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Expressive language abilities of boys with idiopathic autism spectrum disorder and boys with fragile X syndrome + autism spectrum disorder: Cross-context comparisons

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Abstract

Background and aims: Understanding the unique expressive language profiles of children with phenotypically similar, but distinct neurodevelopmental disorders, such as idiopathic autism spectrum disorder and fragile X syndrome with a co-diagnosis of autism spectrum disorder (fragile X syndrome + autism spectrum disorder), has both clinical and theoretical implications. However, comparative studies of these two clinical groups have been limited, and results have been inconsistent, partially as a result of different assessment methods being utilized. Thus, the current study compared the expressive language profiles of boys with idiopathic autism spectrum disorder and boys with fragile X syndrome + autism spectrum disorder and examined whether a similar linguistic profile emerged across different language sampling contexts: a semi-structured conversation and the Autism Diagnostic Observation Schedule.

Methods: Eighteen boys with autism spectrum disorder ($M_{age} = 13.25$ years) and 19 boys with fragile X syndrome + autism spectrum disorder ($M_{age} = 12.19$ years), matched on autism spectrum disorder symptom severity and similar in terms of chronological age and mean length of utterance, participated in this study. Boys produced two language samples: one semi-structured conversation and one taken from the Autism Diagnostic Observation Schedule. Language samples were coded for talkativeness, lexical diversity, mean length of utterance, intelligibility, and repetitive or perseverative language.

Results: Analyses revealed that boys with autism spectrum disorder produced language samples that were more lexically diverse and intelligible, and that included less topic perseveration compared to boys with fragile X syndrome + autism spectrum disorder. With regards to sampling context, boys in both groups were more talkative and produced longer and more intelligible utterances in their conversation sample compared to their Autism Diagnostic

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Observation Schedule sample. However, boys with autism spectrum disorder and fragile X syndrome + autism spectrum disorder used a higher proportion of topic perseveration during the conversation sample.

Conclusions: Overall, we found similarities as well as distinctions in the expressive language profiles of boys with fragile X syndrome + autism spectrum disorder and boys with idiopathic autism spectrum disorder. Moreover, the Autism Diagnostic Observation Schedule sample elicited a relatively different language profile than the conversation sample for boys in both groups.

Implications: These findings help to further elucidate the unique language phenotypes of boys with idiopathic autism spectrum disorder and boys with fragile X syndrome + autism spectrum disorder. Moreover, our findings indicate that multiple language samples may be needed to obtain a comprehensive account of a child's expressive language ability.

Keywords

Autism spectrum disorder, fragile X syndrome, expressive language, language sampling

The ability to express oneself through language is an essential part of the human experience. Through the development of spoken language, children are able to communicate their wants and needs, express their thoughts and feelings, and engage in interactions with others. Expressive language skills are also predictive of academic outcomes (e.g. reading and writing; Berninger & Abbott, 2010). As a result, impairments in expressive language can greatly impact these processes, and lead to difficulties throughout the lifespan. Children with autism spectrum disorder (ASD) have core impairments in social communication; however, outside of social communication, expressive structural language abilities vary greatly.

Expressive language difficulties, however, are not unique to children with ASD. Language delays or impairments are common across children with other neurodevelopmental disorders, such as fragile X syndrome (FXS). FXS is the leading inherited cause of intellectual disability and is the result of a single gene mutation (*FMR1*) on the X chromosome (Verkerk et al., 1991). FXS is an interesting comparison group for children with ASD because there is a high comorbidity between FXS and ASD (FXS + ASD). Depending on the diagnostic measures used, recent rates have estimated that 27–75% of males with FXS have a co-diagnosis of ASD (FXS + ASD; e.g. Bailey et al., 2008; Clifford et al., 2007; Klusek et al., 2014a). Thus, comparative studies between children with FXS + ASD and idiopathic ASD provide a unique opportunity to investigate whether ASD influences expressive language in a similar way in children who also have FXS. Therefore, the present study explored whether boys with FXS + ASD have a distinct expressive language profile compared to boys with idiopathic ASD and examined whether this differed depending on sampling context.

Expressive language in children with idiopathic ASD and children with FXS

Autism spectrum disorder

ASD is diagnosed on the basis of impairments in social communication and restricted interests and repetitive behaviors (American Psychiatric Association, 2013). Although not a part of the diagnostic criteria, delays in language acquisition are present in the majority of young children with ASD; in fact, delayed language abilities are often the primary reason for initial diagnostic referral (Richards et al., 2016). While pragmatic language deficits are considered universal, the presentation of expressive language impairments in vocabulary and grammar are more variable in verbal children with ASD (Tager-Flusberg et al., 2011). Specifically, a substantial number of children with ASD (as many as 60%) demonstrate impairments in one or more of these domains compared to children with typical development (TD), and many children present with uneven expressive language profiles (e.g. better vocabulary than grammar; see Boucher, 2012; Tager-Flusberg et al., 2011). For example, research has shown that many children with ASD often produce a fewer number of different words in spontaneous language samples than chronological age-matched peers with TD (Condouris et al., 2003), but are similar to children with TD when matched on nonverbal mental age or receptive language (Eigsti et al., 2007). In terms of grammar, though, studies have shown that many children with ASD use less complex and diverse sentences compared to chronological age-matched and mental age-matched children with TD (Bacon et al., 2019; Eigsti et al., 2007; Park et al., 2012).

Fragile X syndrome

Expressive language impairments are also common in FXS. Like other X-linked disorders, males are differentially affected compared to females, and the effects are often more severe in males because they do not have an additional unaffected X chromosome to help to buffer the effects of the mutated X chromosome (Loesch et al., 2002). Due to these sex differences, the present study and the following discussion are focused on males with FXS. In general, males with FXS have greater deficits in language production than comprehension (Roberts et al., 2001), and all aspects of expressive language are delayed relative to chronological age expectations, including pragmatics, vocabulary, and grammar (e.g. Martin et al., 2013; Roberts et al., 2001). Moreover, studies typically show that the expressive vocabulary and grammatical abilities of boys with FXS are impaired compared to younger children with TD matched on nonverbal mental age (e.g. Kover et al., 2012; Martin et al., 2013). Spontaneous expressive language in males with FXS has also been characterized as more rapid, unintelligible, and perseverative or repetitive than mental age-matched peers with TD (Klusek et al., 2014b; Kover et al., 2012; Martin et al., 2012).

Yet, these findings depend in some cases on the ASD severity of boys with FXS. In fact, research suggests that boys with FXS + ASD present with a rather distinct phenotype of social behavior, with similarities to ASD in the general population (Budimirovic et al., 2006; Hernandez et al., 2009). In terms of language, differences in expressive language are often even more pronounced for boys with FXS + ASD, who tend to have greater pragmatic deficits (Klusek et al., 2014b; Losh et al., 2012; Martin et al., 2013, 2017), speech that is less intelligible (Kover & Abbeduto, 2010), and language that is more perseverative (Martin et al., 2012, 2018) compared to boys with FXS-Only. In terms of vocabulary and grammar, boys with FXS + ASD often do not differ significantly from boys with FXS-Only (e.g. Kover et al., 2012; McDuffie et al., 2012; Price et al., 2008; Roberts et al., 2007a, 2007b). However, several studies found that after controlling for nonverbal mental age, boys with FXS + ASD had significantly lower expressive vocabulary and grammatical abilities compared to boys with TD, whereas the performance of boys with FXS-Only did not differ from TD controls in some cases, suggesting a negative influence of ASD beyond the effect of FXS (Price et al., 2008; Roberts et al., 2007a, 2007b).

Comparisons between boys with idiopathic ASD and boys with FXS + ASD

Given the differences in language ability between boys with FXS-Only and FXS + ASD, and the number of boys with FXS who meet criteria for a comorbid ASD classification, previous work has examined if boys with FXS + ASD have a distinguishable language phenotype compared to idiopathic ASD. Although limited, research on expressive language abilities in these groups has been mixed, making it difficult to discern the areas of linguistic overlap and distinction in boys with FXS + ASD and idiopathic ASD. For example, after controlling for nonverbal cognition, ASD severity, and chronological age, Thurman et al. (2017) found that boys with FXS + ASD had better standardized expressive vocabulary scores than boys with idiopathic ASD. However, the boys with FXS + ASD and idiopathic ASD did not differ on a standardized measure of grammar. Similarly, after controlling for nonverbal mental age, receptive vocabulary, and expressive vocabulary and grammar, others have found that boys with FXS + ASD and idiopathic ASD do no differ in terms of the overall frequency (Martin et al., 2018) or rating of perseverative language (Klusek et al., 2014b). Interestingly, Martin et al. (2018) found that boys with FXS + ASD and ASD had a higher frequency of perseveration than boys with FXS-Only but not boys with TD. In contrast, Klusek et al. (2014b) found that ratings of perseverative language were similar across boys with FXS-Only, FXS + ASD, and ASD, and all groups were more perseverative than boys with TD.

