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Language Comprehension Profiles of Young Adolescents With Fragile X Syndrome

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Purpose: In this study, the authors sought to characterize the language phenotype of fragile X syndrome (FXS), focusing on the extent of impairment in receptive syntax, within-syndrome variability in those impairments in relation to gender, and the syndrome specificity of those impairments.
Method: The Test for Reception of Grammar, Version 2 (Bishop, 2003), was used to examine the overall receptive syntactic skills of adolescents with FXS (*n* = 35; 30 males, 5 females), adolescents with Down syndrome (DS; *n* = 28; 18 males, 10 females), and younger typically developing (TD) children (*n* = 23; 14 males, 9 females) matched on nonverbal cognition. Performance on specific grammatical constructions and error types was examined for a subset of matched participants.
Results: Participants with FXS had overall receptive syntax scores that were lower than those of the TD participants but higher than those of the participants with DS; however, there was no difference in performance between the FXS and DS groups when females were excluded. Grammatical constructions that were especially difficult for participants with FXS and those with DS were identified, especially relative clause constructions and reversible constructions requiring attention to word order encoded by syntactic features.
Conclusion: The current findings have implications for understanding the nature of the language learning difficulties of individuals with FXS and for language interventions.

Key Words: adolescents, developmental disorders, language, syntax

Fragile X syndrome (FXS), the most common form of inherited intellectual disability, results from a mutation in the *FMR1* gene located on the X chromosome (Crawford, Acuna, & Sherman, 2001). It is estimated that 1 in 4,000 males and 1 in 6,000–8,000 females are affected with FXS (Centers for Disease Control and Prevention [CDC], 2010). Because it is an X-linked condition, FXS is more common in males than females, and males are more severely affected, on average, than females (Crawford et al., 2001). The cognitive abilities presumed to be important for language (e.g., auditory memory) are typically impaired or delayed in individuals with FXS (Ornstein et al., 2008). Consequently, most individuals with FXS have language impairments, although there is wide variability in the extent of the impairment even within each gender (Abbeduto, Brady, & Kover, 2007). It is not clear, however, whether impairments are variable across the different components of language. The aim of the current study was to further characterize the language phenotype of FXS. The focus was on receptive syntax, with the goals being to examine the extent and source(s) of impairments in receptive syntax, within-syndrome variability in those impairments in relation to gender, and the syndrome specificity of the impairments through a comparison with Down syndrome (DS).

The majority of previous research on language in individuals with FXS has focused on the expressive domain (Finestack & Abbeduto, 2010; Finestack, Palmer, & Abbeduto, 2012; Levy, Gottesman, Borochowitz, Frydman, & Sagi, 2006; Mazzocco et al., 2006; Murphy & Abbeduto, 2007; Price et al., 2008). Most of this research has relied on broad summary measures such as mean length of utterance (MLU) in spontaneous language samples or total scores on standardized tests (Abbeduto et al., 2007). Some studies, however, have assessed more specific aspects of expressive language, such as the use of complex clauses (Levy et al., 2006) or various elements of narrative structure (Finestack et al., 2012), with the goal being to provide a more detailed and clinically useful picture of language development in individuals with FXS. In general, individuals with FXS display delays in expressive language relative to their levels of

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nonverbal cognitive ability, with expressive syntax posing especially serious challenges (Finestack & Abbeduto, 2010; Finestack, Sterling, & Abbeduto, 2013; Levy et al., 2006; Price et al., 2008). It is important to extend these findings to the receptive modality in FXS, because the extent and profile of impairments has been found to be quite different across modalities in some disorders (Volden et al., 2011). Variation in impairments across modalities can provide insights into mechanisms and potential intervention targets.

Unfortunately, there have been only two studies of language comprehension in individuals with FXS. In the first study, Abbeduto et al. (2003) compared the receptive language skills of male and female adolescents and young adults with FXS without comorbid autism to those of typically developing (TD) children matched on nonverbal mental age (NVMA). Participants were assessed using the Test for Auditory Comprehension of Language—Revised (TACL–R; Carrow-Woolfolk, 1985), which includes subtests to measure vocabulary, grammatical morphology, and multiword syntactic patterns. Abbeduto et al. found that there were no differences in age-equivalent scores on any of the three subtests between the participants with FXS and the TD participants, suggesting that vocabulary, grammatical morphology, and syntax more generally keep pace with nonverbal cognition in adolescents and young adults with FXS. In the second study, Price, Roberts, Vandergrift, and Martin (2007) focused only on males with FXS and used the more recent Test for Auditory Comprehension of Language—Third Edition (Carrow-Woolfolk, 1998). Price et al. also found that these participants' performance was equally delayed across the domains of vocabulary, grammatical morphology, and syntax; however, they also found that boys with FXS regardless of comorbid autism status had lower age-equivalents than expected based on their nonverbal cognitive levels on all three subtests.

These inconsistent findings regarding performance relative to NVMA across the two studies might relate to the fact that the sample in the Abbeduto et al. (2003) study was older on average and included both males and females. Differences in the two versions of the TACL might have had an impact on the findings as well. In the present study, an attempt was made to clarify the issue by using the Test for Reception of Grammar, Version 2 (TROG–2; Bishop, 2003) rather than the TACL and systematically examining the impact of gender on the findings. In addition, the sample for the present study fell in the middle of, and overlapped with, the samples of Abbeduto et al. and Price et al. (2007) in terms of age.

Any attempt to characterize receptive language must recognize its complex and multifaceted nature. Many standardized assessments of receptive language, including those that purport to examine specific domains of language (e.g., receptive syntax), are seldom sufficient for characterizing profiles of receptive language impairments because they fail to fully probe mastery of specific linguistic elements or constructions. An individual who fails to understand a sentence such as *The man is chasing the dog,* for example, might do so because he or she does not understand the meanings of individual words, fails to recognize that the syntactic frame specifies that the first noun phrase is the agent and the second noun phrase the recipient, or simply cannot maintain the sentence in memory long enough to get to its meaning. Deciding among these (and other alternatives) is likely to require a more careful assessment of performance on specific linguistic constructions or an examination of the types of errors made during processing so as to isolate the source(s) of difficulty (Laws & Bishop, 2003). Isolating the source(s) of receptive language difficulties is important for adequately characterizing the phenotype of FXS as well as for identifying and prioritizing targets for intervention. In the present study, therefore, we examined individuals' comprehension of specific language forms and the types of errors they made in addition to the overall level of receptive language performance.

