU

Figure 2 shows the MLU scores over time for participating children. As was the case for IPSyn scores, initial

rapid linear growth followed by a slowing of the growth rate over time with a decline for some participants can be observed in the trajectories. Parameter estimates obtained from Model 1 indicate that there was significant variability in intercepts across children (see Table 4). In addition, the ICC was .47, indicating that 47% of the variance in MLU scores was due to between-person differences and 53% was due to longitudinal changes over time within the person.

Fixed effects terms for intercept, linear growth, and quadratic growth were included in the initial growth model with random effects for intercept and slope, and modeling was conducted using the same procedures as for IPSyn scores. Attempts to include random slopes in the model resulted in estimation problems that were most likely due to the small sample size. Therefore, only random intercepts were included in the final model. Average MLU score at 48 months (the intercept age) was 2.12 with significant variability in intercepts across participants. There was significant linear growth over time with scores increasing by .02 with every month increase in age on average. There was also a significant negative quadratic term indicating a slowing in the rate of growth each month over time.

Strengths and Weaknesses in Expressive Syntax

In order to determine if there were differences in specific aspects of syntax captured by the IPSyn, we modeled subscale scores over time. Because the maximum possible raw score varied across subscales (24 for Noun Phrases, 34 for Verb Phrases, 22 for Questions/Negations, and 40 for Sentence Structure), we examined percent of possible subscale scores rather than raw scores. The slope for percent Noun Phrases was .0033, the slope for percent Verb Phrases

Figure 1. Index of Productive Syntax (IPSyn) scores over time by gender.

Table 3. Fixed effects estimates, standard errors, and variance–covariance estimates for IPSyn scores with child and maternal predictors.

Parameter	Model 1	Model 2	Model 3
Fixed effects			
Intercept	52.06 (1.84)***	39.22 (2.19)***	39.38 (1.81)***
Level 1			
Linear		.55 (.09)***	.55 (.09)***
Quadratic		–.003 (.001)*	–.003 (.001)*
Level 2 predictors			
Mullen NV on intercept			.31 (.16)
CARS on intercept			–.94 (.35)*
Maternal responsivity on intercept			1.22 (1.00)
Random parameters			
Intercept	80.87 (30.63)**	133.91 (34.46)***	74.18 (21.05)***
Residual	178.63 (25.21)***	56.78 (8.05)***	56.97 (8.11)***
–2 log likelihood	1152.4	1042.6	1023.3
BIC	1163.4	1061.0	1052.6

Note. IPSyn = Index of Productive Syntax; NV = nonverbal; CARS = Childhood Autism Rating Scale; BIC = Bayesian information criterion.

* $p < .05$. ** $p < .01$. *** $p < .001$.

was .0038, and the slope for percent Questions/Negations was .0036. The slope for percent Sentence Structure was somewhat smaller .0024, but still significantly larger than a slope of 0. Follow-up tests using a multivariate multilevel model indicated that the slope of Sentence Structure was significantly smaller than the slope for Noun Phrase, $F(1, 38) = 7.13$, $p = .01$.

Predictors of Expressive Syntax

We included the predictors autism symptoms, non-verbal IQ, and maternal responsivity into our growth models.

These variables did not significantly relate to slopes, but were related to intercepts at 48 months.

Predictors of IPSyn Scores at 48 Months

Autism symptoms. Each child's score on the CARS was entered into the model as a part of the set of three predictors. We found that CARS scores were significantly related to IPSyn scores at 48 months (the intercept used in our model). For every one point above the average CARS score a child scored, the IPSyn score at 48 months was reduced by .94 points. This indicates that more autism symptoms were associated with lower expressive syntax scores.

Figure 2. Mean length of utterance (MLU) scores over time by gender.

Table 4. Fixed effects estimates, standard errors, and variance–covariance estimates for MLU scores with child and maternal predictors.

Parameter	Model 1	Model 2	Model 3
Fixed effects			
Intercept	2.49 (.09)**	2.12 (.11)**	2.12 (.10)**
Level 1			
Linear		.02 (.005)**	.02 (.005)**
Quadratic		–.0002 (.00008)*	–.0002 (.00008)*
Level 2 predictors			
Mullen NV on intercept			.02 (.008)*
CARS on intercept			–.04 (.02)*
Maternal responsivity on intercept			.04 (.05)
Random parameters			
Intercept	.26 (.08)**	.31 (.09)**	.18 (.05)**
Residual	.29 (.04)**	.20 (.03)**	.20 (.03)**
–2 log likelihood	278.6	273.1	225.6
BIC	289.6	280.4	254.9

Note. MLU = mean length of utterance; NV = nonverbal; CARS = Childhood Autism Rating Scale; BIC = Bayesian information criterion.