Other comparative studies have documented poorer expressive language abilities in children with FXS + ASD compared to children with idiopathic ASD. Haebig and Sterling (2017) found that boys with idiopathic ASD had significantly higher expressive vocabulary growth scores than the boys with FXS + ASD who were matched on ASD symptom severity. Also, when boys with FXS + ASD and idiopathic ASD are similar or matched on ASD symptom severity, boys with FXS + ASD demonstrate greater impairments in the production of specific morphemes (i.e. auxiliary *be* verbs; Sterling, 2018) and use more single-utterance perseverations (Friedman et al., 2018). Additionally, analysis of language samples has indicated that the language of boys with FXS + ASD is less intelligible than boys with idiopathic ASD, as well as boys with FXS-Only and TD (Klusek et al., 2014b).

These inconsistencies between studies appear to be partially reflective of differences in matching methods and/or assessment type utilized. In terms of matching, a greater number of language deficits have been identified in boys with FXS + ASD when studies have matched on criteria such as ASD severity, instead of controlling for factors such as nonverbal mental age. The motivation for comparing these groups on ASD severity instead of nonverbal mental age is rooted in that fact that impaired cognition is a core impairment in males with FXS, but not children with idiopathic

ASD. Therefore, comparisons that control for cognition can be difficult to interpret given that intellectual disability is inseparable from the disorder (Dennis et al., 2009). Matching on ASD severity can be informative because this is a behavioral phenotype that is shared by boys with idiopathic ASD and FXS + ASD.

Beyond matching considerations, it is also important to consider the way in which language is being assessed as each technique can serve a different purpose and provide unique information. For example, while standardized assessments can provide a quick appraisal of language performance, many measures tend to provide a single score that aggregates performance across a range of language domains. Some standardized assessments are also prone to floor effects in individuals with intellectual disabilities (Mervis & Robinson, 2005). In contrast, other measures, such as spontaneous language samples, are often more in line with real-world practical contexts and are sensitive to small differences in more narrowly defined, but perhaps clinically meaningful domains of language. As such, language samples may be well-suited for comparing and characterizing the language profiles of children with phenotypically similar developmental disorders. With these points in mind, the first aim of the study was to compare the expressive language profiles of boys with FXS + ASD and idiopathic ASD matched on ASD severity using spontaneous language samples.

Language sampling methods: Using the Autism Diagnostic Observation Schedule

Traditionally, language sampling contexts have included narration as well as conversations elicited through free play or semi-structured interviews with an examiner or a caregiver. More recently, an increasing number of studies have begun to use other assessment tools, such as the Autism Diagnostic Observation Schedule (ADOS, ADOS-2; Lord et al., 1999, 2012), to examine the language abilities of children with FXS and ASD (e.g. Estigarribia, et al., 2011; Klusek et al., 2014b; Martin et al., 2018; Park et al., 2012; Roberts et al., 2007a). Indeed, Tager-Flusberg and expert colleagues (2009) noted the value of using multiple language samples in the assessment process, and recommended the ADOS as a way to collect natural language samples in children with ASD. The ADOS is a diagnostic assessment designed to evaluate ASD symptomology. However, it may also provide a practical way to obtain language samples in individuals with ASD and FXS given that it is often administered as part of a study procedure. The ADOS includes numerous social presses and activities which can provide an excellent opportunity to evaluate pragmatics, as well as semantics, morphology, and grammar (e.g. responses to questions, responses to examiner comments, descriptions of a picture). Moreover, given the semi-structured nature of the ADOS, administration is uniform across children while also allowing the examiner to flexibly follow the child's lead, making it a potential context for obtaining a naturalistic language sample (Tager-Flusberg et al., 2009).

It is currently not well understood though how the linguistic samples obtained from the ADOS may compare to those obtained using more traditional language sampling techniques. It is important to examine this because different language sampling contexts may influence the language that a child produces, which can have implications in both clinical and research settings. For example, previous research has shown that children often produce more grammatically complex sentences during narrative contexts compared to conversational contexts, but children are generally more talkative (producing more words or utterances) in conversational contexts (e.g. Abbeduto et al., 1995; Kover & Abbeduto, 2010; Kover et al., 2012; Murphy & Abbeduto, 2007; Westerveld et al., 2004). Thus, the intrinsic properties of the method selected may yield different information in terms of language performance, and impact the conclusions reached about children's expressive language profiles (Abbeduto et al., 1995; Finestack et al., 2014; Kover et al., 2012; Southwood & Russell, 2004). Given that the structure of the ADOS differs from both a narrative as well as a traditional conversation sample, it would not be surprising for language performance to differ when comparing the ADOS to these other sampling techniques.

Only two studies have compared children's language samples from the ADOS to more traditional sampling contexts to directly assess whether the properties of the ADOS may in fact yield different linguistic profiles. One of these studies included school-age boys with FXS (Martin et al., 2012) and the other included preschool-age children with ASD (Kover et al., 2014). Martin et al. (2012) used a language sample from the ADOS, as well as a separate narrative task, to examine differences in the frequency of perseveration in 6–16-year-old boys with FXS-Only, FXS + ASD, and Down syndrome (DS), and boys with TD between 3 and 6 years of age. Nonverbal mental age was controlled for in the analyses. Boys with FXS-Only and FXS + ASD used a greater frequency of topic perseveration during the ADOS sample compared to the narration task. Moreover, boys with FXS + ASD, DS, and TD used more conversational device perseveration during the ADOS sample (e.g. "hmm", "I don't know", "oh man"). In contrast, boys with TD used more utterance level perseveration during the narrative task than the ADOS. Using a younger sample of preschool children with ASD, Kover et al. (2014) compared a language sample from the ADOS to two conversational play-based language samples (parent–child play and examiner–child play). Assessing a wider-range of expressive language features, they found that children with ASD produced language with more grammatical and semantic complexity, and made more comments and requests in the examiner-and/or parent-child play-based language samples compared to the ADOS. Additionally, the parent-child play-based language samples yielded increased turn-taking utterances compared to the ADOS. As a result, the authors noted concerns that the ADOS may underestimate certain language skills in young children with ASD.

The findings from these two studies (Kover et al., 2014; Martin et al., 2012) provide valuable information about the utility of the ADOS and further suggest that the ADOS may yield a different profile of expressive language than other traditional language sampling techniques for children with neurodevelopmental disorders. However, a number of questions remain. It is still unclear whether the context differences (conversation vs. ADOS) seen in young children with idiopathic ASD (Kover et al., 2014) would also emerge for boys with FXS + ASD across a wide range of expressive language abilities, and in older children with idiopathic ASD. Moreover, the ADOS has yet to be compared to an interview-style or semi-structured conversation (a commonly used technique with school-age children). Additionally, it is still unclear whether similar context effects may emerge for boys with idiopathic ASD and boys with FXS + ASD, or whether context may differentially impact the expressive language produced by boys with idiopathic ASD compared to FXS + ASD. With these points in mind, the second aim of the study was to extend this previous work by comparing the ADOS to a semi-structured conversation in a sample of school-age boys with idiopathic ASD and FXS + ASD. Such comparisons can help to provide insight into whether these common sampling contexts are eliciting the same type of language, or if they are instead more ideally suited to eliciting different kinds of language. In turn, this information may help researchers and clinicians make more informed decisions when selecting language sampling contexts. Finally, by including both clinical groups, we are not only able to explore context differences, but evaluate whether the ADOS could be used to distinguish between the language profiles of children with phenotypically similar, but genetically distinct, neurodevelopmental disorders. This is particularly important to consider given the practical application of using the ADOS as a language sampling context in these populations.

Overview of the present study

The current study compared the expressive language profiles of boys with FXS + ASD and idiopathic ASD matched on ASD severity and evaluated whether similar linguistic characteristics were observed equally across different language sampling contexts. In particular, we explored whether boys' expressive language performance would differ when elicited from a language sample collected from the ADOS compared to a traditional semi-structured conversation sample. To expand on the current literature, we coded for talkativeness, lexical diversity, mean length of utterance (MLU), intelligibility, and perseveration.

As such, this study aimed to address two research questions: (1) are there differences in expressive language performance in boys with FXS + ASD and boys with idiopathic ASD? and (2) how does the expressive language ability of boys with FXS + ASD and idiopathic ASD vary across language sampling contexts (i.e. conversation, ADOS)? In line with previous studies that have used more nuanced types of language assessments (Friedman et al., 2018; Klusek et al., 2014b; Sterling, 2018), we predicted that boys with FXS + ASD would produce language samples that were shorter and less lexically diverse, as well as more unintelligible and perseverative than those of boys with idiopathic ASD. Based on the findings of Kover et al. (2014), it was expected that boys in both groups would be less talkative, use fewer different words, and produce sentences with lower MLU during the ADOS compared to the conversational language sample. Also, we hypothesized that the participants in both groups would be less intelligible and more perseverative during the ADOS (Martin et al., 2012).

