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Research Article

Examining the Language Phenotype in Children With Typical Development, Specific Language Impairment, and Fragile X Syndrome

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Purpose: One aspect of morphosyntax, finiteness marking, was compared in children with fragile X syndrome (FXS), specific language impairment (SLI), and typical development matched on mean length of utterance (MLU).
Method: Nineteen children with typical development (mean age = 3.3 years), 20 children with SLI (mean age = 4.9 years), and 17 boys with FXS (mean age = 11.9 years) completed the Test of Early Grammatical Impairment (TEGI; Rice & Wexler, 2001), and other cognitive and language assessments. Quantitative comparisons on finiteness marking and qualitative comparisons of unscorable (i.e., nontarget) TEGI responses were conducted.
Results: Children with typical development and FXS performed better on finiteness marking than children with SLI. Although unscorable responses were infrequent, boys with FXS produced more unscorable responses than children with typical development and SLI.
Conclusions: Although boys with FXS have language deficits, they performed similarly to MLU-matched typically developing children on finiteness marking. This language profile differs from children with SLI, who present with a delay-within-a-delay profile with finiteness marking delays that exceed delays in MLU. Unscorable responses produced by the boys with FXS may reflect pragmatic deficits, which are prominent in this population. Assessment procedures should be carefully considered when examining language in boys with FXS.

Recent research has been aimed at examining language phenotypes across developmental disorders with a focus on identifying overlapping profiles (Caselli, Monaco, Trasciani, & Vicari, 2008; Finestack, Sterling, & Abbeduto, 2013). This work has theoretical and clinical implications in that it informs the underlying language processes affected by developmental disorders. Comparisons across diagnostic groups have included disorders with known and unknown etiologies as well as those with and without intellectual disability, including fragile X syndrome (FXS), idiopathic autism spectrum disorder (ASD), Down syndrome (DS), and specific language impairment (SLI). For example, individuals with FXS share a similar behavioral phenotype with individuals who have idiopathic ASD (Bailey, Hatton, Skinner, & Mesibov, 2001), including difficulties with language. The language deficits seen in children with SLI have been documented in children with other concomitant disorders, including ASD and DS (Tomblin, 2011). Although some children with FXS have overlapping language phenotypes with children with ASD, to our knowledge, no previous study has directly compared language abilities in children with FXS to those with SLI. Therefore, the current study directly compares one area of language, finiteness, in children with FXS, SLI, and typical development (TD).

Finiteness marking is a notable area of language research that explores the interplay between grammatical morphology and syntactic structure (Schütze, 2004). Finiteness is a component of morphosyntax that marks verb tense agreement (e.g., third person singular *-s*, past tense *–ed*). Syntactic deficits, in particular deficits in finiteness marking, are a hallmark characteristic of SLI (Rice, Wexler, & Cleave, 1995). Emerging literature has identified similar finiteness deficits in children with FXS (Martin, Losh, Estigarribia, Sideris, & Roberts, 2013), yet it is currently unknown if the

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language deficits seen in children with FXS overlap with language deficits observed in children with SLI. The current study addresses this gap.

Specific Language Impairment and Fragile X Syndrome

Specific Language Impairment

Language deficits observed in SLI occur in the absence of hearing impairments, intellectual disability, or emotional or neurological impairments, and the etiology at present is unknown. The prevalence of SLI is roughly 7% among kindergarten children (Tomblin et al., 1997). Children with SLI persistently have lower mean length of utterance (MLU) than their age-matched peers across development (Rice et al., 2010), and difficulty with finiteness marking has been identified as a clinical marker of SLI (e.g., *He walk*, *She go*; Rice & Wexler, 1996). Delays in finiteness marking are also observed in the receptive domain (Rice, Wexler, & Redmond, 1999). Some children with SLI have persistent language deficits that extend into adolescence and adulthood (Rice, Hoffman, & Wexler, 2009), but difficulties with finiteness seem to diminish over time for many children. Rates of finiteness learning seem to be influenced by child characteristics, such as cognitive abilities and earlier language abilities (Rice, Tomblin, Richman, & Marquis, 2004). Although finiteness is the most consistently identified area of language difficulty in children with SLI, other linguistic and nonlinguistic skills can be impaired. For example, some children with SLI have deficits in phonological working memory evidenced by poor nonword repetition and word recall (Ellis Weismer, Evans, & Hesketh, 1999; Gathercole & Baddeley, 1990). Also, some have impairments in statistical learning with weakened ability to track statistical patterns in verbal and nonverbal input (Evans, Saffran, & Robe-Torres, 2009). It is important to note that Tomblin (2011) suggests that children with language impairment and other diagnoses (e.g., DS and ASD) may share language learning styles resulting in language impairments, motivating comparison studies across clinical groups.