In the case of FXS, virtually all aspects of cognition are impaired; thus, there are likely to be numerous contributors to receptive language problems (Abbeduto et al., 2007). Nevertheless, there are two areas of especially severe cognitive impairment in individuals with FXS that are likely to have an important impact on receptive language. First, it has been documented that individuals with FXS are quite poor at processing sequential patterns (Burack et al., 1999; Dykens, Hodapp, & Leckman, 1987). This observation raises the possibility that language forms whose comprehension hinges on attending to the order of linguistic elements will be especially challenging for individuals with FXS. Second, auditory memory is also an area of special weakness (Baker et al., 2011), which raises the possibility that language forms that place particularly heavy demands on auditory memory during comprehension will also be especially challenging for individuals with FXS. In the present study, therefore, special attention was focused on the comprehension of (a) reversible sentences with the prepositions *in* and *on* (e.g., *The duck is on the ball*); (b) reversible subject-verb-object (SVO) sentences (e.g., *The man is chasing the dog*), each of which depends on processing information about word order; (c) sentences containing four lexical elements (e.g., *There is a yellow star and a big flower*); and (d) sentences with subject relative clauses (e.g., *The man that is eating looks at the cat*), each of which places heavy demands on auditory memory. Data on these forms will help illuminate the ways in which other aspects of the FXS cognitive phenotype contribute to language difficulties.

Because individuals with FXS, particularly males, often have an intellectual disability (i.e., an IQ of 70 or less), it is useful to determine the extent to which the FXS language is syndrome specific or common to individuals with an intellectual disability. Such a determination typically requires comparison with individuals who have an intellectual disability of a different etiology (Dykens, Hodapp, & Finucane, 2000). DS provides a useful comparison for FXS in part because the language phenotype of DS has been well described (Abbeduto & McDuffie, 2010). In particular, individuals with DS have especially severe deficits in the area of syntax, with these deficits being seen in both the expressive and receptive modalities (Abbeduto et al., 2003; Chapman, Schwartz, & Kay-Raining Bird, 1991; Glenn & Cunningham, 2005; Laws & Bishop, 2003; Miller, 1988). Moreover, the receptive syntax deficits in DS do not appear to be attributable solely to auditory memory impairments, hearing loss, or other nonlinguistic impairments (Chapman & Hesketh, 2001; Laws & Bishop, 2003); instead, syntactic processing appears to pose its own unique challenges or, at least, to magnify impairments in auditory memory, hearing, or other nonlinguistic domains of processing (Laws & Bishop, 2003).

Current Study

The current study evaluated the possibility that FXS, like DS, is characterized by especially severe impairments in receptive syntax. This possibility was addressed by comparing the receptive syntax performance of individuals with FXS to that of individuals with DS matched on NVMA. The measure of receptive syntax was the TROG–2, which has only recently been used in studies of FXS (McDuffie, Kover, Abbeduto, Lewis, & Brown, 2012; Pierpont, Richmond, Abbeduto, Kover, & Brown, 2011). Four constructions from the TROG–2 were selected for additional in-depth analysis: reversible *in* and *on* sentences, reversible SVO sentences, sentences containing four lexical elements, and sentences with subject relative clauses (see Table 1 for examples of all constructions). These forms were selected because they had been administered to a relatively large number of participants in the study (as part of a larger study) and because they make it possible to determine whether syntactic information per se placed an added burden on the poor sequential processing and limited auditory memory of individuals with FXS.

In particular, comparison of reversible *in* and *on* sentences to reversible SVO sentences is useful because the relationships among words are encoded lexically in the former sentences and syntactically in the latter sentences. Similarly, comparison of four-element sentences to sentences with subject relatives is useful because, despite being of similar length, only the latter are multiclause and, thus, highly syntactically complex (Bishop, 1997; Karmiloff-Smith et al., 1997; van der Lely & Harris, 1990). Finally, the types of comprehension errors committed by the participants were examined to determine whether they had their origin in syntactically or lexically based decisions during comprehension. Examination of such error types has helped uncover similarities and differences between individuals with DS and those with specific language impairment in regard to the source of receptive language difficulties (Laws & Bishop, 2003). Thus, the present study was designed to clarify the extent and nature of receptive impairments in individuals with FXS, thereby yielding insights into the factors that contribute to those impairments and identifying targets and avenues for intervention.

Study Questions and Predictions

- *Are there diagnostic group and gender differences in the overall receptive syntactic skills between young adolescents with FXS and young adolescents with DS?* In light of previous findings on DS (Abbeduto et al., 2003; Chapman et al., 1991; Glenn & Cunningham, 2005, Laws & Bishop, 2003; Miller, 1988), as well as the inconsistent findings for FXS (Abbeduto et al., 2003; Price et al., 2007), we hypothesized that after controlling for nonverbal cognitive ability, young adolescents with FXS would score higher in terms of overall receptive syntactic skills than would young adolescents with DS. No prediction was possible, however, as to how the receptive syntactic skills of young adolescents with FXS would compare to those of TD participants. Additionally, based on the X-linked nature of FXS, we predicted that males with FXS would score lower on overall receptive syntax than females with FXS.

Table 1. Examples of grammatical constructions in the Test for Reception of Grammar, Version 2 (TROG–2; Bishop, 2003).