Methods

Participants

Participants included 19 boys with FXS + ASD and 18 boys with idiopathic ASD between 9 and 16 years of age. All boys with FXS + ASD had the full mutation, confirmed via previous genetic testing, and all boys with idiopathic ASD had prior genetic testing to rule out FXS. Additionally, the boys with ASD had a community diagnosis of ASD, which was provided previously by a physician, neurologist, psychiatrist, or psychologist. All children met ASD criteria on the ADOS (Lord et al., 1999, 2012), as well as the Autism Diagnostic Interview-Revised (Rutter et al., 2003) which is a parent-based interview of children's past and present behaviors. All boys were monolingual English speakers, as indicated by parent report, and were part of a larger study on grammar and language

Table 1. Participant characteristics.

	ASD[d]	FXS + ASD[d]	t (df)	p	Cohen's d	Variance ratio
Chronological age (years)	13.25 (1.91)	12.19 (2.01)	1.64 (35)	.111	.54	1.11
ADOS severity score[a]	7.50 (1.72)	7.21 (1.81)	0.50 (35)	.622	.17	1.12
Average MLU[b]	4.31 (1.70)	3.80 (0.92)	1.18 (35)	.249	.38	0.28
Nonverbal IQ[c]	70.44 (21.46)	47.16 (7.84)	4.43 (35)	.0001	1.11	0.13
PPVT-4 standard score	78.94 (20.20)	60.42 (12.29)	3.39 (35)	.002	1.12	0.40
EVT-2 standard score	80.22 (18.84)	62.58 (11.91)	3.43 (35)	.002	1.44	0.37

ASD: autism spectrum disorder; FXS + ASD: fragile X syndrome + autism spectrum disorder; MLU: mean length of utterance; PPVT-4: Peabody Picture Vocabulary Test, Fourth Edition; EVT-2: Expressive Vocabulary Test, Second Edition.
[a]Autism Diagnostic Observation Schedule.
[b]Average MLU in morphemes from conversation and ADOS language samples.
[c]Nonverbal IQ from brief IQ score from the Leiter International Performance Scale—Revised.
[d]Standard deviations are in parentheses.

assessment in FXS and ASD (e.g. Friedman et al., 2018; 2019; Haebig et al., 2016; Haebig & Sterling, 2017; Sterling, 2018). Participants were recruited from a research registry run by the University of North Carolina at Chapel Hill and the Waisman Center at the University of Wisconsin-Madison, as well as from a national parent listserv, the National Fragile X Foundation, and the Interactive Autism Network. The participants were from 15 different states.

Of the boys with FXS + ASD, 15 were White (78.95%), two were African-American (10.53%), one reported more than one race (5.26%; American-Indian and White), and one reported "other" (5.26%). Two boys with FXS + ASD were reported to be Hispanic or Latino (10.53%) and ethnicity was not reported for two boys (10.53%). Of the boys with idiopathic ASD, 15 were White (83.33%), one was African-American, White, and "other" (5.56%), one was African-American and White (5.56%), and one reported "other" (5.56%). One boy with idiopathic ASD was Hispanic or Latino (5.56%), and ethnicity was not reported for two boys with idiopathic ASD (11.11%).

Group matching

The boys with FXS + ASD were matched to boys with idiopathic ASD on ASD severity following the guidelines outlined in Kover and Atwood (2013). The groups were also similar on chronological age and average MLU in morphemes, which was determined by averaging the MLUs across the conversation sample and ADOS sample. Given that the present study was focused on comparing the expressive language skills of boys with FXS + ASD and boys with idiopathic ASD, MLU was selected as a variable in order to control for variation in language. MLU-based comparisons are common in studies of children with neurodevelopmental disorders and language impairments (e.g. Haebig et al., 2016; Hoover et al., 2012; Levy et al., 2006; Rice et al., 2006; Sterling, 2018). MLU was also selected as a matching variable because it has reliable normative data (e.g. Rice et al., 2010). Thus, MLU-based comparisons allowed the findings from this study to be evaluated within the context of the broader expressive language literature. It should be noted that while several boys had MLUs above 4.0, the mean MLU for the two groups was right around 4, or even less than 4 (mean MLU for boys with ASD = 4.31; mean MLU for boys with FXS = 3.80). The range for MLU was also similar across the groups (ASD: 1.53–6.84; FXS + ASD: 2.13–5.58). Although the interpretation of MLUs that are above 4.0 can be less clear-cut when assessing language complexity, there is psychometric evidence to support the use of MLU beyond 4.0 as an appropriate measure of grammar in childhood as well as adolescence (Channell et al., 2018; Heilmann et al., 2010; Nippold et al., 2005; Rice et al., 2010). See Table 1 for more descriptive and statistical information regarding these matching variables, as well as additional participant characteristics.

Procedure

All testing was completed at the Waisman Center during the course of a single visit. The university's Institutional Review Board approved the study procedures. After obtaining informed written consent from the legal guardian and oral assent from the child, the children completed a number of cognitive and linguistic norm-referenced assessments, a conversational language sample, and the ADOS. The assessments were videotaped and audiotaped for off-line transcription. The participants were given breaks between testing as needed. Each participant was given a small honorarium.

Norm-referenced assessments

Nonverbal IQ was measured using the Leiter International Performance Scale-Revised (Roid & Miller, 1997). The brief IQ composite is computed using four subtests: Figure Ground, Form Completion, Sequential Order, and Repeated Patterns. This test was used to help characterize the sample and benchmark this population within the broader intellectual disabilities' literature.

The participants also completed measures of expressive and receptive vocabulary in order to further describe the sample's language abilities. Specifically, each child was given the Expressive Vocabulary Test, Second Edition, as a measure of expressive vocabulary (Williams, 2007) along with the Peabody Picture Vocabulary Test, Fourth Edition, which measures vocabulary comprehension (Dunn & Dunn, 2007).

Language sampling measures

Conversation sample. Each child completed a 10-minute semi-structured, interview-style conversation with the examiner. Following the procedures outlined by Berry-Kravis et al. (2013), the conversation sample began with a topic related to the participant's specific interest based on parent report. For the remainder of the conversation, the examiner followed a set list of topics and a script for introducing and following up on topics. This helped ensure comparability across boys. Topics included sports, pets, school, hobbies/ interests, families, and vacations. Throughout the conversation, the trained examiner employed language elicitation techniques such as open-ended prompts (e.g. "tell me more about that") to encourage the participant to talk. At the same time, the examiner minimized her use of yes/no questions as well as her own talk.

ADOS sample. As part of an ASD diagnostic battery, each child completed the ADOS, first or second edition (Lord et al., 1999, 2012). As described earlier, the ADOS is a semi-structured assessment of ASD symptoms. The ADOS examiner was either research reliable or training to be research reliable, with a research reliable examiner present for coding. The ADOS has four modules, and selection of the module is based on the child's expressive language level. The boys in this study received either a Module 2 (phrase-level language) or Module 3 (verbally fluent). For our 10-minute ADOS sample, we selected the tasks that overlapped between Modules 2 and 3, which included: description of a picture, joint interactive play, demonstration task, conversation and reporting, make-believe play, and telling a story from a book.

Given that children completed the tasks in different amounts of time, children varied in the number of tasks they completed to reach a 10-minute language sample. However, for all children, the majority of their language sample ($M = 86.44\%$; $SD = 12.82\%$) was taken from description of a picture, joint interactive play, and the demonstration task because these tasks provided ample opportunity for a communicative interchange in a minimally structured setting. If children did not meet the 10-minute mark after these three tasks, conversation and reporting was included, followed by make-believe play, and then telling a story from a book, in order to meet the time requirement. Ten (27.03%) of the children's ADOS samples included three tasks, 15 (40.54%) included four tasks, 10 (27.03%) included five tasks, and two (5.41%) included all six tasks. Boys with FXS + ASD ($M = 4.37$; $SD = 0.89$) were similar to the boys with idiopathic ASD ($M = 3.83$; $SD = 0.79$) on the total number of tasks needed to reach a 10-minute language sample, $t(35) = -1.93$, $p = .062$, $d = .64$.

Transcription and coding

The language samples were transcribed using the Systematic Analysis of Language Transcripts software (SALT; Miller et al., 2011) by trained undergraduate and graduate students. The unit of segmentation was the C-unit, which refers to an independent clause and all subordinate clauses attached to it. All of the expressive language outcomes were calculated out of the complete and intelligible utterances that the child produced, with the exception of percent intelligibility, which was calculated from the total utterances produced by the child. With the exception of perseverative language codes, all of the following language variables were produced using SALT. These variables included:

Talkativeness. Talkativeness was defined as the number of total intelligible utterances the child produced.

Intelligibility. Intelligibility was determined by taking the proportion of the child's intelligible utterances out of the total number of child utterances.

Lexical diversity. Lexical diversity was determined by using the moving-average type–token ratio (MATTR; Covington & McFall, 2010). MATTR is a computational technique used for quantifying lexical diversity that can be interpreted similarly to the normal type token ratio, with larger MATTR scores indicating greater lexical diversity. However, it minimizes the effect of sample length because it estimates the type/ token ratio (TTR) using a moving window, and is a more accurate measure of lexical diversity than TTR (Stills, 2016). For example, for a window length of 100 words, it calculates TTR for words 1–100, 2–101, 3–102, and so on to the end of the sample. The final number is the average of all of the TTRs. MATTR has been used to examine lexical diversity in children and adults with TD (Fergadiotis et al., 2015; Stills, 2016) and adults with speech disorders (Fergadiotis et al., 2013).