Fragile X Syndrome

Children with FXS also have significant impairments in language, including finiteness marking (Sterling, Rice, & Warren, 2012). Fragile X syndrome is the leading inherited cause of intellectual disability. It is a single-gene X-linked disorder (Verkerk et al., 1991). Estimates of the prevalence range from a conservative one in 4,000 boys and one in 8,000 girls for the full mutation (Crawford, Acuña, & Sherman, 2001) to 1 in every 2,500 boys (Fernandez-Carvajal et al., 2009). Boys tend to be more affected than girls because of the X-linked nature of the disorder (Hagerman & Hagerman, 2002); therefore, we will focus solely on boys.

Individuals with FXS often have comorbid diagnoses. The majority of boys have moderate-to-severe intellectual disability (Kover, Pierpont, Kim, Brown, & Abbeduto, 2013; Skinner et al., 2005). Boys with FXS also have a high comorbidity with ASD (McDuffie et al., 2010). Also, many boys with FXS have difficulties with anxiety and social anxiety, which could affect pragmatic skills (Bailey et al., 2001).

As with SLI, deficits in finiteness have been documented in both receptive and expressive domains (Oakes, Kover, & Abbeduto, 2013; Sterling et al., 2012), and expressive syntax appears to be more delayed in FXS (J. E. Roberts, Mirrett, & Burchinal, 2001). Several studies have examined spontaneous morphosyntax and reported less complexity above and beyond what we would expect on the basis of mental age, thereby suggesting a deficit similar to that seen in SLI (Estigarribia, Roberts, Sideris, & Price, 2011; Finestack & Abbeduto, 2010; J. E. Roberts et al., 2007). Sterling et al. (2012) verified previous findings reporting expressive finiteness deficits in boys with FXS using a standardized assessment of expressive finiteness marking skills. Also, on the basis of receptive vocabulary skills, boys with FXS performed below expectations on both third-person singular and past tense finiteness markings. Correct productions of third-person singular and irregular past tense markings correlated with MLU and receptive vocabulary, and regular past tense forms correlated with receptive vocabulary only. Despite correlations between MLU and third-person singular and irregular past tense, Sterling et al. found that finiteness marking in boys with FXS exceeded expectations benchmarked to MLU (Rice et al., 2010; Sterling et al., 2012).

Comparisons Between Clinical Groups

There have been a number of studies comparing different clinical groups with a specific focus on identifying overlapping language phenotypes (e.g., Caselli et al., 2008; Finestack et al., 2013). The language profile seen in children with SLI has been directly compared with children with language impairments resulting from various etiologies, including children with idiopathic ASD who have comorbid language impairments as well as children with known genetic disorders, such as DS (Brock, Norbury, Einav, & Nation, 2008; Caselli et al., 2008). These comparisons are noteworthy given that the children with ASD and DS present with structural language deficits but also with a number of co-occurring deficits. For example, children with DS have intellectual disability and significant language impairments. Caselli et al. (2008) compared the grammatical language abilities of children with DS with children with SLI, matching the two groups on mental age. Children with DS and SLI share a similar profile of finiteness weakness and, in particular, a similar finiteness marking error pattern. Furthermore, Eadie, Fey, Douglas, and Parsons (2002) compared finiteness marking and morphosyntactic errors in children with DS and SLI who were matched on MLU. The children with DS demonstrated deficits in finiteness marking on a sentence imitation task, much like children with SLI. Although performance differed according to the specific finiteness marker, the authors concluded that profiles in finiteness marking overlapped significantly in children with SLI and DS.

In terms of other clinical populations, some children with idiopathic ASD show structural language impairments, including problems with finiteness (Kjelgaard & Tager-Flusberg, 2001; Riches, Loucas, Baird, Charman, & Simonoff, 2010). J. A. Roberts, Rice, and Tager-Flusberg (2004) examined finiteness marking in a large group of children with ASD with groups defined by receptive vocabulary scores. Children with impaired receptive vocabulary were significantly less accurate on a finiteness probe compared with children with borderline impairment. Although the children with ASD who scored in the normal range of receptive vocabulary had better performance compared with the other two groups, they still had significant deficits in finiteness marking, particularly in the past tense form. In comparison with published norms on SLI, the children with ASD with language impairments were comparable to 5-year-old children with SLI, which was on average 3–4 years younger than their chronological age.

