The Development of Morphosyntax in Fragile X Syndrome

BY

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Abstract

An objective of present research across various developmental disorders is the comparison of language phenotypes; one goal is to determine the extent to which there are unique profiles of strengths and weaknesses associated with different disorders. Disorders with similar symptomology, such as autism and fragile X syndrome (FXS) are of particular interest. One area of inquiry within language development is morphosyntax. Morphosyntax is the interplay between grammatical morphology and syntactic structure. It is a known area of weakness for children with Specific Language Impairment as well as children with autism and language impairment. However, little work has been done within the realm of morphosyntax in FXS.

The purpose of this study is to examine morphosyntax in a group of children with fragile X syndrome, which is the most common inherited form of intellectual disability. Approximately 25-45% of males with FXS meet the criteria for a co-diagnosis of autism, and regardless of co-diagnosis, 50-90% of males are reported to display behaviors that are concurrent with autism symptomology. Given the co-morbidity of autism with FXS, an additional purpose of this study is to examine the impact of autism on FXS. Finally, a secondary purpose of this study is to examine the impact of gender on certain cognitive and linguistic variables in FXS.

Thirty-three children with FXS participated in the study: 26 males and 7 females, between 7-16 years of age. Children were asked to complete a number of standardized tests, including measures of morphosyntax, receptive language, and

nonverbal IQ. The examiner completed an autism rating scale, which served as the grouping mechanism for the children in this study: FXS and autism (FXS-A) versus FXS no autism (FXS-NA).

The data indicated that boys with FXS do have a specific deficit in morphosyntax, relative to language comprehension. The presence of autism did have a negative impact on the dependent variables, although there was not a significant difference on all the different types of verbs. Specifically, boys with FXS-A show a greater deficit on past tense markers for irregular verbs compared to boys with FXS-NA. In terms of gender differences, some of the females in this study have low nonverbal IQ, and receptive language scores. Additionally, irregular past tense verbs were a particular problem for this group of females.

This study serves as a direct extension of the literature on children with autism and language impairments (Roberts et al., 2004). The boys in this study demonstrated a similar deficit in terms of morphosyntax. This study, as well as the Roberts et al. (2004) study, highlights the importance of refining the language phenotype of FXS and how language delays are categorized in FXS and autism.

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Communication is a basic biological function, central to most aspects of day-to-day functioning. Humans use communication to relay ideas, basic needs, emotions, knowledge, and a number of other functions to each other. It is an innate ability in human beings, and develops throughout the first several years of life. Given the critical importance of language and communication, disruptions to the linguistic systems can have global effects on functioning. Intellectual disabilities are commonly associated with language and communication delays and deviant development. Among the intellectual disabilities commonly cited, fragile X syndrome (FXS) and autism represent two such instances.

Fragile X syndrome is the most common known inherited cause of intellectual disability, affecting an estimated 1 out of every 4000 males and 1 out of every 8000 females (Crawford, Acuna, & Sherman, 2001; Hagerman, 2002; Turner, Webb, Wake, & Robinson, 1996). It is a single-gene disorder, and diagnosis involves genetic testing to determine the presence of mutations on the long arm of the X chromosome. Autism on the other hand, has an unknown etiology. A diagnosis of autism is not based on genetic testing, but rather a behavioral diagnosis (Lord et al., 2000). There is a high rate of co-morbidity of autism within FXS (25-50% of males), and even those children who do not have symptoms warranting a co-diagnosis display a high degree of autistic-like behaviors (Bailey, Hatton, Mesibov, Ament, & Skinner, 2000; Hatton, 2006; Rogers, 2001). The two disorders share a great deal of overlap in terms of symptomology, making the comparison between them of particular interest.

An objective of present research across various developmental disorders is the comparison of language phenotypes; one goal is to determine the extent to which there are unique profiles of strengths and weaknesses associated with different disorders, relative to explanations based primarily on the presence of an intellectual disability (Warren & Abbeduto, 2007). Morphosyntax has been an area of interest in language disorders due to its central role in linguistic communication. Morphosyntax is the interplay between grammatical morphology and syntactic structure. It refers to the closed class morphemes of a language, specifically, morphemes associated with inflectional morphology (e.g., he walk*s*, he walk*ed*), derivational morphology (e.g., fool, fool*ish*), and words with a specific functions such as articles and auxiliary verbs (Leonard, 2002). In particular, tense and agreement markers have been identified as a unique area of weakness for children with Specific Language Impairment (SLI), and also for some children with autism (Rice, Wexler, & Cleave, 1995; Rice & Wexler, 1996; Roberts, Rice, & Tager-Flusberg, 2004).

The purpose of this study is to examine morphosyntax in a group of children with fragile X syndrome; one reason for this is to create the basis for comparing this aspect of the language phenotype of this disorder to other disorders (i.e., autism and SLI). Tense and agreement morphemes are the primary focus of investigation. Additionally, given the co-morbidity of autism with FXS, an additional purpose of this study is to examine the impact of autism on FXS. Finally, a secondary purpose of this study is to examine the impact of gender on certain cognitive and linguistic variables in FXS.

Fragile X Syndrome

Fragile X is an X-linked disorder. The gene is located in the 5' untranslated region on the long arm of the X chromosome (locus Xq27.3). The gene is called FMR1, and it directs cells to produce the fragile X mental retardation protein (FMRP), which is believed to play an important role in typical brain development and functioning (Rogers, Wehner, & Hagerman, 2001). Research has indicated that FMRP is involved in synaptic maturity and plasticity (Churchill et al., 2002). The FMR1 gene is made up of trinucleotide (CGG) repeats. A normal number of repeats ranges anywhere from 5 to 50. Fragile X syndrome occurs when an individual has an elevated number of CGG repeats (Hagerman, 2002). This increased number of trinucleotides results in excessive methylation of cytosines in the FMR1 promoter region, thereby shutting down transcription of the FMR1 gene into mRNA. This in turn interferes with the translation of FMRP (Reiss & Dant, 2003). Trinucleotide repeats ranging from 50 to 200 signify a premutation carrier. Full mutation occurs when an individual has more than 200 repeats (Bailey et al., 2001; Hagerman, 2002). Since FXS is an X-linked disorder, fathers can pass it to their daughters, while mothers can pass it to their sons or daughters. All daughters of premutation males inherit the gene, but only as carriers. Female carriers have a 50% chance of passing the expanded gene (full mutation form) on to their children. Fragile X is a dynamic gene mutation, meaning that it is unstable and will most likely expand through generations. Males are typically more affected compared to females, since it is an X-linked disorder (Hagerman & Hagerman, 2002).

In addition to substantial cognitive and motor delays, individuals with FXS have a number of speech and language delays including late emergence of first spoken words, problems with intelligibility and delays in both expressive and receptive morphosyntax (Abbeduto & Hagerman, 1997; Sterling & Warren, 2007). Boys with FXS show greater delays in expressive compared to receptive language (Roberts, Mirrett, & Burchinal, 2001). Research has indicated that children with FXS and autism have lower language skills in general than children with FXS no autism (Bailey et al., 1998; Rogers et al., 2001). The work to date has been descriptive in nature, particularly in terms of the more advanced linguistic skills, such as morphosyntax. Although significant work has been conducted on morphosyntactic development in Down syndrome and some research with children with autism, research with children with FXS has been limited. In particular, several studies have excluded children with both FXS and autism, thereby ignoring this large subgroup of children.

Comorbidity of FXS and autism

One reason FXS is of particular interest to researchers is the high co-morbidity with autism. Approximately 25-45% of males with FXS meet the criteria for a co-diagnosis of autism, and regardless of co-diagnosis, 50-90% of males are reported to display behaviors that are concurrent with autism symptomology including hand biting, hand flapping, perseveration in speech, tactile defensiveness, and poor eye contact (Bailey, Hatton, Mesibov, Ament, & Skinner, 2000; Hatton, 2006; Rogers, 2001). Males with both FXS and autism typically have more severe

language and social impairments, as well as lower IQ scores compared with children with FXS without autism (Bailey et al., 1998). Rogers and colleagues have completed a number of studies examining the impact of autism on FXS. They reported that children with comorbid FXS and autism have sensory impairments similar to children with autism without FXS (Rogers, Hepburn, & Wehner, 2003). Imitation skills were more impaired in children with FXS and autism compared to FXS only (Rogers, Hepburn, Stackhouse, & Wehner, 2003). Higher levels of adaptive and problem behaviors also appeared to be more impaired in boys with FXS and autism compared to boys with FXS only (Kau et al., 2004).

Rogers, Wehner, & Hagerman (2001) examined the presence of autism in 24 children (23 boys, 1 girl) with FXS between the ages of 21 and 48 months. Eight of the children in the study were diagnosed with both autism and FXS, whereas the remaining 16, including the one girl, had FXS only. Rogers and colleagues also recruited a group of children with autism but no FXS. The authors found that the children with both FXS and autism scored lower on measures of both expressive and receptive language compared with the children with FXS without autism and the children with autism without FXS. In short, the co-occurrence of both FXS and autism almost inevitably means that communication and social skills will be more severely impaired from early in development onward.

The majority of these studies have utilized the *Autism Diagnostic Observation Scale* (ADOS; Lord et al., 2000) or the *Autism Diagnostic Interview-Revised* (ADI-R; Lord, Rutter, & Le Couteur, 1994). The ADOS and ADI-R represent the current gold

standard for autism diagnosis. However, they are both quite costly in terms of training and time to administer. The *Childhood Autism Rating Scale* (CARS; Schopler, Reichler, & Renner, 1988) is typically used as a screener for autism in the clinical world. Researchers often utilize the CARS in studies, given its relative low cost in terms of training, time and administration. The CARS is composed of a number of subscales focused on behaviors associated with the three core impairments in autism (e.g., verbal and nonverbal communication, repetitive behaviors).

Studies have shown a high correlation between CARS scores and autism diagnosis within samples of children with FXS (Bailey et al., 1998; Bailey et al., 2000). Although it is not a diagnostic tool, it has high internal validity (Eaves & Milner, 1993; Sturney, Matson, & Sevin, 1992; Teal & Wiebe, 1986), and reliability. Given the previous studies findings on the differentiation of the two groups based on the ADOS and the ADI-R it is surprising that there has not been a similar examination of group membership based on the CARS subscales.

Morphosyntax in FXS

The limited work to date on morphosyntactic development in FXS syndrome has extended to both receptive and expressive language and has almost exclusively focused on males. Early work in the 1980s involved case studies, or very small sample sizes, and was generally restricted to comparisons based on Mean Length of Utterance (MLU). This is a very common index of sentence length, but some researchers argue that it is not highly reliable at measuring greater complexity in

morphosyntax, particularly in developmental disabilities (Rondal, Ghiotto, Bredart, & Bachelet, 1988; Scarborough et al., 1991).

Early work by Sudhalter and colleagues (1990) indicated that although morphosyntax is delayed in FXS, it is not deviant from the course of typical development. They predicted that the males with FXS with more perseverative speech would have the most limited expressive morphosyntax, and that there would be an unusual relationship between MLU and their structural complexity (based on the Index of Productive Syntax, IPSyn, evaluated from a language sample). However, the authors did not find a significant relationship between perseverative speech and low syntactic competencies. Additionally, the 12 males with FXS in this study showed a similar pattern in terms of MLU and its relation to grammatical complexity as seen in typical development, just at a slower pace. Individuals in this study were excluded if they had a diagnosis of autism, and the ages of the participants ranged from 5 to 36 years. The language sample served as the sole instrument for language evaluation.

