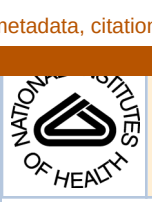



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# Expressive morphosyntax in boys with Fragile X syndrome with and without autism spectrum disorder

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## Abstract

**Background—**Fragile X syndrome (FXS) is the most common inherited cause of intellectual disability, and the most common single gene disorder associated with autism. Language impairments in this disorder are well documented, but the nature and extent of syntactic impairments are still unclear.

**Aims—**To compare the performance of boys with FXS with and without autism spectrum disorder on measures of verb (VM) and noun (NM) morphosyntax with that of typically developing boys of similar non-verbal mental ages.

**Methods & Procedures—**Conversational samples were obtained from 33 boys with FXS with autism spectrum disorder (FXS-ASD), 35 boys with FXS and no ASD (FXS-O), and 46 typically developing boys (TD). Production of verbal and nominal morphosyntax was assessed separately in these two subdomains. A hierarchical linear model compared morphosyntactic scores in all groups after adjusting for non-verbal cognition, articulatory skill, and caregiver education. The model also tested interactions between group and morphosyntactic subdomain.

**Outcomes & Results—**Boys with FXS in both groups scored lower than the TD boys on both measures. The FXS-O and the FXS-ASD groups did not differ on either composite measure. All covariates were significantly related to morphosyntactic scores.

**Conclusions & Implications—**Part of the morphosyntactic impairment in FXS may be attributable to cognitive, environmental, and speech factors. However, it is clear that boys with FXS perform at levels lower than expected from differences in these extra-linguistic factors alone, across both the verb and the noun domains. Clinical interventions should therefore seek to address specific syntactic targets.

### Keywords

Fragile X syndrome; morphosyntax; autism spectrum disorder; X-linked; language disorders; conversation

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## Introduction

Current research on language in developmental disorders is in critical need of detailed language phenotypes to inform studies relating genes, environment, and disorders (McCardle *et al.* 2005). It is particularly important to identify specific subtypes within given disorders to define narrow linguistic phenotypes that facilitate theorizing about the origins of impairment and have the potential to suggest specific intervention targets. Two of the main challenges to adopting an aetiology-specific approach to intervention are the difficulty of defining language phenotypes precisely, and the lack of understanding of individual variation in the development and outcome of these phenotypes (Fidler *et al.* 2007). Within linguistic phenotypes, syntax is a component thought to be largely genetically determined (although this position is highly controversial) and therefore shows promise in light of genotype–phenotype links. Moreover, syntactic difficulties are quite common in developmental populations and have been described in extensive detail for several disorders. In recent years, many studies have pointed out syntactic deficits in specific language impairment (SLI) (Bedore and Leonard 1998, Eadie *et al.* 2002), Down's syndrome (Abbeduto and Chapman 2005, Eadie *et al.* 2002), and high-functioning autism (HFA) (Landa and Goldberg 2005, Roberts *et al.* 2004). Syntax is important, lastly, because improvements in this area translate into improvements in general communication abilities and into better perceptions of individuals with disabilities (Hewitt *et al.* 2005).

Fragile X syndrome (FXS) is a vital neurodevelopmental disorder to study within this conceptual framework. It is the most common inherited cause of intellectual disability, with a high prevalence of one of every 4000 male births and one of every 8000 female births (Sherman 2002). It is second only to Down's syndrome as a genetic cause of intellectual disability. The genetics and molecular biology of FXS are very well known, but their impact on language development is still elusive. Although syntactic problems are generally not considered a hallmark of the language phenotype of FXS, many of the earlier studies on which this conclusion is based (reviewed below) were conducted on small samples, using general measures of expressive language. More recent studies of boys with FXS suggest that expressive syntax is specifically impaired in relation to non-verbal cognitive functioning (Price *et al.* 2008, Roberts *et al.* 2007a). Compounding this lack of consensus in the literature (and partly as a result of it), to date there are no hypotheses about the nature of morphosyntactic impairment in FXS. Such hypotheses are nonetheless crucial to identify specific targets of intervention and to identify which aspects of impairment are related to cognitive deficits and which are possibly syntax specific.

In this study, we seek to refine our understanding of the morphosyntactic phenotype in FXS. We compare the amount of grammatical morphemes produced in conversation by boys with FXS with that of typically developing (TD) boys of similar non-verbal mental age (MA). We assess the contribution of non-verbal cognition, articulatory skill, and primary caregiver education to morphosyntactic production. After controlling for differences due to these predictors, we ask whether morphosyntactic skills are at different levels in different subdomains. Finally, given the high comorbidity of FXS and autism spectrum disorder (ASD), we examine the possibility that these two subpopulations present different linguistic subtypes.

### Phenotype of Fragile X syndrome

FXS is a developmental disorder linked to a mutation on the Fragile X Mental Retardation 1 (FMR1) gene (see above for prevalence). In boys, it is associated generally with mild to severe intellectual disability, and often with hyperactivity and hyper-arousal, attention deficits, social anxiety, and autistic characteristics such as decreased eye contact and repetitive behaviours. Females are less severely affected by the mutation because of the

presence of a second, unaffected copy of the FMR-1 gene on the additional X chromosome. For this reason, we only included boys with FXS in this study, with the TD group excluding girls to preserve gender-matching. Perseveration, tangential language, and poor intelligibility are characteristic of the speech and communication of males with FXS (Abbeduto and Chapman 2005, Levy *et al.* 2006, Sudhalter *et al.* 1991). Articulatory skill in particular can have an effect on morphosyntactic assessment, since many grammatical morphemes are consonantal and/or of short duration. Children with lower phonological accuracy may not be able to produce the required grammatical morphemes in many contexts in connected speech.

**FXS and autism—**Twenty-five per cent of school aged boys with FXS also have autism (Bailey *et al.* 1998). More characteristics of autism in FXS are associated with lower IQs and more severe language and social deficits (Bailey *et al.* 1998, 2000). Lewis *et al.* (2006) argued that comorbid FXS and autism represents a distinct subtype of FXS, with receptive language and theory of mind significantly more impaired in the former. The same study, however, found no differences on expressive language measures between individuals with FXS with autism and those with only FXS. On the other hand, some children with autism without FXS have clear expressive syntax delays. Roberts *et al.* (2004) found that children with autism with low receptive vocabulary scores also had difficulties with tense-marking (past tense *-ed* and third-person singular *-s*). Additionally, Landa and Goldberg (2005) found that children with HFA had lower expressive syntax skills than TD controls matched for chronological age, IQ, gender, and socio-economic status. In order to discern the possible contribution of comorbid autism to expressive morphosyntax challenges in FXS, this analysis includes a separate group of boys with FXS and ASD.