Grammatical complexity. Grammatical complexity was determined using MLU in morphemes. MLU indicates increasing complexity at the phrasal level, clausal level, and level of argument structure (Scott & Stokes, 1995; Scott & Windsor, 2000).

Frequency of perseveration. Given that repetitive or perseverative language is produced by both boys with idiopathic ASD and FXS + ASD (Friedman et al., 2018; Martin et al., 2012, 2018), we were also interested in the frequency of perseverative language. Perseverative language was coded using the Martin et al. (2012) published coding scheme. These codes included utterance, topic, and conversational device perseverations. Utterance-level perseveration was coded when a child repeated a word, phrase (e.g. "It's a bowling pin, bowling pin."), or entire utterance (e.g. "I like football. I like football.") in immediate succession. Topic perseveration was coded when a child repeated a topic, theme, or idea three or more times in an excessive manner at any point throughout the language sample. Although the conversation sample did encourage examiners to ask open-ended follow-up questions about the same topic, utterances were only coded as perseverative if the child continued to discuss the topic after the topic of conversation had changed. Conversational device perseveration was defined as the excessive repetition (three or more instances) of rote or conventional words, phrases, or sayings (e.g. "Oh man", "Cool"). Each perseverative language code was calculated as a proportion (e.g. the number of utterance perseverations divided by the total number of utterances).

Reliability

SALT reliability. Eight (21.62%) of the conversation language samples and eight (21.62%) of the ADOS transcripts were transcribed in SALT independently by a second transcriber. Reliability for SALT variables was calculated line-by-line. In line with other studies (e.g. Hogan-Brown et al., 2013; Lee et al., 2018), overall agreement between primary and reliability coders for the conversational language samples was 83.21% for utterance segmentation, 78.70% for number of morphemes, 80.37% for number of words, 95.70% for unintelligibility, and 87.33% for word identification. Overall agreement between the primary and reliability ADOS transcriptions was 78.24% for utterance segmentation, 79.28% for number of morphemes, 80.89% for number of words, 90.33% for unintelligibility, and 82.33% for word identification. Although reliability was slightly below 80% for several variables, it has been noted that exact agreement between raters of 70% is often sufficient for more complex coding (Hartmann et al., 2004), such as morphemes.

Perseveration reliability. A second independent coder completed perseverative language coding for eight (21.62%) of the conversation language samples and 10 (27.03%) of the ADOS transcripts. Reliability was calculated from these files using Cohen's kappa's. Reliability values for the conversation transcripts were .53 for topic perseverations, indicating moderate agreement, .93 for utterance perseverations, indicating near perfect agreement, and .69 for conversational device perseverations, indicating substantial agreement. Values for the ADOS transcripts were .97 for topic perseverations, indicating near perfect agreement, .79 for utterance perseverations, and .71 for conversational device perseverations, both indicating substantial agreement (Hallgren, 2012).

Data analysis plan

In order to examine group differences (research question one) as well as context differences (research question two) in expressive language ability, we conducted a series of mixed-model ANOVAs. The between-subjects variable was Diagnostic Group (FXS + ASD, idiopathic ASD) and the within-subjects variable was Sampling Context (10-minute conversation sample, 10-minute ADOS sample). Separate mixed-model ANOVAs were run for talkativeness, lexical diversity, intelligibility, and frequency of perseveration. Thus, we present our findings for research questions 1 and 2 together for almost all expressive language variables. However, because MLU was used as a matching variable, we only analyzed context differences (research question 2) in grammatical complexity using a repeated measures ANOVA instead of a mixed-model ANOVA. Given the number of variables analyzed, Holm–Bonferroni corrections were used in follow-up analyses. Effect sizes were interpreted as small ($\eta_p^2 = .01$), medium ($\eta_p^2 = .06$), and large ($\eta_p^2 = .14$; Cohen, 1988).

Although the average IQ of the boys with ASD was also in the intellectual disability range ($M = 70.44$, $SD = 21.46$), there was still a significant difference in nonverbal IQ between boys with FXS + ASD and ASD. Therefore, IQ was considered as a covariate. In line with best-practice guidelines, correlation analyses were first completed to assess whether there was a relationship between nonverbal IQ and the dependent variables (Tabachnik & Fidell, 2001). Correlations were run separately for each diagnostic group. After correcting for multiple comparisons using the Holm–Bonferroni method, analyses revealed that nonverbal IQ was not correlated with any of the dependent variables for boys with FXS + ASD or ASD, $p > .052$.

Moreover, Dennis et al. (2009) discussed that there are theoretical and methodological concerns with controlling for nonverbal IQ in studies of children with neurodevelopmental disorders. As noted earlier, when nonverbal IQ is adjusted for in clinical populations where intellectual disability is a central part of the disorder, findings may be difficult to interpret, given that intellectual disability is inextricable from the disorder. Specifically, Dennis et al. (2009) state that covarying for IQ in groups that inherently differ on IQ can provide a comparison of groups at values that may not exist in nature or are unrepresentative of the population. With regards to the present study, intellectual disability has a significantly higher prevalence in males with FXS (more than 90%; Hessl et al., 2009) than males with idiopathic ASD (15.8–31%; Baio et al., 2018; Ryland et al., 2014). As a result, comparisons that covary IQ may limit the clinical validity and generalizability of the findings. Therefore, we chose not to include nonverbal IQ as a covariate. Given these concerns, others have similarly compared various aspects of language in boys with FXS + ASD and idiopathic ASD without using IQ as a covariate (Friedman et al., 2018, 2019; Haebig & Sterling, 2017; Sterling, 2018).

Results

Comparisons of boys with FXS + ASD and idiopathic ASD across sampling contexts

Table 2 presents the means and standard deviations for all expressive language variables compared across the two groups and the two contexts. We present the findings from each analysis below. Figure 1 highlights the

Table 2. Group differences on the conversation and ADOS language samples.

		ASD		FXS + ASD	
Expressive language variable	Variable definition	Conversation	ADOS	Conversation	ADOS
Talkativeness[a]	Total number of utterances	117.50 (34.04)	96.39 (36.37)	129.63 (31.82)	110.32 (42.10)
Lexical diversity[b]	Moving-average type/token ratio	.61 (.05)	.59 (.06)	.54 (.04)	.54 (.05)
Grammatical complexity[a]	Mean length of utterance	4.65 (2.00)	3.97 (1.56)	4.02 (1.02)	3.57 (0.74)
Intelligibility[a,b,c]	Proportion of intelligible utterances	.99 (.01)	.98 (.03)	.96 (.04)	.88 (.12)
Perseveration					
Utterance	Total utterance level perseverations/total utterances	.01 (.03)	.01 (.02)	.02 (.02)	.02 (.02)
Topic[a,b]	Total topic level perseverations/ total utterances	.15 (.12)	.09 (.15)	.27 (.18)	.13 (.10)
Conversation	Total conversational device perseverations/total utterances	.05 (.06)	.09 (.08)	.06 (.08)	.07 (.08)

[a]Main effect of context.
[b]Main effect of diagnostic group.
[c]Significant interaction between context and diagnostic group.
Note: Standard deviations are in parentheses.

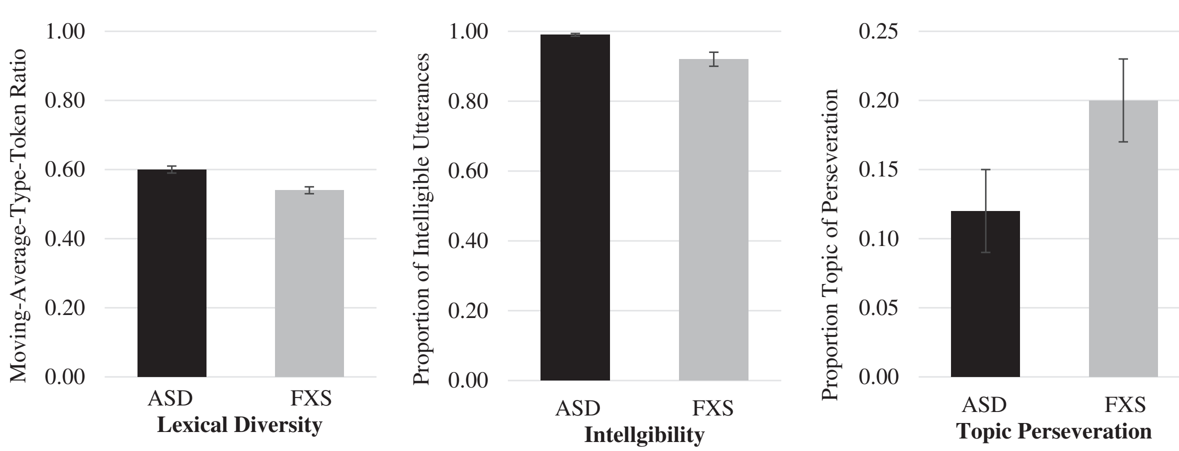

Figure 1. Significant group differences (collapsed across context). Standard error bars are shown.
ASD: autism spectrum disorder; FXS: fragile X syndrome.