Recent work has involved larger sample sizes and a higher level of complexity in terms of analyses. Roberts et al. (2007) examined expressive morphosyntax and vocabulary skills of 35 boys with FXS between the ages of 3 and 14 years of age. Males with FXS and autism were excluded from the study. The authors included a nonverbal mental age match of 27 boys with typical development. Mean length of utterance, number of different words and grammatical complexity based on the *Index of Productive Syntax* (IPSyn; Scarborough, 1990) scores were evaluated based on a language sample of 100 utterances. The IPSyn is a measure of child language

development that yields a score for grammatical complexity. It is important to note that the IPSyn is a measure of emergence, and not mastery of grammatical structures. It was designed to measure the emerging syntactic and morphological complexity in utterances spoken during spontaneous language samples of preschool age children.

The children with FXS in the Robert's et al study showed significant delays in terms of their morphosyntactic skills, compared to the control group. The authors argued that the use of shorter, less complex utterances and the production of significantly fewer words in the FXS group support the theory of an overall expressive language delay, and not a specific syntactic delay. The boys with FXS in this study used less complexity in their noun and verb phrases, as well as sentence structure, but not in their use of questions and negations. The boys with FXS used many different nouns, pronouns, plurals and two-word noun phrases, but fewer complex noun phrases, and bound morphemes. In terms of their use of verb phrases, they used phrases with the copula, present auxiliary and present modals in utterances. However, they rarely used more complex verb phrases such as present and past tense verb markers, past tense copula, auxiliaries, and modals. In terms of their sentence structure, the children commonly used simple conjunctions and simple structures such as subject-verb-object, but only rarely used relative clauses and bi-transitive predicates (predicates with three arguments: subject, direct object and indirect object, e.g., Abby threw the ball to Annie).

Another recent study examining syntactic complexity during conversational language samples found that young boys with FXS do seem to have a syntactic delay,

beyond what is expected for nonverbal mental age. Price, Roberts, Hennon, Berni, Anderson and Sideris (2008) compared the language samples of 35 boys with FXS only, 36 boys with FXS and autism spectrum disorder (ASD), 31 boys with Down syndrome (DS), and 46 boys with typical development. The four groups were matched on nonverbal mental age, calculated from the Brief IQ composite from the *Leiter-R*. The authors compared the four groups on four separate subscales of the IPSyn (Scarborough, 1990). The four subscales included verb phrases, noun phrases, questions/negations, and sentence structure. The boys with FXS, regardless of autism status scored lower than the boys with typical development on three of the four subscales, verb and noun phrases, as well as sentence structure. The boys with FXS only did not score significantly lower than the boys with typical development on the questions/negations subscale, although the boys with FXS and ASD did score significantly lower. The boys with DS and FXS (regardless of autism status) did not score significantly different from each other, with the exception of the questions/negations subscale for the boys with FXS only.

The Roberts et al and Price et al studies indicate that boys with FXS do score lower than nonverbal mental age expectations on expressive morphosyntax. However, it is important to note that their language samples were not designed to elicit specific syntactic structures, and the authors did not use standardized tests for this purpose. It is apparent that the children in these studies have delayed morphosyntactic skills; however, the argument that there is not a morphosyntactic delay is premature given the lack of specific testing for this construct. However these studies serve as an initial

look at this question, and suggest the need for a deeper level of inquiry in terms of the nature of morphosyntactic development in children with FXS.

In another recent study, Price and authors (2007) examined receptive morphology and syntax skills in young boys with FXS and found a significant delay in these skills controlling for nonverbal mental age. The participants were the same boys from the Price et al. (2008) study. They compared the same groups of boys: 35 boys with FXS no autism, 19 boys with FXS and autism, 45 boys with DS, and 40 boys with typical development who were matched on nonverbal mental age. The authors used the *Test for Auditory Comprehension of Language – 3rd Edition* (TACL-3; Carrow-Woolfolk, 1999) to assess receptive vocabulary, grammatical morphemes, as well as multiword syntactic patterns (i.e., active and passive voice, direct and indirect objects). Boys with FXS, regardless of autism status, and boys with DS scored significantly lower on all the subtests compared to the boys with typical development. Boys with FXS did not differ according to autism status on the receptive language portion of the test. In addition, the authors found that nonverbal cognition was not significantly correlated with the TACL-3 for the boys with FXS no autism, although it was correlated with scores in the FXS autism group. The boys with DS and the boys with typical development did show a significant, large correlation between nonverbal IQ and the TACL-3 scores. The authors postulated that perhaps in FXS, linguistic and non-linguistic development is not as closely related as in Down syndrome and typical development.

It is important to note that these findings are not consistent with the findings from other studies. Some studies have found that receptive morphosyntax seems to keep pace with nonverbal cognition in children with FXS (Abbeduto et al., 2003; Madison, George, & Moeschler, 1986). Abbeduto et al. (2003) found that adolescents and young adults with DS had significantly lower scores on receptive morphosyntax scores compared to individuals with FXS matched on nonverbal mental and chronological age, as well as individuals with typical development, also matched on nonverbal mental age. However, the individuals with FXS did not differ significantly on their age-equivalent scores from the typically developing group. There was a positive correlation (r = .70) between nonverbal mental age and receptive morphosyntax. Madison et al. (1986) also found that the language skills, including expressive and receptive morphosyntax seemed to keep pace with nonverbal mental age abilities in their sample of twelve individuals from a single family, although the authors did report that expressive language was a relative strength for the males in the family compared to receptive language.

Paul et al. (1984) reported descriptive data from three case studies involving young males with FXS (note: two of the children were brothers). Although the children's receptive language seemed to keep pace with their nonverbal mental age, the authors did report that their expressive syntax was below that expected for nonverbal mental age and receptive language level (based on MLU). In a later study by Paul et al. (1987), the authors compared the speech and language characteristics of 12 adult males with FXS with the same measures in a group of adult males with

nonspecific forms of intellectual disability, and males with autism, matching the groups on age and IQ. There were no significant differences between the three groups in terms of performance on the measures of expressive morphosyntax. However, scores approaching significance did indicate possible deficits in expressive morphosyntax relative to individuals with nonspecific intellectual disability. In both of the studies mentioned, the sample sizes were very small and the methodologies were general and not precise, perhaps accounting for the lack of significant findings.

Although the work on morphosyntax in FXS has been steadily increasing over the past twenty years, there are a number of unanswered questions. The data that has been presented is as variable as the methods used in the studies. For the most part, the studies reported were descriptive in nature and did not use specific probes for morphosyntax, with the exception of the receptive morphosyntax studies. Some researchers have found a delay, relative to mental age, while others have argued that there is an expressive language delay in FXS, but not a delay specific to morphosyntax. In order to clearly answer this question, it is important to use methods specifically designed to measure morphosyntactic ability, and to use both language sampling and standardized testing. The elimination of children with a co-diagnosis of autism and FXS does eradicate a significant source of variance, but clouds the true picture of the linguistic profile of FXS since such a large portion of males with FXS have autism characteristics. A more in depth morphosyntactic profile of language development in FXS may settle the ambiguity in the expressive morphosyntactic findings.

Gender Differences in FXS

The literature has consistently reported that females with FXS are not as affected compared to males with FXS, reflecting the X-linked nature of this genetic disorder. As noted earlier, FXS is a mutation on the long arm of the X chromosome, which in turn shuts down the production of FMRP. Females have two X chromosomes, unlike males who have one X and one Y. Therefore, females with FXS have one X chromosome with a stable FMR1; instead of the protein being completely shut down, the presence of one stable FMR1 allows for partial production of FMRP (Jin & Warren, 2000; Kaufmann, Abrams, Chen, & Reiss, 1999; Tassone, Hagerman et al., 2000).

While males with FXS are typically reported to have intellectual impairments ranging from the moderate to severe (e.g., IQ of 30-70), females are reported to range from mild intellectual impairment to no noticeable impairments (e.g., IQ of 65-normal; Hagerman & Sobesky, 1989; Bennetto & Pennington, 1996). It should be noted that there are very few studies that include females with FXS, and recent work by Warren and colleagues have reported more severe cognitive impairments in some young females with FXS (Sterling, Brady, & Warren, 2007; Warren et al., in review). Murphy and Abbeduto (2003) noted in their review of language in FXS that based on the limited literature, it is unclear whether females demonstrate the same profile of impaired development as males, but to a lesser degree, or perhaps have their own unique profile.

Murphy and Abbeduto (2007) examined repetitive language in children with FXS. Their purpose was to search for any gender differences, as well as examine the impact of cognitive and linguistic ability on repetitive language. Both narrative and conversational contexts were analyzed. The males in the study produced significantly more repetitive language when using conversational devices, and there were no differences in utterance-level repetition or topic repetition. Conversational devices refer to rote phrases or sayings such as “that’s it”, or “that’s a wrap”, and typically help control the flow of the interaction but do not contribute to the substance of the interaction. It seems that males with FXS rely more on rote phrases in their expressive language, regardless of whether they are engaging in a conversation or telling a story. One point to note is that there were not gender differences on utterance-level repetition and topic repetition; in other words, both groups were engaging in these types of repetitive language. This is the only comparative gender study to date of language in FXS.

Autism

Autism spectrum disorders constitute a group of related neuropsychiatric disorders, including autism, Asperger’s disorder, Pervasive Developmental Disorder not otherwise specified (PDD-NOS), Rett’s disorder, and Childhood Disintegrative Disorder (Volkmar & Klin, 2005). Autism is characterized by impairments in three core domains: impairments in social interaction, impairments in verbal and nonverbal communication, and restricted and repetitive patterns of behavior, with an onset before 36 months of age (American Psychiatric Association, 1994). Autism is

separated from PDD and Asperger's syndrome by age of onset (before 36 months for autism), presence of language or cognitive delay, severity of symptoms, and the domains affected (all three must be affected in order to be diagnosed with autism; Lord & Risi, 2000).

There is evidence of delay in expressive syntax in children with autism. Roberts, Rice and Tager-Flusberg (2004) examined tense marking (third person singular, ex: he walk*s*; past tense –ed, ex: he walk*ed*) in a large group of children (n=62) with autism between the ages of 5-15 years of age. The authors focused on the parallels between children with SLI and children with autism and language impairment. Tense marking has been identified as a clinical marker for SLI in English-speaking children (Rice & Wexler, 1996; Rice, Wexler, & Cleave, 1995; Rice, Wexler, & Hershberger, 1998). Children with SLI frequently omit tense marker morphemes in every day speech, namely third person singular present tense (e.g., he walk*s*), and past tense for both regular and irregular verbs (e.g., he walk*ed, fell*); they also have difficulty producing them in an experimental task (Leonard, Bortolini, Caselli, McGregor, & Sabbadini, 1992; Rice et al., 1995).

The data published from this study came from a larger study, examining a number of linguistic and social cognitive variables in this population of children. The authors used a grouping technique drawn from criteria set forth in a paper by Kjelgaard and Tager-Flusberg (2001). Children were divided into three groups based on scores on the *Peabody Picture Vocabulary Test-III* (PPVT-III; Dunn & Dunn, 1997): normal language (standard scores above 85), borderline (standard scores

between 70 and 84), and impaired (standard scores below 70). Children were given linguistic probes based on early forms of the *Test of Early Grammatical Impairment* (TEGI; Rice & Wexler, 2001), as well as a nonverbal IQ test. The probes of particular interest elicited tense and agreement markers, specifically third person singular *–s* (e.g., he walk*s*), and past tense *–ed* (e.g., he walk*ed*). The children with autism and language impairment made significantly fewer correct responses on the third person singular probe (36.8% correct) compared to both the children with normal (76.3%) and borderline language (61.3%). The same finding was reported for the past tense probe: children with autism and language impairment supplied significantly fewer correct responses (30.6% correct) compared to the other two groups (63.8% and 58.2% respectively). It is noteworthy that even the children with autism in the normal language group performed well below age expectations on both probes.