* $p < .05$. ** $p < .001$.

Nonverbal IQ. The grand mean centered nonverbal IQ was entered as part of the set of three predictors. We found that nonverbal IQ was not significantly related to the IPSyn scores at 48 months.

Maternal responsivity. The grand mean centered average maternal responsivity score obtained across the observation period was also entered as part of the set of three predictors. Average maternal responsivity scores were not significantly related to IPSyn scores at 48 months.

When all three predictors were added together to the model as level two predictors of IPSyn scores, the variance in intercepts was reduced by almost 60 points (45% reduction). Model summaries can be found in Table 3.

Predictors of MLU at 48 Months

Autism symptoms. CARS scores were significantly related to MLU at 48 months (the intercept). For every one point above the average CARS score (i.e., 30) a child scored, the MLU score at 48 months was reduced by .04 points.

Nonverbal IQ. Mullen nonverbal scores were also significantly related to MLU at 48 months. The MLU score was increased by .02 points for every point the child scored above the average Mullen nonverbal score.

Maternal responsivity. Average maternal responsivity scores were not related to MLU scores at 48 months.

When the three predictors were added together to the model as level two predictors of MLU scores, the addition reduced the variance in intercepts by .13 (42% reduction). Model summaries can be found in Table 4.

Gender Influences

Gender Influences on the IPSyn

Girls generally had higher IPSyn scores than boys, with girls accounting for the four highest trajectories (see Figure 1). However, there were not enough girls to

formally test for gender differences among participants. In order to consider how much including girls in the analysis affected the overall model, we re-ran the model excluding all girls. We found significant linear growth over time among boys with FXS with IPSyn scores increasing on average by .60 with every month increase in age. Average IPSyn score at 48 months (the intercept age) was 35. Thus, the rate of growth was slightly larger in the boys-only model, but the intercepts were also somewhat lower in the boys-only model. CARS was no longer a significant predictor of IPSyn scores at 48 months in the boys-only model. Model summaries can be found in online Supplemental Material S1.

Gender Influences on MLU

Similar to the IPSyn, girls generally had higher MLU scores than boys (see Figure 2). However, the small number of girls did not allow us to formally test for gender differences among participants. We ran separate models with boys only in order to informally determine how much the girls influenced the overall models. There was significant linear growth over time with MLU scores increasing on average by .02 with every month increase in age (the same as was observed in the full model). Average MLU score at 48 months (the intercept age) was 1.86, slightly lower than the 2.12 observed in the full sample. Model summaries can be found in online Supplemental Material S2.

Relationship Over Time Between MLU and the IPSyn

Figure 3 shows a scatterplot of the relationship of MLU and IPSyn scores. MLU and IPSyn data from all available observations were included in the graph. Based on past research showing stronger correlations between MLU and syntactic complexity when MLU was less than 3.0 (Rescorla et al., 2000; Scarborough et al., 1991) and initial visual inspection of the data, we looked at correlations

Figure 3. Relation over time between mean length of utterance (MLU) and the Index of Productive Syntax (IPSyn).

between all pairs of scores and also separate correlations for MLUs below 3.0 and above 3.0. The correlation between MLU and IPSyn when MLU was less than 3.0 was .82 and significant ($N = 108, p < .001$). The correlation between MLU and IPSyn when MLU was greater than/equal to 3.0 was .24 and not significant ($N = 31, p = .20$).

Discussion

We examined growth in expressive syntax over a 5-year period in children with FXS with chronological ages between 2 and 10 years, and variability in growth as a function of autism symptoms, nonverbal intelligence, maternal responsivity, and gender. Overall, we found a significant linear growth for both the IPSyn and MLU, and these findings coincide with previously reported findings on growth in expressive syntax among participants with FXS (Martin et al., 2013; Pierpont et al., 2011). In addition to the linear trend, there was a small but significant quadratic trend, with a slowing of the growth rate and a slight decline in later ages for some of the participants. It is possible that these trends mirror the ones observed in adaptive behavior in FXS (Hahn, Brady, Warren, & Fleming, 2015). Future studies with larger sample sizes should examine the influence of specific variables such as cognitive abilities and autistic behavior on observed syntactic declines in children with FXS.

