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Syntactic Ability of Girls with Fragile X Syndrome: Phonological Memory and Discourse Demands on Complex Sentence Use

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Abstract

This study was designed to establish the extent of delay in complex sentence use by females with fragile X syndrome (FXS) and to identify sources of variability among individuals. Females with FXS (n=16; 10;2–15;7) and younger typically developing girls (n=17; 4;1–8;11) were group-wise matched on nonverbal cognition and receptive syntax. Language samples (conversation and narration) yielded syntactic complexity in terms of mean length of C-unit (MLCU) and Developmental Level sentence coding (DLevel; Rosenberg & Abbeduto, 1987). Complex syntax was not weaker than developmental expectations; however, MLCU was lower than expected for age. Phonological memory and verbal working memory correlated with measures of syntactic complexity in narration. Discourse demands may play an important role in the language produced by females with FXS.

The cause of fragile X syndrome (FXS) is a full mutation of the *FMR1* gene of the X chromosome in the form of an expanded repetition of a CGG trinucleotide sequence (Oostra & Willemsen, 2003; Verkerk et al., 1991). In males, FXS is almost invariably associated with significant language impairment and intellectual disability (Abbeduto, Brady, & Kover, 2007). Females with FXS tend to be less severely affected than males with FXS, on average, because they have two X chromosomes, only one of which contains the *FMR1* mutation (Tassone, Hagerman, Chamberlain, & Hagerman, 2000); however, variability in the extent of language and cognitive delay is even greater in females than males with FXS due to X-inactivation (one of the two X chromosomes is randomly inactivated in each cell). Approximately one-third to one-half of females with FXS fall into the range of intellectual disability, whereas others may experience more focused deficits in cognitive abilities such as attention, working memory, and executive function (Chromik et al., 2015; Cornish, Munir, & Cross, 1998; de Vries et al., 1996; Keysor & Mazzocco, 2002). There is evidence of a distinct phenotype across the lifespan for females with FXS; however, most research has addressed difficulties only in specific areas of cognition (e.g., mathematics ability) and social-emotional functioning (e.g., anxiety; Keysor & Mazzocco, 2002; Lesniak-Karpiak,

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Mazzocco, & Ross, 2003; Murphy, Mazzocco, Gerner, & Henry, 2006), with many other aspects of the phenotype, including language, relatively unexplored.

Communication is an important domain that may be vulnerable to impairment in females with FXS. Difficulty with social interaction is a notable aspect of the phenotype for females with FXS (Mazzocco, Baumgardner, Freund, & Reiss, 1998), and one likely to interfere with language learning and use. In particular, within social interactions, females with FXS exhibit more gaze aversion, task avoidance, and poorer speech quality than unaffected siblings during childhood and adolescence (Hessl, Glaser, Dyer-Friedman, & Reiss, 2006). Difficulties with pragmatics (social language use), such as noncontingent or repetitive utterances, have also been observed (Mazzocco et al., 2006; Murphy & Abbeduto, 2007; Turkstra, Abbeduto, & Meulenbroek, 2014). Many contemporary theories of language acquisition posit that language develops on the basis of multiple factors, including cognitive constraints and social-pragmatics (e.g., the emergentist coalition model; Hollich, et al., 2000); therefore, in addition to language use, there is reason to believe that structural aspects of language ability (i.e., language form–including syntax) may also be impaired in females with FXS, although the extent and nature of the impairment is unclear (Abbeduto et al., 2007).

Little research has focused on the structural language abilities of females with FXS. Results from a large scale national survey of parents of 283 females with FXS suggested that not all females acquire the ability to use complex syntax by adulthood (Bailey, Raspa, Holiday, Bishop, & Olmsted, 2009). Very few studies, however, have directly assessed, and specifically reported on, the syntactic ability in females with FXS. Abbeduto et al. (2003) examined language comprehension in terms of receptive vocabulary and syntax in six females with FXS, but, in addition to the small sample size, this study lacked a developmental-level comparison group. More recently, Oakes, Kover, and Abbeduto (2013) reported on the sentence comprehension of adolescents with FXS, including five females with FXS, using a standardized assessment of receptive syntax. Although females with FXS outperformed males with FXS, their standard scores on the TROG-2 were more than two standard deviations below the mean; however, the small sample makes any conclusions tentative. Together, these studies highlight the possibility of substantial delay in receptive syntax for females with FXS, but do not address abilities related to expressive syntax (i.e., production). The purpose of the current study was to establish the extent of delay in expressive syntax for females with FXS relative to typically developing children with similar nonverbal cognition and receptive syntax and relative to a chronological-age based comparison from a normative database, as well as to identify correlates of syntactic ability in females with FXS.

Syntactic Complexity in Typical Development

Complex syntax production–defined as the ability to construct utterances with multiple clauses–begins to emerge quite early in development (Arndt & Schuele, 2012). As skills advance, syntactic complexity becomes critical to academic and social functioning, and it continues to develop through the school-age years, adolescence, and into adulthood (Channell, Loveall, Conners, Harvey, & Abbeduto, 2018; Nippold, Frantz-Kaspar, &

Vigeland, 2017; Nippold, Hesketh, Duthie, & Mansfield, 2005). Utterance length and relative clause use during conversation, for example, increase over the period of 8 to 25 years of age (Nippold et al., 2005). Given this protracted typical developmental trajectory, examining complex syntax use for school-age and adolescent females with FXS can reveal delays or patterns of performance for even mildly affected individuals during a critical window of growth.

One measure by which syntactic complexity can be estimated is mean length of utterance (MLU). However, some data suggest that after the earliest phases of language production (e.g., beyond an MLU of 3.0), the relationship between utterance length and complexity of syntax weakens (Klee & Fitzgerald, 1985; Rescorla, Dahlsgaard, & Roberts, 2000). For example, Klee and Fitzgerald (1985) found that, in two- to three-year-old children, MLU did not correlate with age and it did not discriminate "profiles of grammatical development." Rescorla et al. (2000) found that MLU correlated with IPSyn at age three, but not age four in typical development. Scarborough et al. (1991) compared MLU and IPSyn in typically developing preschoolers and children and adolescents with Down syndrome, autism, or FXS and found that MLU after 3.0 was less closely correlated with IPSyn. Scarborough et al. also found that for those with language delays and neurodevelopmental disorders, MLU overestimated IPSyn, including FXS. The correlation between MLU and IPSyn was .92 for the cases with MLU below 3.0 but only .59 for the cases with MLU over 3.0. MLU overestimated IPSyn quite dramatically for some of these participants. Based on these data, it is critical to consider not only MLU, but also more nuanced measures of syntactic complexity to fully understand the expressive syntactic abilities of children and adolescents —especially those who are likely to have capacity for growth in complex syntax, such as females with FXS.

Syntactic Ability in Females with FXS

Studies on expressive language in females with FXS are sparse. Finestack and Abbeduto (2010) reported that five females with FXS received higher expressive language scores on Developmental Sentence Scoring (DSS; Lee, 1974) than males with FXS in that study. With an explicit focus on females, Sterling and Abbeduto (2012) reported on 21 females with FXS between 7 and 15 years of age. Of these, nine received a nonverbal IQ score below 70 on the Leiter-R Brief IQ subtests and eight more scored at least one standard deviation below the mean on that test. Language samples from conversations were used to generate MLU in morphemes for each participant. In that sample, MLU ranged from 4.00 to 8.75, which was interpreted as near age-expectations for some females, but delayed for others. The failure to include a typically developing comparison group, however, makes firm conclusions about the extent of syntactic delay characteristic of females with FXS difficult. Moreover, there is evidence that the use of a single estimate of syntactic complexity, such as MLU, might overestimate the grammatical proficiency of children or adolescents with language impairments, including FXS, especially as utterance length increases because increased length can be accomplished by stringing together many words, but with a simplistic structure (Komesidou, Brady, Fleming, Esplund, & Warren, 2017; Scarborough, Rescorla, Tager-Flusberg, Fowler, & Sudhalter, 1991).

Recently, Komesidou and colleagues (2017) took into account the limitations of MLU by studying an additional measure of expressive syntax in children with FXS, known as the Index of Productive Syntax (IPSyn; Scarborough, 1990). IPSyn is a coding scheme geared towards the developmental range of 24 to 48 months. It has utility for assessing expressive syntax in children with FXS at those developmental levels (Roberts et al., 2007; Price et al., 2008); thus, these studies have focused on younger children and on males. Komesdiou et al. (2017) included individuals from 32 to 121 months of age (roughly 3 to 10 years), with an average developmental level of 28 months at the first assessment (Komesdiou et al., 2017). Overall, they found that IPSyn and MLU increased over time; autism symptoms predicted IPSyn and MLU, and nonverbal cognition predicted MLU. Although eight females were included in the sample, no analyses focused exclusively on females or compared females to males; yet, the authors do report that only four females outperformed the males. This study also lacked a comparison group, leaving many questions about expressive syntax development in females with FXS.

Given the paucity of research on the linguistic phenotype of females with FXS, there is a need to establish the extent of delay in language ability—especially in syntax given that it is a domain vulnerable to impairment, yet critical for advanced language production—to lay the groundwork for appropriate interventions. In addition, characterizing the range of variability in language ability in multiple discourse contexts and the sources of that variability for females with FXS will be important for understanding the factors that contribute to their language development and use. In the current study, we address these issues by examining spontaneous expressive syntactic ability during conversation and narration in females with FXS in direct comparison to younger girls with typical development who were similar in terms of both nonverbal cognitive ability and receptive syntactic ability, as well as relative to a reference database of typically developing females of similar chronological ages.

Discourse Demands and Effects of Context on Syntactic Complexity

In children with and without language impairments, different discourse contexts present different demands and opportunities for complex syntax use (Abbeduto, Benson, Short, & Dolish, 1995; Miles, Chapman, & Sindberg, 2006; Nippold, 2009). Generally, narrative contexts elicit greater syntactic complexity than conversations (Dollaghan, Campbell, & Tomlin, 1990; Westerveld, Gillon, & Miller, 2004). Even among older typically developing children and adolescents, narratives tend to elicit greater mean length of C-unit and clausal density than conversation (Nippold et al., 2014), and this has been replicated in adults (Nippold et al., 2017). Taken together, these data suggest that complex language is likely to be produced when an individual is provided with opportunities to talk about multifaceted topics, such as mental states, temporally ordered events, and cause and effect—highlighting a connection between syntactic ability and more general knowledge and cognitive processes (Kover, McDuffie, Abbeduto, & Brown, 2012; Nippold, 2009).

Effects of context in individuals with FXS.

For individuals with FXS, the context in which spontaneous expressive language is elicited can have consequences on both the effective use of language for communication and structural language performance (Martin, Roberts, Helm-Estabrooks, Sideris, Assal, & Moskowitz, 2012; Kover et al., 2012). In a sample of older adolescents with FXS, including eight females, Murphy and Abbeduto (2007) found that some aspects of repetitive language, such as inappropriate topic repetition, occurred more in conversation than narration. In a similar vein, Mazzocco and colleagues (2006) found that females with FXS with average-range IQ produced fewer questions that followed from earlier utterances and fewer automatic comments than age-matched typically developing comparison participants during brief conversational encounters. These two studies highlight the demands placed on language use by social interaction and the need to carefully consider the discourse context of assessment (Hessl et al., 2006).