Roberts et al. reported correlations between the two probes and PPVT-III, age and IQ measurements. Correct responses, collapsed across groups, were positively correlated with PPVT-III scores and age. In terms of the past tense probes, correct responses were positively correlated with PPVT-III, age, verbal and nonverbal IQ scores. The authors argued in their discussion that this study continues to elucidate the lack of evidence linking tense marking and nonverbal intelligence. Based on the results, nonverbal IQ accounted for 6% of the variance in performance on the third person singular probe, and 13% of the variance on the past tense probes. They also noted that some children with high nonverbal IQ scores scored poorly on the probes,

while some children with low nonverbal IQ scores performed quite well on the probes.

The data presented indicate similarities between the linguistic profile of children with SLI and children with autism and language impairments. The authors note the possibility of an SLI subgroup within children with autism. Given this evidence, a logical next step would be to compare other groups of children with similar symptomology.

Impact of Nonverbal Intelligence on Morphosyntax

The impact of nonverbal IQ on morphosyntax has been examined in different clinical groups (e.g., SLI, children with intellectual impairments). A series of studies on children with SLI and typical development indicated that nonverbal intelligence was not a significant predictor of performance on measures of grammatical tense markers (Rice, Tomblin, Hoffman, Richman, & Marquis, 2004; Rice, Wexler, & Hershberger, 1998; Rice, Wexler, Marquis, & Hershberger, 2000). The Roberts et al. (2004) study indicated that in a sample of children with autism and language impairment nonverbal IQ was not correlated with third person singular –s. However, it was significantly correlated with the past tense performance.

The impact of nonverbal intelligence on grammatical tense marking has also been examined in children with low nonverbal IQ with and without language impairments. Rice et al. (2004) included three additional groups of children within their analysis of children with SLI: a control group of children (control), children with nonverbal IQ below 85 who had impaired language (NLI), and children with

nonverbal IQ scores below 85 with normal language (low cognition; LC). The same test used in the Roberts et al. (2004) study, the *Test of Early Grammatical Impairment* (TEGI; Rice & Wexler, 2001) was also used in this study. The control group and LC children did not demonstrate impairment in terms of grammatical tense marking, and were not significantly different on the third person singular probe or the past tense probe. However, the children with SLI scored significantly lower compared to the LC and control groups, and the NLI group had the lowest performance across the board.

The LC group's performance indicated that low IQ is not necessarily indicative of linguistic performance. This group mirrored the performance of the children with typical language and cognition in terms of morphosyntax. Additionally, IQ was not significantly correlated with both measures on the TEGI in a group of children with autism. It should be noted that the children with autism in this study displayed a large range in terms of IQ, even within the group of children with language impairment. In fact, the nonverbal IQ scores ranged from 43-102, with a mean IQ of 71.3 (Roberts et al., 2004). Additionally, in the group of children with autism and normal language who performed quite well on the linguistic tasks, there was also a large range of nonverbal IQ scores, 53-153 (mean 95).

Data from several clinical groups, including children with SLI, autism and LC indicate that nonverbal IQ is not a significant predictor of morphosyntax (in particular, tense and agreement markers). It seems that certain aspects of delayed morphosyntax represent a sort of deviant development, and not just a general delay associated with impaired cognition and language abilities. Extending the work to

other groups of individuals with intellectual disabilities may help elucidate this finding.

Specific Aims

The literature to date indicates that certain children demonstrate deficits in morphosyntax development that are not explained by low cognition. The current study extends the research on this issue by including a group of children with FXS. Given the high comorbidity of autism within FXS, and the findings of specific morphosyntactic deficits associated with autism and SLI, the proposed study represents a logical extension. The overall aim of this study is to examine specific morphosyntactic structures (i.e., tense and agreement markers) in a group of children with FXS. Previous research suggests there is most likely an overall deficit in expressive morphosyntax in boys with FXS, but only at a very general level. In other words, previous studies have not examined the characteristics known to be a weakness for children with SLI and children with autism and language impairments (i.e., third person singular and past tense markers). Is the deficit in expressive morphosyntax in FXS evidence of an overall delay in language and cognition? Or do children with FXS, much like the children with autism and language impairments, demonstrate a linguistic profile of relative strengths and weaknesses (tense marking delayed relative to receptive language). If so, children with FXS could demonstrate a profile similar to that of children with SLI and children with autism and language impairments.

Within the realm of this overall aim, the impact of autism on language in FXS is also examined. Although the presence of autism is associated with lower overall language and cognitive scores, researchers do not know if the impairments represent a more impacted profile of development, or whether there are differences in the patterns of impairment in individuals with FXS compared to individuals with FXS and autism. This study examines not only productive morphosyntax, but also receptive language and nonverbal IQ in children with FXS with and without autism.

Finally, the impact of gender will be examined, preliminarily. Females are not well represented in the literature, and little is known about the linguistic and cognitive development of females with FXS. Typically research has reported that females are not as impacted as males but current studies are bringing this question under scrutiny.

Given the overall aim of this study, the following represent the main and secondary research questions and hypotheses for this study.

Main Research Questions and Hypotheses:

1. *Is there a morphosyntactic deficit in fragile X syndrome?*
    a.) *Do children with FXS display a specific deficit in tense marking (third person singular, ex: he walks; past tense –ed, ex: he walked), much like that seen in Specific Language Impairment (SLI) and children with autism and language impairment?*

Based on the similarities between FXS and autism, it is hypothesized that children with FXS will demonstrate deficits in morphosyntax, much like what is seen in children with autism and language impairments; specifically they will demonstrate a deficit in tense and agreement markers. Children with SLI show a distinct profile of strengths and weaknesses, including a protracted weakness in terms of tense and

agreement marking. Other structures of morphosyntax will be examined in this study, but only at a very general level.

2. *Is morphosyntax in general delayed relative to receptive language?*

Given the documented absence of a relationship between nonverbal IQ and morphosyntax, it is hypothesized that children with FXS will show deficits in morphosyntax, but that this will not be related to nonverbal IQ. However, the deficits will be relative to the other language measures, such as receptive language.

3. *Does the presence of autism in FXS impact the development of morphosyntax?*
    a) *Do children with both FXS and autism display a similar development of morphosyntax?*

In terms of the presence of autism, studies have documented the negative impact of autism on FXS (e.g., lower IQ, lower expressive and receptive language). Therefore, it is likely that children with comorbid FXS and autism will show greater deficits in terms of morphosyntax. However, given the literature on autism, it seems likely that the children with both FXS and autism and FXS only will show the same types of delays, but the presence of autism will impact these variables to a greater degree.

Secondary Research Questions and Hypotheses:

1. *Do females with FXS exhibit a specific deficit in morphosyntax?*
    a) *Specifically, do females with FXS demonstrate specific deficits in tense and agreement markers for the third person singular and past tense verbs endings?*
2. *Do females with FXS exhibit delays in their receptive language, and/or nonverbal IQ?*

Females with FXS were included, but only as a preliminary investigation to this topic. The work on FXS in females is limited, particularly in terms of language

development. Typically the literature has reported that females are not as affected, so it is likely that the females will not exhibit delays in terms of morphosyntax, receptive language and/or nonverbal IQ. However, given the evidence of some delays in cognition and the existence of repetitive language in some females with FXS (Murphy & Abbeduto, 2007) it does warrant examination into more specific aspects of linguistic development.

3. *Do the children with FXS show the same pattern of impairments as the children in the Roberts et al. (2004) study using the same set of PPVT-IV groups?*

In order to benchmark the children in this study within the broader literature, the children will be divided into the same groups based on PPVT-IV scores as the children with autism in the Roberts et al. (2004) paper. In other words, the children will be grouped according to level of language impairment: normal, borderline, and impaired language. It is hypothesized that the children in this study will show the same pattern of impairments compared to the children with autism.

Methods

Participants

The participants in this study included both boys (n = 26) and girls (n = 7) with the full mutation of FXS. Participants were excluded if they had not had genetic testing to confirm the diagnosis of FXS. The children ranged in age from 7 to 16 years. The mean age for the boys was 10 years 7 months, and the mean age of the girls was 11 years 3 months. The children were monolingual, English speakers, as indicated by parent report. Participants were recruited from state based support groups and a national parent listserv. Table 1 describes the participants and their demographic information. This was a sample of convenience and the participants in the study were from 16 states all over the country. The sample was virtually all white, and the children came from well educated homes.

Table 1

Participant Information

	Boys (n = 26)	Girls (n = 7)
Chronological Age	M = 10;7 SD = 2.64	M = 11;4 SD = 2.50
Child Race/Ethnicity		
Hispanic or Latino	1	0
White	25	7
Maternal Education (years completed)	M = 15.2 SD = 2.17	M = 14.4 SD = 2.23

P*rocedures*

The assessments were completed in the participants' home. All data were collected in the course of a single visit lasting 1 ½ to 3 hours. After obtaining informed consent from the legal guardian, and oral assent from the child, a number of different standardized tests as well as a language sample were completed (see table 2 for information about the main research questions and tests; table 3 for means and standard deviations). The assessment was videotaped using a digital video recorder mounted on a tripod. Participants were given breaks between testing as needed, and were rewarded for the completion of tasks with stickers. Each participant was given a ten dollar gift card at the conclusion of the visit.

Standardized Tests

Syntactic forms. The *Test of Early Grammatical Impairment* (TEGI; Rice & Wexler, 2001) was one of the dependent variables in this study. Three of the subtests were given to each participant: articulation of word final consonants, third person singular probe, and the past tense probe. After an initial training session, children were shown a picture and then asked to generate a sentence using the target structure (e.g., children shown a picture of a dentist; target answer: He cleans your teeth). Responses were scored and criterion scores were computed. The TEGI is a frequently used test in research involving children with Specific Language Impairment (SLI). It is sensitive and specific to the development of morphosyntax, and has been used for both children with normal and impaired cognition. Rice and colleagues have found that children with SLI have trouble mastering tense marking, specifically third person

singular and past tense –ed. Rice has argued that this delay in tense marking can be used as a clinical marker for language impairment in this group (Rice, Tomblin, Hoffman, Richman, & Marquis, 2004; Rice et al., 1998).

The scores for the TEGI are presented in percentage forms; in other words, 80 represents 80% correct on the subscale of interest. It is important to note that the TEGI scores are based on responses to scorable items, and not necessarily all the items on the subscale. For example, the third person singular probe is comprised of 10 items. Only verbs with an overt third person singular marker are included in the score. Children might provide a verb such as "can" which does not have an overt tense marker. This would be considered an unscorable response. If the child had 7 responses including verbs with over tense markers, then the percentage would be calculated based on these 7 responses: 5 correct, 2 incorrect would yield a score of 5/7 which is 71.4% correct.

The *Clinical Evaluation of Language Fundamentals-Fourth Edition* (CELF-4; Semel, Wiig, & Secord, 2003) was the second measure of morphosyntax. Although the CELF-4 consists of a number of subtests, only the word structures subtest was given. This subtest measures a number of grammatical morphemes, including plural –s, third person singular –s, present and past tense verb forms, possessive nouns, possessive, reflexive, and subjective pronouns, etc. The word structure subtest includes norms for 5-8-year-old children. However, the manual notes that this subtest can be given to an older child who appears to be functioning developmentally at a

younger level. This test was standardized with children with typical development, with children with autism, and with children with intellectual disabilities.

A *language sample* was completed with each participant. The samples were concluded once the child reached a minimum of 100 non-imitative utterances (approximately 20-25 minutes depending on the child). A standard set of conversation topics were presented. The questions were drawn from the experimental interview protocol outlined in a paper by Evans and Craig (1992). Questions focused on three topics: family, school, and preferred after-school activities (see the Appendix for the protocol). Sample questions included “Tell me about your family.”, “Let’s talk about your school.”, “Tell me about it.”, and “What types of things do you like to do when you are not in school?” The children were prompted based on their answers.