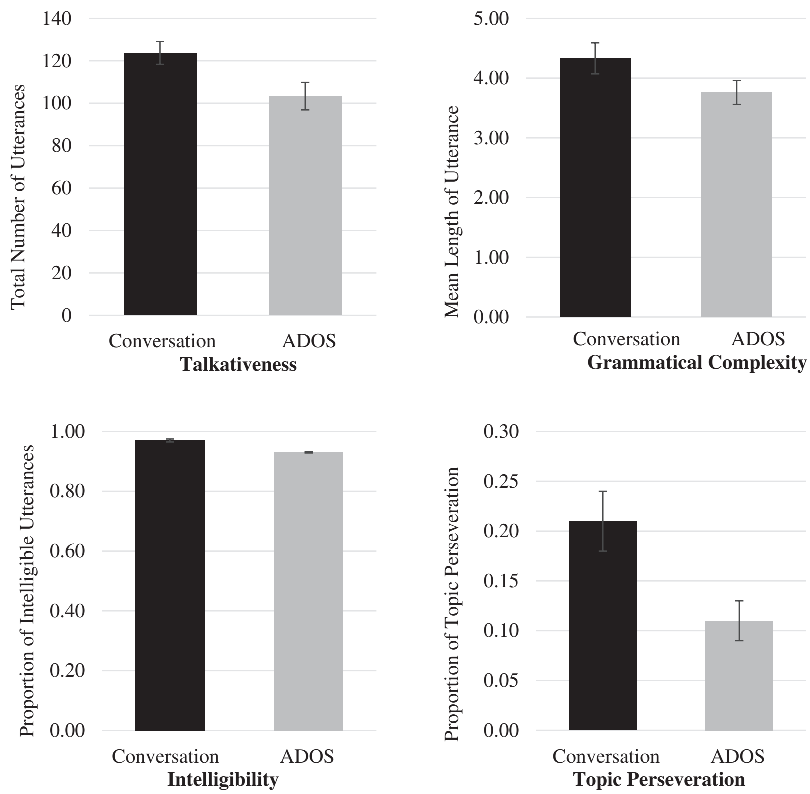

Figure 2. Significant context differences (collapsed across group). Standard error bars are shown.
ADOS: Autism Diagnostic Observation Schedule.

significant group differences and Figure 2 highlights the significant context differences.

Talkativeness. The mixed-model ANOVA revealed that boys produced significantly more utterances in the conversation sample than in the ADOS sample, $F(1, 35) = 16.35$, $p = .0001$, $\eta_p^2 = .32$. There was no main effect of diagnostic group, $F(1, 35) = 1.44$, $p = .238$, $\eta_p^2 = .04$, or significant interaction between diagnostic group and sampling context, $F(1, 35) = .03$, $p = .859$, $\eta_p^2 = .001$.

Lexical diversity. Analyses revealed that boys with idiopathic ASD had more lexically diverse language samples (i.e. greater MATTRs) than boys with FXS + ASD, $F(1, 35) = 11.87$, $p = .001$, $\eta_p^2 = .25$. No differences in lexical diversity were found between the conversation and ADOS samples, $F(1, 35) = 1.74$, $p = .196$, $\eta_p^2 = .05$. Similarly, no significant interaction was present, $F(1, 35) = 0.76$, $p = .391$, $\eta_p^2 = .02$.

Grammatical complexity. In terms of grammatical complexity, children's utterances were more complex during the conversation sample than the ADOS sample, $F(1, 36) = 13.46$, $p = .001$, $\eta_p^2 = .27$.

Intelligibility. Analyses revealed a significant interaction between sampling context and diagnostic group, $F(1, 35) = 6.51$ $p = .015$, $\eta_p^2 = .16$, as well as significant main effects of sampling context, $F(1, 35) = 10.54$, $p = .003$, $\eta_p^2 = .23$, and diagnostic group, $F(1, 35) = 13.06$, $p = .001$, $\eta_p^2 = .27$. Follow-up tests with Holm–Bonferroni corrections revealed that the utterances of boys with idiopathic ASD were more intelligible than the utterances of children with FXS + ASD in both the conversation, $t(21.51) = 3.11$, $p = .013$, and ADOS sampling contexts, $t(19.77) = 3.30$, $p = .009$. Moreover, both boys with idiopathic ASD and boys with FXS + ASD produced a higher proportion of intelligible speech during the conversation than the ADOS (respectively, $t(17) = -2.22$, $p = .040$; $t(18) = -3.02$, $p = .014$). However, there seems to be a larger difference between context for the boys with FXS + ASD than the boys with ASD.

Perseveration. In terms of topic perseveration, analyses revealed a significant main effect of sampling context, $F(1, 35) = 12.08$, $p = .001$, $\eta_p^2 = .26$, as well as a significant main effect of diagnostic group, $F(1, 35) = 5.06$, $p = .031$, $\eta_p^2 = .13$, but not a significant interaction, $F(1, 35) = 2.12$, $p = .155$, $\eta_p^2 = .06$. These results show that both groups used more topic perseveration during the conversation than the ADOS, but boys with idiopathic

ASD used less topic perseveration overall than boys with FXS + ASD. At the utterance level, no significant differences were found in terms of sampling context, $F(1, 35) = 1.66$, $p = .205$, $\eta_p^2 = .05$, or group, $F(1, 35) = 2.20$, $p = .147$, $\eta_p^2 = .06$. Similarly, the proportion of conversational device perseveration did not differ as a function of context, $F(1, 35) = 2.80$, $p = .090$, $\eta_p^2 = .08$, or group, $F(1, 35) = 0.27$, $p = .620$, $\eta_p^2 = .01$. Moreover, no interactions were found between group and context for either utterance, $F(1, 35) = 0.64$, $p = .428$, $\eta_p^2 = .02$, or conversational device perseverations, $F(1, 35) = 0.98$, $p = .330$, $\eta_p^2 = .03$.

Discussion

Understanding the individual language profiles of children with phenotypically similar, but distinct neurodevelopmental disorders, such as idiopathic ASD and FXS + ASD, has both clinical and theoretical implications. Such comparisons can inform our understanding of how ASD impacts language, and whether uniquely tailored language interventions may be needed for these clinical groups (Abbeduto & Murphy, 2004). However, studies comparing the expressive language profiles of the two groups have been limited, and results have been inconsistent, partially as a reflection of different assessment methods being utilized (Haebig & Sterling, 2017; Klusek et al., 2014b; Martin et al., 2018; Sterling, 2018; Thurman et al., 2017). Thus, the purpose of the present study was to compare the expressive language profiles of boys with FXS + ASD and boys with idiopathic ASD, and to examine whether a similar linguistic profile emerged across two different language sampling contexts that are commonly used in the literature (i.e. ADOS, semi-structured conversation; e.g. Estigarribia et al., 2011; Finestack et al., 2014; Friedman et al., 2018; Price et al., 2008).

Examination of group differences in expressive language

Our findings revealed that boys with FXS + ASD generally demonstrated poorer expressive language skills in several domains compared to boys with idiopathic ASD despite being matched on ASD severity and having similar MLUs and chronological age. Although groups did not differ in their overall talkativeness, boys with FXS + ASD had less lexically diverse language samples (i.e. lower MATTRs). This finding aligns with Haebig and Sterling (2017) who found that boys with FXS + ASD demonstrated lower standardized expressive vocabulary growth scores compared to ASD severity matched boys with idiopathic ASD. Additionally, we found that the speech of boys with FXS + ASD was less intelligible than that of boys with idiopathic ASD. Even though intelligibility differed between the groups, and poor intelligibility is often considered a characteristic of FXS, intelligibility was quite high in our sample. For both groups, the average proportion of intelligible utterances was at or above 88% across all sampling contexts. Using a language sample collected from the ADOS, Barnes et al. (2009) also found that the percentage of intelligible words was over 80% for a group of 5–15-year-old boys with FXS + ASD. As a result, our findings suggest that intelligibility may not be a linguistic characteristic that should be the primary clinical concern for boys with FXS + ASD, at least for boys within these age ranges and when using these types of sampling contexts.

Although the groups did not differ in terms of utterance or conversational device perseveration, boys with FXS + ASD did perseverate more on topics, themes, and ideas throughout the language samples compared to the boys with idiopathic ASD. This is somewhat at odds with the results of Friedman et al. (2018) who found that boys with FXS + ASD used a significantly greater frequency of single-utterance perseveration than boys with idiopathic ASD. However, their analyses also indicated that the difference between clinical groups for topic-level perseveration approached significance and a large effect size was present ($d = .82$). There are several possible reasons why we did not find a group difference in single-utterance perseveration in the current study. First, our findings are collapsed across two different sampling contexts (ADOS, conversation), whereas Friedman et al. (2018) found this pattern of results when simply using a conversation sample. Secondly, because of the number of dependent variables in the study, we chose to measure utterance level perseverations that included one or more repetitions, whereas Friedman et al. (2018) categorized utterance level perseverations under two categories (i.e. one repetition, more than two repetitions) and analyzed these variables separately. This discrepancy further highlights the need to consider the sampling context as well as the specific facet of language that is measured.