**Morphosyntax in Fragile X syndrome—**The research on the morphosyntactic skills of individuals with FXS is still very limited. Paul *et al.* (1984) measured the mean length of utterance in morphemes (MLU) and the Developmental Sentence Scores (DSS; Lee 1974) of three young boys aged 10;0, 10;6, and 13;9. They reported overall delays in syntax relative to non-verbal MA and receptive language, with verb marking, sentence embedding, and conjoining particularly affected. Levy *et al.* (2006) found that Hebrew-speaking pre-pubertal boys with FXS without autism produced fewer complex clauses in conversation, but also fewer errors on grammatical agreement and past tense marking than TD boys matched on MLU. Sudhalter *et al.* (1991), on the contrary, argued against syntactic impairments beyond cognitive level. They studied the expressive syntax in play-based conversation of 19 males with FXS (no autism) between 5 and 36 years and Vineland Communication Age equivalents from 3 to 9. They found that the relationship between Index of Productive Syntax (IPSyn; Scarborough 1990) scores and MLU was curvilinear (with greater gains in IPSyn scores at lower MLUs) and similar to that observed in TD preschoolers. The authors interpreted this as suggesting that delays in morphosyntactic development were partly attributable to the participants' cognitive level. Two recent studies take issue with this conclusion. Roberts *et al.* (2007a) compared the syntax and vocabulary skills of 35 boys with FXS (but no autism) with 27 TD boys at similar MA levels. They found that when the effects of non-verbal cognition level, maternal education level, and speech intelligibility were removed, boys with FXS scored lower than TD boys on MLU and most IPSyn scales.

A follow-up study by Price *et al.* (2008) compared the MLU and IPSyn scores (total and four subscales) of 35 boys with FXS and no ASD, 36 boys with FXS and ASD, 31 boys with Down's syndrome, and 46 TD boys. After controlling for non-verbal MA and years of maternal education, they found that both FXS groups scored lower than the TD group but higher than the group with Down's syndrome. On the Noun Phrases, Verb Phrases, and Sentence Structure IPSyn subscales, both FXS groups scored lower than the TD group and did not differ from the group with Down's syndrome. No differences were found between

the two FXS groups. Note that both Roberts *et al.* (2007a), and Price *et al.* (2008) examined syntax primarily via a type-frequency measure like the IPSyn. In this study, we will assess syntax via token-frequency measures, to supplement the information obtained in earlier studies and attempt to clarify the extent and nature of the syntactic impairment.

### Study goals

To summarize, recent research strongly suggests that expressive syntax is globally impaired beyond nonverbal cognition in FXS. However, different measures, sampling methods, and small sample sizes in many studies, coupled with a relative scarcity of relevant literature, prevent a broader consensus on the syntactic impairments in FXS to date. Moreover, theories addressing the causes of impairment in FXS are lacking. One comparison that has proven fruitful in disorders like SLI and Down's syndrome is that between verb and noun morphosyntax, although sometimes (especially for clinical purposes in SLI) the difference is conceptualized as a comparison between tense-expressing and non-tense-expressing 'control' morphemes. We believe that this comparison is useful as well in FXS, because it provides a finer degree of granularity of analysis than previous studies. Furthermore, verbal and nominal morphosyntax are two coherent syntactic subdomains, possibly underlain by different learning mechanisms and different relationships to general cognition.

The objective is therefore to provide token frequency measures in conversation of these two distinct domains in FXS. The more detailed and finegrained our language phenotype descriptions, the better prepared we are to investigate specific genes and gene/environment interactions that yield given linguistic phenotypes (critical for studying the genetic basis of language disorders in atypical populations), as well as to inform assessment and intervention. Furthermore, in light of previous research that indicates syntactic deficits in children with ASD (Roberts *et al.* 2004), exploration of the impact of comorbid ASD and FXS is warranted. Two questions guided our analysis:

- Do boys with FXS with and without ASD score lower than TD boys on both morphosyntactic composite measures, or is the impairment confined to one of the two domains?
- Do boys with FXS and ASD score lower on one or both morphosyntactic measures than the boys with FXS only, indicating that comorbidity with ASD significantly impacts syntax?

The present analysis expands on the work of Roberts *et al.* (2007a) and Price *et al.* (2008) by analysing the production of morphosyntactic forms via composites based on token-frequency measures rather than via a type-frequency measure such as the IPSyn or a general, non-specific measure of syntactic complexity such as MLU. Token counts provide a direct measure of the quantity of morphosyntactic production. In addition, it expands on Roberts *et al.* (2007a) by examining separately the language of boys with FXS and ASD and that of boys with FXS but no ASD. Finally, unlike in previous studies, we controlled for speech differences in articulatory skill, as well as for any pre-existing cognitive (MA) and environmental (caregiver education) differences.

## Materials and methods

### Participants

All the boys who participated in this study were recruited for a larger ongoing longitudinal project (Roberts *et al.* 2007b). Boys with FXS were eligible if they were 16 years or younger, and had an MLU greater than 1.1 and an expressive vocabulary of at least 40 words. The TD boys were between 2 and 6 years of age and showed a distribution of developmental ages for non-verbal cognitive abilities on the Brief IQ composite of the Leiter

International Performance Scale—Revised (Leiter-R; Roid and Miller 1997) that was similar to those of the other two groups ($F(2, 110) = 1.55$, $p = 0.22$).

All participants used spoken English as their primary mode of communication and resided in homes where English was the primary language spoken. Hearing threshold screening was conducted using an audiometer at 500, 1000, 2000, and 4000Hz, and all boys' thresholds were below 30 dB in the better ear. The Behavioral Institutional Review Board at the University of North Carolina at Chapel Hill reviewed and approved study protocols annually. Informed consent was given by the parent or guardian at study entry.

Table 1 displays summary measures for background characteristics and covariates for boys in each of the four diagnostic groups.

**Boys with Fragile X syndrome—**All participants with FXS (FXS-O and FXS-ASD) had been diagnosed with the full mutation of the disorder by DNA analysis. They were recruited from ongoing longitudinal studies of children with FXS and had been referred from paediatric offices, genetic clinics, or developmental clinics in the south-eastern United States. A total of 68 boys with FXS participated in the study. The *Autism Diagnostic Observation Schedule* (ADOS; Lord *et al.* 2001), a standardized observation of communicative and social behaviour in children, was used to determine autism status in the boys with FXS. It is composed of a 45-minute series of structured and semi-structured interactions with an examiner that provide contexts to elicit behaviours characteristic of autism. Scores were determined by trained examiners through videotapes of the ADOS sessions. We administered three modules of the Autism Diagnostic Observation Schedule based on each child's developmental and language levels (Lord *et al.* 2001): Module 1 for children who were using single words to simple phrases (eleven participants, zero TD, five FXS-O, and six FXS-ASD); Module 2, for children who ranged from using three-word phrases to fluency (77 participants, 34 TD, 22 FXS-O, and 21 FXS-ASD); and Module 3, for children and young adolescents who were verbally fluent (26 participants, twelve TD, eight FXS-O, and six FXS-ASD). Each child received a categorical score of ‘no autism,’ ‘spectrum,’ or ‘autism’, with the latter two scores combined to form a single group (FXS-ASD).[1] Reliability was calculated on 16% of the interactions, equalling 0.89 for the individual items (range = 0.83–0.96) and 0.93 for diagnosis (range = 0.81–1.00).