Receptive vocabulary. The *Peabody Picture Vocabulary Test Fourth Edition* (PPVT-IV; Dunn & Dunn, 2007) was used to measure receptive vocabulary. Participants are asked to point to a visual representation of a word spoken by the examiner. The PPVT-IV is a standardized test; age equivalent scores can be calculated based on results. In addition, the PPVT-III was used in the Roberts et al. (2004) to divide children with autism into language subgroups.

Nonverbal intelligence. The *Leiter Test of Nonverbal Intelligence* (Leiter-R; Roid & Miller, 1997) served as the measure of nonverbal cognition. In order to compute a Brief IQ composite, four subtests were administered: Figure Ground, Form Completion, Sequential Order, and Repeated Patterns. Individuals were asked to find

an item in a picture, choose the next item within a sequence, or arrange items in a pattern. The test took approximately 25 to 45 minutes to administer.

Autistic behaviors. The *Childhood Autism Rating Scale* (CARS; Schopler, Reichler, & Renner, 2002) was scored for all the children in the study. The CARS is a 15-item scale. The examiner completed this rating sheet after the assessment was completed. Each item ranges from one to four, with a score of one being within normal limits, and four as severely abnormal for age. Total scores are based on the sum of the 15 items. A score below 30 is considered nonautistic, scores from 30 to 36.5 are considered mildly to moderately autistic, and scores above 37 are considered severely autistic. Although scores from the CARS did not serve as a diagnosis of autism, the test is often used for research purposes and has documented reliability (Bailey, Hatton, Skinner, & Mesibov, 2001; Sevin, Matson, Coe, & Fee, 1991). This scale was used to group the boys in the study into two groups: boys with FXS who had scores below 30 (fragile X no autism; FXS-NA), and boys who had CARS scores above 30, on the autism spectrum (fragile X autism; FXS-A).

The *Social Responsiveness Scale* (SRS; Constantino, 2004) served as a second indicator of presence of autistic behaviors. The measure consists of 65 parent report items. The questions are grouped into five subscales: social awareness, social cognition, social communication, social motivation, and autistic mannerisms. A total raw score yields a T-score; in addition, each subscale also yields a T-score. The total T-scores mirror the same grouping systems as the CARS, ex: a score above 76 is

considered in the severe range, while a score of 60-75 is in the mild to moderate range. Any total T-score below 59 is considered in the normal range.

The standardized tests were scored on-line and then verified via the video recordings in the lab. Standardized scores were then calculated and both raw and standardized scores were entered into SPSS. Data entry was verified by an additional research assistant. The language samples were transcribed, coded, and analyzed using the Systematic Analysis of Language Transcripts (SALT; Miller & Chapman, 2000). Transcription guidelines were based on the SALT conventions manual. Bound morphemes were coded using the standard SALT conventions (e.g., She walk/3s to the store"). Word codes were used to identify errors at the word and utterance level (e.g., He were [EW:was] happy), irregular verbs (e.g., I hurt [IV] my elbow), copula BE (e.g., You are [cop] happy), auxiliary BE (e.g., He was [aux] running), and modal verbs (e.g., She should [modal] go to the store).

Trained research assistants transcribed and coded all language samples. Reliability was completed on all of the transcripts, and the checks were completed at both the word and code level. The independent reliability transcriber selected a random sample within the transcript of 100 child utterances to complete the word and code reliability. Word reliability was set at 85%, and consensus reliability was required for any transcript that did not meet criteria. The average percent agreement for word reliability was 87% and the range was from 80 to 98%. The consensus reliability was completed by the independent transcriber who noted all word disagreements using the transcript and the video, and then the disagreements were

settled by a third party. The coding reliability was completed after the word reliability; this was set at 85%. The average percent agreement for coding reliability was 92% and the range was from 80 to 100%. The consensus reliability procedures were similar for the codes. An independent transcriber noted all disagreements using the transcripts, and disagreements were then settled by a third party.

Table 2

Standardized Measures Linked with Main and Secondary Research Questions

	TEGI	CELF	PPVT-IV	Leiter	CARS/ SRS
Is there a morphosyntactic deficit in fragile X syndrome?	X	X			
Is morphosyntax in general delayed relative to receptive language?	X	X	X	X	
Does the presence of autism in FXS impact the development of morphosyntax?	X	X			X
Do females with FXS exhibit a specific deficit in morphosyntax?	X	X			
Do females with FXS exhibit delays in their receptive language and/or nonverbal IQ?			X	X	
Do the children with FXS show the same pattern of impairments as the children in the Roberts et al. (2004) study?	X		X		

Table 3

Means and Standard Deviations for all Test Variables

Measures	Males	Females
Language:		
PPVT-IV		
Mean	65.46	89.43
SD	*14.97*	*12.65*
TEGI – 3S		
Mean	54.32	91.11
SD	*38.17*	*16.02*
TEGI – Past Tense		
Mean	42.30	88.09
SD	*33.44*	*18.54*
CELF – WS		
Mean	13.24	25.00
SD	*8.99*	*8.08*
MLU		
Mean	3.09	6.74
SD	*1.06*	*1.38*
Autism:		
CARS		
Mean	27.71	17.29
SD	*4.33*	*2.67*
SRS		
Mean	77.96	59.33
SD	*10.81*	*16.91*
Nonverbal IQ:		
Leiter – R Brief IQ		
Mean	51.46	76.17
SD	*11.92*	*17.41*

Results

In order to answer the main and secondary research questions, the analyses were completed at various levels: descriptive statistics, *t* tests comparing boys with CARS scores above 30 and those with CARS scores below 30, a discriminant function analysis, a matching analysis for gender, and a series of univariate ANOVAs to examine group differences based on PPVT-IV groups.

The main research question for this study was: *Is there a morphosyntactic deficit in fragile X syndrome?* Therefore, the first level of analysis was the descriptive examination of the measures of morphosyntax. Tables 4-6 present the means and standard deviations for the boys on the Test of Early Grammatical Impairment (TEGI; Rice & Wexler, 2001). Table 4 reports the percentages of responses on the third person singular and past tense probes. The boys were grouped within the table by autism status as indicated by the CARS scores. A score above 30 on the CARS is considered on the autism spectrum (FXS and autism; FXS-A), while a score below 30 is considered nonautistic (FXS no autism; FXS-NA). Although scores within the autistic range can be categorized as “mild to moderate” or “severe autism”, these categories were collapsed for the analyses. For the majority of the analyses, the boys were grouped according to autism status.

Table 4

Percentage of Correct Responses on TEGI Probes (boys only)

	Third person singular	Past Tense	Regular Past Tense	Irregular Past Tense	Irregular Past Finite
FXS-NA					
N = 14					
Mean	65.66	52.30	48.86	45.54	55.86
SD	*35.00*	*35.31*	*39.44*	*33.51*	*33.95*
FXS-A					
N = 12					
Mean	38.76	29.60	38.35	16.38	18.53
SD	*41.50*	*27.33*	*35.93*	*26.36*	*29.06*

Tables 5-6 represent the type of responses given by each subgroup of boys with FXS on the third person singular and past tense probes on the TEGI. The *bare stem* column indicates when a tense marker was dropped. The *other verb* column indicates when the child provided a verb for which tense and agreement markers are not overtly expressed in English (e.g., He *can* walk). Additionally, instances where the child did not respond were also noted. The TEGI past tense probe includes both regular (e.g., She walk*ed*) and irregular verbs (e.g., She *ran*). Table 6 includes columns for both the regular verbs and irregular verbs. Additionally, over-regularization of irregular verbs in the past tense is also noted (e.g., *He *catched* the ball).

Table 5

Percentage of Responses on Third Person Singular Probes (Boys only)

Group	Correct	Bare Stem	Other Verb	No Response
FXS-NA				
Mean	58.57	17.14	12.86	11.43
SD	*29.83*	*19.39*	*12.04*	*28.78*
FXS-A				
Mean	30.00	19.17	20.83	30.00
SD	*36.18*	*22.34*	*21.51*	*45.53*

Table 6

Percentage of Responses on Past Tense Probes (Boys only)

Group	Regular Correct	Regular Bare Stem	Irregular Correct	Irregular Over Regular	Irregular Bare Stem	Other Verb	No Response
FXS-NA							
Mean	48.57	35.71	39.29	9.82	29.46	9.52	8.73
SD	*39.39*	*27.93*	*31.34*	*11.16*	*24.81*	*12.22*	*26.93*
FXS-A							
Mean	25.83	26.67	15.63	2.08	34.38	14.35	33.33
SD	*27.12*	*29.02*	*28.76*	*4.87*	*32.91*	*21.12*	*49.07*

Correlations were run to look at the relationship between the independent and dependent variables. The relationship between the CELF word structures subtest, TEGI third person singular probe, and TEGI past tense probe was examined with respect to language comprehension and nonverbal IQ. The boys were again divided into two groups based on scores from the CARS.

Table 7

Correlations between Language Comprehension and Measures of Morphosyntax (Boys only)

	PPVT-IV	Nonverbal IQ
FXS-NA		
CELF	.615*	-.357
TEGI-3S	.625*	-.094
TEGI Past Tense	.700**	-.319
FXS-A		
CELF	.690*	.097
TEGI-3S	.739**	.159
TEGI Past Tense	.507	-.096

Comorbidity of autism

The issue of autism in FXS was one of the main research questions. In other words, *Does the presence of autism in FXS impact the development of morphosyntax?* Specifically, does the presence of autism impact the dependent variables in this study, and furthermore, do the children also show differences in the independent variables based on autism status. Before looking at the two groups of boys, a series of correlations were run to examine the relationship between the autism measures and both the independent and dependent variables. Tables 8 and 9 present these correlations. A series of *t* tests were completed in order to compare boys with FXS-NA to the boys with FXS-A. The first step was to compare the independent variables, nonverbal IQ and language comprehension. The FXS-NA and FXS-A boys were not significantly different in terms of their nonverbal IQ scores $t(20) = .464$, $p = .648$, $d = .192$ (FXS-NA: $M = 52.43$, $SD = 12.15$; FXS-A: $M = 50.10$, $SD = 12.10$). They also

did not differ significantly in terms of language comprehension, $t(24) = 1.59$, $p = .126$, $d = .628$ (FXS-NA: $M = 69.64$, $SD = 15.63$; FXS-A: $M = 60.58$, $SD = 13.11$). However, there was a moderate effect size, indicating that perhaps with a larger sample, the difference might be significant.