Examination of context differences in expressive language

Despite both language samples being 10 minutes in length, boys in both groups were more talkative, producing more utterances, in the conversation sample compared to the ADOS sample. One explanation for this difference could be that the semi-structured conversation provided more instances that necessitated a response, and as such the examiner may have provided a higher level of scaffolding in this context. Moreover, because the conversation sample was flexibly structured around a list of topics, starting with a topic of interest, boys may have been more engaged during the semi-structured conversation than during the ADOS. In contrast, during the ADOS, the examiner tries to engage the child in play and social routines that the child may not be motivated to participate in. Additionally, it is important to note that during many portions of the ADOS, the child is also able to engage with toys or pictures; this is not the case in the conversation sampling context. In line with this result, we also found that conversation samples were more grammatically complex than the ADOS samples. Given that there were no props or pictures used, conversation samples may have required or encouraged more complete or elaborate utterances, resulting in the longer MLUs. Additionally, both groups produced a higher frequency of intelligible utterances in the conversation sample than the ADOS sample. However, as previously discussed, intelligibility was still high in both contexts.

In contrast, boys with FXS + ASD and idiopathic ASD produced a greater frequency of topic perseverations in the conversation sample compared to the ADOS. More specifically, the proportion of topic perseverations almost doubled in the conversational context (21%) compared to the ADOS (11%). One possible explanation for this may be that the changing toys/props in the ADOS could have served as an external signal to change topics rather than to perseverate on a specific topic. However, contrary to our findings, Martin et al. (2012) found that boys with FXS + ASD used more topic perseverations and conversational device perseverations in the ADOS compared to a narrative sample. Therefore, different profiles or degrees of perseveration seem to emerge in different contexts for boys with FXS + ASD and ASD. This is important to consider given that topic perseveration is a hallmark characteristic in individuals with FXS and ASD and it is important to quantify this variable in addition to the lexical and grammatical variables that are most often quantified and analyzed in language samples.

Nevertheless, not all dimensions of language differed between contexts. In addition to finding similarities in utterance or conversational device perseveration between contexts, the boys also produced language samples that were similar in their lexical diversity. This is at odds with Kover et al. (2014) who found that preschool-age children with ASD produced fewer different words during the ADOS than their play-based language samples. However, a number of methodological differences may contribute to this discrepant finding including the specific type of conversational sample used (semi-structured conversation vs. play), the age of the participants (school-age vs. preschool), and the specific way in which lexical diversity was measured (MATTR vs. number of different words). Finally, while context differences did emerge, the relative lack of interaction effects between group and context suggests that the ADOS distinguished between boys with FXS + ASD and idiopathic ASD in the same manner as the conversation sample.

Limitations and future directions

Although we believe our findings have important clinical and theoretical implications, several limitations related to sampling and methodology must be noted. First, sample sizes were relatively small for both groups which may have precluded our ability to detect significant interactions between diagnostic group and context. However, FXS is a rare disorder, and our sample size is similar to those of published studies (McDuffie et al., 2012; Sterling, 2018). Second, our study did not include a group of boys with FXS-Only or a group of boys with TD, which limits our ability to understand the extent to which an ASD diagnosis impacts expressive language in FXS. Although nonverbal IQ was not significantly related to expressive language performance, research should also consider comparing boys with FXS-Only and FXS + ASD to boys with idiopathic ASD with and without co-occurring intellectual disability to better understand the impact of IQ on language.

In terms of language sampling procedures, it is important to note that transcription reliability fell slightly below 80% (range = 78–95%) for several conversation variables. Moreover, we selected the activities for the ADOS sample for several reasons (e.g. overlap between modules, opportunity for minimally structured communication, and based on previous literature). However, it is possible that a different combination of tasks or a longer language sample may have elicited a different linguistic profile. Relatedly, the number of tasks completed during the 10-minute ADOS sample varied across children, which could have influenced language performance. Future work should assess whether group differences in expressive language hold when using all of the ADOS tasks that overlap between Modules 2 and 3. Finally, while MLU has been found to strongly correlate with other measures of grammar (Condouris et al., 2003; Rice et al., 2006), it does not provide a thorough representation of grammatical development, particularly at older ages and with more advanced MLU. Thus, future studies should consider using the Index of Productive Syntax (Scarborough, 1990), Developmental Sentence Scoring (Lee & Canter, 1971), or clause density (e.g. Keller-Bell & Abbeduto, 2007; Nippold et al., 2008; Scott & Stokes, 1995) to provide a more in-depth analysis of how grammar may be influenced by diagnosis and sampling context.

Conclusions and implications

Using spontaneous language sampling, we found that there were overlapping language features (i.e. talkativeness, utterance, and conversational device perseveration), as well as distinctions (i.e. lexical diversity, intelligibility, and topic-level perseveration) in the expressive language phenotypes of boys with FXS + ASD and boys with idiopathic ASD. These findings help to further elucidate the unique linguistic profiles of each clinical group. Moreover, we found that the ADOS sample elicited a relatively different language profile for boys with FXS + ASD and idiopathic ASD than the conversation sample. A conversation with one person using a semi-structured list of topics elicited one type of result, whereas the ADOS, which uses various tasks and manipulatives, produced a different profile of language in terms of talkativeness, grammatical complexity, intelligibility, and topic-level perseveration. Thus, our findings highlight that selecting an appropriate sampling technique is essential, particularly when working with children with complex disorders. Researchers should keep this information in mind when interpreting language measures collected from the ADOS or a conversation sample. Finally, given that the different properties of a sampling context may impact the conclusions reached about children's expressive language profiles, if it is possible, it may be best to collect more than one language sample using multiple sampling techniques to obtain a complete picture of a child's linguistic ability (Abbeduto et al., 1995; Tager-Flusberg et al., 2009).

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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*, 270–288.

Abbeduto, L., & Murphy, M. M. (2004). Language, social cognition, maladaptive behavior, and communication in Down syndrome and fragile X syndrome. In M. L. Rice & S. F. Warren (Eds.), *Developmental language disorders: From phenotypes to etiologies* (pp. 77–96). Lawrence Erlbaum Associates.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). American Psychiatric Publishing.

Bacon, E. C., Osuna, S., Courchesne, E., & Pierce, K. (2019). Naturalistic language sampling to characterize the language abilities of 3-year-olds with autism spectrum disorder. *Autism*, *23*, 699–712. https://doi.org/10.1177/1362361318766241

Bailey, D. B., Raspa, M., Olmsted, M., & Holiday, D. B. (2008). Co-occurring conditions associated with FMR1 gene variations: Findings from a national parent survey. *American Journal of Medical Genetics, Part A*, *146*, 2060–2069. https://doi.org/10.1002/ajmg.a.32439

Baio, J. L., Wiggins, L., Christensen, D. L., Maenner, M. J., Daniels, J., Warren, Z., Kurzius-Spencer, M., Zahorodny, W., Robinson Rosenberg, C., White, T., Durkin, M. S., Imm, P., Nikolaou, L., Yeargin-Allsopp, M., Lee, L. C., Harrington, R., Lopez, M., Fitzgerald, R. T., Hewitt, A., ... Dowling, N. F. (2018). Prevalence of autism spectrum disorder among children aged 8 years – Autism and developmental disabilities monitoring network, 11 sites, United States, 2014. *MMWR Surveillance Summaries*, *67*, 1–2. https://doi.org/10.15585/mmwr.ss6706a1

Barnes, E., Roberts, J., Long, S. H., Martin, G. E., Berni, M. C., Mandulak, K. C., & Sideris, J. (2009). Phonological accuracy and intelligibility in connected speech of boys with fragile X syndrome or Down syndrome. *Journal of Speech, Language, and Hearing Research*, *52*, 1048–1061. https://doi.org/10.1044/1092-4388(2009/08-0001)

Berninger, V. W., & Abbott, R. D. (2010). Listening comprehension, oral expression, reading comprehension, and written expression: Related yet unique language systems in grades 1, 3, 5, and 7. *Journal of Educational Psychology*, *102*, 635–651. https://doi.org/10.1037/a0019319

Berry-Kravis, E., Doll, E., Sterling, A., Kover, S. T., Schroeder, S. M., Mathur, S., & Abbeduto, L. (2013). Development of an expressive language sampling procedure in fragile X syndrome. *Journal of Developmental & Behavioral Pediatrics*, *34*, 245–251. https://doi.org/10.1097/DBP.0b013e31828742fc

Boucher, J. (2012). Research review: Structural language in autistic spectrum disorder – Characteristics and causes.