**Boys with Fragile X syndrome only (FXS-O)—**Thirty-five boys with FXS had no ASD. Their chronological ages ranged from 2;10 to 14;4 (mean = 9;11), and their non-verbal MA ranged from 2;2 to 6;8 (mean = 5;0). Eighty-six per cent of the boys were Caucasian, 11% were African-American, and 3% were of other ethnic backgrounds. Maternal[2] education levels ranged from 12 to 20 years (mean = 14;2).

**Boys with Fragile X syndrome with autism spectrum disorder (FXS-ASD)—**Thirty-three boys with FXS also had ASD. Their chronological ages ranged from 3;6 to 13;11 (mean = 8;7), and their non-verbal MA ranged from 2;5 to 5;11 (mean = 4;8). Ninety-one per cent of these participants were Caucasian, 6% were African-American, and 3% were of other ethnic backgrounds. Maternal education levels ranged from 12 to 20 years (mean = 15;2).

[1]To be classified in the FXS-ASD group, each child had to meet the autism spectrum cut-off score for each subtotal: Communication and Social, and the autism spectrum cut-off for the Communication plus Social total. For Module 1, communication cut-off is 2 or higher, social interaction is 4 or higher, and total is 7 or higher. For Module 2, the respective cut-offs are 3, 4, and 8. For Module 3, they are 2, 4, and 7.

[2]All primary caregivers were mothers in the sample. See below.

**Typically developing boys (TD)—**Forty-six TD boys participated in the study, with a mean MA equivalent on the Leiter-R Brief IQ of 4;9 and a range of 2;10–7;1. Their chronological ages ranged from 2;1 to 6;7 (mean = 4;6). Maternal education ranged from 12 to 20 years (mean = 16;5). Seventy-two per cent of the boys were Caucasian, 15% were African-American, and 13% were of other ethnic backgrounds. TD boys were recruited from childcare centres, paediatric offices, and schools in North Carolina. We excluded any boys reported to have a history of developmental disability, ASD, speech or language difficulties, or receiving speech and language therapy. Standardized speech and language tests were given as part of the larger assessment, and any TD boy was excluded from the study if he scored more than 1.5 standard deviations below the mean on any of these tests. Any TD boy who received a score of 'autism' or 'spectrum' on the ADOS was excluded.

## Procedures

Participants were tested at the FPG Child Development Institute, at their school, or in their home, depending on parental preference. All sessions were videotaped with a Sony Digital8 video camera (DCR-TVR27) and audiotaped with a portable Digital Auditory Tape TASCAM (TD-P1) recorder and a Shure WBH headset microphone system.

**Language samples—**Trained research assistants transcribed 100 usable spontaneous child utterances from videotapes of ADOS sessions using CLAN software and CHAT conventions (MacWhinney 2000).[3] We examined the activities from the ADOS that elicited more natural conversation: make-believe play, demonstration task, picture description, blocks, and wordless book reading.[4] Partially or fully unintelligible utterances, exact self-repetitions (possibly more common in FXS due to perseveration), parts of routines (for example, reciting the alphabet), exact imitations of examiner utterances, and yes/no responses to questions were excluded from analysis.

**Transcription reliability—**A second researcher with extensive transcription training listened to the audiotapes to verify and correct all original transcripts. A third researcher then independently verified and corrected via audiotape a randomly selected 12% ($n$ = 14) of the original transcripts for reliability. Morpheme-to-morpheme agreement was then calculated for the verified transcripts of the second and third researchers. Overall agreement was 91%. Agreement was 85% for the transcripts of boys with FXS-O ($n$ = 4, 11% of FXS-O group); 85% for boys with FXS-ASD ($n$ = 5, 15% of FXS-ASD group); and 95% for TD boys ($n$ = 5, 11% of TD group) (Price *et al.* 2008).

## Covariates

**Non-verbal cognition—**We used the Brief IQ composite of the *Leiter International Performance Scale—Revised* (Leiter-R; Roid and Miller 1997) to measure non-verbal cognition. We administered four subtests: Figure Ground (identifying figures or designs within a complex stimulus), Form Completion (recognizing an object from its fragmented parts), Sequential Order (selection of the next item in a logical sequence of items), and Repeated Patterns (supplying the missing portion of a repeated pattern). The Leiter-R is standardized on 1719 individuals, aged 2–20 years, and has a test–retest coefficient of 0.96 for the Brief IQ composite and alpha reliability coefficients of 0.75–0.88 for the four

[3]All children included had at least 100 utterances. The number of utterances analysed was capped at 100 because (1) equal sample lengths minimize the likelihood of finding spurious differences for token measures of language, and (2) many children did not have many more usable utterances. We believe this is not problematic since it is customary to do so for other measures of syntax.

[4]For Module 1, most utterances were produced during free play, a pretend birthday party, a snack break, and interactions using bubbles and a balloon. For Modules 2 or 3, utterances came from make-believe play with toys, a joint interactive play, a book activity, a pretend birthday party, a snack, and bubbles and/or balloon interactions.

subtests. It has adequate validity, with correlations with other frequently used IQ tests from 0.85 to 0.86. We computed age equivalents and Brief IQ scores for each child using published norms.

**Articulatory skill—**We measured skill in production of consonant sounds using the Goldman–Fristoe Test of Articulation— Second Edition (GFTA-2; Goldman and Fristoe 2000) Sounds-in-Words subtest. The GFTA-2 is designed to elicit single-word responses to stimulus pictures and measures the ability to produce all of the English consonants in the initial, medial, and final positions in common words. Very high levels of reliability, adequate content validity, and adequate construct validity are reported in the manual. The boys' speech was transcribed by two trained speech–language pathologists using narrow transcription guidelines. The percentage agreement between the two transcribers for broad transcription was 92.4% for Fragile X syndrome (range = 78.4–98.8%), and 92.5% for typically developing boys. For narrow transcription, the average percentage agreement between two transcribers was 87.4% for FXS (range = 75.1– 95.2%), and 88.7% for typically developing boys.

**Primary caregiver education—**We measured this as total years of education completed by participants' primary caregivers at the time of enrolment. In the sample, all primary caregivers were mothers, so we will refer to this variable as ‘maternal education’ in the remainder of the paper. This measure was a proxy for the effect of environmental and family factors on verbal and non-verbal skills, since studies have shown that higher levels of maternal education correlate with more advanced speech and language development in TD children and children with developmental disabilities (Campbell *et al.* 2003, Dollaghan *et al.* 1999).

## Measures of morphosyntax

The measures of morphosyntactic skill are counts of produced morphemes grouped in two subdomains: verb and noun morphosyntax. This comparison is widely used for investigating syntactic deficits. It originated in Rice and Wexler's (1996) who compared performance on tense-related morphemes to a heterogeneous group of control morphemes that were not related to tense, in children with SLI and typical controls. Since then, several studies have used composite groups of morphemes to measure syntactic production in TD children, children with SLI, and children with Down's syndrome (Bedore and Leonard 1998, Eadie *et al.* 2002, Leonard *et al.* 1999, Rice 1998). Aggregate measures capture a whole syntactic subdomain in one variable, in addition to limiting the number of statistical comparisons (Eadie *et al.* 2002), and including variables whose individual frequency would otherwise be too low for analysis. However, no standard definition of composites has yet emerged. Although showing considerable overlap, the morpheme subsets used vary depending on the goals of different studies.