Table 8

Correlations between Autism Measures and Independent Variables (boys only)

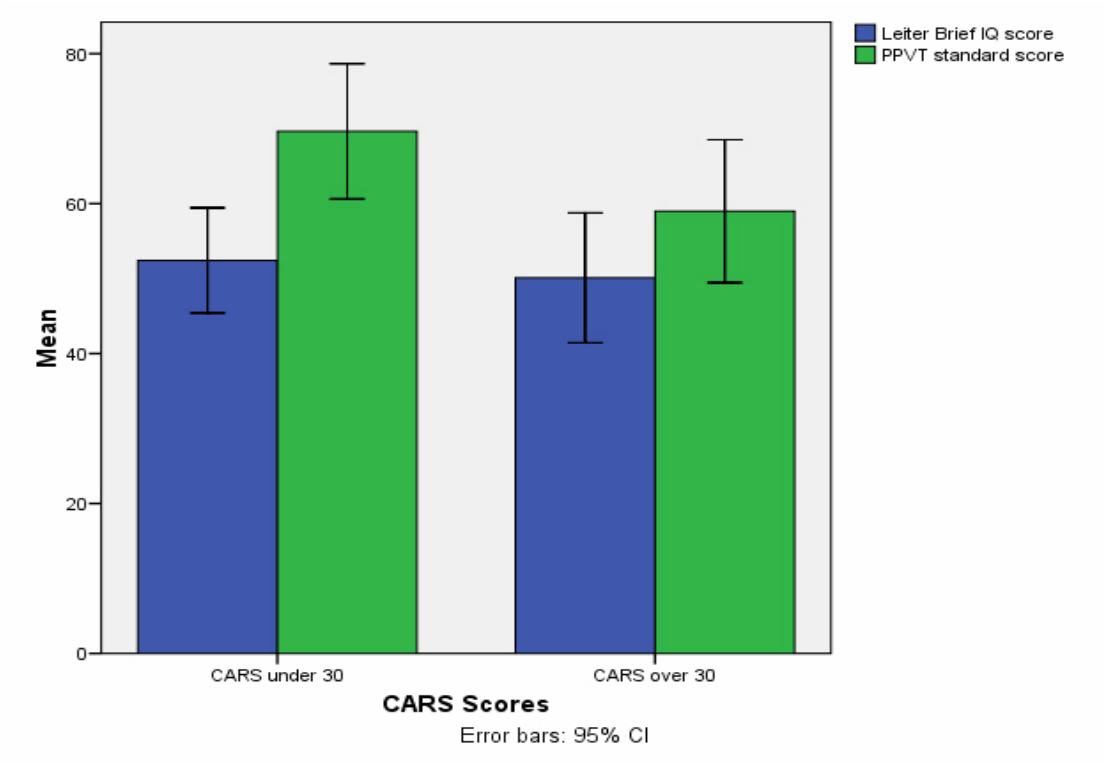
Independent Variables	CARS Total Score	SRS Total Score
PPVT-IV	-.363	-.045
Leiter-R	-.052	.184
Chronological Age	.001	-.205

Table 9

Correlations between Autism Measures and Dependent Variables (boys only)

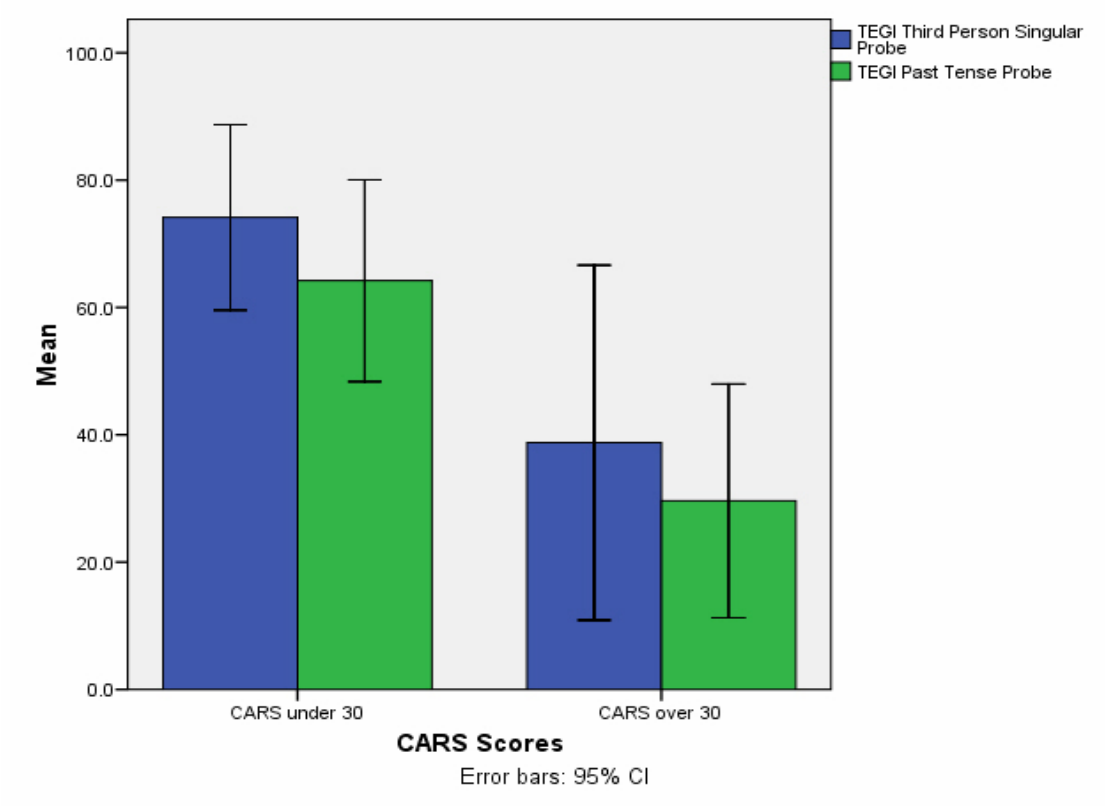
Dependent Variables	CARS Total Score	SRS Total Score
CELF	-.474*	-.128
TEGI-3S	-.452*	-.290
TEGI Past Tense	-.572**	-.237
TEGI Regular Past	-.383	-.223
TEGI Irregular Past	-.661**	-.170
TEGI Irregular Past Finite	-.688**	-.211

Figure 1. Mean scores on nonverbal IQ and language comprehension grouped by autism status (boys only)



The dependent variables were also compared. The scores on the CELF were marginally significantly different from each other, $t(23) = 1.98$, $p = .060$, $d = .796$ (FXS-NA: $M = 16.21$, $SD = 8.52$; FXS-A: $M = 9.45$, $SD = 8.45$). The effect size for this *t* test was .796, indicating a large effect. The third person singular probe did not yield a significant difference, $t(19) = 1.76$, $p = .092$, $d = .701$ (FXS-NA: $M = 65.66$, $SD = 35.00$; FXS-A: $M = 38.76$, $SD = 41.50$), however it did yield a medium effect size. The past tense probe yielded a nonsignificant difference with a medium effect size, $t(23) = 1.75$, $p = .093$, $d = .730$ (FXS-NA: $M = 52.26$, $SD = 34.31$; FXS-A: $M = 29.62$, $SD = 27.33$).

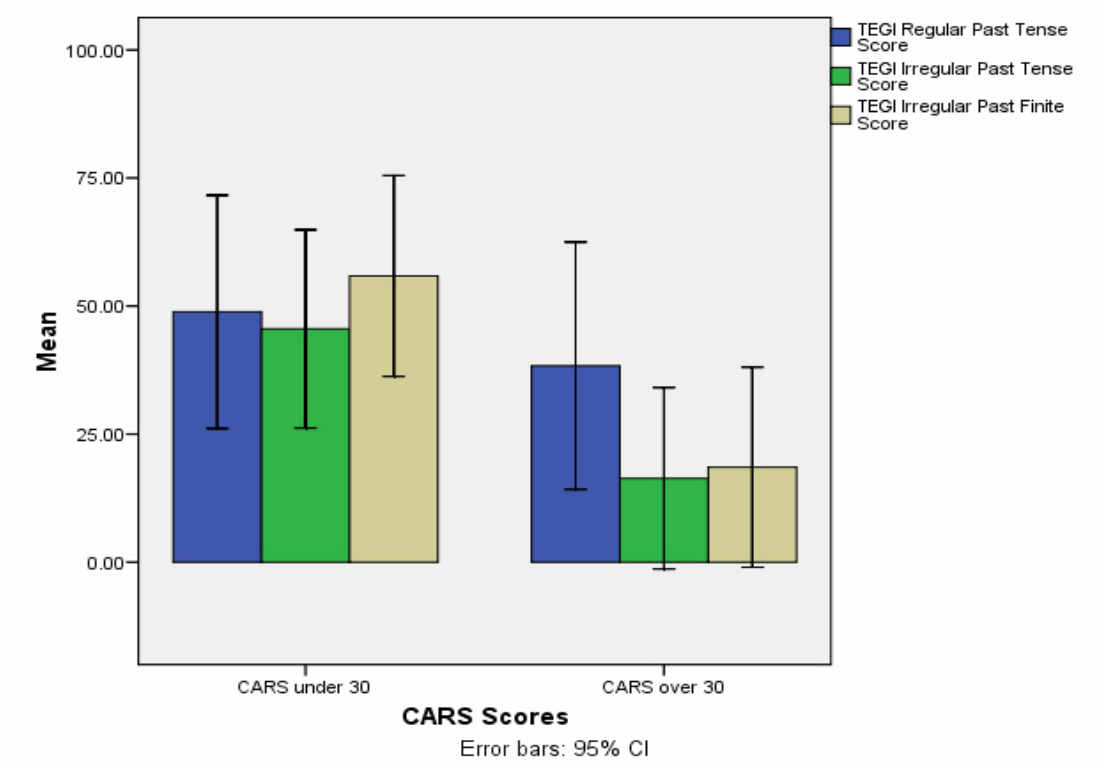
Figure 2. Mean scores on TEGI tasks grouped by autism status (boys only)



The three remaining subscales from the past tense probe of the TEGI were also compared using *t* tests. The regular past tense verbs did not yield a significant difference, and only a small effect was observed, $t(23) = .69$, $p = .499$, $d = .279$ (FXS-NA: $M = 48.86$, $SD = 39.44$; FXS-A: $M = 38.35$, $SD = 35.94$). The two groups of boys did show a significant difference on the irregular past tense verbs with a large effect, $t(23) = 2.37$, $p = .027$, $d = .967$ (FXS-NA: $M = 45.54$, $SD = 33.51$; FXS-A: $M = 16.38$, $SD = 26.36$). The groups also demonstrated a significant difference with a large effect on irregular past finite verbs, $t(23) = 2.90$, $p = .008$, $d = 1.182$ (FXS-NA:

M = 55.86, *SD* = 33.95; FXS-A: *M* = 18.53, *SD* = 29.06). In each case, the boys without autism scored significantly higher than the boys with both FXS and autism.

Figure 3. Mean scores on past tense probes grouped by autism status (boys only)



Discriminant Function Analysis

The next step in the analyses was to determine if the observed deficit in morphosyntax could discriminate the children with FXS in this study on the basis of autism status. A two group discriminant function analysis was performed using four measures of morphosyntax as predictors of membership in the two groups (FXS-A and FXS-NA). The predictors were performance on the third person singular probe

(TEGI), performance on the regular past tense verbs (TEGI), performance on the irregular past finite verbs (TEGI), and performance on the word structures subtest (CELF). The two groups were based on the cutoff scores from the CARS: under 30 = no autism (FXS-NA), and above 30 = autism (FXS-A). Both genders were included, due to the small overall sample size.

Thirty-three children with FXS were included in this analysis. One case was dropped due to missing data. There was a significant association between the groups and predictors χ^2 (4) = 15.26, p = .04. Box's M indicated that there were no violations of homogeneity, F(10, 1934) = 1.130, p = .335. The loading matrix of correlations between predictors and the discriminant function indicated that the predictors used in the analysis were able to distinguish between children with FXS-NA and FXS-A approximately 81.3% of the time. The FXS-A group scored significantly lower on all the tests compared to the FXS-NA group (see tables 3-4 for means and *SD*s).

Table 10

Results of Discriminant Function Analysis of CARS Groups

	Correlations of Predictor Variables with Discriminant Function		Pooled Within-Group Correlation among Predictors		
Predictor Variable	*1*	Univariate *F*(1, 30)	TEGI Regular Past Tense	CELF	TEGI Irreg. Past Finite
TEGI 3S	.58	7.19	.75	.82	.61
TEGI Regular Past Tense	.35	2.59		.83	.65
CELF	.62	8.43			.68
TEGI Irregular Past Finite	.89	17.12			
Canonical *R*	.65				
Eigenvalue	.73				

The classification results indicated that 81.3% of the cases were correctly classified according to autism group; this is a good level of classification overall, with more than two thirds of the cases correctly classified.