Journal of Child Psychology and Psychiatry and Allied Disciplines, *53*, 219–233. https://doi.org/10.1111/j.1469-7610.2011.02508.x

Budimirovic, D. B., Bukelis, I., Cox, C., Gray, R. M., Tierney, E., & Kaufmann, W. E. (2006). Autism spectrum disorder in fragile X syndrome: Differential contribution of adaptive socialization and social withdrawal. *American Journal of Medical Genetics Part A*, *140A*, 1814–1826. https://doi.org/10.1002/ajmg.a.31405

Channell, M. M., Loveall, S. J., Conners, F. A., Harvey, D. J., & Abbeduto, L. (2018). Narrative language sampling in typical development: Implications for clinical trials. *American Journal of Speech-Language Pathology*, *27*, 123–135. https://doi.org/10.1044/2017_AJSLP-17-0046

Clifford, S., Dissanayake, C., Bui, Q. M., Huggins, R., Taylor, A. K., & Loesch, D. Z. (2007). Autism spectrum phenotype in males and females with fragile X full mutation and premutation. *Journal of Autism and Developmental Disorders*, *37*, 738–747. https://doi.org/10.1007/s10803-006-0205-z

Cohen, J. (1988). *Statistical power analysis for the behavioral sciences* (2nd ed.). Erlbaum.

Condouris, K., Meyer, E., & Tager-Flusberg, H. (2003). The relationship between standardized measures of language and measures of spontaneous speech in children with autism. *American Journal of Speech-Language Pathology*, *12*, 349. https://doi.org/10.1044/1058-0360(2003/080)

Covington, M. A., & McFall, J. D. (2010). Cutting the gordian knot: The moving-average type-token ratio (MATTR). *Journal of Quantitative Linguistics*, *17*, 94–100. https://doi.org/10.1080/09296171003643098

Dennis, M., Francis, D., Cirino, P., Barnes, M., & Fletcher, J. (2009). Why IQ is not a covariate in cognitive studies of neurodevelopmental disorders. *Journal of International Neuropsychological Society*, *15*, 331–343. https://doi.org/10.1017/S1355617709090481

Dunn, L. M., & Dunn, D. M. (2007). *Peabody Picture Vocabulary Test – 4th edition* (4th ed.). NCS Pearson.

Eigsti, I. M., Bennetto, L., & Dadlani, M. B. (2007). Beyond pragmatics: Morphosyntactic development in autism. *Journal of Autism and Developmental Disorders*, *37*, 1007–1023. https://doi.org/10.1007/s10803-006-0239-2

Estigarribia, B., Roberts, JE., 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. https://doi.org/10.3109/13682822.2010.487885

Fergadiotis, G., Wright, H. H., & Green, S. B. (2015). Psychometric evaluation of lexical diversity indices: Assessing length effects. *Journal of Speech Language and Hearing Research*, *58*, 840. https://doi.org/10.1044/2015_JSLHR-L-14-0280

Fergadiotis, G., Wright, H. H., & West, T. M. (2013). Measuring lexical diversity in narrative discourse of people with aphasia. *American Journal of Speech-Language Pathology*, *22*, S397–S408. https://doi.org/10.1044/1058-0360(2013/12-0083)

Finestack, L. H., Payesteh, B., Disher, J. R., & Julien, H. M. (2014). Reporting child language sampling procedures. *Journal of Speech, Language, and Hearing Research*, *57*, 2274–2279. https://doi.org/10.1044/2014_JSLHR-L-14-0093

Friedman, L., Lorang, E., & Sterling, A. (2019). The use of demonstratives and personal pronouns in fragile X syndrome and autism spectrum disorder. *Clinical Linguistics & Phonetics*, *33*, 420–436. https://doi.org/10.1080/02699206.2018.1536727

Friedman, L., Sterling, A., & Barton-Hulsey, A. (2018). Gaze avoidance and perseverative language in fragile X syndrome and autism spectrum disorder: Brief report. *Developmental Neurorehabilitation*, *21*, 137–140. https://doi.org/10.1080/17518423.2018.1424264

Haebig, E., & Sterling, A. (2017). Investigating the receptive-expressive vocabulary profile in children with idiopathic ASD and comorbid ASD and fragile X syndrome. *Journal of Autism and Developmental Disorders*, *47*, 260–274. https://doi.org/10.1007/s10803-016-2921-3

Haebig, E., Sterling, A., & Hoover, J. (2016). Examining the language phenotype in children with typical development, specific language impairment, and fragile X syndrome. *Journal of Speech, Language, and Hearing Research*, *59*, 1046–1058. https://doi.org/10.1044/2016_JSLHR-L-15-0185

Hallgren, K. A. (2012). Computing inter-rater reliability for observational data: An overview and tutorial. *Tutorials in Quantitative Methods for Psychology*, *8*, 23–34. https://doi.org/10.20982/tqmp.08.1.p023

Hartmann, D. P., Barrios, B. A., & Wood, D. D. (2004). Principles of behavioral observation. In M. Hersen (Ed.), *Comprehensive handbook of psychological assessment* (*Vol. 3*, pp. 108–137). John Wiley & Sons.

Heilmann, J. J., Miller, J. F., & Nockerts, A. (2010). Using language sample databases. *Language, Speech, and Hearing Services in Schools*, *41*, 84–95. https://doi.org/10.1044/0161-1461(2009/08-0075)

Hernandez, R. N., Feinberg, R. L., Vaurio, R., Passanante, N. M., Thompson, R. E., & Kaufmann, W. E. (2009). Autism spectrum disorder in fragile X syndrome: A longitudinal evaluation. *American Journal of Medical Genetics, Part A*, *149*, 1125–1137. https://doi.org/10.1002/ajmg.a.32848

Hessl, D., Nguyen, D. V., Green, C., Chavez, A., Tassone, F., Hagerman, R. J., Senturk, D., Schneider, A., Lightbody, A., Reiss, A. L., Hall, S. (2009). A solution to limitations of cognitive testing in children with intellectual disabilities: The case of fragile X syndrome. *Journal of Neurodevelopmental Disorders*, *1*, 33–45. https://doi.org/10.1007/s11689-008-9001-8

Hogan-Brown, A. L., Losh, M., Martin, G. E., & Mueffelmann, D. J. (2013). An investigation of narrative ability in boys with autism and fragile X syndrome. *American Journal on Intellectual and Developmental Disabilities*, *118*, 77–94. https://doi.org/10.1352/1944-7558-118.2.77

Hoover, J. R., Storkel, H. L., & Rice, M. L. (2012). The interface between neighborhood density and optional infinitives: Normal development and specific language impairment. *Journal of Child Language*, *39*, 835–862. https://doi.org/10.1017/S0305000911000365

Keller-Bell, Y. D., & Abbeduto, L. (2007). Narrative development in adolescents and young adults with fragile X syndrome. *American Journal on Mental Retardation*, *112*, 289–299. https://doi.org/10.1352/0895-8017(2007)112[289:NDIAAY]2.0.CO;2

Klusek, J., Martin, G. E., & Losh, M. (2014a). Consistency between research and clinical diagnoses of autism among boys and girls with fragile X syndrome. *Journal of Intellectual Disability Research*, *58*, 940–952. https://doi.org/10.1111/jir.12121

Klusek, J., Martin, G. E., & Losh, M. (2014b). A comparison of pragmatic language in boys with autism and fragile X syndrome. *Journal of Speech, Language, and Hearing Research*, *57*, 1692–1707. https://doi.org/10.1044/2014_JSLHR-L-13-0064

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. https://doi.org/10.1111/j.1365-2788.2010.01255.x

Kover, S. T., & Atwood, A. K. (2013). Establishing equivalence: Methodological progress in group-matching design and analysis. *American Journal on Intellectual and Developmental Disabilities*, *118*, 3–15. https://doi.org/10.1352/1944-7558-118.1.3

Kover, S. T., Davidson, M. M., Sindberg, H. A., & Ellis Weismer, S. (2014). Use of the ADOS for assessing spontaneous expressive language in young children with ASD: A comparison of sampling contexts. *Journal of Speech, Language, and Hearing Research*, *57*, 2221–2233. https://doi.org/10.1044/2014_JSLHR-L-13-0330

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*, 1022–1039. https://doi.org/10.1044/1092-4388(2011/11-0075)

Lee, L. L., & Canter, S. M. (1971). Developmental Sentence Scoring: A clinical procedure for estimating syntactic development in children's spontaneous speech. *Journal of Speech and Hearing Disorders*, *36*, 315–340. https://doi.org/10.1044/jshd.3603.315

Lee, M., Martin, G. E., Hogan, A., Hano, D., Gordon, P. C., & Losh, M. (2018). What's the story? A computational analysis of narrative competence in autism. *Autism*, *22*, 335–344. https://doi.org/10.1177/1362361316677957

Levy, Y., Gottesman, R., Borochowitz, Z., Frydman, M., & Sagi, M. (2006). Language in boys with fragile X syndrome. *Journal of Child Language*, *33*, 125–144. https://doi.org/10.1017/s030500090500718x

Loesch, D. Z., Huggins, R. M., Bui, Q. M., Epstein, J. L., Taylor, A. K., & Hagerman, R. J. (2002). Effect of the deficits of fragile X mental retardation protein on cognitive status of fragile X males and females assessed by robust pedigree analysis. *Journal of Developmental and Behavioral Pediatrics*, *23*, 416–423. https://doi.org/10.1097/00004703-200212000-00004

Lord, C., Rutter, M., DiLavore, P. C., & Risi, S. (1999). *Autism diagnostic observation schedule*. Western Psychological Services.