The Verb Morphosyntax composite (VM) is a count of words and affixes that express tense.[5] It includes regular past tense -*ed*, regular third-person singular -*s*, and finite non-negative forms of copula *BE*, auxiliary *BE*, auxiliary *DO*, and auxiliary *HAVE*. We excluded negative contractions that conflate syntactic and semantic information (Mabel Rice, personal communication). Noun Morphosyntax (NM) is a measure of overall production of morphological forms related to nouns. It counts tokens of the articles *a*/*an* and *the*, the possessive -'*s*, and regular plurals, and it is identical to the noun morphology composite used in Bedore and Leonard (1998).

[5]We did not include the two aspectual markers progressive -*ing* and perfect (generally -*en*), because they are usually not included in verb composites (or in tense composites, for obvious reasons).

When morphosyntax is measured this way, the composites are usually percentages of production of morphemes in 'obligatory contexts' (OCs). Measures based on OCs are undoubtedly useful under elicitation conditions, where the grammatical targets are known and the number of OCs can be manipulated. On the other hand, their use in conversational samples is not without problems: children's productions may not give enough clues to determine intended targets (Balason and Dollaghan 2002). In addition, Brown (1973: 257) points out that OCs become rarer the younger the children. In fact, OCs for grammatical morphemes are usually very infrequent in spontaneous language samples (Balason and Dollaghan 2002), making estimates extremely unreliable.[6] Furthermore, Hewitt *et al.* (2005) comment on the difficulty of determining OCs in conversation and report their percentage coder agreement as 67%. In short, even though use of a measure of OCs would facilitate comparison with earlier studies, its methodological problems render its applicability suspect for analysing conversation.

Nevertheless, analysing conversational samples is vital because it constitutes data collected in the most ecologically valid way possible, limiting extraneous task demands (Stromswold 1996). For example, measures from conversation help estimate the practical importance in children's language of theoretically central morphemes (like the regular past tense -ed and the third present singular -s). Token counts provide a more direct measure of the quantity of morphosyntactic production, information that is usually absent from statistical analyses (see, for example, Chapman *et al.* 1998, who report frequencies of production of grammatical morphemes in 12-minute narratives samples but provide no statistical analysis). Recently, token measures (in the form of percentages) have been used by Levy *et al.* (2006) for past tense verbs, noun plus complement clauses, and complex sentences. Cleave *et al.* (2007) also used token measures to analyse verbal and non-verbal syntax in SLI and Down's syndrome. In addition, token-frequencies are important as a measure of the likelihood of entrenchment and learning of forms. Frequency of use correlates with entrenchment of forms. Fewer productions mean less practice and possibly slower learning rates.

Count measures, however, are affected by unequal numbers of opportunities for production of a morpheme. To bypass this problem, in this study we had equal length conversational samples (100 utterances for each subject). Furthermore, these were obtained from a standardized assessment (The Autism Diagnostic Observation Schedule: ADOS). Still, inflation of token counts from children producing the same type repeatedly cannot be controlled with our measures (but note that this is also the case with OC measures obtained from conversation). For that reason, we supplemented out token-frequency model with several quantitative and qualitative analyses. Specifically, we (1) compared production of verbal and nominal tokens across the different groups, (2) regressed VM and NM on diagnostic group, covarying the number of verb and noun tokens for each participant, and (3) qualitatively examined outlying participants that had low numbers of types but high numbers of tokens.

**Coding—**The first author (blind to the diagnosis for each participant) coded the data automatically via Excel macros, and subsequently verified the coding utterance by utterance manually. Coding was relatively straightforward, and whether to credit certain forms was decided on an individual basis. Plural forms were not given credit when part of a brand name ('Skittles'), when a singular corresponding form does not exist or is uncommon ('pants'), or in general when thought to be learned as a whole ('lots' in 'lots of'). No past participles ending in *-ed* were given credit because they carry aspect, not tense, even though they are homophonous with the past tense forms. 'Supposed to' and 'used to' were not

[6]For example, assuming the true percentage being estimated is 50%, with three OCs (the minimum required by Eadie *et al.* 2002) the 90% confidence level interval ranges from 24% to 76%.

credited either, because they are arguably fixed forms in the adult language. *DO* and *HAVE* were not given credit when used as main verbs.

A trained research assistant recoded 15 (13%) randomly selected transcripts by hand (without using macros). Reliability was very high, as shown by Shrout and Fleiss's (1979) ICC(2, 1) intraclass correlation coefficients: 0.89 for VM and 0.93 for NM.

### Data analysis strategy

A hierarchical linear model (HLM) tested between group differences on the two composite measures of grammatical morphemes, VM and NM. The primary predictor was diagnostic group, a categorical variable with three levels: FXS boys without autism (FXS-O), FXS boys with autism (FXS-ASD), and typically developing boys (TD). Three covariates were included: non-verbal MA, measured by the Leiter-R Age Equivalent score, articulatory skill, measured via the GFTA-2 as the percentage of consonants correct, and maternal education, coded as the total number of completed years of education. We calculated Pearson's $r$ to examine correlations among the three covariates and the two outcome measures for each diagnostic group, to help us identify possible collinearity between variables and interpret the HLM model.

We created an indicator variable ('composite') with two levels in each model corresponding to the dependent variables, VM and NM, and we treated it as a repeated measure to account for the non-independence of the measurements within each child. The interaction between diagnostic group and composite addressed whether the different measures behave differently between groups. A random intercept was included in the model, but all other effects were fixed. The model was estimated under REML with an unstructured covariance matrix. Even though mild non-normality was present, normal probability plots and examination of residuals revealed no problems with the assumption of multivariate normality. Subsequent pairwise tests tested differences between the TD group and the two FXS groups, and between FXS-O and FXS-ASD. The APA Task Force on Statistical Inference strongly recommends reporting effect size measures. Given the current debate about effect sizes obtained from HLM models, we report the size of mean differences as a percentage of the maximum score in the groups being compared.

## Results

### Descriptive analyses

**Occurrences of individual grammatical morphemes—**Means and standard deviations for occurrences of individual morpheme measures for each of the three groups are shown in table 2. It is sorted in decreasing order of mean values for the TD group to facilitate comparisons of relative frequencies. The rightmost column details the composites to which each form contributes. Even though they do not enter in the VM composite, we include negative forms of verbal morphemes here to illustrate the relative importance of omitting these data.

**Typically developing (TD)—**The two most common morphemes are articles and *BE*. This latter's frequency is carried almost exclusively by the word forms with unmarked polarity ('+': *am, are, is, was, were*, and their contractions). Next in frequency are plurals, auxiliary *DO*, and third singular -*s*, in that order. Auxiliary *HAVE* and possessive -'*s* are rare. The only auxiliary whose negative form is more frequent than the unmarked one is *DO*. Past -*ed* is rather more infrequent than expected given its theoretical importance as a tense marker.