Table 11

Predicted CARS Group Membership

		Predicted Group Membership	
	CARS score groups	CARS under 30	CARS over 30
Count	CARS under 30	17	4
	CARS over 30	2	9
%	CARS under 30	81.00	19.0
	CARS over 30	18.20	81.80

Gender Differences

A secondary set of two research questions focused on gender differences in FXS. First, *Do females with FXS exhibit a deficit in morphosyntax?* Second, *do females with FXS exhibit delays in their receptive language, and/or nonverbal IQ?* Seven girls with FXS participated in the study. Table 12 presents descriptive information about the girls' performance on both the dependent and independent variables. The same set of correlations performed for the boys in the study was completed for the girls. The pattern of significant correlations was the same for the girls as for the boys, with language comprehension yielding significant results, and nonverbal IQ not significantly correlating with the dependent measures (see table 13 for more information).

Table 12

Percentage of Correct Responses on TEGI Probes (Girls only)

	Third person singular	Past Tense	Regular Past Tense	Irregular Past Tense	Irregular Past Finite
Girls					
Mean	91.11	88.09	87.14	75.60	90.47
SD	*16.02*	*18.54*	*29.84*	*29.53*	*14.18*

Table 13

Correlations between Language Comprehension and Measures of Morphosyntax (Girls only)

	PPVT-IV	Nonverbal IQ
Girls		
CELF	.768*	.615
TEGI-3S	.833*	.680
TEGI Past Tense	.409	.099

In order to examine gender differences, a comparison was made with a subset of the boys in the study. The overall means displayed by the girls compared to the boys regardless of autism status indicated as expected that the girls were not as delayed compared to the boys. In order to verify this, seven males matched on language comprehension at the group level were selected from the overall sample. A number of independent samples *t* tests were computed in order to examine differences between the boys and girls with FXS. Since the girls with FXS in this study did not score on the autism spectrum on the CARS, only boys with scores below 30 (FXS-NA) were included in the matched group.

Table 14 provides information about the boys vs. girls on the independent variables, including language comprehension, nonverbal IQ, and the two autism measures. The groups were not significantly different on language comprehension or the two autism measures, but they were significantly different on the nonverbal IQ measure.

Table 14

Gender Differences on the Independent Variables

Variable	Boys n = 7	Girls n = 7
Matching Variable: PPVT-IV *t*(8) = -1.57, *p* = .16	*M* = 81.43 *SD = 4.69*	*M* = 89.43 *SD = 12.65*
Additional Variables: Leiter Nonverbal IQ* *t*(11) = -2.99, *p* = .012	*M* = 52.71 *SD = 10.53*	*M* = 76.17 *SD = 17.41*
Childhood Autism Rating Scale* *t*(12)= 4.14, *p* = .002	*M* = 24.29 *SD = 3.59*	*M* = 17.29 *SD = 2.67*
Social Responsiveness Scale* *t*(11)= 2.31, *p* = .047	*M* = 78.14 *SD = 11.36*	*M* = 59.33 *SD = 16.91*

Figure 4. Gender differences on language comprehension and nonverbal IQ

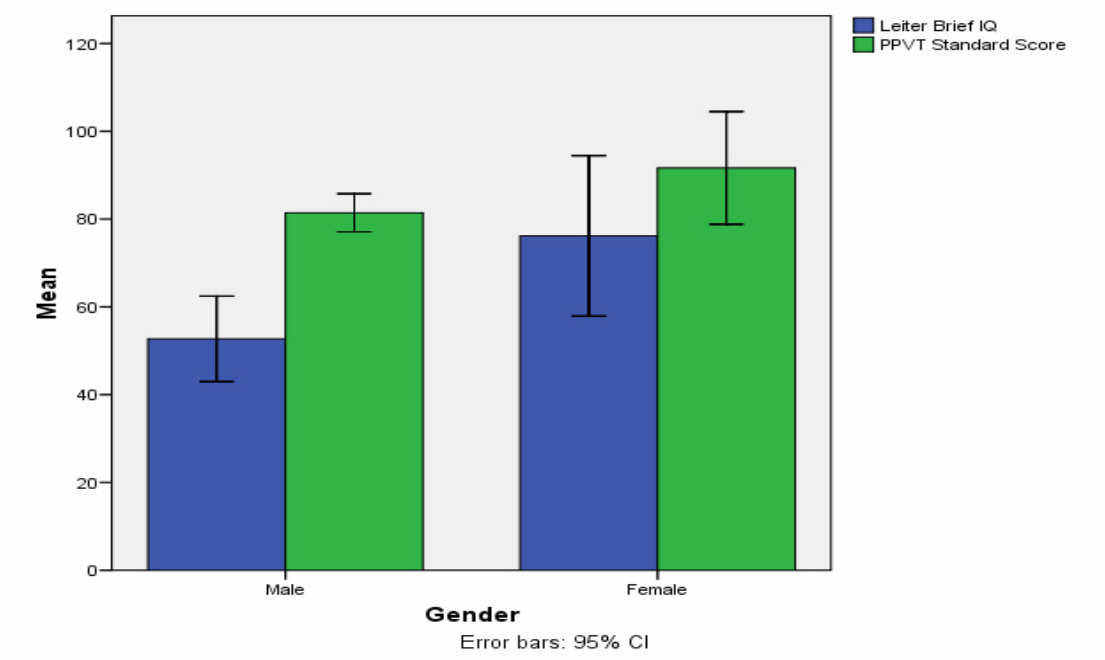
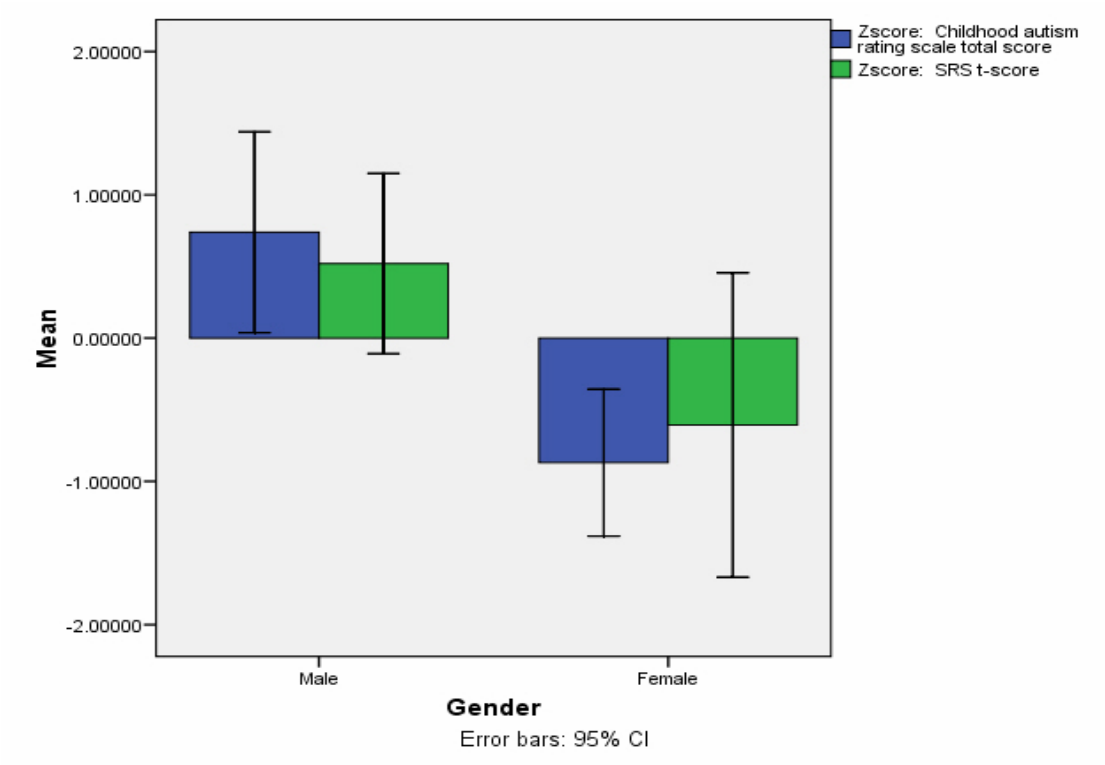


Figure 5. Gender differences on CARS and SRS total score (z-scores)



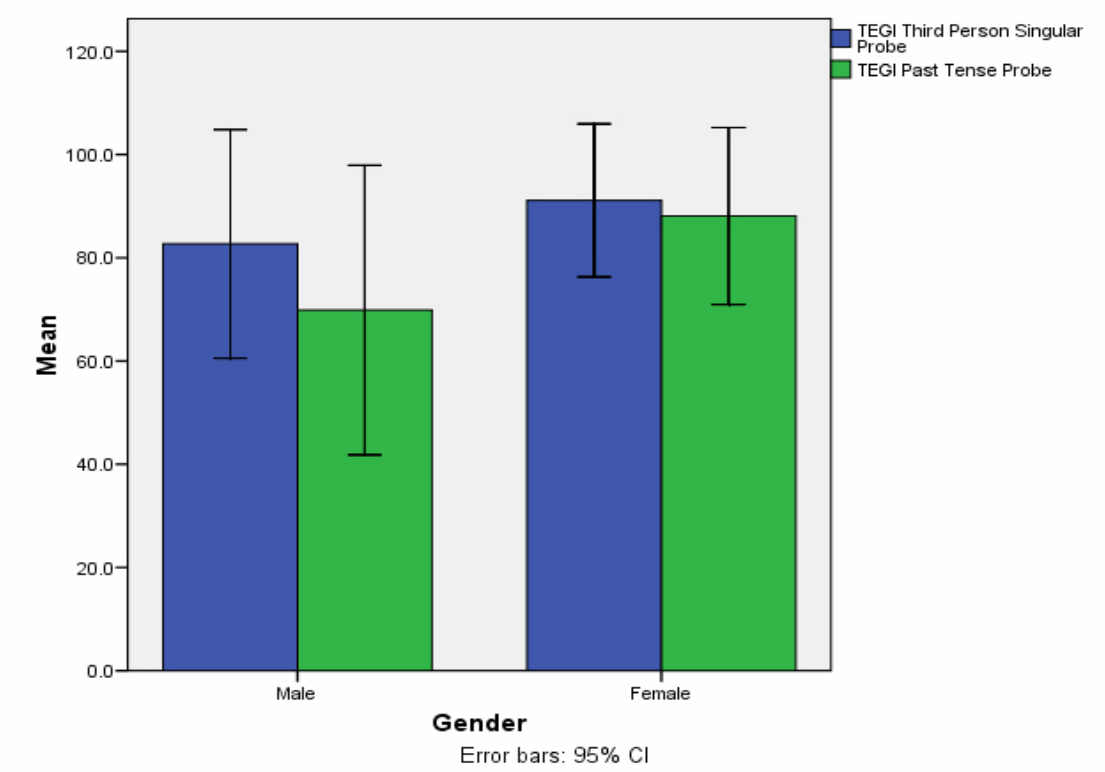
A series of independent sample *t* tests were also completed to examine gender differences on the dependent variables. The two groups were not significantly different on any of the measures of morphosyntax. Table 15 provides means and standard deviations on the dependent variables.