Block	Construction	Example item
A	Two elements	*The sheep is running.*
B	Negative	*The fork is not big.*
C	Reversible *in* and *on*	*The duck is on the ball.*
D	Three elements	*The girl pushes the box.*
E	Reversible subject–verb–object (SVO)	*The man is chasing the dog.*
F	Four elements	*There is a yellow star and a big flower.*
G	Relative clause in subject	*The man that is eating looks at the cat.*
H	Not only X but also Y	*The man is not only running but also pointing.*
I	Reversible above and below	*The cup is below the star.*
J	Comparative/absolute	*The flower is longer than the comb.*
K	Reversible passive	*The cow is chased by the girl.*
L	Zero anaphor	*The book is on the scarf and is blue.*
M	Pronoun gender/number	*They are carrying him.*
N	Pronoun binding	*The girl sees that the lady is pointing at her.*
O	Neither nor	*The girl is neither pointing nor running.*
P	X but not Y	*The man but not the horse is jumping.*
Q	Postmodified subject	*The scarf on the shoe is blue.*
R	Singular/plural inflection	*The cat chases the ducks.*
S	Relative clause in object	*The man pushes the cow that is standing.*
T	Center-embedded sentence	*The sheep the girl looks at is running.*

- *Are there diagnostic group differences in the comprehension of sentences differing in length, reversibility, and clause embedding betweeen young adolescents with FXS and young adolescents with DS?* Due to the cognitive and auditory memory deficits found in both individuals with FXS (Baker et al., 2011; Ornstein et al., 2008) and those with DS (Chapman & Hesketh, 2001; Laws & Bishop, 2003), we hypothesized that both group profiles would reflect special difficulty with syntax and, thus, word order and clause embedding.
- *Are there diagnostic group differences in the pattern of lexical and syntactic errors across these sentence distinctions (i.e., length, reversibility, and syntactic complexity) between young adolescents with FXS and young adolescents with DS?* Due to the cognitive and auditory memory deficits found in both individuals with FXS (Baker et al., 2011; Ornstein et al., 2008) and those with DS (Chapman & Hesketh, 2001; Laws & Bishop, 2003), we hypothesized that the group profiles would reflect special difficulty with syntactically based errors relative to lexically based errors.

Method

Participants

Participants were drawn from a larger longitudinal project on language development in FXS and DS that involved four annual visits. During each of the four annual visits, participants completed a battery of language and cognitive measures. The current study used only a subset of these measures, most of which were obtained at the Time 1 (T1) assessment. Three groups participated in the larger study: 53 children and adolescents with FXS, ages 10;2 (years;months) to 16;0 at T1; 30 children and adolescents with DS, ages 10;2 to 15;9 at T1; and 56 TD children, ages 3;1 to 8;9 at T1. Ninety percent of the participants identified as Caucasian, 4% as African American, 4% as Hispanic, and 2% as "other." Regarding maternal educational level, 42% of the participants' mothers had graduated from high school, 43% had graduated from college, and 15% had graduated with an advanced degree.[1] Although other articles have been published based on this larger project (Kover, McDuffie, Abbeduto, & Brown, 2012; McDuffie et al., 2010; McDuffie et al., 2012; Pierpont et al., 2011), none has focused on the questions of interest in the present study.

Participants with FXS and DS were recruited nationally using a variety of sources, including a university recruitment registry, Internet websites and listservs, newspaper advertisements, and postings/flyers at parent meetings. TD participants were largely recruited locally using posters and flyers in public places and a university research registry. Enrollment criteria included English as the native language, use of three-word phrases at least occasionally, and no significant uncorrected vision or hearing impairments, all of which were determined from parent report. The parents of the TD children indicated that their child was not receiving special education services at the time of participation, with the exception of limited speech articulation treatment. The parents of the adolescents with FXS or DS provided diagnostic confirmation, generally through copies of medical records.

Individuals with more than a mild hearing loss (i.e., pure-tone air conduction threshold of 30 dB HL or higher in each ear averaged across 500, 1000, and 2000 Hz) at T1 were excluded from the current study, which resulted in the exclusion of one participant with DS. For one participant with FXS, hearing could not be evaluated at T1 or T2 (1 year after T1) because of his level of cooperation and functioning; however, his hearing was successfully evaluated at T3 (2 years after T1), and he was found to meet the hearing criterion; thus, he was included in the current study.

Several within-group sibling pairs (n = 8) participated in the larger longitudinal study; however, only one sibling from same-gender sibling pairs was included in the current study. This resulted in the exclusion of five TD participants. Opposite-gender sibling pairs were not excluded from the current study because gender was a factor of interest in some of the analyses. Scores from standardized measures of language and cognition, described later, were obtained during T1 for all but two participants with FXS and one TD participant. For these three participants, the tasks of interest were either not completed in a standardized way or not attempted because of noncompliance, or the participant completed less than half of the measure of nonverbal cognition. In these cases, data from the first valid measures (at T2 or T3) were substituted.

Procedure

Written consent was given by all parents before participation in the study. Testing sessions took place in a quiet room and lasted between 4 and 8 hr over the course of 2 days, with breaks taken as needed. A variety of standardized and experimental measures of language and cognition were administered as part of the larger longitudinal study. As previously mentioned, only a subset of these measures formed the basis for the current study.

Measures

Receptive language. Participants were assessed using the TROG–2, which is a standardized measure that is used to examine syntactic comprehension skills. According to the manual, internal consistency of the TROG–2 is .877 (N = 896). Correlations between the TROG–2 and subtests from the Clinical Evaluation of Language Fundamentals—Preschool, UK Edition (CELF–P^{UK}; Linguistics Concepts subtest; Wiig, Secord, & Semel, 2000) and the Clinical Evaluation of Language Fundamentals—Third, UK Edition (CELF–3^{UK}; Concepts and Directions subtest; Semel, Wiig, & Secord, 2000) reveal moderate levels of correlation (.582 and .525, respectively; Bishop, 2003).