In terms of structural language ability, effects of context on the expression of syntactic ability have been demonstrated in adolescent males with FXS. In particular, differences in MLU between a structured conversation context and a narrative context were more pronounced in the FXS sample than in a typically developing sample, controlling for nonverbal cognitive ability (Kover et al. 2012). This finding suggests that the context for sampling expressive language could meaningfully impact the conclusions that are drawn about the language abilities of individuals with neurodevelopmental disorders and demonstrates the need to consider the demands of the language sampling task for females with FXS. In the present study, we directly assessed the complex sentence production of females with FXS in two distinct discourse contexts (conversation and narration), thereby taking into account the possibility that discourse-level factors might impact their complex syntax in important ways.

Cognitive Predictors of Linguistic Ability

A pressing issue related to understanding the language of females with FXS is identifying sources of the individual variability in structural language skills that has been documented by descriptive studies (e.g., Sterling & Abbeduto, 2012). Based on the research on typical development and those with other neurodevelopmental disorders, several specific cognitive skills have been identified as predictors of syntactic ability. Studies of aging in adults without developmental difficulties have suggested that aspects of phonological memory or working memory, indexed by digit span and backward digit span tasks, respectively, relate to expressive syntactic complexity (Kemper & Sumner, 2001). Here, we follow Pierpont et al. (2011) in distinguishing between maintenance/rehearsal (phonological memory) and manipulation (verbal working memory) of phonological representations (Baddeley, Gathercole, & Papagno, 1998; Baddeley, 2003; Gathercole, Pickering, Ambridge, & Wearing, 2004). For example, a digit span task that requires recall of digits presented auditorally would tap phonological memory; a backwards digit span task that requires recalling digits in the reverse order of their presentation is thought to tap verbal working memory (Gathercole & Pickering, 2001).

Kemper and Sumner (2001) hypothesized that auditory (phonological) memory constrains the extent to which lexical units and their hierarchical relations can be held in mind to allow for the formulation of complex sentences, such as those that contain embedded clauses. Pointing to a similar cognitive constraint on syntactic ability in adolescents with a neurodevelopmental disorder, phonological memory has been shown to be associated with the MLU of expressive language samples elicited during narration by individuals with Down syndrome (Laws, 2004). Thus, phonological memory and verbal working memory are candidate constructs for explaining individual differences in complex syntax production across development.

Extending this research to individuals with FXS, Pierpont et al. (2011) found that phonological memory and verbal working memory predicted growth in vocabulary and syntactic ability in male adolescents with FXS, assessed with standardized norm-referenced language measures. For females with FXS, the relations between these aspects of memory and language did not hold; however, Pierpont et al. interpreted the difference in findings between males and females to suggest that adolescent females with FXS were beyond the developmental period in which these memory factors contribute to language abilities, whereas the language development of adolescent males with FXS was less advanced and thus, still constrained by the limits of phonological and verbal working memory. A limitation of the Pierpont et al. study, however, was the use of only gross measures of language domains (i.e., one raw score from an omnibus standardized test of expressive syntax). Previous research has demonstrated the utility of more fine-grained measures of expressive syntax, particularly those drawn from spontaneous language samples, for detecting diagnostic group differences in language ability (Kover & Abbeduto, 2010; Levy, Gottesman, Borochowitz, Frydman, & Sagi, 2006; Price et al., 2008).

The potential role of phonological memory in shaping the language abilities of individuals with FXS is further supported by work using spontaneous language samples. One such study evaluated story retelling from a picture book in terms of narrative macrostructure (Estigarribia et al., 2011). For boys with FXS and boys with Down syndrome, word recall (a measure of phonological memory) was significantly associated with story grammar performance, controlling for nonverbal cognitive ability, syntactic complexity, and parental education. No effect of word recall performance was observed for typically developing children. Examining a different assessment context designed for the observation of autism symptoms, Estigarribia et al. (2012) found that diagnostic group, nonverbal cognitive ability, and phonological memory (assessed with nonword repetition) each independently accounted for variability in expressive syntax ability among boys with FXS, Down syndrome, or typical development. No interactions were significant among predictors, which the authors interpreted as meaning that the effects of phonological memory were similar across groups. Finally, Sterling and Abbeduto (2012) examined spontaneous language samples in their report on females with FXS; however, they reported only MLU and examined only age and nonverbal IQ as putative correlates of syntactic ability, neither of which was predictive. Overall, previous research suggests that there are considerable individual differences in structural language ability (i.e., complex syntax production) among females with FXS and that phonological memory and verbal working memory may explain some of this variability, although this has yet to be tested directly.

Research Questions

In the present study, we sought to characterize the syntactic complexity of the expressive language of females with FXS, in comparison to typical development. We used mean length of C-unit (MLCU) as a broad indicator of syntactic complexity. A C-unit is defined as an independent clause with its modifiers (Loban, 1976). The C-unit is a more appropriate unit of analysis than the "utterance" for individuals with more advanced skills (Abbeduto et al., 1995). The primary measure of complex syntactic production was based on Developmental Level (DLevel) Coding (Rosenberg & Abbeduto, 1987). We selected DLevel coding to align with the more advanced language skills of older school-age girls and adolescents with FXS. In addition to examining specific syntactic constructions, it has been used widely with individuals with neurodevelopmental disorders or intellectual disability, and it has been used to identify relationships among sources of individual variability in syntax and cognition (Cheung & Kemper, 1992; Kemper & Sumner, 2001; Lu, 2009; Snowdon, 1996).

We addressed four research questions: **(1)** Does the effect of context on syntactic complexity differ for females with FXS compared to those with typical development? We predicted that the narrative context would elicit greater complexity than conversation from both groups than would conversation. **(2)** Do females with FXS produce sentences with less syntactic complexity than would be expected for their levels of nonverbal cognitive ability and syntactic comprehension or than would be expected for their chronological ages? For MLCU and DLevel variables, we directly compared syntactic complexity in females with FXS to younger girls with typical development matched group-wise on nonverbal cognition and level of language comprehension. For MLCU, we also utilized a reference database to allow a chronological-age comparison. We hypothesized that complexity would be lower than expected based on age- and developmental-/language-level. **(3)** Are individual differences in syntactic complexity among females with FXS related to phonological memory or verbal working memory, even when accounting for nonverbal cognition? We expected phonological memory and verbal working memory to correlate with complexity, especially in narration. **(4)** Are differences in DLevel performance between conversational and narrative contexts related to phonological memory and verbal working memory? We conducted exploratory analyses to generate hypotheses about potential predictors of differential performance across discourse contexts.

Method

Participants

The data were drawn from the first annual assessment of a larger longitudinal project on language development. Sixteen females with FXS between the ages of 10;2 and 15;7 (*M* = 12.04; *SD* = 1.47) participated. Note that only one participant was over the age of 13 years. The participants with FXS displayed a range of symptoms of autism, with calibrated severity scores on the Autism Diagnostic Observation Schedule (ADOS) ranging from 1 to 9 and a range of FMRP expression as well (see Table 1 and Figure 1). Four participants with FXS met the cut-off for ASD based on updated algorithms (Gotham et al., 2007); an additional participant met on the original published algorithm for Module 4. No exclusions were made on the basis of autism symptom severity because we were interested in characterizing the broad linguistic phenotype associated with FXS in females. Twenty-three females with typical development between the ages of 3 and 8 years from the same study were available for comparison. Participants with FXS were recruited nationally; participants with typical development were recruited locally. A legal guardian provided consent and the participant gave assent prior to testing. The participant samples overlap with samples reported in other studies drawing participants from the same larger project, but these studies had different foci (Del Hoyo Soriano, Thurman, Harvey, Brown, & Abbeduto, 2018; Oakes et al., 2013; Pierpont et al., 2011; Sterling & Abbeduto, 2012; Thurman, Kover, Brown, Harvey, & Abbeduto, 2017).

Of the 23 typically developing girls who participated in the larger study, one was missing a nonverbal IQ score and both language samples, four scored over two standard deviations over the mean on nonverbal IQ (i.e., > 130), and one failed to complete one of the two language sampling activities of interest (conversation). Removing these six cases left 17 girls with typical development, 4;1 – 8;11 (*M* = 6.41; *SD* = 1.69), including three siblings. These 17 typically girls served as the developmental (i.e., nonverbal cognition- and receptive syntax-matched) comparison group.

Comparisons Between FXS and Typical Development

Developmental comparison.—This study examined performance of participants with FXS using two typically developing comparisons. The primary comparison involved matching participants with FXS group-wise to establish equivalence on nonverbal cognitive developmental level and level of syntactic comprehension to typically developing participants from the same study. These typically developing participants were directly assessed as part of this study and are described throughout the Method and Results. The purpose of this comparison was to isolate any differences in complex syntax to syntactic production, rather than nonverbal cognition or general language comprehension (see Table 1). The equivalence of these groups is described below in Results.

Chronological-age comparison.—A secondary comparison was made between participants with FXS and a normative reference database. We compared MLCU from conversation and narration samples of the females with FXS to the average MLCU produced in language samples collected under similar sampling conditions from typically developing females of overlapping chronological ages. We used the SALT normative database (Miller & Iglesias, 2015) to select a sample of transcripts to provide a chronological age comparison of MLU to the females with FXS. The comparison transcripts (26 to 179 transcripts, depending on the type narrative elicitation; i.e., student selects a story, Gillam narrative) were selected on the basis of elicitation prompt (e.g., conversation, narration), age (10 to 13 years of age), gender (female), and duration of language sample (10 minutes for conversation; entire transcript for narration). Although the reference database could not address relative DLevel performance, the purpose of this comparison was to test whether utterance length in females with FXS is shorter than expected based on chronological age, if not developmental and comprehension level. No additional data were available based on the reference database; see Results for Research Question 2.

Procedures

Participants were tested in a quiet room in a university research center. All testing for any given participant was conducted over a period of one to two days, typically with the same examiner administering all measures to any given participant.

Measures

Nonverbal cognition.—The Leiter International Performance Scales-Revised Brief IQ subtests (Roid & Miller, 1997) were administered. The Brief IQ screener yields standard scores, age-equivalents, and growth scores, the latter of which provide a metric of absolute ability akin to age-equivalents, but without the former's methodological limitations (Mervis & Klein-Tasman, 2004). Standard scores, growth scores, and age-equivalents are reported in Table 1.

Syntactic comprehension.—Receptive grammatical ability was assessed with the Test for Reception of Grammar, Version 2 (TROG-2; Bishop, 2003). Each of twenty grammatical constructions is tested in a block of four consecutive items, with the blocks increasing in developmental difficulty of the target syntactic construction as the test progresses. The TROG-2 yields standard scores and raw scores, which reflect the number of blocks passed, with a pass defined as all four items testing a particular construction being answered correctly. Data from the TROG-2 are presented in Table 1.