Influence of Tools Used to Assess Syntactic Skills

Assessment Tools

Finiteness marking can be evaluated using standardized assessments as well as analyses from language samples. The assessment method used can influence children's response patterns and, therefore, our impressions of the severity of their language impairments (Finestack & Abbeduto, 2010; Kover, Davidson, Sindberg, & Ellis Weismer, 2014; Kover, McDuffie, Abbeduto, & Brown, 2012). There have been several studies on children with FXS using more gross measures, such as MLU, and microanalysis of morphosyntactic complexity measured in language samples (e.g., Index of Productive Syntax; Scarborough, 1990; e.g., Price, Roberts, Hennon, Berni, & Anderson, 2008; Developmental Sentence Scoring; Lee, 1974; e.g., Finestack & Abbeduto, 2010). However, a potential downfall of analyzing language samples is that the absence of a specific tense marker does not necessarily mean that a child lacks knowledge of that particular tense marker (Hadley, Rispoli, Holt, Fitzgerald, & Bahnsen, 2014). For example, a child who only produces the present tense in a language sample may still be able to produce past tense grammatical forms. Structured obligatory contexts are often necessary to observe specific grammatical tense forms, particularly in children with complex phenotypes.

Standardized measures of syntax, in contrast, create obligatory contexts in order to assess various target forms. Many of these assessments are omnibus measures that provide a limited number of opportunities on which to judge the child's knowledge of the target form (e.g., Clinical Evaluation of Language Fundamentals–Preschool; Wiig, Secord, & Semel, 2004). Having a limited number of test items per grammatical form can lead to the overestimation or underestimation of knowledge of a specific grammatical marker. The Test of Early Grammatical Impairment (TEGI; Rice & Wexler, 2001) directly targets particular finiteness markers in an obligatory context and provides several test items to allow for a more accurate picture of grammatical abilities. To be specific, the TEGI includes probes to assess the following English finiteness markers: third-person singular, past tense (regular and irregular), *be* auxiliary and copula, and *do* verbs. Only responses in which the child produces or attempts the target grammatical form are included in the correct or incorrect score, and other forms are categorized as unscorable.

Response Type

Although obligatory contexts provide targeted tests of knowledge in a particular language form, not all responses correspond to the item prompt. Responses that use nontarget forms are considered incorrect or unscorable. Treating a nontarget response as incorrect identifies limitations in a child's ability to produce the target form but yields a score that reflects responses in which the target form was not attempted. A large number of unscorable responses conversely can result in a subtest score that is based on very few items for which the target form was attempted. Such a score may provide a skewed picture of language skills. For instance, a child who produces only two responses that are scorable could receive a score of 100%, 50%, or 0% mastery of the specific finiteness marker. Given that the child failed to produce the target form for the majority of the test items in this example, the score can either greatly overestimate or underestimate the child's true abilities.

An evaluation of unscorable responses has the potential to provide valuable information that is otherwise disregarded, yet very few studies have considered them. J. A. Roberts et al. (2004) reported that children with ASD often produced responses that contained other verb tenses, such as present progressive *–ing*, echolalic responses, and omissions of verbs. Sterling et al. (2012) found that the modal verb was the most common type of unscorable response produced by boys with FXS during a third-person singular elicitation task (e.g., *He can run*). Unscorable responses produced in the other TEGI subtests were not discussed. It is more important to note that neither study compared unscorable responses produced by children with TD or SLI. Children with TD and SLI produce unscorable responses, but the forms that they produce may demonstrate a different compensatory strategy (e.g., progressive *–ing* vs. omissions of a verb). This has not been examined in the literature. The current study extends previous work by directly comparing the production of finiteness markers across children with FXS, SLI, and TD who were matched on MLU. We also characterized and compared unscorable responses across the three groups. Our research questions were the following:

1. Do children with FXS, SLI, and TD differ in finiteness marking?
2. Do children across the three groups differ in the percentage of unscorable responses produced in the TEGI?
3. Are there patterns of overlap within unscorable responses across the diagnostic groups and the TD group?