The TROG–2 consists of 20 blocks (A – T), each testing a specific grammatical construction. The order of

[1]Missing for one participant with FXS.

administration of the blocks reflects their relative order of difficulty for the normative sample. Examples of the grammatical constructions are shown in Table 1. Each block contains four test items. The vocabulary included in the TROG–2 was restricted to simple words in order to minimize lexical influences on comprehension performance (Bishop, 2003). For each item in the TROG–2, the examiner read a sentence that referred to one of four drawings, and the participant's task was to point to the one drawing that corresponded to the meaning of the sentence. The distracter drawings, or foils, differed by either a lexical or grammatical element. Choosing a drawing that differed from the target by a lexical element (e.g., a noun, verb, or adjective) reflected a lexical error, whereas choosing a drawing that differed by a grammatical element (e.g., a function word, word order, or inflection) reflected a grammatical error. For example, an item testing the reversible SVO construction took the form, *The man is chasing the dog.* A lexical distracter for this item depicted a man chasing a ball; a grammatical distracter depicted a dog chasing a man. The foils for a few grammatical constructions (e.g., the four-element construction) contained only lexical distracters, meaning that only lexical errors were possible. Most blocks were constructed so that both lexical and grammatical distracters or only grammatical distracters were included.

Consistent with the manual and standardization of the TROG–2 (Bishop, 2003), testing began at the first item in Block A and was discontinued after five consecutive failed blocks, with one or more incorrect responses in a block constituting failure. The total number of blocks passed was used to calculate the standard and age-equivalent scores. Although using the total number of blocks passed is useful to determine an individual's overall receptive language ability, a finer level of analysis of an individual's performance on the TROG–2 is possible by examining the number of items answered correctly in each block as opposed to whether the block was failed or passed (McDuffie et al., 2012). In the current study, the total number of items answered correctly (rather than blocks passed) was used in the analyses of overall performance and performance on the grammatical constructions of interest.

Nonverbal cognition. Participants were assessed using the Brief IQ subtests of the Leiter International Performance Scale—Revised (Leiter–R; Roid & Miller, 1997): Figure Ground, Form Completion, Sequential Order, and Repeated Patterns. These subtests measure visualization and fluid reasoning skills and yield a nonverbal IQ score, an age-equivalent score, and a growth score. The Leiter–R is fully nonverbal; examiners use pantomime and nonverbal cues to explain the task, and participant responses are given by either pointing or with shapes or cards.

Analyses

Separate analyses were conducted to address the research questions relating to (a) overall performance, (b) performance on specific grammatical constructions, and (c) patterns of error types. Before the analyses of interest, participants were compared group-wise on nonverbal cognitive ability using Leiter–R growth scores. Growth scores were used instead of age-equivalent scores because the former provides a measure of the individual's skills that are assessed by the items on the Leiter–R rather than by comparing the participants' abilities to individuals in the same age group (Roid & Miller, 1997). Whereas standardized norm-referenced scales may not always provide a detailed look at the actual skills and growth of an individual, especially for individuals who are functioning lower than their same-age peers, growth scores reflect the absolute level of an individual's ability and may detect small differences in ability, which is particularly useful for individuals with neurodevelopmental disorders.

Conceptually, growth scores are similar to age-equivalent scores but without the latter's psychometric limitations. After initial exclusions based on hearing loss and sibling status, TD children (n = 12) whose nonverbal IQ scores were above 130 (2 *SD*s from the mean), as well as TD children (n = 3) and one adolescent with FXS whose total growth scores were high relative to the other participants (i.e., higher than 500), were excluded from the current study so that a group-wise match could be achieved. This resulted in samples of 52 participants with FXS, 29 participants with DS, and 36 TD participants. Based on these groups, participants with FXS and TD participants were well matched on Leiter–R growth scores $t(86) = 0.14$, $p = .890$, $d = 0.03$, whereas participants with DS differed from those with FXS and TD, $t(79) = 3.5$, $p = .001$, $d = 0.85$, and $t(63) = 3.2$, $p = .002$, $d = 0.82$, respectively. Participants with growth scores higher than 475 were also excluded (n = 31) in order to allow acceptable overlap in growth scores from the Leiter–R.

Thus, the analysis addressing the first research question included a final sample of 35 participants with FXS, 28 participants with DS, and 23 TD participants. Characteristics of the participants included in this analysis are shown in Table 2. *T* tests revealed that there were no significant differences in nonverbal cognitive ability (i.e., Leiter–R growth scores) between the participants with FXS and TD, $t(56) = -0.419$, $p = .677$, $d = 0.11$; TD and DS, $t(49) = 0.163$, $p = .871$, $d = 0.05$; or FXS and DS, $t(61) = .645$, $p = .522$, $d = 0.16$. There were five, nine, and 10 females in the FXS, TD, and DS groups, respectively.

As a result of the standardized administration of the TROG–2, in which testing is discontinued after five consecutive failed blocks, not all participants were administered the construction containing four lexical elements (Block F) or the construction containing a subject relative clause (Block G). Therefore, only participants who completed each of the blocks of interest (i.e., reversible *in* and *on,* reversible SVO, four elements, and relative clause in subject) were included in analyses of those specific grammatical constructions and their accompanying error patterns. Thus, analyses addressing the second and third research questions included 23 participants with FXS, 22 with DS, and 22 TD, as shown in Tables 3 and 4. The groups in these analyses were well matched on Leiter–R growth scores: FXS and DS, $t(43) = -0.14$, $p = .893$, $d = 0.04$; FXS and TD, $t(43) = -0.50$,

Table 2. Participant characteristics by group and gender.

	DS				FXS				TD			
	Females (n = 10)		Males (n = 18)		Females (n = 5)		Males (n = 30)		Females (n = 9)		Males (n = 14)	
Characteristic	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
Chronological age	12.7	1.4	12.9	1.9	11.0	1.0	12.8	1.8	4.7	0.5	4.6	1.1
Nonverbal IQ[a]	45.0	8.6	41.8	5.9	54.6	7.7	42.5	5.9	110.3	7.7	107.4	12.6
Leiter–R growth score	465.1	7.1	460.6	7.3	468.6	4.5	462.5	7.2	463.9	9.9	461.7	9.8
Leiter–R age-equivalent	5.3	0.7	4.7	0.7	5.6	0.5	4.9	0.7	5.1	0.4	4.8	1.0

Note. Leiter–R = Leiter International Performance Scale—Revised (Roid & Miller, 1997).
[a]One participant with DS completed only three out of four subtests of the Leiter–R Brief IQ subtests; therefore, scores from the three subtests were averaged to obtain estimated scores.

p = .620, d = 0.15; and DS and TD, $t(42) = -0.60$, p = .551, d = 0.03. As a result of the small number of females in each group, gender was not included in the analyses addressing these research questions.