Expressive language samples.—Participants engaged in two structured language samples in which the content and examiner behavior was highly prescribed: conversation and narration of a wordless picture book (Kover et al., 2012). Conversations were ten minutes in length with the exception of one participant with FXS and one participant with typical development: for these two participants, the conversations were nine minutes long. The examiner (a trained female graduate-level research assistant) raised a number of topics in order from a list, lingering on each subject as long as it yielded talk from the participant. The examiner asked open-ended questions and strove to limit the amount of her talk. The examiner's introduction of topics and follow-ups were scripted. For narration, the participant told the story either of *Frog Goes to Dinner* (Mayer, 1974) or *Frog on his Own* (Mayer, 1973) as she looked at the book page by page after viewing the book silently first. Examiner scaffolding was scripted and limited to the first few pages. *Frog Goes to Dinner* was told by 9 of the 16 participants with FXS and 9 of the 17 participants with typical development. There were no significant differences on the variables of interest (MLCU, proportion of complex sentences, average syntactic complexity—each defined below) across the books within groups; two books were utilized because they were counterbalanced across visits in the longitudinal study. Narratives were complete (i.e., something relevant was said on each page of the book) for all but one (typically developing) participant, who provided a partial narrative (i.e., said something relevant on at least three-quarters of the pages of the book).

Transcription.: Language samples were audio recorded and transcribed by a highly trained and reliable individual, who was not the examiner, according to procedures from Abbeduto et al. (1995), using SALT software (Miller & Iglesias, 2012). Child language was segmented into communication units (C-units; Loban, 1976). A C-unit is defined as an independent clause and its modifiers. All transcripts were checked for accuracy by a second transcriber who provided feedback which was then reviewed and incorporated as needed by the original transcriber prior to data analysis. In addition to that checking and verification process for every transcript, independent inter-transcriber agreement was calculated in the larger project for 23 participants (4 overlapping with the current study) for a total of 39 transcripts (6 overlapping with the current study). Inter-transcriber agreement was high across participant groups and language sampling contexts, including C-unit segmentation, intelligibility, word identification, and morpheme-level variables (i.e., averaging 89%).

MLCU.: Using SALT, MLCU was defined as mean length of C-unit in morphemes for the analysis set of complete and intelligible utterances.

DLevel coding.: Each C-unit produced by the participants during conversation and narration was coded for syntactic complexity by a trained and reliable individual. The coding scheme was based on the Developmental Level system (DLevel), which was first proposed for use with older individuals with intellectual disabilities (Rosenberg & Abbeduto, 1987). In addition to the constructions of interest (D1 through D6; see below), several codes were applied that were not the focus of analysis in the present paper. We extended the published coding system to include distinctions between complete simple sentences (one-clause sentences; assigned a code of D0) and sentence fragments (utterances without a main clause; assigned a code of DFrag), given the younger ages and developmental levels of participants in the current study. Combinations of complex constructions within a single utterance received a code of D7; however, we did not analyze the presence of D7 codes because examining combinations of structures would complicate interpretability of the findings and the focus was on production of particular syntactic structures. Each sentence was also judged for grammaticality (i.e., containing grammatical errors or not) to assess the extent to which participants attempted complex grammatical forms that were in the process of being acquired (ungrammatical forms received a code of DU).

The DLevel codes of interest (D1 through D6) are presented in Table 2. These DLevel codes are thought to be ordered in an approximate developmental sequence: early-acquired complex sentence constructions receive a score of D1 and the most complex receive a score of D6. The DLevel variables of interest were proportions of syntactically complex C-units and average level of complexity. We calculated the proportion of complex C-units (i.e., D1 through D6 altogether) relative to the total number of linguistic utterances produced: fragments, simple sentences, or complex sentences (i.e., D1 through D6 altogether). In addition to this proportion, we calculated an average complex sentence DLevel score. The average syntactic complexity score was calculated as a weighted average of the complex C-units produced based on their coded level (e.g., the number of D1 utterances + 2 × the number of D2 utterances + …+ 6 × the number of D6 utterances/total complex utterances). This average syntactic complexity score has been used extensively as an indication of level of expressive syntactic ability (Cheung & Kemper, 1992; Kemper, Marquis, & Thompson, 2001; Kemper, Rice, & Chen, 1995; Lu, 2009; Snowdon, 1996).

For these two DLevel variables (i.e., proportion complex; average syntactic complexity), a score was calculated separately for conversation and for narration, as well as for both language samples combined (hereafter, *total* score). That is, variables of interest included: proportion of complex sentences (conversation, narration, total) and average syntactic complexity (conversation, narration, total). Finally, to address the role of discourse demands associated with language sampling context, we also calculated an exploratory difference score between average DLevel complexity during narration and conversation. See Table 3.

Inter-rater agreement was calculated for approximately 25 percent of coded transcripts included in the current analyses, including 10 narrations (3 from participants with FXS) and 10 conversations (6 from participants with FXS). Agreement, calculated on coders' first codes for a given C-unit, was good both overall for each group, as well as when considered separately by context (kappas from .80 – 1.00 are generally interpreted as very good or excellent; Fleiss, 1981). For participants with FXS, overall kappa was .97; kappa for conversation was .97, and for narration was .95. For the participants with typical development, overall kappa was .97; kappa for conversation .97 and for narration was .96.

Putative memory predictors for participants with FXS.—Participants with FXS completed two subtests of the Working Memory Test Battery for Children (WMTB-C; Pickering & Gathercole, 2001) to assess phonological memory and verbal working memory, respectively. Although standard scores have preferred psychometric properties, limitations of the standardization WMTB-C sample (i.e., floor effects, reduced variability due to wide age bands, age bands omitted from the norming sample) would compromise the ability to detect variability; thus, standard scores are reported for descriptive purposes only (see Table 1). As dictated by the manual, for participants tested at ages not in the norming sample, standard scores were assigned based on the age band directly lower than the child's age (e.g., participants 11;9 to 12;9 received standard scores developed based on children from 10;7 to 11;8). All inferential analyses including WMTB-C performance are based on raw scores.

Phonological memory.: The Digit Recall subtest of the WMTB-C taps phonological memory and requires the participant to repeat sequences of digits of increasing length produced by the examiner. Standard scores (for descriptive purposes) and raw scores (number of digit sequences correctly recalled; for analysis) were computed.

Verbal working memory.: The WMTB-C Backward Digit Recall subtest served as a measure of verbal working memory. This subtest requires mental manipulation in addition to holding digits in mind. Standard scores (descriptive) and trials correct (number of digit sequences recalled; for analysis) were computed. The task was not scored for one participant because she was unable to understand the task requirements; as such, analyses including verbal working memory trials correct are limited to 15 participants. For three girls who were unable to repeat any sequence of numbers in a backwards order, only a raw score of zero was given because no standard scores are associated with raw scores lower than one.

Analysis Plan

Equivalence between groups (FXS, developmental comparison) by way of group-wise matching was established with *t*-tests, effects sizes, and variance ratios on the primary matching variables (Kover & Atwood, 2013). Group differences on other foundational aspects of language production between the participants with FXS and the developmental comparison group for typical development were tested using *t*-tests.

Prior to the primary research questions, we examined correlations (1) among DLevel measures of complex syntax in conversation and narration and (2) between average syntactic complexity and selected participant characteristics.

Effects of context on syntactic complexity (Research Question 1) were addressed with a Group X Context ANOVA, with follow-up *t*-tests comparing conversation and narration for each group. Extent of delay in syntactic complexity in females with FXS (Research Question 2) was addressed with *t*-tests comparing their performance to (a) the developmental-level comparison for MLCU and DLevel scores and (b) the chronological-age reference database comparison for MLCU. Phonological and verbal working memory were tested as correlates of syntactic complexity in females with FXS (Research Question 3) using Spearman's rho (r_s) with $n - 2$ degrees of freedom, with one-tailed *p*-values given that positive correlations were anticipated. These were followed up by partial correlations controlling for Leiter-R growth scores. Exploratory analyses examined the correlations among phonological and verbal working memory with syntactic complexity differences between conversation and narration (Research Question 4).

Results

Establishing Equivalence Between Groups

Developmental comparison.—Recall that participants with FXS were group-wise matched to a directly assessed comparison group of younger typically developing participants.

As expected, these groups differed on nonverbal IQ from the Leiter-R, $p < .001$, $d = 3.46$, but not absolute level of ability (i.e., Leiter-R Brief IQ growth scores), $t(31) = 0.41$, $p = .683$, $d = 0.14$, variance ratio = 0.47. The groups also did not differ in level of syntactic comprehension reflected by number of blocks passed on the TROG-2, $t(31) = 0.56$, $p = .578$, $d = 0.19$, variance ratio = 0.89. Given this, these groups can be considered sufficiently equivalent in nonverbal cognitive ability and syntactic comprehension (Kover & Atwood, 2013; Mervis & Klein-Tasman, 2004).

In addition to the group-wise matching variables, these participants with FXS and typical development were also similar in basic aspects of their expressive language. The groups did not differ significantly in the number of C-units contained in the language samples for either the total number of C-units produced or the number of complete and intelligible C-units produced in conversation, $p = .253$ and $p = .347$, respectively, or narration, $p = .486$ and $p = .487$, respectively. See Table 4. Females with FXS did not differ from the participants with typical development in total proportion of simple sentences produced in conversation, $t(31) = .44$, $p = .666$, $d = 0.15$, or narration, $t(31) = 1.32$, $p = .196$, $d = 0.46$. Although this latter result was not significant, the effect size suggests a potential decrease in simple sentence production by females with FXS during narration. The groups also did not differ in proportion of sentence fragments in conversation, $t(31) = .11$, $p = .913$, $d = 0.03$, or narration, $t(31) = 1.69$, $p = .102$, $d = 0.59$. Again, the effect size for sentence fragments in narration is worth noting, given a potential higher rate of fragments for females with FXS in this context relative to typical development. Finally, the groups did not differ in proportion of C-units with grammatical errors in conversation, $t(31) = -.14$, $p = .889$, $d = 0.05$, or narration, $t(31) = .14$, $p = .888$, $d = 0.05$.

Chronological-age comparison.—Equivalence with the chronological-age reference database comparison was established solely using selection criteria of language samples from the reference database (i.e., age; sampling context; transcript length). Thus, group-wise matching was not completed in the same manner as with the directly assessed developmental level comparison because no data were available from the reference database outside of the average MLU of the selected transcripts.

Preliminary Analyses

Correlations among DLevel measures of syntax: Proportion and average level.—For participants with FXS and those with typical development, the proportions of complex utterances in each context were not correlated with each other, $r_s = .21$, $p = .222$, and, $r_s = .02$, $p = .474$, respectively. Likewise, average syntactic complexity from conversation and narration were not significantly correlated for participants with FXS, $r_s = .03$, $p = .45$, or typical development, $r_s = .14$, $p = .298$. Because conversation and narration scores were not correlated for either group, we report analyses separately for conversation, narration, and total performance averaged across contexts for proportion of complex utterances and average syntactic complexity.