In addition to overall growth, we examined performances across the four subscales of the IPSyn (Noun Phrases, Verb Phrases, Questions/Negations, Sentence

Structure). Significant linear growth trends over time were observed for all four subscales. However, the slope for Sentence Structure was smaller than other slopes, suggesting that children in our sample were increasing their sentence structure skills at a slower rate than other areas of syntax. These findings are not surprising as the Sentence Structure subscale contains several late-developing syntactic structures (e.g., relative clauses, *wh*- clauses, multiple verb phrases, passives) and may be especially sensitive to syntactic delays in children with FXS.

In terms of gender differences, our examination was limited to descriptions of relative performances by boys and girls, due to the low number of girls in our sample. Consistent with previous findings indicating higher language abilities in girls with FXS (Abbeduto et al., 2003; Finestack & Abbeduto, 2010; Pierpont et al., 2011), most girls in our sample demonstrated higher syntactic abilities than boys, although individual differences were noted. That is, only half of the female participants outperformed the male participants. These findings appear to be in agreement with those reported by Sterling and Abbeduto (2012) who also noted delays in expressive syntactic abilities in 10% of their female participants, despite a generally high mean in MLU. Our results showed similar growth patterns for models with boys only and for the full sample that included girls; however, the intercepts were slightly lower in the boys-only models. Thus, on average, boys are scoring lower at 48 months of age than girls but grow at a similar rate to girls.

We also examined variability in syntactic growth related to nonverbal intelligence, autism symptoms, and maternal responsivity. In terms of nonverbal intelligence, we found the Mullen nonverbal scores significantly predicted MLU. This finding coincides with the one reported by Price et al. (2008), suggesting that growth in expressive syntax is tied to nonverbal cognitive development. It is important to note that autism symptomology was found to be a significant predictor of syntactic growth in children with FXS, even after considering children's nonverbal IQ levels. Children who exhibited more autism symptoms (i.e., higher CARS scores) at the time of a child's first observation had lower scores on MLU and the IPSyn at 48 months of age than children who exhibited fewer autism symptoms (i.e., lower CARS scores). Our findings conflict with those reported by Price et al. (2008) and Estigarribia et al. (2011) who found no significant differences between expressive syntax in boys with FXS with autism compared with those with FXS without autism. Our findings also conflict with those reported by Martin et al. (2013) who found a significant effect of autism severity for pragmatic language skills over time, but not for lexical or syntactic skills. One possible explanation for this discrepancy is the fact that we used the CARS to determine the autism symptoms in our FXS sample, whereas previous reported studies used the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, Delavore, & Risi, 2001). The results from the boys-only models coincide with previously reported findings (Estigarribia et al., 2011; Price et al., 2008); CARS was

no longer a significant predictor for the IPSyn. Yet, it is likely that the effect of CARS became nonsignificant due to lower statistical power in the boys-only sample.

We did not find any significant association between maternal responsivity and syntactic growth in children with FXS. Maternal responsivity has been previously related to vocabulary outcomes in children with FXS (Brady et al., 2014; Warren et al., 2010), but it was not predictive of syntactic outcomes in the current study. That is, children with more responsive mothers did not appear to have a general advantage in syntactic development. This implies that vocabulary development in FXS groups is more sensitive to environmental influences, such as maternal input, than syntactic development. Our findings were somewhat surprising because they do not align with generally positive relationships between responsivity and syntactic development reported in many past studies completed with children with typical and atypical language development. For example, specific types of input parents provided to their TD children positively correlated with MLU (Barnes, Gutfreund, Satterly, & Wells, 1983), and noun or verb phrases (Hoff-Ginsberg, 1986; Huttenlocher, Vasilyeva, Cymerman, & Levine, 2002). Similar findings were also reported for children with language delays by Yoder (1989), who found that maternal use of informational-seeking questions (e.g., yes/no questions or *wh*-questions) predicted later use of auxiliary verbs. Specific measures of parental input appear to better predict child-specific syntactic skills than our general measure of maternal responsivity, perhaps because they account for differences in types of input such as “follow in comments” that reflect coattentional states between mother and child. Further research is needed to determine whether a relationship might exist between specific maternal behaviors and production of syntactic structures in children with FXS, rather than a general responsive interaction style.