Results

Overall Performance

The first research question was addressed using a 3 (group: FXS, DS, TD) × 2 (gender: male, female) analysis of variance (ANOVA), with number of items answered correctly on the TROG–2 as the dependent variable, to test for overall group differences and to establish whether the effect of gender differed across the groups. Partial eta squared (η_p^2), a measure of effect size, was calculated and was interpreted using the values of .01 (small effect), .06 (medium effect), and .14 (large effect; Cohen, 1988). Scores derived from performance on the TROG–2 are shown in Table 5. There was a significant main effect of group, $F(2, 80) = 17.7$, $p < .001$, $\eta_p^2 = .31$; a significant main effect of gender, $F(1, 80) = 7.6$, $p = .007$, $\eta_p^2 = .09$; and a significant Group × Gender interaction, $F(2, 80) = 3.4$, $p = .038$, $\eta_p^2 = .08$. TD participants scored higher than participants with FXS, $p < .001$, and higher than participants with DS, $p < .001$. In addition, participants with FXS scored higher than those with DS, $p = .019$.

Although females scored higher than males, the effect of gender was qualified by the interaction with group, which was examined using separate ANOVAs for each group. For participants with FXS, analyses yielded a significant effect of gender, $F(1, 33) = 16.5$, $p < .001$, $\eta_p^2 = .33$, but no significant effect of gender was found for TD participants, $F(1, 21) = .011$, $p = .917$, $\eta_p^2 < .01$, or for those with DS, $F(1, 26) = 3.7$, $p = .066$, $\eta_p^2 = .12$. To examine whether females accounted for the main effect of group, a separate univariate ANOVA with group was conducted only for males, with number correct as the dependent variable. Analyses yielded a significant effect of group, $F(2, 59) = 22.8$, $p < .001$, $\eta_p^2 = .44$. Male participants with TD scored significantly higher than both male participants with FXS, $p < .001$, and those with DS, $p < .001$; however, no significant difference was found between male participants with FXS and those with DS, $p = .648$.

Performance on Specific Constructions

To address the second research question, performance on the four grammatical constructions was analyzed using a 3 (group: FXS, DS, TD) × 4 (grammatical construction: reversible *in* and *on,* reversible SVO, four elements, and relative clause in subject) repeated measures ANOVA, with grammatical construction as the repeated measure and the number of items answered correctly as the dependent variable. Analyses yielded a significant main effect of group, $F(2, 64) = 15.4$, $p < .001$, $\eta_p^2 = .33$; a significant main effect of grammatical construction, $F(3, 192) = 11.0$, $p < .001$, $\eta_p^2 = .15$; and a significant Group × Grammatical Construction interaction, $F(6, 291) = 3.8$, $p < .001$, $\eta_p^2 = .11$ (see Figure 1). TD participants scored higher than participants with FXS

Table 3. Characteristics, by group, of participants who completed the four grammatical constructions of interest.

	DS (n = 22)		FXS (n = 23)		TD (n = 22)	
Characteristic	*M*	*SD*	*M*	*SD*	*M*	*SD*
Chronological age	13.0	1.7	12.8	1.8	4.7	0.9
Nonverbal IQ	43.4	7.5	43.3	6.9	108.6	11.5
Leiter–R growth score	464.4	5.9	464.1	5.7	463.1	7.6
Leiter–R age-equivalent	5.1	0.6	5.1	0.6	5.0	0.8

Table 4. TROG–2 scores, by group, of the participants who completed the four grammatical constructions of interest.

	DS (n = 22)		FXS (n = 23)		TD (n = 22)	
Characteristic	*M*	*SD*	*M*	*SD*	*M*	*SD*
Standard score	55.0	0.0	55.0	0.0	101.9	12.7
Age-equivalent	4.1	0.2	4.1	0.2	5.1	1.3
Total blocks passed	3.0	1.4	3.1	1.3	6.9	3.8
Total items passed	25.5	7.8	24.7	5.8	46.4	16.3

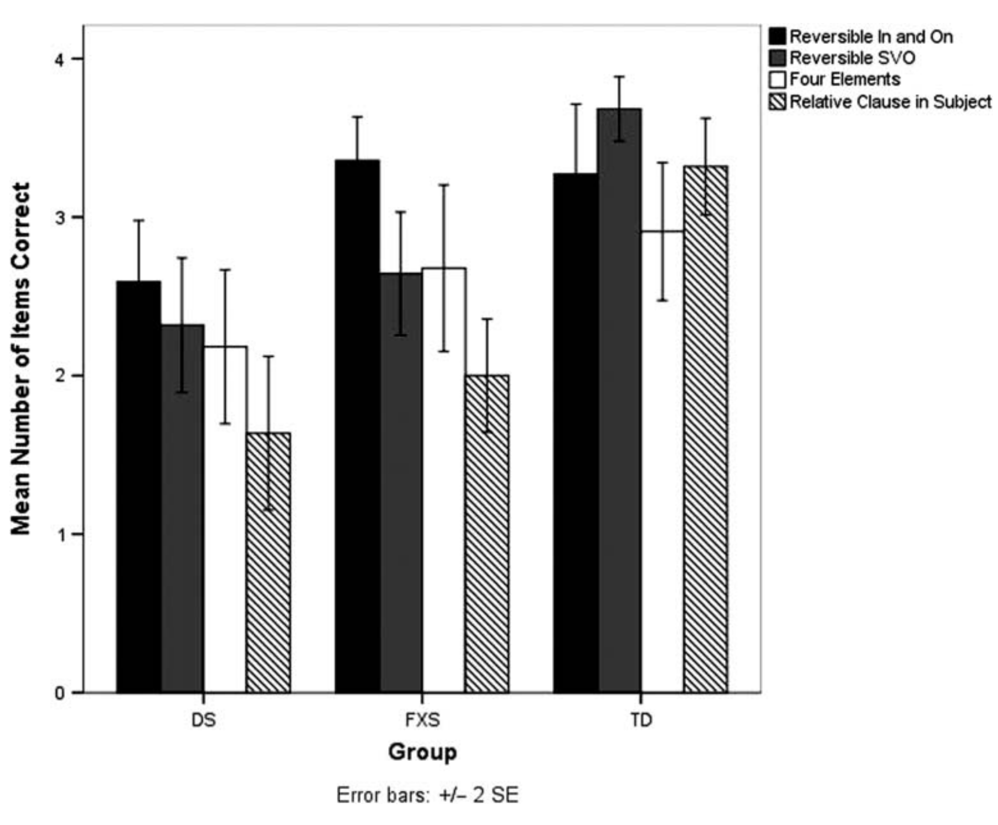
Table 5. TROG–2 scores, by group and gender, of participants who completed the four grammatical constructions of interest.