On the basis of descriptive findings presented by Sterling et al. (2012), we predicted that children with FXS and TD would perform significantly better on finiteness markers than MLU-matched children with SLI. Furthermore, we predicted that children with FXS would produce more unscorable responses than children with TD and SLI, possibly because of pragmatic weaknesses (J. A. Roberts et al., 2004). We also predicted that unscorable responses would be more frequent for those finiteness markers that are later acquired (e.g., *be* verbs are mastered later in development than third-person singular).

Methods

Participants

Fifty-six children participated (TD: n = 19, SLI: n = 20, FXS: n = 17). The children with TD and SLI participated in a larger study (Hoover, Storkel, & Rice, 2012). The children with FXS were a subsample of participants from a larger study examining finiteness marking. To be included in the current study, children had to be standard American English speakers, pass the phonological probe in the TEGI (which assessed word final /*s z t d*/—the sounds used to produce finiteness markers), and complete all three subtests on the TEGI. In addition, children with TD were required to score within the normal range on the TEGI subtests and have normal intellectual abilities. Children with SLI were required to have been previously identified as having language impairment by a speech-language pathologist or have delayed expressive grammatical performance relative to age expectations for both MLU and TEGI subtests and have normal intellectual abilities and normal hearing. All of the children with SLI performed below age expectations on morphosyntax, but, as indexed by vocabulary scores in Table 1, only some also had significant weaknesses in vocabulary knowledge.

The participants who met the inclusionary criteria were then selected on the basis of the MLU scores from a language sample. MLU-based comparisons are common in the SLI literature (e.g., Leonard, McGregor, & Allen, 1992; Rice, Buhr, & Nemeth, 1990). These comparisons not only control for important confounds, such as utterance length, but also allow researchers to systematically examine the specific pattern of overall grammatical development in children with language impairments (Rice, Redmond, & Hoffman, 2006). There were 36 boys with FXS in the larger study; of those children, eight could not complete all three subtests of the TEGI because of limited language abilities or because they did not understand the tasks, four did not pass the phonological probe, and one was a nonstandard American English speaker. Twenty-three children with FXS met the inclusionary criteria; however, after completing group-wise matching on MLU, only 17 were retained for the current study. All boys with FXS met criteria for an ASD diagnosis on the Autism Diagnostic Observation Schedule (Lord et al., 2012). One participant with TD was excluded in order to match the groups on MLU. Overall, participants with TD, SLI, and FXS were well matched on MLU, $F(2, 53) = 0.23$, $p = .796$, $\eta_p^2 = .01$. Groups also met matching criteria in follow-up pairwise comparisons with p values greater than 0.5 (Kover & Atwood, 2013; Mervis & Klein-Tasman, 2004; see Table 1 for participant characteristics).

Language and Cognitive Assessments

Two different assessments of nonverbal cognition were used. The children with TD and the children with SLI completed the Reynolds Intellectual Assessment Scale (Reynolds & Kamphaus, 2003). The children with FXS completed the Leiter International Performance Scale–Revised (Roid & Miller, 1997). Receptive vocabulary was assessed with the Peabody Picture Vocabulary Test–Fourth Edition (Dunn & Dunn, 2007). Children with TD and children with SLI completed a play-based language sample, and boys with FXS completed the conversation-based language sample, both of which were deemed to be age-appropriate tasks. In the 30-min play-based language sample, the examiner and child played with age-appropriate toys, and the examiner engaged in toy talk and provided the child with several opportunities to communicate (Hadley & Walsh, 2014). During the 10-min conversation sample, the examiner followed a semistructured list of conversation topics and used language elicitation techniques (e.g., open-ended questions, comments). Language samples were transcribed and coded following the Systematic Analysis of Language Transcript procedures (Miller, Andriacchi, & Nockerts, 2011) by trained and reliable undergraduate and graduate student coders. Only complete and intelligible utterances were used to calculate the morpheme-level MLU. Utterances were segmented according to c-units (number of complete and intelligible utterances: TD M = 206.3, SD = 84.4, range 24–354; SLI M = 208.1, SD = 70.1, range 40–363; FXS M = 142.3, SD = 25.8, range 96–189). For reliability coding, we randomly selected 20% of the children and had an independent trained coder in the lab retranscribe the language samples from the audio recording of the sample. Interjudge word agreement was 90% (SD = 4%) for the SLI group, 90% (SD = 3%) for the TD group, and 83% (SD = 9%) for the FXS group. Interjudge agreement for the presence or absence of bound morphemes was 89% (SD = 2%) for the SLI group, 89% (SD = 2%) for the TD group, and 81% (SD = 9%) for the FXS group. We also calculated Krippendorff's alpha values to assess inter-rater reliability; scores of one indicate perfect reliability, and scores of zero indicate an absence of reliability. Krippendorff's alpha for word agreement was TD = 0.999, SLI = 0.988, and FXS = 0.962. Krippendorff's alpha for bound morphemes was TD = 0.917, SLI = 0.994, and FXS = 0.861. Last, Krippendorff's alpha for MLU was TD = 0.946, SLI = 0.979, and FXS = 0.986.