Table 15

Gender Differences on Measures of Morphosyntax

Variable	Boys n = 7	Girls n = 7
CELF Word Structures *t*(12)= -.860, *p* = .406, *d* = .460	*M* = 21.43 *SD = 7.44*	*M* = 25.00 *SD = 8.08*
TEGI Third Person Singular *t*(12)= -.773, *p* = .457, *d* = .413	*M* = 82.70 *SD = 23.95*	*M* = 91.11 *SD = 16.02*
TEGI Past Tense *t*(12)= -1.36, *p* = .206, *d* = .724	*M* = 69.89 *SD = 30.33*	*M* = 88.09 *SD = 18.54*
TEGI Regular Past Tense *t*(12)= -1.29, *p* = .224, *d* = .687	*M* = 64.13 *SD = 36.78*	*M* = 87.14 *SD = 29.84*
TEGI Irregular Past Tense *t*(12)= -.722, *p* = .485, *d* = .386	*M* = 63.01 *SD = 35.48*	*M* = 75.60 *SD = 29.50*
TEGI Irregular Past Finite *t*(9)= -1.25, *p* = .234, *d* = .670	*M* = 76.01 *SD = 27.01*	*M* = 90.47 *SD = 14.18*

Figure 6. Gender differences in two subscales from TEGI



PPVT-IV Groups

The boys in this study were also compared based on their PPVT-IV scores. Roberts et al. (2004) used a grouping criterion for their children with autism based on PPVT-IV standard scores. The children in the study were divided into three groups: normal language (PPVT-IV scores above 85), borderline language (PPVT-IV scores between 70-84), and finally impaired language (PPVT-IV scores below 70). In order to more closely compare the boys with FXS to the children from this study, the boys were divided into groups based on this same grouping criterion. Two boys scored in the normal range, 9 in the borderline, and 14 in the impaired group. Three of the boys in the borderline range had scores above 30 on the CARS, while 6 scored below 30.

Nine of the boys in the impaired group had scores above 30 on the CARS, while 5 scored below 30.

A series of univariate ANOVAs were completed to compare group differences on the dependent variables comparing PPVT-IV groups. There was a significant main effect for PPVT-IV group for the third person singular probe on the TEGI, $F(1, 22) = 8.51$, $p = .002$, $\eta_p^2 = .436$. Given that there were only two boys who scored in the normal group, they were excluded from all follow-up analyses. A *t* test revealed that the boys in the borderline group scored significantly higher on the third person singular probe, $t(21) = 3.34$, $p = .003$, $d = 1.49$ (borderline: $M = 75.10$, $SD = 23.26$; impaired: $M = 30.56$, $SD = 35.27$).

There was also a significant main effect for PPVT-IV group for the past tense probe on the TEGI, $F(1, 22) = 8.39$, $p = .002$, $\eta_p^2 = .433$. A follow-up *t* test indicated that the boys in the borderline group performed significantly better compared to the boys in the impaired group, $t(21) = 2.69$, $p = .014$, $d = 1.17$ (borderline: $M = 56.41$, $SD = 24.75$; impaired: $M = 25.49$, $SD = 28.15$). The other three subscales within the TEGI past tense probe yielded a significant main effect of PPVT-IV group with a similar finding in terms of the borderline group showing a distinct advantage over the impaired group. The regular past tense probe indicated a significant main effect of group, $F(1, 22) = 5.06$, $p = .016$, $\eta_p^2 = .315$. The follow-up test again indicated an advantage for the borderline group, $t(21) = 2.17$, $p = .042$, $d = .95$ (borderline: $M = 58.57$, $SD = 28.91$; impaired: $M = 27.86$, $SD = 35.53$).

The irregular past tense verbs yielded a significant main effect for PPVT-IV group, $F(1, 22) = 5.23$, $p = .014$, $\eta_p^2 = .322$. However, the follow-up tests indicated there was not a significant difference between the borderline and impaired groups, $t(21) = 1.58$, $p = .130$, $d = .65$ (borderline: $M = 39.89$, $SD = 33.40$; impaired: $M = 20.27$, $SD = 26.18$). The irregular finite composite on the past tense probe also yielded a main effect of PPVT-IV group, $F(1, 22) = 7.01$, $p = .004$, $\eta_p^2 = .389$. The follow-up *t* tests did indicate a significant difference between the borderline and impaired groups of boys, $t(21) = 2.20$, $p = .040$, $d = .92$ (borderline: $M = 51.59$, $SD = 32.67$; impaired: $M = 22.97$, $SD = 29.11$).

Discussion

The overall aim of this study was to examine morphosyntax in a group of children with FXS with an in depth focus on tense and agreement markers. The main research question was: *Is there a morphosyntactic deficit in fragile X syndrome?* It was hypothesized that children with FXS would exhibit a deficit in tense and agreement markers, and that this profile would look much like that of the children with autism and language impairments as reported in the Roberts et al. (2004) study. The results from the TEGI indicated that the boys with FXS did show a specific deficit in terms of third person singular and past tense marking. The overall mean for the boys in this study was 54% accuracy on the third person singular probe, and 42% accuracy on the past tense probe. Children in this age range should be performing at near-perfect accuracy.

Given that expressive language delays are well documented in the FXS literature (Abbeduto & Hagerman, 1997; Roberts et al., 2001) the most striking finding was the relative strength of receptive language within this sample of boys. The mean score for the boys was 65.46. Although this is in the delayed range, the sample included males within the normal range (scores between 37-88). Additionally, the mean age equivalent on the PPVT-IV was 5 years 9 months. Based on the standardized results from the TEGI, children at this age should be performing at near-perfect accuracy (Rice & Wexler, 2001). In other words, the deficit in morphosyntax would not be expected given the relatively good performance on the PPVT-IV, and

the general developmental age range they were performing at. Instead, the boys were performing well below what would be expected.

It was the case that the boys were inconsistently omitting tense and agreement markers. The boys with FXS-NA omitted the third person singular marker 17% of the time, but correctly marked tense and agreement 59% of the time. The boys with FXS-A omitted the third person singular marker 19% of the time, but used it correctly 30% of the time. In terms of the past tense probes, the boys with FXS-NA omitted the past tense marker 36% of the time for regular verbs (49% correct), and 29% of the time for irregular past tense (39% correct). The boys with FXS-A omitted the tense and agreement marker 27% of the time for regular verbs (26% correct), and 34% for irregular verbs (16% correct). Taking into account the large standard deviations associated with each of these variables, it is still noteworthy that the boys in this sample were displaying a pattern of optional deletions of tense and agreement markers for both third person singular and past tense. This mirrors what is reported in the SLI literature (Rice, Wexler, & Cleave, 1995; Rice & Wexler, 1996).

One issue to note is the wide range of scores in terms of scorable and unscorable responses on the TEGI probes (see tables 5-6 for means and standard deviations). The boys in this study did have a number of unscorable responses, or occasionally would not give a response. As noted previously, unscorable responses included verbs without overt tense markers (e.g., He *can* walk). The boys with FXS-A did not respond to the probes approximately 30% of the time on both the third person singular and past tense probes. Although they were able to give a number of

scorable responses, it is noteworthy that this is a different pattern than what is seen in typical development as well as language impairment without autism (Rice et al., 1995).

The boys with FXS seem to be in an extended optional infinitive stage of morphosyntax. The Extended Optional Infinitive account (EOI; Rice et al., 1995; Rice & Wexler, 1996) has been one of the main theories of SLI for the last ten years. It has its basis in Wexler's Optional Infinitive (OI) account (Wexler, 1994), which is based on children with normal language development. In normal development, children go through a phase where they treat tense and agreement marking as optional, although it is obligatory in adult grammar. The basic premise of EOI is that children with SLI seem to get "stuck" in an optional infinitive stage, whereby they follow the same basic course of morphosyntactic development as children with normal language abilities, but their transition out of the OI period is protracted (Rice et al., 1995; Rice & Wexler, 1996). It could be the case with FXS that the boys get stuck in this phase, without transitioning out, or perhaps for an even longer period of time. An examination of adult grammar in individuals with FXS would be necessary in order to examine this question.

Performance on the morphosyntactic variables was highly correlated with language comprehension scores. Given the wide range of scores on the PPVT-IV, this is not a surprising finding. However, nonverbal IQ was not correlated with any of the dependent measures. This complements the Roberts et al. (2004) study, as well as the work on children with low cognition but normal tense and agreement marking (Rice

et al., 2004; Rice et al., 2000); specifically, the literature has reported that there are a number of children with robust language systems, but impaired IQ. Despite the smaller sample size, the lack of an effect size indicates that even with more participants, nonverbal IQ is unlikely to be significantly correlated to performance on the measures of morphosyntax.

Comorbidity of autism

The impact of autism is a central issue in this study due to its very high comorbility with FXS in males. Consequently, one of the research questions was: *Does the presence of autism in FXS impact the development of morphosyntax?* The literature has reported that males with both FXS and autism are more affected cognitively and behaviorally than males with FXS only (Bailey et al., 1998). In terms of language comprehension and nonverbal IQ, this study does not support previous findings that boys with both FXS and autism were more impaired in terms of their communication, cognitive performance, and adaptive behaviors (Bailey et al., 1998; Bailey et al., 2000; Roberts et al., 2001; Turk & Graham, 1997). In the present study the boys with FXS-A were not significantly different in terms of their nonverbal IQ scores than the boys with FXS-NA. The effect size was .192, which is below what is considered a small effect, indicating that this finding was not due to a limited sample size. Although the two groups also did not differ on language comprehension, the effect size was medium, and the means indicate that the boys with FXS-A did score lower compared to the boys with FXS-NA.

Previous studies comparing the status of autism in FXS have not excluded males that are nonverbal (e.g., Bailey et al., 1998; Bailey et al., 2000; Cohen, 1995). However, individuals who were nonverbal were excluded from the present study since they would not have been able to complete the tasks for the dependent variables. Perhaps it was the nonverbal males with FXS-A who drove the significant findings in previous studies. In any case, it is noteworthy that among boys with FXS who are verbal, nonverbal IQ was not a differentiating factor in terms of their autism status.

In terms of the morphosyntax variables, the FXS-A group did not score significantly lower on the third person singular and past tense probes on the TEGI. However, both probes yielded a medium effect size of at least .70. On the past tense probe, there was a mean difference of 23 percentage points between the two groups. Although the findings were not significant (possibly due to the sample size), the proportional size of this differences suggests there may be meaningful differences between the two groups. In terms of the irregular verb composites on the past tense probes, the boys with FXS-A scored significantly lower compared to the boys with FXS-NA on the irregular past and irregular past finite verbs, with large effect sizes. The boys with FXS-A made more errors on the irregular past and irregular past finite verbs compared to the boys with FXS-NA. This group produced a bare stem 34% of the time for irregular verbs; yet instances of over-regularization were only 2% in this group. The boys with FXS-A overall showed a similar pattern of errors, just at a greater rate compared to the boys with FXS-NA.

In terms of autism group predictors, the discriminant function analysis indicated that the CELF, third person singular, and regular and irregular past tense finite probes from the TEGI taken together were good predictors of group membership. In other words, poorer performance on the dependent variables indicated membership in the autism group. It should be noted that girls with FXS were included in the analysis, due to the small sample size. Overall, these four predictors were able to classify 81% of cases correctly. The means and standard deviations, as well as the *t* tests demonstrated that boys with FXS-A do have a distinct weakness in terms of morphosyntax. The discriminant function analysis is particularly interesting relative to the finding that nonverbal IQ does not discriminate between boys with FXS-A and boys with FXS-NA (although previous studies have indicated it does discriminate between the two groups). Additionally, the language comprehension scores were not significantly different between the two groups, although this could change with an increase in sample size. However, the difference between the means was only 9 points (FXS-A: 60.58, FXS-NA: 69.64). Conversely, in terms of the measures of morphosyntax, the boys with FXS-NA typically had scores 20 percentage points higher than the boys with FXS-A.