**Fragile X syndrome (FXS, FXS-ASD)—**Like for TD boys, articles and *BE* were the most frequently occurring morphemes for the two groups with FXS (For FXS-O, *BE* is more common than articles). Next are plurals. Production of third singular -*s* seems remarkably low, compared with either the positive or negative forms of *DO*. As well as for the TD population, for our groups with intellectual disability possessive -'*s* and auxiliary *HAVE* occur very rarely.

**Occurrences for composite measures—**Unadjusted means and standard deviations for total occurrences for each composite measure are shown in table 3.

**Correlation matrices—**Correlations among all variables are provided to help interpret the regression model. Table 4 shows non-verbal MA was correlated with morphosyntax in all groups, $r = 0.38–0.52$, all $p < 0.01$, except for the FXS-ASD group, where no correlations were significant at the 0.5 level. Maternal education was generally not significantly correlated with our outcome measures, except with VM in the TD group, $r = 0.34$, $p < 0.05$. Articulation is correlated with outcome measures in most cases. None of the correlations between covariates raises concerns about collinearity. The outcome measures are moderately correlated in all groups.

**Multilevel model results—**One multivariate outlier was removed prior with analysis (see the appendix), after which model assumptions were satisfactorily met, as indicated by normal probability and residual plots. The analysis indicated significant main effects for composite, $F(1, 109) = 37.71$, $p < 0.0001$, and diagnostic group, $F(2, 109) = 10.37$, $p < 0.0001$. The effect of composite simply reflects the fact that scores for NM are higher than scores for VM. This is to be expected, since on average there will be more noun phrases than verb phrases in a given utterance.[7] There was also a significant interaction between composite and diagnostic group, $F(2, 109)$, $p = 0.01$, indicating that this NM advantage is moderated by diagnostic. Specifically, NM scores are usually higher than VM scores, except in the FXS-O group, where there is no clear NM advantage. In addition, there were significant effects of all covariates in all groups: MA, $F(1, 109) = 13.09$, $p = 0.0005$, articulatory skill, $F(1, 109) = 11.56$, $p = 0.001$, and maternal education, $F(1, 109) = 4.18$, $p = 0.04$. Table 5 shows model parameters.

Table 6 shows the estimated mean number of morphemes in 100 utterances for each composite by diagnostic group. Between-group comparisons of both composites are reported as superscripts, with different superscripts indicating significantly different means, without adjustment since only two comparisons were performed.

- Do boys with FXS with and without ASD score lower than TD boys on both morphosyntactic composite measures, or is the impairment confined to one of the two domains? The TD mean for VM is higher than that for both FXS groups combined ($diff = 11.42$, $t(109) = 4.85$, $p < 0.0001$). This difference is 20% of the maximum VM score (58). The TD mean for NM is also higher ($diff = 6.51$, $t(109) = 2.77$, $p = 0.007$), this difference being 8.5% of the maximum NM score (77).
- Do boys with FXS and ASD score lower on one or both morphosyntactic measures than the boys with FXS only, indicating that comorbidity with ASD significantly impacts syntax? No. In fact, the boys with FXS-ASD scored on average higher than those with FXS-O in NM, although this difference is not significant ($p = 0.23$). The difference in VM (in the other direction) did not reach significance either ($p =$

[7]Simple sentences with transitive verbs, for example, have two noun phrases for one verb phrase. Also, noun phrases can occur in isolation in utterances, but verbs alone are rarer.

0.15). However, a post-hoc comparison revealed that the FXS-ASD mean for VM is significantly lower than the TD mean (*diff* = 8.56, $t(109) = 3.17$, $p = 0.002$, a 15% decrease in scores), suggesting a detrimental effect of comorbid ASD beyond the effect of FXS on verb morphosyntax.

## Supplementary type-frequency and lexical analyses

We were concerned that our token-frequency measures would be unduly sensitive to fewer opportunities for grammatical morpheme production in the FXS groups (although this would be the case also for percentages of production in OCs derived from conversational samples —see the discussion of this above). Another possible problem is that high VM and NM scores might be driven by a few lexically based forms that do not reflect true productivity, and might inflate some of our participants' scores. The decision to exclude repetition of utterances from the sample does go some way towards alleviating this problem. Nonetheless, we undertook several supplementary quantitative and qualitative analyses of our data.

First, we compared the range of verbal and nominal tokens in each group. An analysis of variance (ANOVA) indicated no effect of group, $F(3, 141) = 1.34$, $p = 0.26$. (No differences were found even after controlling for MA, maternal education, and articulation.) This indicates that all groups had, on average, the same number of opportunities to produce verbal and nominal morphology.

Additionally, we regressed VM and NM on diagnostic group and covaried the number of noun and verb tokens in each boy's conversational sample, to control for opportunities of morpheme production. There is a main effect of group on VM, $F(3, 137) = 18.39$, $p < 0.0001$, but not of noun and verb tokens, $F(1, 137) = 0.22$, $p = 0.64$, or an interaction effect, $F(3, 141) = 0.36$, $p = 0.78$. Similarly, there is a main effect of group on NM, $F(3, 137) = 13.12$, $p < 0.0001$, but not of noun and verb tokens, $F(1, 137) = 0.42$, $p = 0.52$, or an interaction effect, $F(3, 141) = 0.26$, $p = 0.85$. Therefore, it is not the case that lower scores on our composites are related to lower counts of nouns and verbs and therefore fewer opportunities for production. Scatterplots of VM and NM against noun and verb tokens, especially in the groups with intellectual disability, indicated that low scores are associated with a range of values for noun and verb counts.

We then plotted the VM and NM scores against the number of different morpheme types in each composite. The type-frequency for VM ranged from one to twelve; that for NM, from zero to three. (Type frequencies are bounded by the total number of morphemes we looked at in each composite; the restricted ranges make a regression analysis unadvisable, especially for NM.) We examined scatterplots of token- against type-frequency for VM and NM, divided into quadrants determined by the median values for each variable. We identified in the figure for VM participants 9 and 131 with low type-frequency scores and relatively high token-frequency scores, indicating possible item-based productivity. Participant 9 is in the TD group. He has a VM score of 46, for seven different verb word forms: the forms of *BE are, is*, and the contractions (*a*)'*m*, (*a*)'*re*, (*i*)'*s*; third singular -*s*; and the verb form *do*.

But the instances of *is* are different, some being a copula and others an auxiliary:

> This is where I put the ladder at!
>
> Dada is sleeping.

The instances of *do* and *are* are generally also different:

> How do you take the ladder out?

Do you put batteries in here?

And so are the verbs to which the third singular morpheme *-s* attaches:

And the little one goes right here!

It opens.

And this guy sits right here.

The dinosaur sleeps in here.

It is apparent that there is quite a range of productive uses of these morphemes.

Participant 131 has FXS-O. He has a VM score of 42 tokens, for six different verb word forms: third singular *-s*, (*a*)'*m*, (*a*)'*re, is*, (*i*)'*s*, (*ha*)'*ve*. Here is a sample of his verb morphosyntax uses:

It goes on top.