Correlations with participant characteristics.—To better understand the relationship between syntactic complexity and its foundational abilities, we examined correlations among age, nonverbal IQ, MLCU, and average DLevel syntactic complexity. Because we included chronological age and IQ, correlations were tested separately for each group. See Table 5. For the typically developing girls, MLCU in conversation and narration were each correlated with age. In contrast, for participants with FXS, age was not correlated with any scores. Instead, nonverbal IQ was related to narrative MLCU and narrative average syntactic complexity level for females with FXS. Only for participants with FXS was narrative MLCU positively correlated with narrative average syntactic complexity.

Research Question 1: Differences Between Conversation and Narration

Based on a 2 × 2 ANOVA for group and context, the interaction between context and group was not significant for MLCU, proportion of complex C-units, or average syntactic complexity score (i.e., the effect of context did not differ between groups), ps> .60. There was a main effect of context for MLCU, $F(1, 31) = 41.99$, $p < .001$, partial eta squared = .58. Both females with FXS, $t(15) = -3.54$, $p = .003$, $d = 0.89$, and typically developing girls, $t(16) = -6.25$, $p < .001$, $d = 1.51$, produced higher MLCU during narration than conversation. In contrast, the proportion of complex utterances produced did not differ between narration and conversation for females with FXS, $p = .482$, or typically developing girls, $p = .184$. The average syntactic complexity score also did not differ between narration and conversation for females with FXS, $p = .500$, or typically developing girls, $p = .305$. In summary, performance only differed significantly between conversation and narration for MLCU. The effect of context did not differ between groups.

Research Question 2: Extent of Delay

Mean length of C-unit: Developmental-level comparison.—Females with FXS and younger typically developing girls matched group-wise on nonverbal cognition and receptive syntax did not differ on MLCU during conversation, $t(31) = 0.37$, $p = .715$, $d = 0.13$, or MLCU during narration, $t(31) = .70$, $p = .488$, $d = 0.25$.

Mean length of C-unit: Chronological age-based comparison to reference database.—The mean MLU of the reference database comparison samples was compared using a one-sample *t*-test to the performance of females with FXS. In conversation, MLCU did not differ from the reference database mean of 6.69, $p = .341$; however, MLCU of the females with FXS ($M = 8.14$) during narration was lower than that produced in the reference database, regardless of the specific narrative prompt selected (student selects a story, Gillam narrative tasks, narrative retell; reference means ranged from 9.51 to 11.27), *p*s < .025. Thus, syntactic complexity as indexed by MLCU, during narration in particular, is delayed beyond age expectations in females with FXS.

DLevel performance: Developmental-level comparison.—Focusing on constructions coded D1 to D6, we directly compared the total proportion of complex sentences averaged across contexts between the females with FXS and the younger typically developing girls group-wise matched on nonverbal cognition and receptive syntax. The groups did not differ, $t(31) = 0.37$, $p = .714$, $d = 0.13$. The groups also did not differ in the proportion of complex sentences when examining conversation and narration separately, *p*s > .87. With the exception of D5 in conversation, all sentence types appeared in at least one language sample in each context in both groups. In addition, the raw frequency of each sentence construction was similar across groups with conversation and narration combined; however, participants with FXS produced more D1 utterances than those with typical development, although the difference was not significant, $t(31) = 1.64$, $p = .111$, $d = .57$; see Figure 2).

For total average level of syntactic complexity, the groups did not differ $t(31) = 0.74$, $p = .464$, $d = 0.26$. This was also true when considering conversation and narration separately, $t(31) = 1.11$, $p = .276$, $d = 0.39$ and $t(31) = 0.66$, $p = .516$, $d = 0.23$, respectively. No differences were found between groups for any DLevel measures.

In summary, the participants with FXS did not differ from the developmental comparison group in terms of MLCU or DLevel performance. In contrast, the length of unit for participants with FXS was shorter in narration than anticipated based on chronological age.

Research Question 3: Putative Predictors of Expressive Syntactic Complexity

For the females with FXS, we examined the relationship between the dependent measures of interest and the putative predictors: phonological memory (digit recall) and verbal working memory (backward digit recall). Correlations are presented in Table 6. In the narrative context, MLCU was significantly correlated with both digit recall score and backward digit recall score. See Figure 3. In the conversation context, the correlations were not significant.

Regarding DLevel performance, digit recall raw scores positively correlated with the proportion of complex sentences in narration and with total proportion of complex sentences averaged across both contexts. Backward digit raw score correlated with proportion of complex sentences in narration. In addition, average syntactic complexity during narration was positively correlated with digit recall and backward digit recall. Again, see Figure 3.

When controlling Leiter-R growth scores, the partial correlations between MLCU narrative and digit recall, $r_{s,ab.c} = .69$, $p = .002$, and backward digit recall, $r_{s,ab.c} = .82$, $p < .001$, remained significant. Total proportion of complex utterances was correlated with digit recall, $r_{s,ab.c} = .46$, $p = .042$, controlling nonverbal cognition.

In summary, phonological and verbal working memory positively relate to MLCU and DLevel proportion of complex sentences, especially in narration, even when controlling for level of nonverbal ability in females with FXS.

Research Question 4: Correlates of Differences Between Contexts (Exploratory Analyses)

Given that discourse demands could have important effects on language, we examined the score representing the difference in DLevel average syntactic complexity between narration and conversation for females with FXS. The difference score was positively correlated with MLCU during narration for females with FXS, $r_s(14) = .67$, $p = .002$. Again, see Table 5.

The differences in DLevel average syntactic complexity between contexts was significantly positively related to Digit Recall raw scores, r_s (14) = 0.66, $p = .003$, and Backward Digit Recall raw scores, $r(13) = 0.61$, $p = .008$. Again, see Table 6. When controlling for nonverbal cognition, the results were similar: $r_{s,ab.c} = .56$, $p = .015$ and $r_{s,ab.c} = .54$, $p = .024$, for digit recall and backward digit recall, respectively. The distribution of difference scores in relation to digit recall raw scores is depicted in Figure 4.

In summary, the difference in DLevel syntactic complexity between narration and conversation was positively related to phonological and verbal working memory for females with FXS, even when controlling for nonverbal cognitive ability.

Discussion

The purpose of this study was to establish the extent of delay in, and the effects of context on, the use of complex syntactic structures in spontaneous language in females with FXS, as well as to identify predictors of individual variability in syntactic complexity.

Presence of Delay in Expressive Syntactic Complexity

In direct comparison to younger typically developing girls with similar nonverbal cognitive skills and receptive syntactic ability, we failed to detect weaknesses in average syntactic complexity for females with FXS based on either MLCU or DLevel performance. However, the results of this study do not suggest that the use of complex syntax is unimpaired in females with FXS. In comparison to similar-age typically developing girls, females with FXS produced language with lower mean length (lower MLCU), relative to a reference database of typically developing children of similar chronological ages (Miller & Iglesias, 2015). That is, the spoken language of females with FXS was comprised of utterances significantly shorter than would be expected on the basis of their age. These limitations in utterance length could have implications for specific aspects of complexity as well. Although an age-matched sample of transcripts was not available for D-Level coding, we speculate that a comparison of DLevel scores with age-matched adolescents would reveal less frequent use of complex language and perhaps a less varied repertoire of complex grammatical forms. Previous literature has shown wide variability in the receptive and expressive syntactic skills of females with FXS, with some skills falling below age-expectations for many individuals (Oakes et al., 2013; Sterling & Abbeduto, 2012). Thus, our findings suggest that females with FXS, as a group, experience significant delays in expressive syntax, but those delays are in line with the delays observed in other facets of language and cognition.

In the context of the broader FXS phenotype, expressive syntax is severely impaired in males with FXS (Abbeduto, Brady, & Kover, 2007). For example, syntactic complexity as indexed by MLCU is lower in nonverbal-age matched boys with FXS than typically developing boys in conversation and narration (Kover et al., 2012). Narrative abilities may be an area of particular weakness in males with FXS (Estigarribia et al., 2011), although not all aspects of narrative macro- and micro-structure abilities are impaired relative to developmental-level expectations in individuals with FXS (Finestack, Palmer, & Abbeduto, 2012). Thus, the present findings raise the possibility that expressive syntax is less of a problem area for females with FXS than for males with FXS, although progress for both is constrained by nonverbal cognitive achievements, as is also the case for other neurodevelopmental disorders, such as Down syndrome or autism spectrum disorder (Hogan-Brown, Losh, Martin, & Mueffelmann, 2013).

Correlates of Syntactic Complexity: Phonological Memory and Verbal Working Memory

Among females with FXS, we observed that phonological memory and verbal working memory correlated with syntactic ability in narration, in terms of MLCU, the proportion of complex utterances produced, and average level of syntactic complexity. That is, the ability to hold auditory information in mind and to manipulate it (during the WMTB-C) was associated with the length and sophistication of grammatical structures produced during a narrative elicitation with picture support. In contrast, phonological memory and verbal working memory did not correlate with syntactic complexity in conversation. In an exploratory analysis, the difference between average complexity in narration and conversation was associated with phonological memory and verbal working memory in females with FXS. This pattern of findings suggests that the demands of a narrative discourse task, including the production of utterances with increased syntactic complexity in response to a wordless picture book, are supported by cognitive processing skills that involve maintenance and manipulation of auditory information.

Pierpont and colleagues (2011) found that these aspects of cognition predicted change in syntactic ability for males, but not females with FXS. In the current study, we identified a concurrent relationship between these skills and expressive syntax for females with FXS, which could be attributed to greater sensitivity of tasks geared towards spontaneous expressive language or fine-grained aspects of syntactic ability coded from such tasks (e.g., Estigarribia et al., 2012) relative to typical standardized tests of language. This also suggests that although phonological memory and verbal working memory might not be significantly contributing to language learning in females with FXS during this developmental period, they might still impact language *use*—especially when meeting particular demands of discourse. Indeed, a relationship between phonological memory and narrative production has also been identified for males with FXS. Among boys with FXS, phonological memory (short-term memory for words) is correlated with production of story grammar elements during narrative recall (Estigarribia et al., 2011). Thus, the association between phonological memory and narrative ability is a feature of the phenotype shared between males and females with FXS.

An additional factor to consider for females with FXS is the role of social anxiety and pragmatic demands on expressive language. It is possible that females with better phonological memory and verbal working memory skills were most likely to be affected by social anxiety provoked by the demands of a reciprocal conversation, depressing the syntactic complexity produced during that context. In other words, one might expect that individuals with more advanced cognitive abilities would be more attuned to a social interaction and its social demands. In fact, social cognition is predicted both by spoken language abilities and executive function in females with FXS (Turkstra et al., 2014). Social anxiety is a salient aspect of the FXS phenotype and is likely to influence social interaction, pragmatic language, and language production (Keysor & Mazzocco, 2002). Other aspects of the FXS phenotype, such as ADHD symptoms and IQ, predict social problems and socialization skills in females with FXS (Chromik et al., 2015). Much remains to be understood regarding the relationships among cognitive, linguistic, and behavioral aspects of the FXS phenotype.