TEGI

The TEGI (Rice & Wexler, 2001) is a clinical tool for the identification, screening, and diagnosis of grammatical

Table 1. Participant characteristics.

Characteristic	FXS *n* = 17 boys		SLI *n* = 20 (13 boys, seven girls)		TD *n* = 19 (9 boys, 10 girls)	
	Mean	(*SD*)	Mean	(*SD*)	Mean	(*SD*)
Chronological age (years)	11.9	(1.7)	4.9	(0.7)	3.3	(0.3)
Cognition (standard score)[a]	47.9	(7.4)	111.0	(16.1)	118.5	(13.9)
Mental age[a]	5.3	(0.7)	5.3	(1.1)	4.3	(0.7)
MLU	3.8	(1.1)	4.0	(0.7)	3.9	(0.8)
Receptive vocabulary (standard score)[b]	59.5	(15.5)	95.8	(11.2)	113.7	(10.9)
Receptive vocabulary (raw score)[b]	100.7	(23.5)	73.5	(20.4)	64.5	(13.0)
Third-person probe (% correct)[c]	82.2	(25.4)	36.5	(25.7)	50.8	(16.3)
Past tense probe (% correct)[c]	63.7	(24.8)	28.0	(20.1)	46.9	(17.5)
Be/do probe–be (% correct)[c]	82.6	(24.4)	50.1	(23.9)	77.3	(17.3)
Be/do probe–do (% correct)[c]	42.8	(34.2)	14.2	(19.3)	44.1	(32.0)
Child race			Frequency			
Caucasian		15	15		18	
African American		1	1		0	
More than one race		1	4		1	

Note. FXS = fragile X syndrome; SLI = specific language impairment; TD = typical development; MLU = mean length of utterance.
[a]Leiter International Performance Scale–Revised (Roid & Miller, 1997) or Reynolds Intellectual Assessment Scale (Reynolds & Kamphaus, 2003). [b]Peabody Picture Vocabulary Test–Fourth Edition (Dunn & Dunn, 2007). [c]Test of Early Grammatical Impairment (Rice & Wexler, 2001) subtest percentage correct scores exclude items with unscorable responses and no response, according to the manual.

deficits in children between the ages of 3 and 8 years. The TEGI examines finite verb morphology. It offers the benefit of having several test items for each of the targeted finiteness markers and allows for flexibility in the specific verb that is used for each item (e.g., *A firefighter helps people. He sprays water on the fire.*). It primarily has been used to examine language abilities in children with SLI (Hoover et al., 2012; Redmond, Thompson, & Goldstein, 2011); however, it also has been used with children who have impaired cognition or other developmental delays (Rice et al., 2004; J. A. Roberts et al., 2004; Sterling et al., 2012).

The TEGI consists of four subtests: third-person singular probe, past tense probe, be/do probe, and grammaticality judgment. The first three subtests provide a prompt that elicits a sentence containing an obligatory context for a given finiteness marker (e.g., *Here the girl is cleaning. Now she's done. What did she do?*). The last subtest is a receptive measure that requires children to judge if utterances provided by the examiner are grammatically correct. Given the emphasis of expressive finiteness productions indicated in our research questions, we only report performance on the third-person singular probe, the past tense probe, and the be/do probe.