He licks Jordan.

I'm going on the moon.

They're watching soccer.

We're missing something.

Where is the remote?

Oh, there it is.

This rocket's stuck on the moon.

He's getting puppy kisses.

There is even one error of commission:

How it spins?

Again, there is no evidence of lexically based productivity here.

Participant 127, in the FXS-ASD group, has a rather high score on NM (33) driven solely by the presence of articles. The articles, however, attach to different nouns: *the baby, a soccer ball, the Mommy, the dog*, a *CD*, just to mention a few examples.

Moreover, children with low composite scores have pervasive morpheme omissions, as far as it can be ascertained from the conversational context. These few examples are from participant 115 with FXS-O:

I take (the) forks.

There (are) (the) cars.

It (is) my turn.

We conclude our token-based measures do not reflect either fewer opportunities for morpheme production in the groups with intellectual disability, or lexically based production, but rather genuine differences in the amount of verbal and nominal morphosyntax produced in conversation.

## Discussion

The goal in this paper was to determine whether boys with Fragile X syndrome (FXS) with and without autism spectrum disorder (ASD) have deficits in verbal and nominal morphosyntax when compared with typically developing (TD) boys of similar non-verbal mental ages. The study expanded on extant descriptions of morphosyntactic skill in FXS based on mean length of utterance (MLU) (Roberts *et al.* 2007a) and type-frequency measures (IPSyn) (Price *et al.* 2008, Roberts *et al.* 2007a), with detailed token-frequency measures for two subsets of syntactic morphemes: verb morphosyntax (VM) and noun morphosyntax (NM). For the first time we included a measure of consonant articulation in TD and FXS to ensure that putative differences in syntax scores were not due to differing articulatory skills.

Measuring morpheme production in conversation is a key component of analysing syntactic skills. Elicitation and other experimental paradigms are invaluable in assessing language, but only conversation (and perhaps narratives) has maximum ecological validity. Measures of production frequency during interaction, in addition, yield information about the actual relative importance of different syntactic elements. In this study, token frequencies of *-ed* are unexpectedly low, even in the TD group. Therefore, although regular past tense *-ed* is clearly a theoretically very important morpheme, in practical terms it may not occur very often in conversation (at least at the mental ages studied). Additionally, the inclusion of some morphemes of rare occurrence in composite measures (*HAVE*, possessive *-'s*) might not be useful when assessing language in conversation, regardless of the actual measure used. In contrast, deficits in article production, or copula and auxiliary *BE*, are likely to be both more visible and to have a higher impact on message understandability.

The findings constitute evidence that the expressive morphosyntax of boys with FXS is delayed beyond nonverbal MA expectations, contra Sudhalter *et al.* (1991) and in agreement with newer research (Price *et al.* 2008, Roberts *et al.* 2007a). Boys with FXS, regardless of ASD status, scored lower on average than TD controls on all measures. These differences subsist after controlling for maternal education level and consonant articulation skill, both of which had significant overall effects on morphosyntax. We addressed (and dismissed) concerns that findings of a syntactic deficit in FXS may be measurement artefacts.

Not unexpectedly, given the known difficulties of other clinical populations in this area (for example, SLI, DS), verbal morphosyntax is impaired in FXS, with or without ASD. This is consistent with Price *et al.'s* (2008) finding that both boys with FXS-O and FXS-ASD groups differ from TD boys on the Verb Phrases subscale of the IPSyn, a measure including modal and negative forms that were not part of our VM composite.

Boys with FXS with and without ASD also have particular impairments in the nominal domain. Articles and plural *-s* are the first and fourth most frequent morphemes in the sample, respectively. Therefore, future studies should test (perhaps experimentally) the hypothesis that these two morphemes are particularly impaired in FXS.

The hypothesis of a difference between FXS-O and FXS-ASD was based on studies of children with autism (but no FXS) and FXS separately (Bailey *et al.* 2000) and of high-functioning autistic children and adolescents (Roberts *et al.* 2004, Landa and Goldberg 2005). Lewis *et al.* (2006) suggested FXS-ASD represents a distinct subgroup of FXS, but reported no differences between individuals with FXS with autism and those with only FXS on expressive language measures. The study is consistent with these results. But significantly lower levels (tested post-hoc) in VM for FXS-ASD with respect to TD values suggest a difference in performance between the two FXS groups (albeit small), with those with FXS and ASD performing at lower levels than those with FXS only. Even though the

sample sizes represent a considerable improvement over previous studies on language in FXS, we still have limited power to detect moderate differences. Importantly, 68% of boys with FXS were within 1 point of the Communication ASD cut-off, 35% within 1 point of the Social ASD cut-off, and 31% within 1 point of the Communication plus Social ASD cut-off. (Children have to exceed all three to be diagnosed as having ASD.) Hence, about one-quarter to one-third of boys with FXS are classified into different groups, but their ADOS scores are quite similar. One important caveat is that these scores cannot be used as a continuous measure of autism severity. Future studies (including our own) should avail themselves of the new ADOS scoring algorithms that allow a continuous assessment of severity (Gotham *et al.* 2009).

In addition, given that Price *et al.* (2008) found boys with FXS-ASD produced a narrower range of questions and negations than TD boys, future studies should examine the performance of boys with FXS with and without ASD on syntactic negation (and indeed, on all forms) with targeted elicitation procedures. Even though such morphemes may be more perceptually salient (most are free-standing morphemes, and they sometimes carry stress), encode more salient semantic material (for example, the meaning of possibility carried by *CAN* is presumably more transparent than third-person singular present), and are clearly lexically generated (Eadie *et al.* 2002), the population with FXS may be unable to benefit from these putative advantages.

### What explains morphosyntactic impairment?

There is no direct evidence from this study that verbal and nominal morphology in FXS are impaired to different degrees, or are related differently to other cognitive, speech, or environmental aspects. Therefore, the results are consistent with a general syntactic impairment, the mechanism of which might be shared across all domains. However, a significant interaction between group and morphosyntactic subdomain was detected. Specifically, in boys with FXS-O, the difference between NM and VM is much smaller than in the other groups. This might be due to a more severe impairment in nominal morphosyntax, relative to verbal morphosyntax. This is consistent with our descriptive finding that FXS-O is the only group where the most common morphemes are not articles, but forms of copula and auxiliary *BE*. Such a pattern is unusual in disorders, and any hypothesis about mechanisms giving rise to this profile must await further confirmation with other methodologies.

As mentioned above, there are currently no theories of syntactic impairment in FXS. One possibility is that these difficulties are specific to syntax, in a way similar to Gopnik's (1990) *missing feature* hypothesis for SLI. Alternatively, although we did not test phonological working memory directly, boys with FXS have known impairments in this domain. Deficits in morphosyntax may therefore reflect short-term memory problems that would result in inability to construct appropriate morpho-phonological representations (Caselli and Stefanini 2006). At any rate, we found no evidence of a specific impairment in the verbal subdomain as opposed to the nominal subdomain. Rather, explanations should account for a widespread syntactic deficit in FXS, across many morphemes with disparate syntactic (verbal and nominal), morphological (bound and free), and phonetic (salient and reduced) properties, and possibly for a supplemental impairment in the verbal domain resulting from comorbid ASD.