Effects of Language Sampling Context

Importantly, the relationship between phonological memory and syntactic complexity would not have been observed had language been sampled only in a conversational context. Whereas conversational language samples might elicit more repetitive language in individuals with FXS (Murphy & Abbeduto, 2007), narrative tasks are more likely to elicit the upper bounds of syntactic ability (Abbeduto et al., 1995; Kover et al., 2012). In the current study, we found that MLCU was higher in narration than conversation for both groups. This aligns with a body of literature that has demonstrated similar effects of sampling context on MLU in both typically developing children and children with language impairments with or without intellectual disabilities (Abbeduto et al., 1995; Dollaghan et al., 1990). Unlike for MLCU, proportion or level of complex syntax use did not differ between conversation and narration. At the group level, overall differences in the proportion of complex utterances or the average syntactic complexity were not detected. In exploratory analyses, we found that the difference score in average syntactic complexity was possibility correlated with narrative MLCU for females with FXS, but negatively correlated with narrative and conversational MLCU for typically developing girls. This could suggest that the discourse demands function differently across groups. The limitations of MLU, particularly in failing to align with syntactic complexity per se at more advanced language levels and for individuals with FXS should be kept in mind (Scarborough et al., 1990; Klee & Fitzgerald, 1985, Rescorla et al., 2003). Overall, MLU may be an index of syntactic ability that is particularly sensitive to the demands of sampling contexts at this developmental level.

It is possible that larger language samples containing more utterances would have yielded significant differences in syntactic ability between contexts based on the DLevel variables. Nonetheless, our findings generally align with those from typically developing children and individuals with language impairment. In a study of school-age children with SLI or typical language development, it was possible to detect the use of many complex grammatical forms in both groups from a 100-utterance conversational sample; however, differences between groups were not found for the use of complex sentence attempts (Marinellie, 2004). Thus, the language samples collected in the current study were likely sufficient to allow opportunities for complex syntactic forms, but future research might address whether other language sampling contexts provide an even better developmental match for adolescent females with FXS and yield stronger effects of context (e.g., expository discourse; Heilmann et al., 2014).

Clinical Implications for Assessment

These data suggest that multiple contexts and multiple measures for assessing spoken language might be optimally informative. Clinically, taking into account the social nature of language sampling will be important for clinicians and researchers to consider as they benchmark the skills of females with FXS to the performance of typically developing individuals in the literature, especially for those individuals with FXS who experience social anxiety.

Strengths, Limitations, and Future Directions

This study is the first to report a relation between distinct cognitive abilities such as phonological memory and specific aspects of expressive syntax in females with the FXS full mutation. One strength of this study was the use of two sampling contexts and a fine-grained coding scheme designed specifically to extract information about a wide range of complex grammatical forms. It is possible, however, that other characterizations of spontaneous syntax production would yield more information regarding the strengths and weaknesses of young adolescents with FXS. That is, the DLevel coding scheme was created for older individuals and adults with mild impairments (Rosenberg & Abbeduto, 1987). Coding schemes such as those proposed by Arndt-Barako and Schuele (2012) are specifically designed to capture emerging abilities. Arndt-Barako and Schuele (2012) give credit for complex syntax that is produced without a complete sentence, unlike the DLevel coding scheme, which requires a complete sentence for an utterance to be considered for a complex syntax code. A coding scheme more sensitive to emerging syntactic abilities would be worthwhile to consider for future work on adolescents with FXS. Additional sampling contexts, such as expository discourse, which is academically relevant and related to cognitive ability, should also be explored in FXS (Lundine et al., 2018). Finally, characteristics of the participant samples (e.g., the above-average nonverbal skills of the typically developing comparison group; the sample sizes, although large relative to other studies of females with FXS) do limit the generalizability of the findings. Likely related to this is the fact that some relationships were in the opposite direction from what was expected (see Tables 5 and 6) and analyses could have been underpowered; indeed, some effect sizes were moderate despite not reaching statistical significance and this should be taken into account in interpreting the findings, given the limitations of relying exclusively upon statistical significance in interpretation.

Future research might also address predictors of language and communication ability in females with the *FMR1* premutation. There is growing evidence of a distinct cognitive phenotype in adult females with the premutation that includes both working memory impairments (Grigsby et al., 2014; Shelton et al., 2015) and impairments in pragmatic and linguistic skills – with implications for language development in their children (Klusek et al., 2016; Losh et al., 2012; Wheeler et al., 2014). In brief language samples from mothers of children with FXS who have the fragile X premutation, Sterling and colleagues (2013) have identified an increased rate of dysfluencies, hypothesized to be related to executive function difficulties associated with the phenotype. Connecting cognitive processing with language performance will offer insight into the phenotype and its developmental patterns, and may also have implications for treatment. Future research should further address how phonological memory and verbal working memory might constrain linguistic abilities in ways that vary across discourse tasks for individuals with neurodevelopmental disorders.

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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. Ment Retard, 33(5), 279–288. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/7476250 [PubMed: 7476250]

Abbeduto L, Brady N, & Kover ST (2007). Language development and fragile X syndrome: Profiles, syndrome-specificity, and within-syndrome differences. Mental Retardation and Developmental Disabilities Research Reviews, 46, 36–46. 10.1002/mrdd

Abbeduto L, Brady N, & Kover ST (2007). Language development and fragile X syndrome: profiles, syndrome-specificity, and within-syndrome differences. Mental Retardation and Developmental Disabilities Research Reviews, 13(1), 36–46. 10.1002/mrdd.20142 [PubMed: 17326110]

Abbeduto L, Murphy MM, Cawthon SW, Richmond EK, Weissman MD, Karadottir S, & O'Brien A (2003). Receptive language skills of adolescents and youong adults with Down syndrome or fragile X syndrome. American Journal on Mental Retardation, 108(3), 149–160. [PubMed: 12691594]

Arndt KB, & Schuele CM (2012). Multiclausal Utterances Aren't Just for Big Kids: A Framework for Analysis of Complex Syntax Production in Spoken Language of Preschool- and Early School-Age Children. Topics in Language Disorders, 33(2), 125–139. Retrieved from http://eric.ed.gov/?id=EJ1013676

Baddeley AD (2003). Working memory and language: an overview. Journal of Communication Disorders, 36(3), 189–206. [PubMed: 12742667]

Baddeley AD, Gathercole S, & Papagno C (1998). The phonological loop as a language learning device. Psychological Review, 105(1), 158–173. Retrieved from <Go to ISI>://000071480000007 [PubMed: 9450375]

Bailey DB, Raspa M, Holiday D, Bishop E, & Olmsted M (2009). Functional skills of individuals with fragile x syndrome: a lifespan cross-sectional analysis. Am J Intellect Dev Disabil, 114(4), 289–303. 10.1352/1944-7558-114.4.289-303 [PubMed: 19642710]

Bishop DVM (2003). Test for Reception of Grammar, Version 2. London: The Psychological Corporation.

Channell MM, Loveall SJ, Conners FA, Harvey DJ, & Abbeduto L (2018). Narrative language sampling in typical development: Implications for clinical trials. American Journal of Speech-Language Pathology, 27(1), 123 10.1044/2017_AJSLP-17-0046 [PubMed: 29222570]

Cheung H, & Kemper S (1992). Competing complexity metrics and adults' production of complex sentences. Applied Psycholinguistics, 13(01), 53 10.1017/S0142716400005427

Chromik LC, Quintin E-M, Lepage J-F, Hustyi KM, Lightbody AA, & Reiss AL (2015). The Influence of Hyperactivity, Impulsivity, and Attention Problems on Social Functioning in Adolescents and Young Adults With Fragile X Syndrome. Journal of Attention Disorders, 1087054715571739–. 10.1177/1087054715571739

Cornish KM, Munir F, & Cross G (1998). The nature of the spatial deficit in young females with Fragile-X syndrome: a neuropsychological and molecular perspective. Neuropsychologia, 36(11), 1239–1246. http://doi.org/S0028393297001620 [pii] [PubMed: 9842768]

de Vries BB, Wiegers AM, Smits AP, Mohkamsing S, Duivenvoorden HJ, Fryns JP, … Niermeijer MF (1996). Mental status of females with an FMR1 gene full mutation. American Journal of Human Genetics, 58(5), 1025–1032. [PubMed: 8651263]

Dollaghan CA, Campbell TF, & Tomlin R (1990). Video narration as a language sampling context. Journal of Speech and Hearing Disorders, 55(3), 582. [PubMed: 2381199]

Estigarribia B, Martin GE, & Roberts JE (2012). Cognitive, environmental, and linguistic predictors of syntax in fragile X syndrome and Down syndrome. Journal of Speech, Language and Hearing Research, 55(6), 1600–1612.

Estigarribia B, Martin GE, Roberts JE, Spencer A, Gucwa A, & Sideris J (2011). Narrative Skill in Boys with Fragile X Syndrome with and without Autism Spectrum Disorder. Applied Psycholinguistics, 32(2), 359–388. 10.1017/S0142716410000445 [PubMed: 21516264]

Finestack L, Palmer M, & Abbeduto L (2012). Macrostructural Narrative Language of Adolescents and Young Adults With Down Syndrome or Fragile X Syndrome. American Journal of Speech-Language Pathology, 21(1), 29 10.1044/1058-0360(2011/10-0095) [PubMed: 22049405]

Gathercole SE, Pickering SJ, Ambridge B, & Wearing H (2004). The structure of working memory from 4 to 15 years of age. Developmental Psychology, 40(2), 177–90. 10.1037/0012-1649.40.2.177 [PubMed: 14979759]

Gathercole S, & Pickering S (2001). Working Memory Test Battery for Children. Psychological Corporation.