Lord, C., Rutter, M., DiLavore, P. C., Risi, S., Gotham, K., & Bishop, S. L. (2012). *Autism Diagnostic Observation Schedule, Second Edition (ADOS-2)*. Western Psychological Services.

Losh, M., Martin, G. E., Klusek, J., Hogan-Brown, A. L., & Sideris, J. (2012). Social communication and theory of mind in boys with autism and fragile X syndrome. *Frontiers in Psychology*, *3*, 1–12.

Martin, G. E., Barstein, J., Hornickel, J., Matherly, S., Durante, G., & Losh, M. (2017). Signaling of noncomprehension in communication breakdowns in fragile X syndrome, Down syndrome, and autism spectrum disorder. *Journal of Communication Disorders*, *65*, 22–34. https://doi.org/10.1016/j.jcomdis.2017.01.003

Martin, G. E., Bush, L., Klusek, J., Patel, S., & Losh, M. (2018). A multimethod analysis of pragmatic skills in children and adolescents with fragile X syndrome, autism spectrum disorder, and Down syndrome. *Journal of Speech, Language, & Hearing Research*, *61*, 3023–3037. https://doi.org/10.1044/2018_JSLHR-L-18-0008

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 and Communication Disorders*, *48*, 432–443. https://doi.org/10.1111/1460-6984.12019

Martin, G. E., Roberts, J. E., Helm-Estabrooks, N., Sideris, J., Vanderbilt, J., & Moskowitz, L. (2012). Perseveration in the connected speech of boys with fragile X syndrome with and without autism spectrum disorder. *American Journal on Intellectual and Developmental Disabilities*, *117*, 384–399. https://doi.org/10.1352/1944-7558-117.5.384

McDuffie, A., Kover, S., Abbeduto, L., Lewis, P., & Brown, T. (2012). Profiles of receptive and expressive language abilities in boys with comorbid fragile X syndrome and autism. *American Journal on Intellectual and Developmental Disabilities*, *117*, 18–32. https://doi.org/10.1352/1944-7558-117.1.18

Mervis, C. B., & Robinson, B. F. (2005). Designing measures for profiling and genotype/phenotype studies of individuals with genetic syndromes or developmental language disorders. *Applied Psycholinguistics*, *26*, 41–64. https://doi.org/10.1017/S0142716405050058

Miller, J. F., Andriacchi, K., & Nockerts, A. (2011). *Assessing language production using SALT software: A clinician's guide to language sample analysis*. SALT Software, LLC.

Murphy, M. M., & Abbeduto, L. (2007). Gender differences in repetitive language in fragile X syndrome. *Journal of Intellectual Disability Research*, *51*, 387–400. https://doi.org/10.1111/j.1365-2788.2006.00888.x

Nippold, M. A., Hesketh, L. J., Duthie, J. K., & Mansfield, T. C. (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. https://doi.org/10.1044/1092-4388(2005/073)

Nippold, M. A., Mansfield, T. C., Billow, J. L., & Tomblin, J. B. (2008). Expository discourse in adolescents with language impairments: Examining syntactic development. *American Journal of Speech-Language Pathology*, *17*, 356–366. https://doi.org/10.1044/1058-0360(2008/07-0049)

Park, C. J., Yelland, G. W., Taffe, J. R., & Gray, K. M. (2012). Morphological and syntactic skills in language samples of pre school aged children with autism: Atypical development? *International Journal of Speech-Language Pathology*, *14*, 95–108. https://doi.org/10.3109/17549507.2011.645555

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. https://doi.org/10.1044/1092-4388(2008/001)

Rice, M. L., Redmond, S. M., & Hoffman, L. (2006). Mean length of utterance in children with specific language impairment and in younger control children shows concurrent validity and stable and parallel growth trajectories. *Journal of Speech, Language, and Hearing Research*, *49*, 793. https://doi.org/10.1044/1092-4388(2006/056)

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. https://doi.org/10.1044/1092-4388(2009/08-0183)

Richards, M., Mossey, J., & Robins, D. L. (2016). Parents' concerns as they relate to their child's development and later diagnosis of autism spectrum disorder. *Journal of Developmental and Behavioral Pediatrics*, *37*, 532–540. https://doi.org/10.1097/DBP.0000000000000339

Roberts, J. E., Hennon, E. A., Price, J. R., Dear, E., Anderson, K., & Vandergrift, N. A. (2007a). Expressive language during conversational speech in boys with fragile X syndrome. *American Journal on Mental Retardation*, *112*, 1–17. https://doi.org/10.1352/0895-8017(2007)112[1:ELDCSI]2.0.CO;2

Roberts, J. E., Mirrett, P., & Burchinal, M. (2001). Receptive and expressive communication development of young males with fragile X syndrome. *American Journal on Mental Retardation*, *106*, 216–230. https://doi.org/10.1352/0895-8017(2001)106<0216:RAECDO>2.0.CO;2

Roberts, J. E., Price, J., Barnes, E., Nelson, L., Burchinal, M., Hennon, E. A., Moskowitz, L., Edwards, A., Malkin, C., Anderson, K., Misenheimer, J., Hooper, S. R. (2007b). Receptive vocabulary, expressive vocabulary, and speech production of boys with fragile X syndrome in comparison to boys with Down syndrome. *American Journal on Mental Retardation*, *112*(3): 177–193.

Roid, G., & Miller, L. (1997). *Leiter International Performance Scale—Revised*. Stoelting.

Rutter, M., LeCouteur, A., & Lord, C. (2003). *Autism Diagnostic Inventory, Revised*. Western Psychological Services.

Ryland, H. K., Hysing, M., Posserud, M. B., Gillberg, C., & Lundervold, A. J. (2014). Autistic features in school age children: IQ and gender effects in a population-based cohort. *Research in Autism Spectrum Disorders*, *8*, 266–274. https://doi.org/10.1016/j.rasd.2013.12.001

Scarborough, H. S. (1990). Index of productive syntax. *Applied Psycholinguistics*, *11*, 1–22. https://doi.org/10.1017/S0142716400008262

Scott, C. M., & Stokes, S. L. (1995). Measures of syntax in school-age children and adolescents. *Language, Speech & Hearing Services in Schools*, *26*, 309–319. https://doi.org/10.1044/0161-1461.2604.309

Scott, C. M., & Windsor, J. (2000). General language performance measures in spoken and written narrative and expository discourse of school-age children with language learning disabilities. *Journal of Speech Language and Hearing Research*, *43*, 324. https://doi.org/10.1044/jslhr.4302.324

Southwood, F., & Russell, A. F. (2004). Comparison of conversation, freeplay, and story generation as methods of language sample elicitation. *Journal of Speech, Language, and Hearing Research*, *47*, 366–376. https://doi.org/10.1044/1092-4388(2004/030)

Sterling, A. (2018). Grammar in boys with idiopathic autism spectrum disorder and boys with fragile X syndrome plus autism spectrum disorder. *Journal of Speech Language and Hearing Research*, *61*, 857–869. https://doi.org/10.1044/2017_JSLHR-L-17-0248

Stills, M. M. (2016). *Language sample length effects on various lexical diversity measures: An analysis of Spanish language samples from children* [Unpublished doctoral dissertation]. Portland State University.

Tabachnik, B. G., & Fidell, L. S. (2001). *Using multivariate statistics (4th ed.)*. Allyn and Bacon.

Tager-Flusberg, H., Edelson, L., & Luyster, R. (2011). Language and communication in autism spectrum disorders. In D. Amaral, G. Dawson, & D. Geschwind (Eds.), *Autism spectrum disorders* (pp. 172–185). Oxford University Press.

Tager-Flusberg, H., Rogers, S., Cooper, J., Landa, R., Lord, C., Paul, R., Rice, M., Stoel-Gammon, C., Wetherby, A., Yoder, P. (2009). Defining spoken language benchmarks and selecting measures of expressive language development for young children with autism spectrum disorders. *Journal of Speech, Language, and Hearing Research*, *52*, 643–652. https://doi.org/10.1044/1092-4388(2009/08-0136)

Thurman, A. J., McDuffie, A., Hagerman, R. J., Josol, C. K., & Abbeduto, L. (2017). Language skills of males with fragile X syndrome or nonsyndromic autism spectrum disorder. *Journal of Autism and Developmental Disorders*, *47*, 728–743. https://doi.org/10.1007/s10803-016-3003-2

Verkerk, A. J. M., Pieretti, M., Sutcliffe, J. S., Fu, Y. H., Kuhl, D. P., Pizzuti, A., Reiner, O., Richards, S., Victoria, M. F., Zhang, F. P. (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*, 905–914. https://doi.org/10.1016/0092-8674(91)90397-H

Westerveld, M. F., Gillon, G. T., & Miller, J. F. (2004). Spoken language samples of New Zealand children in conversation and narration. *Advances in Speech Language Pathology*, *6*, 195–208. https://doi.org/10.1080/14417040400010140

Williams, K. T. (2007). *Expressive Vocabulary Test* (2nd ed.). Pearson Assessments.