We were interested in determining if we could replicate previous findings (Rescorla et al., 2000; Scarborough et al., 1991) that showed that the relationship between MLU and IPSyn was stronger for those with MLU less than 3.0. This distinction has potential implications for clinical decisions and interpreting research findings across studies. For example, relying on MLU as a measure of complexity may not be valid above a certain level such as 3.0. Indeed, we found a significant correlation between the two measures for MLU below 3.0, whereas the correlation weakened at higher MLU levels. These findings reflect previously suggested limitations of MLU to accurately predict syntactic skills at later stages of language development (Scarborough et al., 1991). MLU appears to overestimate syntactic development when utterances get longer but not necessarily more complex. For example, both of the following utterances taken from our language samples: (a) “want do the face now” and (b) “I know where it goes” have five morphemes; however, the second utterance is clearly more complex (e.g., *wh*- clause). Hence, the IPSyn appears to be more sensitive in detecting problems with grammatical complexity. However, our findings are not directly

comparable with those of Scarborough et al. (1991) or Rescorla et al. (2000), because unlike these earlier studies, we included partially intelligible utterances. Although only 13.18% utterances contained unintelligible words, this could result in a slightly lower MLU. In addition, the IPSyn itself may have limits to the range of interpretation. For example, Oetting et al. (2010) reported that the developmental changes in IPSyn plateau after the age of 4 years in children without intellectual disability. Future research needs to better examine the sensitivity of the index with older individuals whose language may be similar to younger children.

It should be noted that we used MLU in morphemes in our analysis and did not use MLU per C-unit as in some other studies (cf. Kover, McDuffie, Abbeduto, & Brown, 2012). Variability due to C-units versus total utterances did not appear to be an issue, because children seldom used simple compound sentences that could inflate MLU (Abbeduto, Benson, Short, & Dolish, 1995). Indeed, only 0.3% of utterances in our sample were simple compound.

Strengths and Limitations

A primary strength of the current study is the examination of syntactic growth in children with FXS over a 5-year period, a longer time period than previously reported (Martin et al., 2013; Pierpont et al., 2011). This allowed us to gain an extensive look of how FXS affects growth in expressive syntax and relative strengths and weaknesses, both of which are important questions in our attempt to better understand the language phenotype in FXS. In addition, the extent to which syntactic growth varies as a function of autism symptoms, nonverbal intelligence, and maternal responsivity is important in terms of informing clinicians on prioritizing and treating expressive language in FXS.

We also note some limitations to the current study. Because of the small number of girls with FXS in our sample, we were not able to properly examine syntactic development in girls and gender differences. In addition, although we did not find significant differences in child and maternal language after exchanging the free play context with the craft activity context, we acknowledge that it was not ideal in terms of controlling our language sampling. Moreover, the contexts used for language sampling may have limited the opportunities for children with FXS to produce advanced syntactic structures. It has been suggested that “sampling contexts vary in the extent to which they elicit the upper bound of an individual’s linguistic ability” (Kover et al., 2012, p. 1023). The contexts we sampled (e.g., conversation, free play) between the child and an adult might not support the production of complex syntactic structures. The use of narrative contexts can be an alternative in eliciting complex syntactic productions from children with FXS, because of the higher proportion of complex sentences embedded in narrative content (Abbeduto et al., 1995).

Another limitation is the absence of a comparison group. Several studies have compared performances by children with FXS to TD children and children with different

etiologies (Estigarribia et al., 2011; Martin et al., 2013; Price et al., 2008). Such comparisons allow one to identify patterns that may be associated with a particular etiology. Our focus on developmental trajectories within FXS does not allow for such comparisons or identification of phenotypic profiles. Finally, we were unable to consider any intervention effects on children's syntactic growth. Most of the children included in this sample were receiving some intervention; however, we were not able to obtain details about the amount and the type of the different interventions that may have affected some of the children's trajectories.

Clinical Implications

Several implications for assessment and intervention for children with FXS arise from the current findings. First, our findings reveal that children with FXS in general make significant progress in their syntactic abilities over time, although such progress appears to be slower for children with comorbid autism symptoms. These findings have important implications for guiding intervention practices in the area of expressive syntax for children with FXS. For example, children with comorbid FXS and autism may require more extensive interventions targeting syntactic abilities. Clinicians who have children with FXS on their caseload should consider autism symptoms as a possible variable associated with increased risk of syntactic impairments. In addition, clinicians need to consider the variability in language abilities among girls diagnosed with FXS. Given the X-linked nature of the disorder, some clinicians may not expect to find significant problems in the expressive language of girls with FXS. However, our findings showed that only half of the girls in our sample performed noticeably better than the boys, suggesting the necessity for assessing and possibly treating expressive syntax problems in girls as well as boys with FXS.