Grigsby J, Cornish K, Hocking D, Kraan C, Olichney JM, Rivera SM, … Jacquemont S (2014). The cognitive neuropsychological phenotype of carriers of the FMR1 premutation. Journal of Neurodevelopmental Disorders, 6(1), 22 10.1186/1866-1955-6-28 [PubMed: 25170346]

Heilmann J, Malone TO, D. B, J. C, K. K, B. LL, … G. W (2014). The Rules of the Game: Properties of a Database of Expository Language Samples. Language Speech and Hearing Services in Schools, 45(4), 277 10.1044/2014_LSHSS-13-0050

Hessl D, Glaser B, Dyer-Friedman J, & Reiss AL (2006). Social behavior and cortisol reactivity in children with fragile X syndrome. J Child Psychol Psychiatry, 47(6), 602–610. 10.1111/j.1469-7610.2005.01556.x [PubMed: 16712637]

Hogan-Brown AL, Losh M, Martin GE, & Mueffelmann DJ (2013). An investigation of narrative ability in boys with autism and fragile X Syndrome. Am J Intellect Dev Disabil, 118(2), 77–94. 10.1352/1944-7558-118.2.77 [PubMed: 23464607]

Kemper S, Marquis J, & Thompson M (2001). Longitudinal change in language production: effects of aging and dementia on grammatical complexity and propositional content. Psychology and Aging, 16(4), 600–14. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/11766915 [PubMed: 11766915]

Kemper S, Rice K, & Chen Y-J (1995). Complexity metrics and growth curves for measuring grammatical development from five to ten. First Language, 15(44), 151–166. 10.1177/014272379501504402

Kemper S, & Sumner A (2001). The structure of verbal abilities in young and older adults. Psychol Aging, 16(2), 312–322. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/11405318 [PubMed: 11405318]

Keysor CS, & Mazzocco MM (2002). A developmental approach to understanding Fragile X syndrome in females. Microsc Res Tech, 57(3), 179–186. 10.1002/jemt.10070 [PubMed: 12112455]

Klusek J, McGrath SE, Abbeduto L, Roberts JE, V. BD, E. C-W, … J. YP (2016). Pragmatic Language Features of Mothers With the *FMR1* Premutation Are Associated With the Language Outcomes of Adolescents and Young Adults With Fragile X Syndrome. Journal of Speech Language and Hearing Research, 59(1), 49 10.1044/2015_JSLHR-L-15-0102

Komesidou R, Brady NC, Fleming K, Esplund A, & Warren SF (2017). Growth of Expressive Syntax in Children With Fragile X Syndrome. Journal of Speech Language and Hearing Research, 60(2), 422 10.1044/2016_JSLHR-L-15-0360

Kover ST, & Abbeduto L (2010). Expressive language in male adolescents with fragile X syndrome with and without comorbid autism. Journal of Intellectual Disability Research, 54(3), 246–265. 10.1111/j.1365-2788.2010.01255.x [PubMed: 20146742]

Kover ST, & Atwood AK (2013). Establishing equivalence: Methodological progress in group-matching design and analysis. American Journal on Intellectual and Developmental Disabilities, 118(1), 3–15. 10.1352/1944-7558-118.1.3 [PubMed: 23301899]

Kover ST, McDuffie A, Abbeduto L, & Brown WT (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(4), 1022–1038. 10.1044/1092-4388(2011/11-0075)

Laws G (2004). Contributions of phonological memory, language comprehension and hearing to the expressive language of adolescents and young adults with Down syndrome. J Child Psychol Psychiatry, 45(6), 1085–1095. 10.1111/j.1469-7610.2004.t01-1-00301.x [PubMed: 15257665]

Lesniak-Karpiak K, Mazzocco MMM, & Ross JL (2003). Behavioral assessment of social anxiety in females with Turner or fragile X syndrome. Journal of Autism and Developmental Disorders, 33(1), 55–67. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/12708580 [PubMed: 12708580]

Levy Y, Gottesman R, Borochowitz Z, Frydman M, & Sagi M (2006). Language in boys with fragile X syndrome. J Child Lang, 33(1), 125–144. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/16566323 [PubMed: 16566323]

Losh M, Klusek J, Martin GE, Sideris J, Parlier M, & Piven J (2012). Defining genetically meaningful language and personality traits in relatives of individuals with fragile X syndrome and relatives of individuals with autism. American Journal of Medical Genetics Part B: Neuropsychiatric Genetics, 159B(6), 660–668. 10.1002/ajmg.b.32070

Lu X (2009). Automatic measurement of syntactic complexity in child language acquisition. International Journal of Corpus Linguistics, 14(1), 3–28. Retrieved from http://cat.inist.fr/?aModele=afficheN&cpsidt=21236932

Lundine JP, Harnish SM, McCauley RJ, Blackett DS, Zezinka A, Chen W, & Fox RA (2018). Adolescent summaries of narrative and expository discourse: Differences and -predictors. Language Speech and Hearing Services in Schools, 49(3), 551 10.1044/2018_LSHSS-17-0105

Marinellie SA (2004). Complex syntax used by school-age children with specific language impairment (SLI) in child–adult conversation. Journal of Communication Disorders, 37(6), 517–533. 10.1016/j.jcomdis.2004.03.005 [PubMed: 15450439]

Mayer M (1973). Frog On His Own. New York: Puffin Books.

Mayer M (1974). Frog Goes to Dinner. New York: Puffin Books.

Mazzocco MMM, Baumgardner T, Freund LS, & Reiss AL (1998). Social Functioning Among Girls with Fragile X or Turner Syndrome and Their Sisters. Journal of Autism and Developmental Disorders, 28(6), 509–517. 10.1023/A:1026000111467 [PubMed: 9932237]

Mazzocco MM, Thompson L, Sudhalter V, Belser RC, Lesniak-Karpiak K, & Ross JL (2006). Language use in females with fragile X or Turner syndrome during brief initial social interactions. Journal of Developmental and Behavioral Pediatrics, 27(4), 319–328. http://doi.org/00004703-200608000-00007 [pii] [PubMed: 16906008]

Mervis CB, & Klein-Tasman BP (2004). Methodological issues in group-matching designs: α levels for control variable comparisons and measurement characteristics of control and target variables. Journal of Autism and Developmental Disorders, 34(1), 7–17. Retrieved from http://www.springeronline.com [PubMed: 15098952]

Miles S, Chapman R, & Sindberg H (2006). Sampling context affects MLU in the language of adolescents with Down syndrome. Journal of Speech, Language, and Hearing Research: JSLHR, 49(2), 325–37. 10.1044/1092-4388(2006/026)

Miller J, & Iglesias A (2012). Systematic Analysis of Language Transcripts (SALT). Middleton, WI: SALT Software, LLC.

Murphy MM, & Abbeduto L (2007). Gender differences in repetitive language in fragile X syndrome. Journal of Intellectual Disability Research, 51, 387–400. http://doi.org/DOI 10.1111/j.1365-2788.2006.00888.x [PubMed: 17391255]

Murphy MM, Mazzocco MM, Gerner G, & Henry AE (2006). Mathematics learning disability in girls with Turner syndrome or fragile X syndrome. Brain Cogn, 61(2), 195–210. http://doi.org/S0278-2626(06)00002-9 [pii]10.1016/j.bandc.2005.12.014 [PubMed: 16500739]

Nippold MA (2009). School-age children talk about chess: does knowledge drive syntactic complexity? Journal of Speech, Language, and Hearing Research: JSLHR, 52(4), 856–71. 10.1044/1092-4388(2009/08-0094)

Nippold MA, Frantz-Kaspar MW, Cramond PM, Kirk C, Hayward-Mayhew C, & MacKinnon M (2014). Conversational and narrative speaking in adolescents: examining the use of complex syntax. Journal of Speech, Language, and Hearing Research: JSLHR, 57(3), 876–86. 10.1044/1092-4388(2013/13-0097)

Nippold MA, Frantz-Kaspar MW, & Vigeland LM (2017). Spoken language production in young adults: Examining syntactic complexity. Journal of Speech Language and Hearing Research, 60(5), 1339 10.1044/2016_JSLHR-L-16-0124

Nippold MA, Hesketh LJ, Duthie JK, & Mansfield TC (2005). Conversational Versus Expository Discourse: A Study of Syntactic Development in Children, Adolescents, and Adults. Journal of Speech, Language, and Hearing Research, 48, 1048–1064.

Oakes A, Kover ST, & Abbeduto L (2013). Language comprehension profiles of young adolescents with fragile X syndrome. American Journal of Speech-Language Pathology / American Speech-Language-Hearing Association, 22(4), 615–26. 10.1044/1058-0360(2013/12-0109)

Oostra BA, & Willemsen R (2003). A fragile balance: FMR1 expression levels. Human Molecular Genetics, 12(2), 249–257.

Pickering SJ, & Gathercole SE (2001). The Working Memory Test Battery for Children. London: The Psychological Corporation.

Pierpont EI, Richmond EK, Abbeduto L, Kover ST, & Brown WT (2011). Contributions of phonological and verbal working memory to language development in adolescents with fragile X syndrome. Journal of Neurodevelopmental Disorders, 3(4), 335–47. 10.1007/s11689-011-9095-2 [PubMed: 21993552]

Price JR, Roberts JE, Hennon EA, Berni MC, Anderson KL, & Sideris J (2008). Syntactic complexity during conversation of boys with fragile X syndrome and Down syndrome. J Speech Lang Hear Res, 51(1), 3–15. 10.1044/1092-4388(2008/001) [PubMed: 18230852]

Roid GH, & Miller LJ (1997). Leiter International Performance Scale-Reviced: Examiner's Manual. Wood Dale, IL: Stoelting Co.

Rosenberg S, & Abbeduto L (1987). Indicators of Linguistic Competence in the Peer Group Conversational Behavior of Mildly Retarded-Adults. Applied Psycholinguistics, 8(1), 19–32. Retrieved from <Go to ISI>://A1987G970200002

Scarborough HS, Rescorla L, Tager-Flusberg H, Fowler AE, & Sudhalter V (1991). The Relation of Utterance Length to Grammatical Complexity in Normal and Language-Disordered Groups. Applied Psycholinguistics, 12(1), 23–45. Retrieved from <Go to ISI>://A1991FF05000002

Shelton AL, Cornish KM, Godler DE, Clough M, Kraan C, Bui M, & Fielding J (2015). Delineation of the working memory profile in female FMR1 premutation carriers: The effect of cognitive load on ocular motor responses. Behavioural Brain Research, 282, 194–200. 10.1016/j.bbr.2015.01.011 [PubMed: 25591477]

Snowdon DA (1996). Linguistic Ability in Early Life and Cognitive Function and Alzheimer's Disease in Late Life. Journal of the American Medical Association, 275(7), 528 10.1001/jama.1996.03530310034029 [PubMed: 8606473]

Sterling A, & Abbeduto L (2012). Language development in school-age girls with fragile X syndrome. Journal of Intellectual Disability Research, 56(10), 974–83. 10.1111/j.1365-2788.2012.01578.x [PubMed: 22676254]

Sterling AM, Mailick M, Greenberg J, Warren SF, & Brady N (2013). Language dysfluencies in females with the FMR1 premutation. Brain and Cognition, 82(1), 84–9. 10.1016/j.bandc.2013.02.009 [PubMed: 23523717]

Tassone F, Hagerman RJ, Chamberlain WD, & Hagerman PJ (2000). Transcription of the FMR1 gene in individuals with fragile X syndrome. Am J Med Genet, 97(3), 195–203. 10.1002/1096-8628(200023)97:3<195::AID-AJMG1037>3.0.CO;2-R [PubMed: 11449488]

Turkstra LS, Abbeduto L, & Meulenbroek P (2014). Social Cognition in Adolescent Girls With Fragile X Syndrome. American Journal on Intellectual and Developmental Disabilities, 119(4), 319–339. 10.1352/1944-7558-119.4.319 [PubMed: 25007297]

Verkerk AJ, Pieretti M, Sutcliffe JS, Fu YH, Kuhl DP, Pizzuti A, … et al. (1991). Identification of a gene (FMR-1) containing a CGG repeat coincident with a breakpoint cluster region exhibiting length variation in fragile X syndrome. Cell, 65(5), 905–914. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/1710175 [PubMed: 1710175]

Westerveld MF, Gillon GT, & Miller JF (2004). Spoken language samples of New Zealand children in conversation and narration. International Journal of Speech-Language Pathology, 6(4), 195–208. 10.1080/14417040400010140