Gender Differences

A secondary set of two research questions focused on gender differences in FXS. First, *Do females with FXS exhibit a deficit in morphosyntax?* Second, *do females with FXS exhibit delays in their receptive language, and/or nonverbal IQ?* The literature has reported that females are not as affected compared to males with

FXS (Hagerman & Sobesky, 1989; Bennetto & Pennington, 1996). Perhaps due to the X linked nature of the disorder, very few studies have been completed with females, particularly at the young ages. The test scores in this study for the seven girls showed a wide range of nonverbal IQ and language comprehension. The mean nonverbal IQ score was 76.17, compared to 51.46 for the boys. Scores below 70 are considered in the intellectual impairment range (APA; DSM-IV). The IQ scores for the seven girls were 52, 60, 79, 83, 83, and 100 indicating that two girls were intellectually impaired (one borderline).

The language comprehension mean score for the girls was at 89.43. Based on norms from the PPVT-IV, scores below 85 are considered in the disordered range (Dunn & Dunn, 2007). The PPVT-IV scores for the girls were 68, 76, 90, 95, 97, 98, and 102, again indicating that two females were within the disordered range. In terms of the dependent variables, the females, as expected, scored higher than the males on all of the subscales on the TEGI and the CELF. The mean scores on the TEGI were 91% and 88% for the third person singular and past tense probes. However, the standard deviations, much like the males with FXS, were large, 16.02 and 18.54 respectively. Additionally, the females did show impairment on the irregular past tense verbs, with a mean score of 75.60. Given the range of ages, the females should have been scoring at near perfect accuracy on all subscales. The irregular past tense verbs were a particular problem for the males in this study as well, regardless of autism status.

The comparison done between the full set of girls and subset of boys indicated that even when matched at the group level on language comprehension, the girls scored significantly higher on nonverbal IQ. Due to the fact that none of the girls in this study had CARS scores on the autism spectrum, only boys with CARS scores below 30 were selected for the comparison. However, the boys still scored significantly higher on the CARS and SRS measures compared to the females. The literature has reported that even when males with FXS do not qualify for a co-diagnosis of autism, behaviors concurrent with autism are typically reported (Bailey et al., 2000; Hatton, 2006; Rogers, 2001). This is consistent with the FXS-NA males who scored significantly higher on the CARS and SRS compared to the females, even though their total scores were below 30. Published studies to date on FXS and autism have not included females. It seems that females with FXS in this study, regardless of language abilities, do not exhibit the same types of autistic symptoms compared to males with FXS.

In terms of the morphosyntactic variables, there were not significant differences on any of the subscales between the boys and girls with FXS. It should be noted that there were medium effect sizes based on Cohen's definitions (medium = .5; Cohen, 1988) for the past tense probe, and within that probe specifically for regular past tense and irregular past finite verbs. The effect size was small for the irregular past tense verbs, d = .386. The females had the lowest mean scores on this subscale; most likely this is why the effect size was small. The CELF word structures subtest and the third person singular probe on the TEGI also yielded small effect sizes, but

they were both above .4 (CELF, d = .460; third person singular, d = .413). Although significant differences were not found between these two groups, this is probably due again to the small sample size. The effect sizes indicate that perhaps with a larger sample size, significant differences would have been found.

These findings are noteworthy in part because the girls were matched to the boys on language comprehension. Although their language comprehension scores were not significantly different, their morphosyntactic skills, appeared to be different, even if not statistically so. Additionally, their nonverbal IQ scores were quite different. This again supports the finding that language comprehension appears to be relatively intact in boys with FXS, particularly those with CARS scores below 30. Additionally, for this small sample, the boys displayed a higher degree of symptoms concurrent with autism compared to the females by both observer and parent report.

PPVT-IV Groups

The final secondary research question for this study was: *Do the children with FXS show the same pattern of impairments as the children in the Roberts et al. (2004) study using the same set of PPVT groups?* The Roberts et al. (2004) study divided the children with autism into three groups based on their PPVT-IV scores (e.g., normal, borderline, and impaired). In order to benchmark this sample within the literature, comparisons were performed based on the same PPVT groups as the Robert's study. Two males were excluded from this analysis because they scored within the normal range on the PPVT-IV. A series of ANOVAs indicated that the boys in the impaired group scored significantly lower compared to the boys in the borderline group on

third person singular and past tense probes. Within the past tense probe, the boys with impaired language scored significantly lower on the regular past tense verbs and the irregular past finite verbs, but were not significantly different on the irregular past tense composite.

This finding replicates in a different population the finding reported in the Roberts et al. paper. In fact, the boys with autism in the impaired group from the Roberts et al. study had a mean score of 36.8% correct on the third person singular probe, and the boys with autism in the borderline group were 61.3% correct. The boys with FXS in the impaired group had a mean score of 30.56%, while the boys with FXS in the borderline group had a mean score of 75.10%. The patterns are similar. In terms of the past tense probes, Roberts et al. reports means scores of 58.2% correct for the boys with autism and borderline language, and a mean of 30.6% for the impaired group. In the present study the mean score for the boys with FXS in the borderline group on the past tense probe was 56.41%, and the boys with FXS in the impaired language had a mean score of 25.49%. The pattern looks much the same, particularly for the boys with FXS in the impaired language group.

The poorer performance of the boys with FXS in the impaired language group cannot also be explained by the presence of autism. Nine boys with FXS were in the borderline language group; three of these boys had CARS scores above 30. Fourteen of the boys were in the impaired group; nine of them had CARS scores above 30. The two boys with scores in the normal range both had CARS scores below 30. Therefore it is not the case that the boys in the impaired group were also the boys with FXS-A.

Perhaps this is why there were not significant differences on PPVT-IV scores between the FXS-A and FXS-NA groups. The boys in the impaired group did show a distinct disadvantage compared to the boys in the borderline group on the dependent variables.

Roberts et al. (2004) suggests that perhaps there is a subset of children with autism who display an SLI-like language profile. Although expressive language is clearly delayed in this sample of boys with FXS, it does seem that the boys with lower language comprehension scores, with autism or with FXS, are displaying more errors on tense and agreement markers.

No relationship was found between nonverbal IQ and the dependent variables within this study, suggesting they are not related. Scores on both the CELF word structures subscale and the third person singular probe were significantly correlated with language comprehension scores in boys with FXS-A and FXS-NA. Nonverbal IQ was not significantly correlated with any of the dependent variables. Previous studies have indicated that nonverbal IQ is not correlated with performance in morphosyntax (Rice et al., 2004; Rice et al., 2000; Roberts et al., 2004). This study lends further support to the absence of a relationship between nonverbal IQ and morphosyntax.

Limitations

This study did have a small sample size, particularly in terms of the girls. A larger sample size would obviously allow for more fine tuned analyses. However, the sample size for both the boys and girls was *relatively* large for a study of FXS.

Additionally, a gold standard for diagnosing autism was not utilized (e.g., ADOS, ADI-R), although the CARS is frequently used in research studies (Bailey et al., 1998; Bailey et al., 2000) and at the very least is a valid measure of severity of autistic symptoms. An important difference in this study compared to previous work (Bailey et al., 1998; Bailey et al., 2000; Cohen, 1995) was the exclusion of males who are nonverbal. This could be viewed as both a limitation (in a comparative sense) and an improvement on previous work.

The language sample did not yield all the information hoped for. A conversation based language sample was employed, in lieu of a play based language sample. This type of language sample was selected, given the large age range of the participants (8-16 years). However, it was difficult to keep the children on task while the sample was being collected, and elicit more than a one- to two-word response. Many of the children also exhibited a high level of anxiety during the collection of the sample. Although the children often seemed to forget about the video camera during the testing, they were often preoccupied with it during the language sample. Additionally, some language samples had to be cut short, and thus did not yield the required number of utterances to calculate a valid MLU. In short, the language sample was less informative than the standardized tests.

The word structures subtest from the CELF also did not yield consistent results. The children in this study seemed to struggle with some of the tasks in terms of understanding what was being asked of them. Additionally, this subtest includes one to two items for different types of grammatical structures making it sometimes

difficult to decipher whether the children really did have a deficit or simply did not understand the question for the item. For instance, one question on the CELF focuses on derivation of adjectives. The teaching prompt is the word “dirty” and the test item is for “lucky”. The majority of the children did not need the teaching prompt for this question; they were able to give the correct answer without any training. However, they did not respond to the test item correctly; therefore they did not receive credit although they used the structure correctly on the teaching prompt.

Conclusions

The data from this study indicate that boys with FXS do have a specific deficit in morphosyntax, relative to language comprehension. By the age of 5, children should be performing at 90% accuracy minimum on both the third person singular and past tense probes (Rice & Wexler, 2001; Roberts et al., 2004). The boys in this study scored around the age of 5 years 8 months developmentally in terms of their receptive language skills. However, their performance on all dependent measures was well below 90%.

The presence of autism did have a negative impact on the dependent variables, although there was not a significant difference on all the different types of verbs (i.e., regular versus irregular). Specifically, boys with FXS-A show a greater deficit on past tense markers for irregular verbs compared to boys with FXS-NA. The means on all the variables from the *Test of Early Grammatical Impairment* demonstrate a clear advantage for the boys with FXS-NA. Nonverbal IQ was not statistically different

between the FXS-NA and the FXS-A groups, and the lack of a small effect size indicates that this would not be a significant finding even with a larger sample.

Females with FXS have been rarely studied to date. The few studies that have included females, with the exception of the recent Murphy and Abbeduto (2007) study, have focused on issues dealing with depression, social anxiety, etc. associated with higher levels of cognitive functioning. However, even with this very limited sample size, there were significant findings indicating a need to examine females with FXS more closely. It is clear that some of the females in this study have low nonverbal IQ, and receptive language scores. Additionally, irregular past tense verbs were a particular problem for this group of females.

Fragile X syndrome as noted earlier is a single gene disorder. Although there is still much to learn about how it impacts neural development, it is easier to determine the impact on neural functioning compared to disorders such as autism with unknown etiology. However, autism and FXS share a striking number of characteristics, including similar deficits in language and intellectual development. Studies such as the current one also indicate that autism is impacting development above and beyond FXS only. Drawing similarities between the two disorders, as well as examining the special impact that autism has on language and cognitive variables within FXS could help elucidate aspects of neural development that are specifically targeted by autism. This study although descriptive in nature, lays the groundwork for developing the language phenotype for FXS, as well as FXS and autism, which

provide the basis for future studies “why” the presence of autism has a unique impact on FXS.

This study extends the previous work on the morphosyntax abilities of children with intellectual disabilities by employing a more sophisticated set of measures. It is an extension of the work on children with autism and language delays, given that it asks the same questions, but with a different although related population of children. This study, as well as the Roberts et al. (2004) study, highlights the importance of refining the language phenotype of FXS and how language delays are expressed in FXS and autism.

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Appendix

Interview Protocol

Interview Protocol
Sterling Dissertation

Examiner begins with, "Now I'd like to talk with you for a few minutes. I am going to ask you a few questions about your family, school and stuff you like to do. Do you have any questions?"
Record language sample with video camera and tripod. Collect at least 15 minutes of data. Do not use yes/no questions during the language sample, and try to spend at least 5 minutes on each topic.

First topic: Family

1. "Tell me about your family."
    a. Prompts: What does your mother do? What does your father do? How many brothers and sisters do you have?

Second topic: School

2. "Let's talk about your school. Tell me about it."
    a. Prompts: What is your teacher like? Tell me about your favorite subjects? Tell me about your friends.

Third topic: After-school and leisure activities

3. "What types of things do you like to do when you are not in school?"
    a. Prompts: Tell me about your favorite television shows. Tell me about your favorite books. What do you like to do with your friends?

Final Question: "Is there anything else you want to tell me about?"

[1]Questions and Topics adopted from Evans and Craig (1992).