Another implication arising from the current findings is the consideration of the IPSyn in clinical practice. The IPSyn appears to provide a useful tool for the assessment of emerging expressive syntax in children with FXS. Language sampling is widely recognized as an optimum method to obtain information about a child's expressive language in natural communicative contexts, in contrast to basic standardized assessments that may not indicate specific areas of potential strengths and weaknesses in expressive syntax. The IPSyn can provide a detailed summary of the language sample relatively fast, and guide further assessment and selection of appropriate intervention goals. That is, the clinician could analyze a child's language sample with the IPSyn, identify areas of weakness (e.g., negation, complex sentences), and follow up with additional assessments (e.g., direct probing) to determine whether the child produces specific syntactic structures under obligatory contexts.

Conclusion

The syntactic abilities of children with FXS have recently received some attention; but, research on syntactic

development over time and sources of variation has been limited. The present study brings important evidence of the developmental course of expressive syntax in FXS, as the positive trajectories we observed indicate that children's expressive syntax continues to develop over time, yet, variability in growth was observed relative to autism symptoms and nonverbal intelligence. Future research is needed to compare syntactic growth rates in children with FXS with those of TD children and identify interventions that can significantly improve these rates, particularly for children who are at increased risk for poor syntax due to severe autism symptoms. Although maternal responsivity as measured in this study did not have an impact on syntactic growth, future research should consider more specific aspects of maternal responsivity, such as follow in comments and expansions, which may be related to syntax. Our findings regarding girls also suggests the need to evaluate, and if necessary, treat syntactic production in all children with FXS.

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References

- Abbeduto, L., Benson, G., Short, K., & Dolish, J. (1995). Effects of sampling context on the expressive language of children and adolescents with mental retardation. *Mental Retardation*, 33, 279–288.
- Abbeduto, L., Brady, N., & Kover, S. T. (2007). Language development and fragile X syndrome: Profiles, syndrome-specificity, and within-syndrome differences. *Mental Retardation and Developmental Disabilities Research Reviews*, 13, 36–46. doi:10.1002/mrdd.20142
- Abbeduto, L., Murphy, M. M., Cawthon, S. W., Richmond, E. K., Weissman, M. D., Karadottir, S., & O'Brien, A. (2003). Receptive language skills of adolescents and young adults with Down or fragile X syndrome. *American Journal on Mental Retardation*, 108, 149–160.
- Barnes, S., Gutfreund, M., Satterly, D., & Wells, G. (1983). Characteristics of adult speech which predict children's language development. *Journal of Child Language*, 10, 65–84.
- Brady, N., Warren, S. F., Fleming, K., Keller, J., & Sterling, A. (2014). Effect of sustained maternal responsivity on later vocabulary development in children with fragile X syndrome. *Journal of Speech, Language, and Hearing Research*, 57, 212–226. doi:10.1044/1092-4388(2013/12-0341)
- Brown, R. (1973). *A first language*. Cambridge, MA: Harvard University Press.
- Crawford, D. C., Acuña, J. M., & Sherman, S. L. (2001). FMR1 and the fragile X syndrome: Human genome epidemiology review. *Genetics in Medicine*, 3, 359–371.