Moreover, we know intelligibility to be impaired in the FXS population. One aspect of speech in FXS that likely contributes to this is variability in rate and speech rhythm. The measure of articulation (per cent consonants correct from the GFTA-2) may not have successfully captured this phenomenon. Consequently, future studies should examine the effect of lower intelligibility measured independently of articulatory skill. Estigarribia

(2010) presented data showing approximately 60% of the variance in syntax scores in three groups (TD, FXS, and Down's syndrome) is attributable to diagnostic group, cognitive measures (including phonological working memory), and speech intelligibility, but that the remaining 40% is possibly specifically syntactic.

What is certain is that lower token-frequencies of morpheme production in conversation have a potential effect on slowing down rates of growth for grammatical morphology. In usage-based theories where practice and entrenchment are important components of learning, one can see how these two would be affected if children are producing some morphosyntactic elements only sporadically. This could mean a lower strength of inflected forms to compete with uninflected ones, and therefore give rise to higher rates of errors of omission. In this sense, morpheme omission is to some extent a self-perpetuating practice.

In any case, further qualitative studies of morphosyntactic production are needed to address the implicated language processes directly. Comparisons of means cannot reveal directly the underlying processes that reveal children's competence in production. For instance, is inconsistent syntactic performance systematic, with production of a certain morpheme perhaps only found with certain lexical items or is it random across lexical items? The former scenario would suggest rote-learning/associative memory may compensate for the lack of generalized production rules. We took some steps toward ruling out lexical effects in our analysis, but a larger item-based analysis is still needed. Clearly, some partial knowledge is present that drives the inconsistent use of morphosyntax, but how best to characterize that knowledge is at present unclear.

## Clinical implications

Improvements in syntactic skill have the potential to improve general communication and also to improve the perception of the individuals with intellectual disability (Hewitt et al. 2005). Even though this study is not focused on intervention, the results suggest that expressive syntax is an area that should be targeted in boys with FXS, regardless of their autism status. We believe the implementation of specific syntactic training would be fruitful, since differences in morphosyntax subsist even after controlling for environmental and speech factors. Some syntax remediation procedures have recently been shown to lead to relative success in individuals with Down's syndrome (Camarata et al. 2006, Hewitt et al. 2005). It is unclear exactly how syntactic skill in FXS differs from the profile in Down's syndrome, although Price et al. (2008) indicates that the severity of syntactic impairment is greater in the latter.

These suggestions notwithstanding, we want to emphasize that implications for intervention and treatment should be guided by further research into (1) the causes of syntactic impairment in FXS, and (2) intervention studies in this population.

## Strengths and limitations

There were several strengths to this study. The analysis makes a much needed contribution to the sparse literature on the expressive morphosyntactic abilities of children with FXS. We documented impairments in both verbal and nominal morphosyntax beyond non-verbal cognition expectations, after the effects of maternal education and articulatory skills were removed. We used large sample sizes in each diagnostic group, especially in the lower-incidence FXS-ASD group. In addition, we separately investigated language ability in both a population with FXS only and a population with FXS and ASD.

One common limitation of language studies in populations with intellectual disability is the wide range of chronological ages (CA) involved, especially when a comparison group of TD

individuals matched for cognitive or language level is included. However, CA does not predict any variance in VM or NM in our data, after controlling for MA.

Although we did not find clear differences between the two FXS populations, in future analyses we plan to compare FXS groups with groups with ASD only. Needless to say, different ways of categorizing the boys with FXS who score in the spectrum range, but not in the autism range (for instance, a direct comparison between FXS-O and boys with FXS and full-blown autism), or using the new ADOS algorithm scores (Gotham *et al.* 2009) as a continuous predictor may reveal patterns of differences missed in our analysis.

A model of the interaction between general cognitive deficits, phonological memory deficits, intelligibility deficits, and expressive language, was beyond the scope of this study. This is crucial in view of the well-known intelligibility problems in FXS (Barnes *et al.* 2006). (Even though Roberts *et al.* (2007a) found that intelligibility was not a significant predictor of syntactic skill in FXS.) Estigarribia (2010) presented a model including putative predictors of syntax in these populations that begins to address the important issue of within-group variability.

Even though the ADOS is not specifically designed for sampling naturalistic language, we analysed parts of the assessment (such as make-believe play) that elicited more natural conversation, and the same parts were analysed for all groups. Using a semi-structured assessment helps elicit language from developmental populations for which unstructured interaction does not draw out enough linguistic behaviour. Nonetheless, replication of these results with less structured interactions is desirable. Future studies should include different sampling methods such as standardized tests, elicited production, and elicited imitation, so that specific morphological forms can be sampled and analysed. Knowing what children's intended targets are also allows a better assessment of errors of omission and commission than what is possible from naturalistic interaction. In addition, longer samples are needed to allow analysis of infrequent individual morphological forms. The current study is neutral with respect to different theories of syntactic competence. Note, first, that there is currently no theoretically principled account of language impairments in FXS. The goal was not to provide such a theory here. We regard theory-neutral analyses as an unavoidable first step in mapping out little-known aspects of given disorders.

In-depth studies of morphosyntactic use (including analysis of overgeneralization errors) are needed to examine whether there are systematic differences in morpheme use that cannot be captured by a comparison of means. The issue of development over time also remains to be studied. Finally, comparisons with other groups with language impairment or developmental disabilities (for example, SLI, Williams' syndrome) would further clarify commonalities and differences between FXS and other disorders.

### What this paper adds

It is not known whether morphosyntactic impairment is present in boys with Fragile X syndrome across different syntactic subdomains, and what other characteristics predict syntactic skill. Following a tradition of research in syntax in developmental disorders, we examine verb and noun morphosyntax production. Verbal and nominal morphosyntax are two coherent syntactic subdomains, possibly underlain by different learning mechanisms and different relationships to general cognition. Fine-grained descriptions of language phenotypes are crucial to investigate specific genes and gene/environment interactions that yield given linguistic phenotypes, as well as to inform assessment and intervention. We furthermore explore the impact of comorbid ASD and FXS. We conclude by

recommending investigations into the causes of these impairments as a crucial next step in informing theory and clinical practice.

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## Appendix: Multivariate outlier

Normal probability and residual plots identified one multivariate outlier. This participant belonged to the TD group. His chronological age was 4;1, and his MA was 4;2. His VM score was 41, and his NM score 118.

This observation was removed in the final HLM model reported here. The general pattern of results, however, did not change.