	DS				FXS				TD			
	Females (*n* = 10)		Males (*n* = 18)		Females (*n* = 5)		Males (*n* = 30)		Females (*n* = 9)		Males (*n* = 14)	
Variable	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
Standard score	55.0	0.0	55.0	0.0	59.8	6.7	55.0	0.0	100.8	12.8	100.9	14.2
Age-equivalent	4.1	0.2	4.0	0.2	4.7	0.9	4.1	0.2	4.9	0.9	5.2	1.5
Total blocks passed	3.1	1.6	2.2	1.6	5.6	3.6	2.6	1.7	6.4	3.3	6.6	4.5
Total items passed	26.8	9.5	20.1	8.6	41.6	19.4	21.7	8.1	44.3	14.9	45.1	19.7

and those with DS, *p*s < .001. In addition, participants with FXS scored higher than participants with DS, $p = .045$. Analyzing the effect of grammatical construction, sentences with a subject relative clause were significantly more challenging than all other constructions, *p*s < .001, except four-element sentences, $p = .157$. On average, reversible *in* and *on* sentences resulted in significantly more correct answers than four-element sentences, $p = .001$. Reversible SVO sentences were significantly easier than four-element sentences, $p = .043$. Reversible SVO sentences were somewhat more difficult than reversible *in* and *on* sentences, but this difference was not significant, $p = .161$.

To further investigate the interaction between group and grammatical construction, we conducted tests of simple effects for each grammatical construction. For reversible *in* and *on* sentences, there was a main effect of group, $F(2, 64) = 4.1$, $p = .02$, $\eta_p^2 = .12$. Participants with DS passed significantly fewer items in the reversible *in* and *on* sentences than TD participants and those with FXS, *p*s = .015, whereas participants with TD and FXS did not perform significantly differently from one another on average, $p = .97$. For reversible SVO sentences, there was also a main effect of group, $F(2, 64) = 15.5$, $p < .001$, $\eta_p^2 = .33$. TD participants passed significantly more items in reversible SVO sentences than either participants with FXS or participants with DS, *p*s < .001. Performance on reversible SVO sentences for individuals with DS and those with FXS did not differ, $p = .440$. For four-element sentences, the analysis yielded no main effect of

Figure 1. Items passed for the four grammatical constructions of interest.



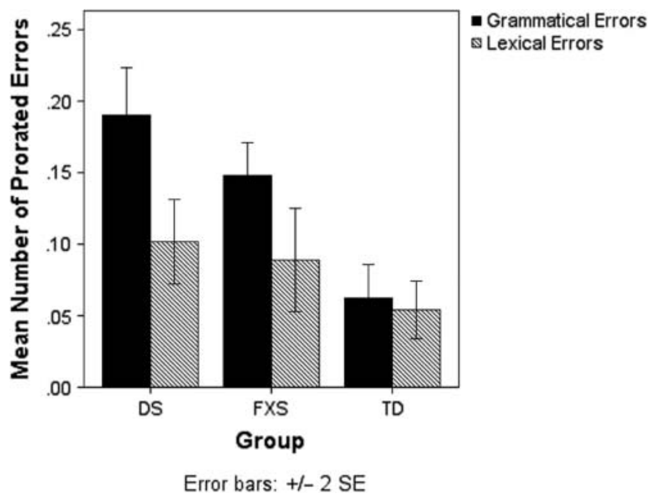
group, $F(2, 64) = 2.1$, $p = .134$, $\eta_p^2 = .06$. Analysis of subject relative clause sentences revealed a main effect of group, $F(2, 64) = 20.6$, $p < .001$, $\eta_p^2 = .39$. TD participants correctly answered more items in sentences with subject relative clauses than did participants with FXS or DS, *p*s < .001, whereas participants with FXS and those with DS did not perform significantly differently from one another, $p = .162$.

Patterns of Error Type

To address the third research question, we examined patterns of errors by comparing the number of lexical and grammatical errors made for the items testing the four constructions of interest (i.e., reversible *in* and *on* sentences, reversible SVO sentences, sentences with four lexical elements, and sentences with subject relative clauses) for participants who completed those blocks. Again, because of the small number of females in each group, gender was not included in the analysis. Across the four grammatical constructions, it was possible to make 27 grammatical errors and 21 lexical errors. To account for this difference and allow for comparison across the two types of errors, the number of errors was prorated such that the number of errors of each type was divided by the total possible number of errors. The prorated sum of lexical errors and the prorated sum of grammatical errors committed in the context of the four constructions, shown in Table 6, were the dependent variables for each participant. Errors were then analyzed using a 3 (group: FXS, DS, TD) × 2 (error type: lexical, grammatical) repeated measures ANOVA, with error type as the repeated measure. A main effect of group, $F(2, 64) = 13.7$, $p < .001$, $\eta_p^2 = .30$; a main effect of error type, $F(1, 64) = 37.7$, $p < .001$, $\eta_p^2 = .37$; and a significant Group × Error Type interaction, $F(2, 64) = 7.6$, $p < .001$, $\eta_p^2 = .19$, emerged (see Figure 2). On average, participants made more grammatical errors than lexical errors. As would be expected based on their overall performance, TD participants made fewer errors than those with FXS or DS, $p = .001$ and $p < .001$, respectively. There was no significant difference between the number of errors made by participants with FXS and those with DS ($p = .056$).