Wheeler AC, Bailey DB, Berry-Kravis E, Greenberg J, Losh M, Mailick M, … Hagerman P (2014). Associated features in females with an FMR1 premutation. Journal of Neurodevelopmental Disorders, 6(1), 18 10.1186/1866-1955-6-30 [PubMed: 25057328]

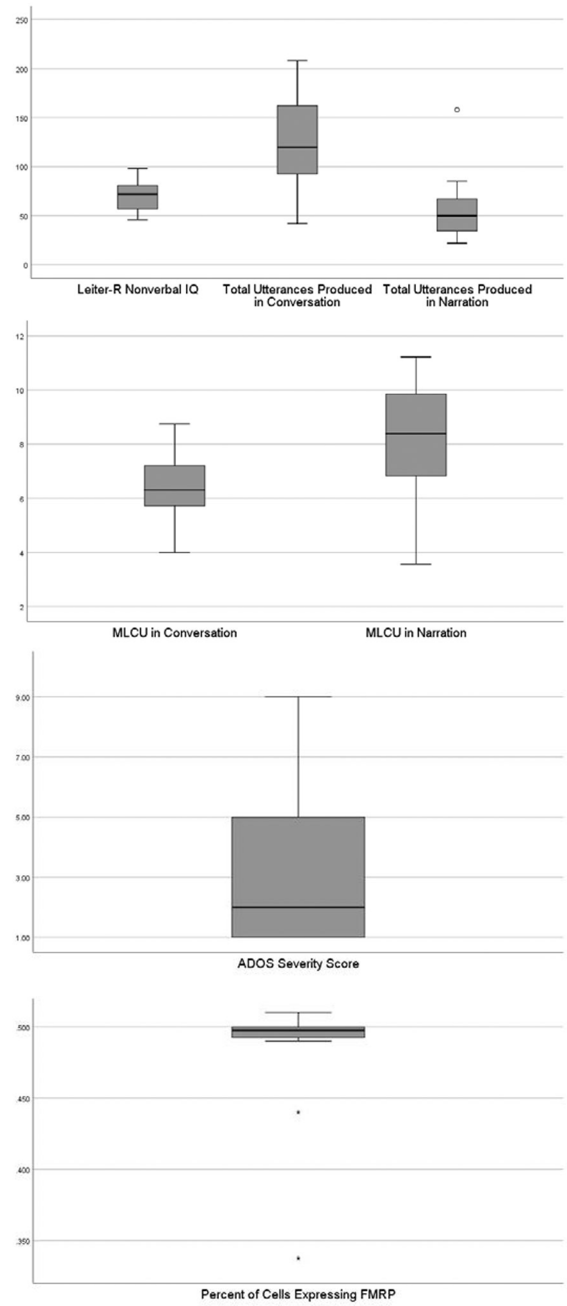

Figure 1.

Boxplots for participant characteristics for females with FXS.

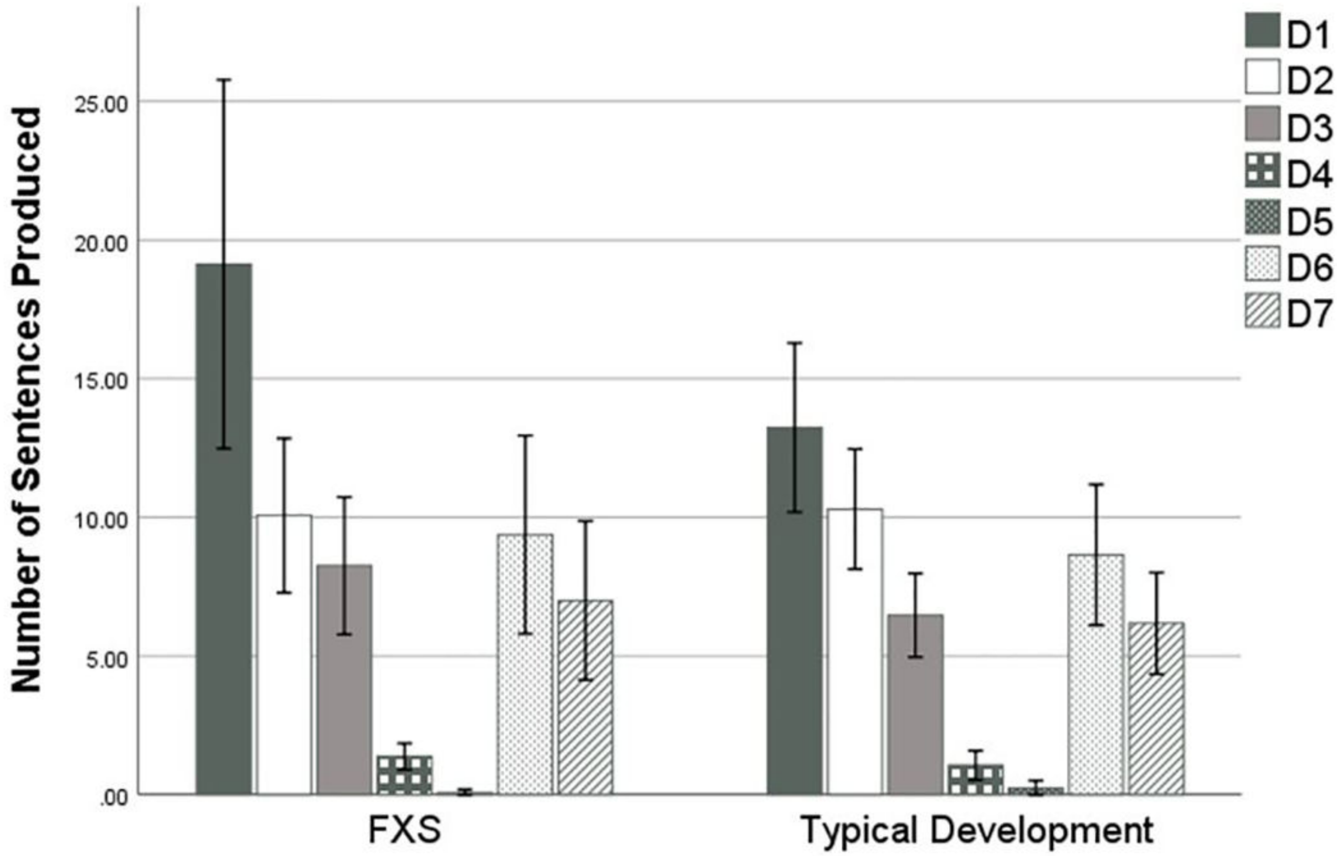

Figure 2.
Number of complex sentences produced by each group for conversation and narration combined. Error bars show +/− 2 standard errors.

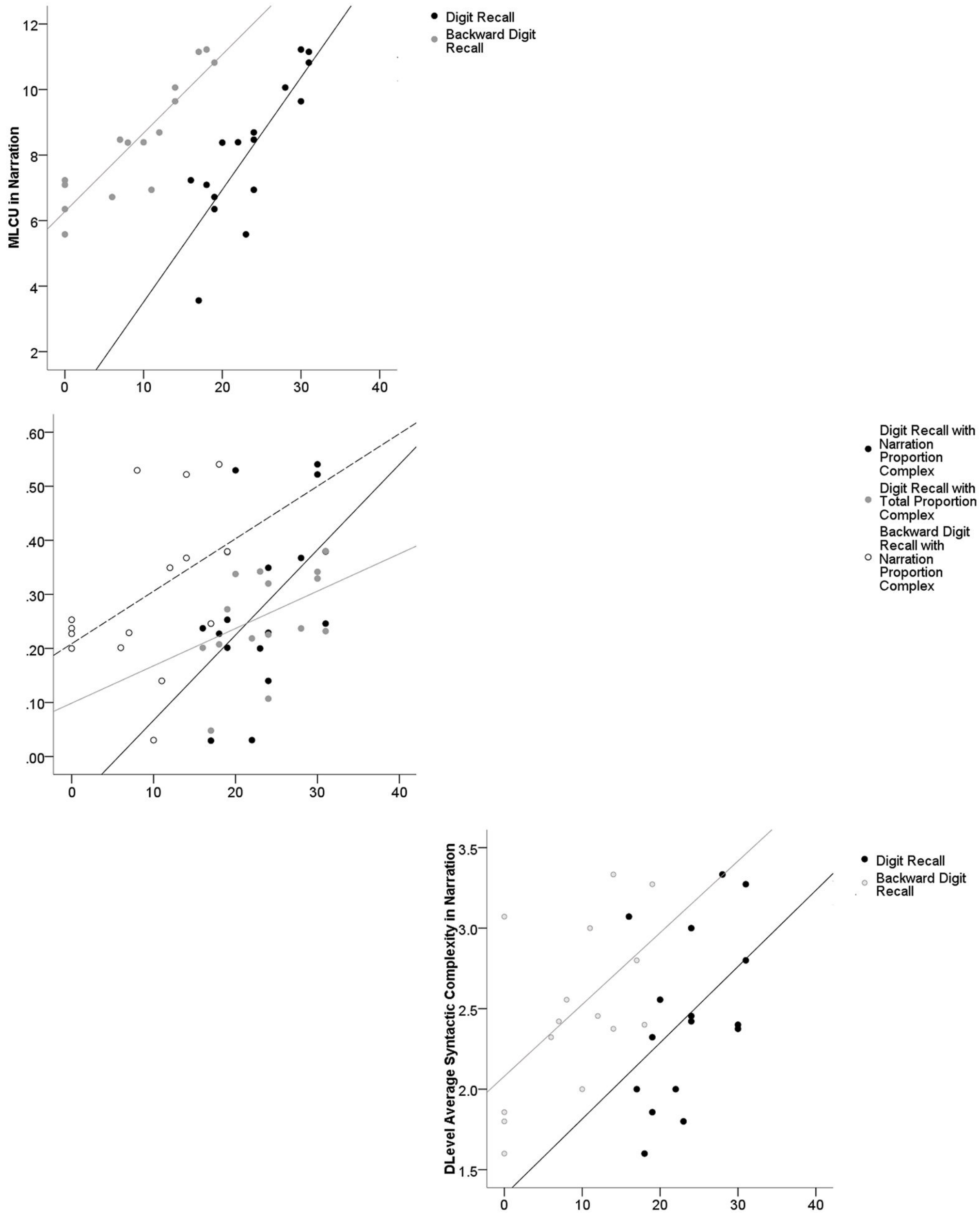

Figure 3.

Scatterplots of phonological memory and verbal working memory with MLCU in narration (mean length of C-unit; top panel), DLevel proportion complex C-units (middle panel), and DLevel average complexity (bottom panel) for females with FXS.