- Eisenberg, S. L., Fersko, T. M., & Lundgren, C. (2001). The use of MLU for identifying language impairment in preschool children: A review. *American Journal of Speech-Language Pathology*, 10, 323–342.
- Estigarribia, B., Roberts, J. E., Sideris, J., & Price, J. (2011). Expressive morphosyntax in boys with fragile X syndrome with and without autism spectrum disorder. *International Journal of Language & Communication Disorders*, 46, 216–230. doi:10.3109/13682822.2010.487885
- Finestack, L. H., & Abbeduto, L. (2010). Expressive language profiles of verbally expressive adolescents and young adults with Down syndrome or fragile X syndrome. *Journal of Speech, Language, and Hearing Research*, 53, 1334–1348. doi:10.1044/1092-4388(2010/09-0125)
- Finestack, L. H., Richmond, E. K., & Abbeduto, L. (2009). Language development in individuals with fragile X syndrome. *Topics in Language Disorders*, 29, 133–148.
- Finestack, L. H., Sterling, A. M., & Abbeduto, L. (2013). Discriminating Down syndrome and fragile X syndrome based on language ability. *Journal of Child Language*, 40, 244–265. doi:10.1017/S0305000912000207
- Fisch, G. S., Holden, J. J., Carpenter, N. J., Howard-Peebles, P. N., Maddalena, A., Pandya, A., & Nance, W. (1999). Age-related language characteristics of children and adolescents with fragile X syndrome. *American Journal of Medical Genetics*, 83, 253–256.
- Hagerman, R. J., & Hagerman, P. J. (Eds.). (2002). *Fragile X syndrome: Diagnosis, treatment, and research*. Baltimore, MD: John Hopkins University Press.
- Hahn, L. J., Brady, N. C., Warren, S. F., & Fleming, K. K. (2015). Do children with fragile X syndrome show declines or plateaus in adaptive behavior? *American Journal on Intellectual and Developmental Disabilities*, 120, 412–432.
- Hewitt, L. E., Hammer, C. S., Yont, K. M., & Tomblin, J. B. (2005). Language sampling for kindergarten children with and without SLI: Mean length of utterance, IPSYN, and NDW. *Journal of Communication Disorders*, 38, 197–213. doi:10.1016/j.jcomdis.2004.10.002
- Hoff-Ginsberg, E. (1986). Function and structure in maternal speech: Their relation to the child's development of syntax. *Developmental Psychology*, 22, 155–163.
- Huttenlocher, J., Vasilyeva, M., Cymerman, E., & Levine, S. (2002). Language input and child syntax. *Cognitive Psychology*, 45, 337–374.
- Karmiloff-Smith, A. (2012). Forward: Development is not about studying children: The importance of longitudinal approaches. *American Journal on Intellectual and Developmental Disabilities*, 117, 87–89.
- Klee, T., & Fitzgerald, M. D. (1985). The relation between grammatical development and mean length of utterance in morphemes. *Journal of Child Language*, 12, 251–269.
- Kover, S. T., & Abbeduto, L. (2010). Expressive language in male adolescents with fragile X syndrome with and without comorbid autism. *Journal of Intellectual Disability Research*, 54, 246–265.
- Kover, S. T., McDuffie, A., Abbeduto, L., & Brown, W. T. (2012). Effects of sampling context on spontaneous expressive language in males with fragile X syndrome or Down syndrome. *Journal of Speech, Language, and Hearing Research*, 55, 1022–1038. doi:10.1044/1092-4388(2011/11-0075)
- Lachiewicz, A. M. (1995). Females with fragile X syndrome: A review of the effects of an abnormal FMR1 gene. *Mental Retardation and Developmental Disabilities Research Reviews*, 1, 292–297.
- Lee, L. (1974). *Developmental Sentence Analysis*. Evanston, IL: Northwestern University Press.
- Lewis, P., Abbeduto, L., Murphy, M., Richmond, E., Giles, N., Bruno, L., & Schroeder, S. (2006). Cognitive, language and social-cognitive skills of individuals with fragile X syndrome with and without autism. *Journal of Intellectual Disability Research*, 50, 532–545.
- Lord, C., Rutter, M., Delavore, P. C., & Risi, S. (2001). *Autism Diagnostic Observation Schedule*. Los Angeles, CA: Western Psychological Services.
- Martin, G. E., Losh, M., Estigarribia, B., Sideris, J., & Roberts, J. (2013). Longitudinal profiles of expressive vocabulary, syntax and pragmatic language in boys with fragile X syndrome or Down syndrome. *International Journal of Language & Communication Disorders*, 48, 432–443. doi:10.1111/1460-6984.12019
- McDuffie, A., Abbeduto, L., Lewis, P., Kover, S., Kim, J. S., Weber, A., & Brown, W. T. (2010). Autism spectrum disorder in children and adolescents with fragile X syndrome: Within-syndrome differences and age-related changes. *American Journal on Intellectual and Developmental Disabilities*, 115, 307–326.
- Miller, J., & Iglesias, A. (2012). Systematic Analysis of Language Transcripts (SALT) [Computer software]. Middleton, WI: SALT Software LLC.
- Mullen, E. (1995). *Mullen Scales of Early Learning: AGS edition*. Circle Pines, MN: AGS.
- National Fragile X Foundation. (2015). *Fragile X prevalence*. Retrieved from <https://fragilex.org/fragile-x/prevalence/>
- Noldus Information Technology. (2008). The Observer XT (Version 8.0) [Computer software]. Wageningen, the Netherlands: Author.
- Oetting, J. B., Newkirk, B. L., Hartfield, L. R., Wynn, C. G., Pruitt, S. L., & Garrity, A. W. (2010). Index of Productive Syntax for children who speak African American English. *Language, Speech, and Hearing Services in Schools*, 41, 328–339. doi:10.1044/0161-1461(2009/08-0077)
- Philofsky, A., Hepburn, S. L., Hayes, A., Hagerman, R., & Rogers, S. J. (2004). Linguistic and cognitive functioning and autism symptoms in young children with fragile x syndrome. *American Journal of Mental Retardation*, 109, 208–218.
- Pierpont, E. I., Richmond, E. K., Abbeduto, L., Kover, S. T., & Brown, W. T. (2011). Contributions of phonological and verbal working memory to language development in adolescents with fragile X syndrome. *Journal of Neurodevelopmental Disorders*, 3, 335–347.
- Price, J. R., Roberts, J. E., Hennon, E. A., Berni, M. C., Anderson, K. L., & Sideris, J. (2008). Syntactic complexity during conversation of boys with fragile X syndrome and Down syndrome. *Journal of Speech, Language, and Hearing Research*, 51, 3–15. doi:10.1044/1092-4388(2008/001)
- Rescorla, L., Dahlsgaard, K., & Roberts, J. (2000). Late-talking toddlers: MLU and IPSyn outcomes at 3;0 and 4;0. *Journal of Child Language*, 27, 643–664.
- Rescorla, L., & Turner, H. L. (2015). Morphology and syntax in late talkers at age 5. *Journal of Speech, Language, and Hearing Research*, 58, 434–444. doi:10.1044/2015_JSLHR-L-14-0042
- Rice, M. L., Smolik, F., Perpich, D., Thompson, T., Rytting, N., & Blossom, M. (2010). Mean length of utterance levels in 6-month intervals for children 3 to 9 years with and without language impairments. *Journal of Speech, Language, and Hearing Research*, 53, 333–349. doi:10.1044/1092-4388(2009/08-0183)