To investigate the significant Group × Error Type interaction, we conducted separate 2 (group) × 2 (error type) ANOVAs. Analyses revealed significant Group × Error Type interactions for all pairs of groups. In contrast to TD participants, who made very few errors of the grammatical or lexical type, participants with FXS not only made more errors overall but also showed increased grammatical errors relative to lexical errors, $F(1, 43) = 27.4$, $p < .001$, $\eta_p^2 = .39$. Participants with FXS and those with DS did not perform significantly differently in terms of number of errors overall; however, relative to participants with FXS, participants with DS made a significantly greater number of grammatical errors, $F(1, 43) = 4.4$, $p = .041$, $\eta_p^2 = .09$.

Figure 2. Mean number of prorated grammatical and lexical errors committed for the four grammatical constructions of interest.



In an exploratory descriptive analysis, the mean raw frequency of specific types of grammatical and lexical errors was investigated. Of course, not all error types were possible for each construction; thus, only verb and adjective lexical errors and word order and function word grammatical errors could be examined. No statistical tests were performed because of the uneven opportunities for error types among constructions. On average, participants with FXS and those with DS made more grammatical errors of the word order type than TD participants in reversible SVO sentences (1.22, 1.59, 0.32, respectively) and sentences with subject relative clauses (2.04, 2.14, 0.64, respectively). On reversible *in* and *on* sentences, participants with DS made a greater number of grammatical errors of the function word type than TD

Table 6. Mean number of grammatical and lexical errors by group and grammatical construction.

	DS (*n* = 22)		FXS (*n* = 23)		TD (*n* = 22)	
Grammatical construction	**Grammatical errors**	**Lexical errors**	**Grammatical errors**	**Lexical errors**	**Grammatical errors**	**Lexical errors**
Reversible *in* and *on*	1.36	—	0.74	—	0.73	—
Reversible SVO	1.59	0.09	1.22	0.26	0.32	0.00
Four elements	—	1.82	—	1.57	—	1.09
Relative clause in subject	2.14	0.23	2.04	0.04	0.64	0.05

Note. A dash indicates that errors of that type were not possible for items testing that grammatical construction.

and FXS participants (0.55, 0.13, 0.09, respectively). For four-element sentences, only lexical errors were possible, and the pattern of errors across participant groups was in line with their overall performance (i.e., 1.82 lexical errors by those with DS compared to 1.57 by participants with FXS and 1.09 by TD participants).

Discussion

The purpose of the present study was to examine the comprehension of specific syntactic constructions and types of errors made by males and females with FXS to gain a more nuanced understanding of the linguistic phenotype of this population. In addition, greater knowledge of the syntactic comprehension skills of individuals with FXS can aid in the development of appropriate interventions.

Overall Performance for Syntactic Comprehension

The first research question focused on overall levels of syntactic comprehension in terms of differences as a function of diagnostic group and gender. TD children achieved higher total TROG–2 scores for items passed than did young adolescents with FXS. This result conflicts with findings from Abbeduto et al. (2003) for older adolescents and young adults with FXS, who did not differ significantly from NVMA-matched TD children on the TACL–R. Instead, results of the current study are in line with those from Price et al. (2007), which included a younger group of males with FXS and DS. The current finding, that young adolescents with FXS achieved overall scores that were lower than the scores of the younger NVMA-matched TD children, suggests that the overall syntactic comprehension skills of young adolescent males and females with FXS is lower than expected based on nonverbal cognitive ability. It may be that in late adolescence or even young adulthood, individuals with FXS catch up in terms of receptive language skills; however, longitudinal research is needed to investigate this further.

The young adolescents with FXS achieved overall scores that were higher than the scores of the young adolescents with DS, suggesting that the syntactic comprehension skills of young adolescents with FXS are not as severely affected as they are in young adolescents with DS (Abbeduto et al., 2003; Price et al., 2007). However, results from the Group × Gender interactions for each diagnostic group revealed a gender difference only for the FXS group, with females outperforming males. This finding suggests that females with FXS may be accounting for the higher performance of participants with FXS relative to DS. When females were excluded from the analysis, male adolescents with FXS and DS did not perform significantly differently from one another. Thus, males with FXS appear to have receptive syntax deficits as severe as those seen in individuals with DS, which is a condition long recognized for having especially serious syntactic impairments. Clearly, syntax must be a target of intervention for males with FXS.

Despite the superior performance of females relative to males, it is noteworthy that the average TROG–2 standard score obtained by females with FXS was almost 3 *SD*s below average, which is consistent with the claim that language poses serious challenges for many females with FXS (Sterling & Abbeduto, 2012). We chose to include females with FXS because they are a relatively understudied group compared to males with FXS; however, these findings suggest that because of females' distinct levels of delay, future studies may do well to examine the performance of males and females with FXS separately in order to ensure an accurate picture of the FXS phenotype in relation to other neurodevelopmental disorders. In the following discussions of the second and third research questions, in which gender was not a factor, findings were interpreted for males and females together because diagnostic groups were matched on NVMA and therefore had quantitatively similar profiles.

Performance on Specific Grammatical Constructions

The second research question focused on performance differences on specific grammatical constructions as a function of diagnostic group. The constructions examined were reversible sentences with the prepositions *in* and *on* (e.g., *The duck is on the ball*), reversible SVO sentences (e.g., *The man is chasing the dog*), sentences containing four lexical elements (e.g., *There is a yellow star and a big flower*), and sentences with subject relative clauses (e.g., *The man that is eating looks at the cat*). These constructions were chosen based in part on the fact that they had been administered to most of the participants in the longitudinal study from which the data were drawn, ensuring an adequate sample size. At the same time, however, these constructions were interesting because they tested dimensions of receptive syntax (i.e., reversibility and length) that are likely to be especially sensitive to the sequential processing and auditory memory deficits of FXS.