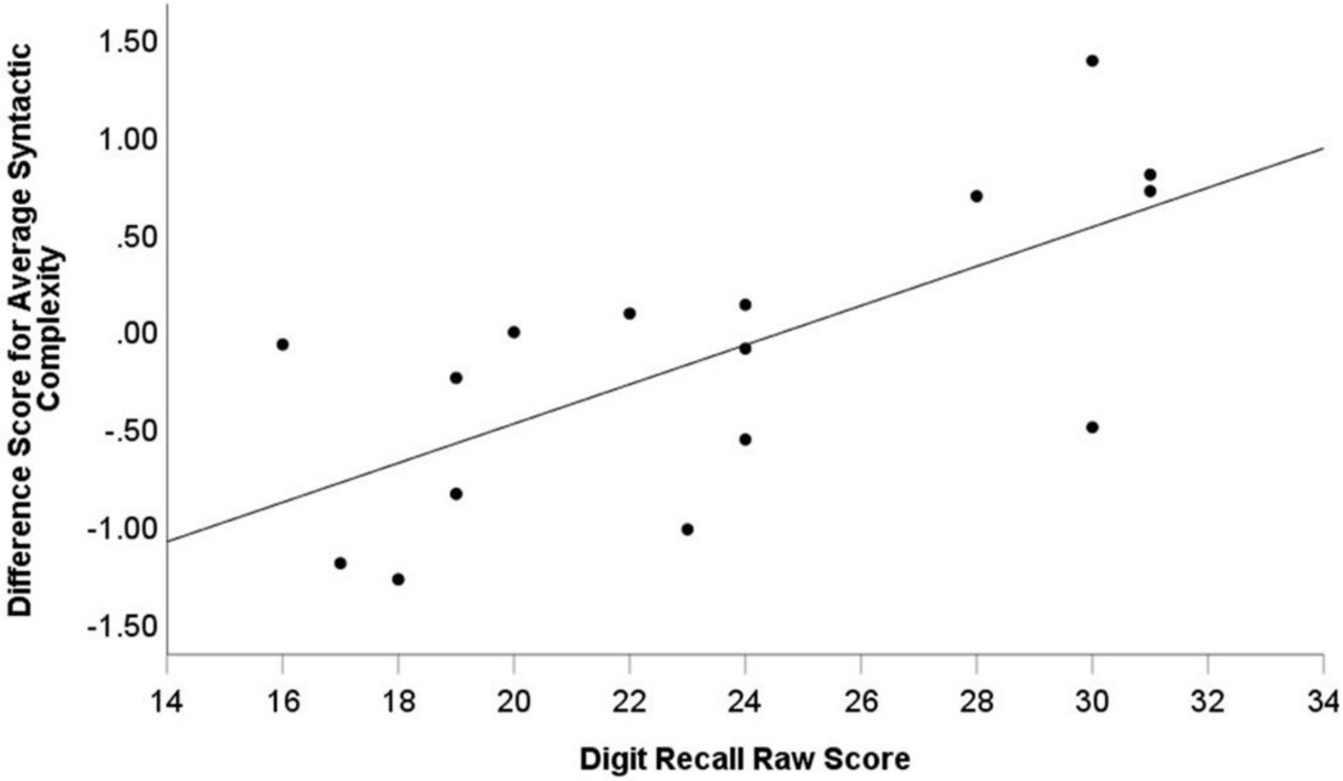

Figure 4.
Scatterplot displaying average syntactic complexity difference scores between narration and conversation with phonological memory for females with FXS.

Table 1

Participant Characteristics

	Fragile X Syndrome				Typical Development			
	n	**Mean**	**(SD)**	**Range**	***n***	**Mean**	**(SD)**	**Range**
Chronological age	16	12.04	(1.47)	10 – 15	17	6.41	1.69	4 – 9
Leiter-R[a]								
Nonverbal IQ	16	69.56	(15.35)	46 – 98	17	113.00	(9.12)	95–124
Growth score	16	482.06	(11.10)	462 – 502	17	480.06	(16.22)	458 – 505
Age-equivalent	16	7.81	(2.05)	4 – 12	17	7.51	(2.77)	4 – 13
TROG-2								
Standard score	16	76.81	(18.60)	55–111	17	106.88	(13.77)	83–130
Blocks passed	16	10.88	(5.28)	2–19	17	11.94	(5.61)	1–19
Age-equivalent	16	7.17	(2.87)	4–12	17	7.88	(3.30)	4–12
Phonological memory (digit recall)[b]								
Standard score	16	77.56	(12.77)	58–99	-	-	-	-
Raw score	16	23.50	(5.18)	16–31	-	-	-	-
Verbal working memory (backward digit recall)[c]								
Standard score	11	85.27	14.65	64–109	-	-	-	-
Raw score	15	9.07	6.80	0–19	-	-	-	-
% cells expressing FMRP	14	.48	(.05)	.34 – .51	-	-	-	-
Autism symptom severity	14	3.14	(2.77)	1 – 9	-	-	-	-

[a]Assessed with the Leiter-R Brief IQ subtests.

[b]Phonological memory was assessed with the WMTB-C Digit Recall subtest.

[c]Verbal working memory was assessed with the WMTB-C Backward Digit Recall subtest.

Table 2

Developmental Level (D-Level; Rosenberg & Abbeduto, 1987) Coding System for Complex Syntactic Constructions of Interest

D-Level Code	Description	Examples
D1: Infinitives	Early-developing, embedded, or subject-complementizing infinitives.	*I wanna see,* *I want you to quit.*
D2: Wh-clauses and compound subjects	Wh-questions, -clauses and –infinitives. Two or more subjects with the same predicate.	*She'll go when I do.* *Who else did you see?* *John and Mary left early.*
D3: Relative clauses and object noun phrase complements	Relative clauses that modify the entire main clause, indirect object, direct object, or object of the prepositional phrase. Noun phrase introduced by 'that' acting as the complement.	*We talk about things which I like. He bought the hotel where he stayed.* *I don't believe that it will work.*
D4: Gerunds and comparatives	Use of a verb acting as a noun, ending in –ing, as the object of a preposition. A construction expressing a difference.	*You hate my singing.* *We talked about lying.* *John is younger than Becky.*
D5: Subject noun phrase relative clause or complement, and subject nominalization	A relative clause that modifies the subject noun. The noun phrase functions as the subject. A noun formed from another word.	*The man who cleans the rooms left.* *That he should fail to turn up is really annoying.* *Shyness isn't so bad.*
D6: Subordinate conjunctions	Two main clauses conjoined with a subordinating conjunction.	*They will play today, if it doesn't rain.*

Table 3

Syntactic Complexity for MLU and DLevel Scores in Conversation and Narration

	Fragile X Syndrome (*n* = 16)			Typical Development (*n* = 17)		
	Mean	**(SD)**	**Range**	**Mean**	**(SD)**	**Range**
Conversation MLCU	6.37	(1.31)	4–8.75	6.53	(1.20)	5.25–9.05
Narration MLCU	8.14	(2.13)	3.56–11.22	8.61	(1.68)	5.05–11.55
Total proportion of fragments	.23	(.08)	.07-.42	.22	(.09)	.11-.43
Total proportion of simple sentences	.56	(.07)	.44-.65	.57	(.08)	.45-.73
Proportion of complex utterances[a]						
Total proportion complex	.26	(.09)	.05-.38	.25	(.07)	.12-.37
Conversation proportion complex	.25	(.11)	.06-.46	.24	(.09)	.12-.38
Narration proportion complex	.28	(.16)	.03-.54	.29	(.10)	.08-.56
Average syntactic complexity[b]						
Total average complexity	2.63	(.37)	1.92–3.12	2.73	(.42)	2.05–3.42
Conversation average syntactic complexity	2.59	(.59)	1–3.20	2.80	(.49)	1.72–3.79
Narration average syntactic complexity	2.45	(.53)	1.60–3.33	2.60	(.66)	1.67–3.81
Difference score for average syntactic complexity in narration vs. conversation[c]	-.13	(.76)	-1.28–1.38	-.20	(.79)	-1.31–1.78

[a]Proportion complex reflects the proportion of utterances coded D1 through D6.

[b]Average syntactic complexity reflects the average weighted complexity score for utterances coded D1 through D6.

[c]Average complexity differences score reflects the narrative average syntactic complexity score minus the conversation average syntactic complexity score.

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Table 4

Utterances Produced During Expressive Language Samples

	Fragile X Syndrome (*n* = 16)			Typical Development (*n* = 17)		
	Mean	**(SD)**	**Range**	**Mean**	**(SD)**	**Range**
Conversation						
Total C-units[a]	126.88	(48)	42–208	112.35	(18)	70–135
Complete and intelligible C-units[b]	116.81	(47)	35 −200	105.29	(16)	67–123
Narration						
Total C-units	56.94	(33)	22–158	49.53	(27)	22–144
Complete and intelligible C-units	53.75	(30)	22–141	47.00	(25)	19–132

[a]Total number of utterances produced during the language samples.

[b]Number of complete and ineligible utterances produced during the language sample (i.e., the analysis set for mean length of C-unit).

Table 5

Spearman's Rho Correlations among Average DLevel Syntactic Complexity and Characteristics of Participants with FXS and Typical Development

	Age	NVIQ	Con MLCU	Nar MLCU	Total syntactic complexity	Con complexity	Nar complexity	Difference score for average syntactic complexity
Age	-	-.01	-.27	.32	.03	-.01	.27	.17
Nonverbal IQ[a]	.24	-	.42	.53*	.14	-.09	.48*	.41
Conversation MLCU	.55*	.10	-	.31	-.03	.03	.19	.01
Narration MLCU	.74*	.32	.50*	-	-.22	-.44	.57*	.67*
Average syntactic complexity total[b]	.22	-.14	.04	.34	-	-.89*	.39	-.38
Conversation average syntactic complexity	.39	.01	.25	.53*	.91*	-	.03	-.70
Narration average syntactic complexity	-.25	-.34	-.57†	-.27	.43*	.14	-	.63*
Difference score for average complexity[c]	-.36	-.33	-.60*	-.49*	-.02	-.36	.83*	-

Note. Results for girls with FXS appear above the diagonal; correlations for girls with typical development appear below the diagonal.

[a]Assessed with the Leiter-R Brief IQ subtests.

[b]Average D-Level weighted syntactic complexity score for conversation and narration combined.

[c]Average D-Level weighted syntactic complexity score for narration minus conversation.

*$p < .05$.

†$p < .05$ in the unexpected direction.

Table 6

Spearman's Rho Correlations among Phonological Memory/Working Memory and Syntactic Complexity for Participants with Fragile X Syndrome

	Phonological Memory (Digit recall raw score)	**Verbal Working Memory (Backward digit recall raw score)**
Conversation MLCU[a]	.15	.24
Narration MLCU	.81 *	.88 *
Total Proportion Complex[b]	.43 *	.23
Conversation Proportion Complex	.14	-.07
Narration Proportion Complex	.52 *	.54 *
Total average syntactic complexity[c]	-.25	−.02
Conversation average syntactic complexity	-.50 †	-.27
Narration average syntactic complexity	.47 *	.55 *
Difference score for average syntactic complexity[d]	.66 *	.61 *

[a]Mean length of C-unit

[b]Proportion of all utterances that received a code of D1 through D6 combined (averaged) for conversation and narration.

[c]Average weighted complexity score of all utterances coded D1 through D6 combined (averaged) for conversation and narration.

[d]Average weighted syntactic complexity score for narration minus the average weighted syntactic complexity score for conversation.

*$p < .05$.

†$p < .05$ in the unexpected direction.