-
- Roberts, J. E., Hennon, E. A., Price, J. R., Dear, E., Anderson, K., & Vandergrift, N. A.** (2007). Expressive language during conversational speech in boys with fragile X syndrome. *American Journal on Mental Retardation, 112*, 1–17.
- Roberts, J. E., Mirrett, P., & Burchinal, M.** (2001). Receptive and expressive communication development of young males with fragile X syndrome. *American Journal of Mental Retardation, 106*, 216–230.
- Scarborough, H. S.** (1990). Index of Productive Syntax. *Applied Psycholinguistics, 11*, 1–22.
- Scarborough, H. S., Rescorla, L., Tager-Flusberg, H., Fowler, A. E., & Sudhalter, V.** (1991). The relation of utterance length to grammatical complexity in normal and language-disordered groups. *Applied Psycholinguistics, 12*, 23–46.
- Schopler, E., Reichler, R., & Renner, B.** (1988). *The Childhood Autism Rating Scale (CARS)*. Los Angeles, CA: Western Psychological Services.
- Sterling, A., & Abbeduto, L.** (2012). Language development in school-age girls with fragile X syndrome. *Journal of Intellectual Disability Research, 56*, 974–983.
- Sudhalter, V., Maranion, M., & Brooks, P.** (1992). Expressive semantic deficit in the productive language of males with fragile X syndrome. *American Journal of Medical Genetics, 43*, 65–71.
- Sudhalter, V., Scarborough, H. S., & Cohen, I. L.** (1991). Syntactic delay and pragmatic deviance in the language of fragile X males. *American Journal of Medical Genetics, 38*, 493–497.
- Warren, S., Brady, N., Sterling, A., Fleming, K., & Marquis, J.** (2010). Maternal responsivity predicts language development in young children with fragile X syndrome. *American Journal on Intellectual and Developmental Disabilities, 115*, 54–75.
- Yoder, P. J.** (1989). Maternal question use predicts later language development in specific-language-disordered children. *Journal of Speech and Hearing Disorders, 54*, 347–355.