On average, across all participants, comprehension of active declarative sentences with four lexical elements was more difficult than comprehension of reversible sentences, including the locative *in* or *on* and reversible SVO sentences. Although comprehension of these two reversible constructions depends on processing information about word order, the syntax in these constructions is relatively simple, and the sentences are rather short, thereby placing little demand on auditory memory. This difference in performance may have been due to the fact that comprehension of four elements within a sentence places heavy demands on auditory memory independent of syntactic complexity. Interestingly, the four-element construction was tested only with lexical distracters, reinforcing the interpretation that poor performance cannot be attributed to difficulty with the syntax of the construction (Bishop, 2003). Items testing sentences that contained a subject relative clause also were more difficult than the two reversible constructions for all groups. The former sentences containing relative clauses also place heavy demands on auditory memory. Future research should focus on a wider range of constructions and experimentally manipulate auditory memory load to verify the source of difficulty for individuals with FXS regarding the four-element and subject relative sentences.

Adolescents with FXS presented a mixed pattern with regard to comprehension of reversible sentences. That is, adolescents with FXS were as successful on reversible *in* and *on* sentences as were the TD children; however, they were less successful than the TD children on reversible SVO sentences. This difference may be explained by the way the words in these two sentence types are encoded. In reversible *in* and *on* sentences, the relations among the words are encoded by lexical items, whereas in reversible SVO sentences, the relations among the words are encoded by abstract syntactic items. Thus, these data provide evidence that youth with FXS have difficulty with the processing of syntactically encoded information, and this difficulty extends beyond cognitive-level expectations.

Similar to their performance on reversible SVO sentences, young adolescents with FXS had lower mean scores than TD children for forms containing a relative clause in the subject; however, the mean scores of young adolescents with FXS were not different from those of young adolescents with DS. Because this construction is typically mastered later in development, it may be the case that neither adolescents with FXS nor adolescents with DS have fully grasped the syntax of sentences containing an embedded clause. Additionally, it is also possible that adolescents with FXS and adolescents with DS display a "linguistic vulnerability" (Schuele & Nicholls, 2000, p. 581) in terms of acquiring complex syntax.

Taken together, these results suggest that aspects of language comprehension are impaired beyond nonverbal cognitive ability expectations for male and female adolescents with FXS and those with DS. For adolescents with FXS, syntactic comprehension deficits may be related to specific grammatical constructions, particularly those that have high demands for auditory memory or for syntactic processing that does not depend solely on lexical knowledge (i.e., lexical bootstrapping). Experience with, and knowledge of, particular vocabulary might support syntactic comprehension for adolescents with FXS, as evidenced by comprehension of locatives and four-element sentences that apparently keeps pace with nonverbal cognitive development. In contrast, comprehension that relies less on the recognition of lexical units and more on syntactic information per se appears to create a challenge for young adolescents with FXS. For adolescents with DS, lexical comprehension might not be developed enough to serve as a support for syntactic aspects of comprehension, resulting in a more generalized pattern of deficits in language comprehension.

Patterns of Error Types

The third research question focused on differences in the pattern of lexical and grammatical errors across diagnostic groups. To address this question, we examined the frequency of errors that were committed in the context of reversible *in* and *on* sentences, reversible SVO sentences, sentences containing four lexical elements, and sentences with subject relative clauses. Across the four grammatical constructions, young adolescents with FXS and those with DS made a greater number of grammatical errors than lexical errors relative to TD children. This finding is consistent with previous research on adolescents with DS (Laws & Bishop, 2003) and individuals with intellectual disabilities (Abbeduto, Furman, & Davies, 1989), suggesting that poor performance on the TROG–2 was related to difficulty with grammar and not necessarily vocabulary. The current findings suggest that for both young adolescents with FXS and young adolescents with DS, grammatical difficulties may be the main factor driving the lower performance on these constructions.

In addition to the broad categories of errors, specific types of grammatical and lexical errors were examined in the four grammatical constructions of interest. For sentences testing the locatives *in* and *on,* each group made more grammatical errors of the word order type than of the function word type; however, young adolescents with DS made a greater number of grammatical errors of the function word type than both the FXS and TD groups. This finding suggests that there may be differences in the profile of syntactic comprehension deficits of young adolescents with FXS or DS. This could be essential information for language interventionists who are working on receptive grammar with young adolescents with FXS.

For sentences testing reversible SVO and relative clauses, participants made more grammatical errors than lexical errors. Although participants with FXS and those with DS did not greatly differ from each other in their accuracy for reversible active sentences, they made a greater number of grammatical errors of word order type than did TD children. This finding suggests that poor performance on such constructions may be due to a difficulty comprehending the word order of reversible and embedded syntactic forms.

Clinical Implications

The results of the current study have implications for interventions for young adolescent males and females with FXS. Findings regarding overall syntactic comprehension abilities reveal the importance of improving receptive syntax of these individuals. Their poor processing of sequential patterns and weakness in auditory memory appear to be major sources of comprehension problems and may make certain grammatical constructions more difficult than others. Thus, interventions for young adolescents with FXS may need to target grammatical constructions that are high in syntactic processing demands and constructions that rely less on lexical knowledge. It would also be useful to try to improve their general sequential processing skills and auditory memory or to circumvent those areas of weakness when imparting new syntactic knowledge.

Limitations and Future Research

There are several limitations of the current study. First, the current study included a small number of females with FXS. Future studies including a larger sample of females with FXS are needed. Second, the current study did not employ a measure of auditory memory or of sequential processing. Future studies should use such measures to determine the extent to which the mere processing load of certain grammatical constructions or general sequential processing deficits account for the receptive language impairments found in these individuals. Third, the classification of foils for the error type analysis was limited to types of errors that could be consistently characterized across TROG–2 items and constructions. Our understanding of the FXS phenotype would benefit from research that addresses other types of comprehension errors.

Conclusion

The current study revealed important findings regarding the receptive syntactic difficulties of young adolescents with FXS and highlights the need for examining language in finer detail than is often done for individuals with neurodevelopmental disorders. The profile of language comprehension identified for adolescents with FXS suggests that research examining the relationships among lexical and syntactic ability in relation to the broader profile of cognitive impairments over the course of development for youth with genetic sources of intellectual disability will be informative.

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