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## Table 1
### Background characteristics of the study participants[a]

| | FXS-O | FXS-ASD | TD |
|---|---|---|---|
| *N* | 35 | 33 | 46 |
| Chronological age (years; months) | 9;11 (2;11) | 8;7 (2;11) | 4;6 (1;2) |
| Non-verbal mental age (years; months) | 5;0 (0;11) | 4;8 (0;11) | 4;9 (1;0) |
| Maternal education (years; months) | 14;2 (2;4) | 15;2 (2;2) | 16;11 (2;1) |
| Per cent consonants correct | 86% (10%) | 85% (11%) | 88% (10%) |

[a]Note: FXS-O, Fragile X syndrome only; FXS-ASD, Fragile X syndrome with autism spectrum disorder; TD, typically developing.

## Table 2

**Means and standard deviations (SD) of occurrences of individual morphemes in 100 utterances[a]**

| Morpheme | TD (*n* = 46) | FXS-O (*n* = 35) | FXS-ASD (*n* = 33) | Contributes to composite |
|---|---|---|---|---|
| Articles | 28.9 | 18.6 | 18.8 | NM |
| | 10.5 | 11.2 | 8.3 | |
| BE (all) | 25.3 | 21.6 | 18.3 | |
| | 8.1 | 9.7 | 9.5 | |
| BE (+)[b] | 25.1 | 21.5 | 18.2 | VM |
| | 7.9 | 9.6 | 9.5 | |
| Plural -s | 10.4 | 6.1 | 7.7 | NM |
| | 8.2 | 4.2 | 4.9 | |
| DO (all) | 6.2 | 5.2 | 3.9 | |
| | 4.1 | 4.8 | 3.0 | |
| 3rd sg -s | 4.2 | 2.3 | 1.6 | VM |
| | 2.9 | 2.6 | 1.5 | |
| DO (−)[b] | 3.6 | 2.7 | 1.9 | |
| | 2.8 | 2.4 | 2.2 | |
| DO (+)[b] | 2.6 | 2.5 | 1.9 | VM |
| | 2.7 | 3.3 | 2.2 | |
| Past -ed | 2.3 | 1.0 | 1.1 | VM |
| | 2.5 | 2.1 | 1.2 | |
| Poss -s | 0.7 | 0.3 | 0.4 | NM |
| | 0.9 | 0.7 | 10.2 | |
| HAVE (all) | 0.6 | 0.5 | 0.2 | |
| | 0.9 | 0.9 | 0.6 | |
| HAVE (+)[b] | 0.5 | 0.5 | 0.2 | VM |
| | 0.8 | 0.9 | 0.6 | |
| BE (−)[b] | 0.2 | 0.1 | 0.1 | |
| | 0.5 | 0.3 | 0.4 | |
| HAVE (−)[b] | 0.1 | 0.0 | 0.0 | |
| | 0.3 | 0.0 | 0.0 | |

Notes:

[a]FXS-O, Fragile X syndrome only; FXS-ASD, Fragile X syndrome with autism spectrum disorder; TD, typically developing

[b]Counts of unmarked polarity forms are identified as (+); counts of negative polarity forms as (−).

## Table 3

**Means and standard deviations (SD) for each composite measure in 100 utterances[a,b]**

| Composite | TD (*n* = 46) | FXS-O (*n* = 35) | FXS-ASD (*n* = 33) |
|---|---|---|---|
| VM | 32.2 | 25.7 | 21.2 |
| | 10.6 | 11.0 | 10.3 |
| NM | 40.0 | 25.6 | 26.8 |
| | 15.9 | 13.2 | 11.1 |

Notes:

[a] FXS-O, Fragile X syndrome only; FXS-ASD, Fragile X syndrome with autism spectrum disorder; TD, typically developing.

[b] VM, verb morphosyntax; NM, noun morphosyntax.

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**Table 4**

**Within-group Pearson's *r* correlations[a]**

| Group[b] | Covariate[c] | Articulation | Maternal education | VM[d] | NM[d] |
|---|---|---|---|---|---|
| TD | MA | 0.40** | −0.05 | 0.39** | 0.38** |
| | Articulation | | 0.06 | 0.33* | 0.09 |
| | Maternal education | | | 0.33* | −0.03 |
| | VM | | | | 0.50*** |
| FXS-O | MA | 0.62*** | −0.34* | 0.50*** | 0.52*** |
| | Articulation | | −0.22 | 0.66*** | 0.50*** |
| | Maternal education | | | 0.06 | −0.04 |
| | VM | | | | 0.53** |
| FXS-ASD | MA | 0.56*** | −0.26 | 0.30 | 0.14 |
| | Articulation | | 0.12 | 0.40* | 0.42* |
| | Maternal education | | | −0.11 | −0.19 |
| | VM | | | | 0.40* |

Notes:

[a]* $p < 0.05$;

** $p < 0.01$;

*** $p < 0.001$.

[b]FXS-O, Fragile X syndrome only; FXS-ASD, Fragile X syndrome with autism spectrum disorder; TD, typically developing.

[c]MA, Leiter-R Mental Age Equivalent; maternal education, maximum level of maternal education.

[d]VM, verb morphosyntax; NM, noun morphosyntax.

**Table 5**

**HLM model parameters**

| Effect | Group | Composite | Estimate | Standard error | Degrees of freedom (d.f.) | *t* | *p* |
|---|---|---|---|---|---|---|---|
| Intercept | | | 24.56 | 2.01 | 106 | 12.23 | <0.0001 |
| Group | TD | | 8.56 | 2.70 | 109 | 3.17 | 0.002 |
| Group | FXS-O | | 4.11 | 2.86 | 109 | 1.44 | 0.15 |
| Group | FXS-ASD | | 0 | | | | |
| Composite | | NM[a] | 9.88 | 2.30 | 109 | 4.29 | <0.0001 |
| Composite | | VM | 0 | | | | |
| Group*composite | TD | NM | 1.14 | 3.03 | 109 | 0.38 | 0.71 |
| Group*composite | TD | VM | 0 | | | | |
| Group*composite | FXS-O | NM | −7.53 | 3.23 | 109 | −2.33 | 0.02 |
| Group*composite | FXS-O | VM | 0 | | | | |
| Group*composite | FXS-ASD | NM | 0 | | | | |
| Group*composite | FXS-ASD | VM | 0 | | | | |
| Maternal education | | | 0.86 | 0.42 | 109 | 2.04 | 0.04 |
| Mental age | | | 0.34 | 0.09 | 109 | 3.61 | 0.0005 |
| Articulation | | | 34.98 | 10.29 | 109 | 3.40 | 0.0009 |

[a]Note: VM, verb morphosyntax; NM, noun morphosyntax.

## Table 6

### Adjusted least-square means and standard errors[a]

| | TD[b] | FXS-O[b] | FXS-ASD[b] |
|---|---|---|---|
| VM[c] | 33.12[1] (1.78) | 28.67[1,2] (2.05) | 24.56[2] (2.01) |
| NM[c] | 44.14[1] (1.78) | 31.02[2] (2.05) | 34.44[2] (2.01) |

Notes:

[a] Within each row, means not sharing superscripts are significantly different ($p < 0.05$).

[b] FXS-O, Fragile X syndrome only; FXS-ASD, Fragile X syndrome with autism spectrum disorder; TD, typically developing.

[c] VM, verb morphosyntax; NM, noun morphosyntax.