The first three expressive subtests are scored on the basis of the subject–verb agreement for the target finiteness markers. Incorrect scores are assigned when finiteness markers are omitted (e.g., *She dance. Yesterday he play baseball. They happy?*). However, if a nontarget tense (e.g., third-person singular during the past tense probe) or if a nontarget subject is produced within a particular subtest (e.g., plural subject in the third-person singular probe), the response item becomes unscorable. The TEGI does not include unscorable responses in the calculation of finiteness tense accuracy and assigns these responses to a separate category. Subtests differ in the number of test items. Therefore, in order to compare performance across subtests, we calculated the percentage of responses that were correct, incorrect, unscorable, and no responses. For example, when calculating the percentage of correct responses, we divided the number of correct responses by the sum of correct, incorrect, unscorable, and no responses. These scores provided additional information compared with scores derived from the TEGI scoring manual, taking into account unscorable responses and no responses.

Unscorable Coding

Responses were first transcribed and then scored according to the TEGI manual. Afterward, the first and second authors gathered the unscorable responses for the FXS group and identified patterns and created eight descriptive categories to characterize unscorable responses. Unscorable responses were individually assigned a descriptive code (e.g., omission of verb, other verb form; see Table 4 for a full list). Each unscorable response also was judged on grammatical correctness. Item responses from the children in the TD and SLI groups were scored and assigned descriptive unscorable codes and grammaticality codes when appropriate. A secondary coder reviewed each transcript and identified disagreements. The primary coder reviewed the disagreements by checking the audio file once again, and the two coders assigned a consensus code.

Reliability

An independent coder assigned unscorable and grammaticality codes to responses that were scored as unscorable for 30% of the participants within each group (randomly selected). Percentage agreement was calculated for each response that was assigned an unscorable code and grammaticality code. Unscorable code agreement across the three subtests was 86.7%. Agreement for grammaticality codes was 96.0%.

Results

Finiteness Marking

Our first research question asked if children with TD, SLI, and FXS differed in finiteness marking abilities. Linear regression models compared group performance on the mean-centered percentage of correct responses and mean-centered percentage of incorrect responses for each TEGI subtest. Planned orthogonal contrasts followed our hypotheses of group performance. Regression analyses with planned orthogonal contrasts provide an efficient way to test hypotheses without requiring follow-up pairwise comparisons. The first contrast tested if children with FXS and TD produced a significantly different percentage of correct responses and incorrect responses than the children with SLI. The second compared responses between the children with TD and FXS. Figures 1 and 2 depict group performance.

Correct Responses

In the third-person singular probe, children with FXS and TD produced significantly higher percentages of correct responses than children with SLI (t = 4.704, p < .001). In addition, children with TD produced a significantly lower percentage of correct responses with the third-person singular finiteness form than the children with FXS (t = −3.429, p = .001). Group comparisons on the past tense probe responses revealed that children with FXS and TD produced significantly higher percentages of correct responses than children with SLI (t = 4.206, p < .001). Children with FXS and TD produced comparable percentages of correct past tense finiteness markers (t = −1.565, p = .123). Next, performance on *be* verbs (auxiliary and copula) in the be/do probe was compared across the groups. Children with FXS and TD had significantly better performance than the children with SLI (t = 4.174, p < .001). Also, performance did not differ between the children with FXS and TD (t = −0.245, p = .808). Last, performance on *do* items on the be/do probe were compared. Children with FXS and TD had significantly higher percentages of correct responses than children with SLI (t = 2.500, p = .016). Performance did not differ between children with FXS and TD (t = 1.556, p = .126; see Table 2).

Incorrect Responses

In the third-person singular probe, children with FXS and TD produced significantly lower percentages of incorrect responses than children with SLI (t = −6.455, p < .001). In addition, children with TD produced a significantly higher percentage of incorrect responses than children with FXS (t = 5.252, p < .001). In a similar manner, the regression model comparing the groups on the percentage of past tense probe incorrect responses revealed that children with FXS and TD produced significantly lower percentages of incorrect responses than children with SLI (t = −6.159, p < .001). In addition, children with TD produced a higher percentage of incorrect responses than children with FXS (t = 3.085, p = .003). Next, the percentage of incorrect responses on items targeting *be* verbs (auxiliary and copula) in the be/do probe were compared across the groups. Children with FXS and TD produced significantly lower percentages of incorrect responses than the children with SLI (t = −5.061, p < .001). In addition, the percentage of incorrect responses did not statistically differ between the children with FXS and TD (t = 1.598, p = .116). Last, performance on *do* items on the be/do probe were compared. Children with FXS and TD produced a significantly smaller percentage of incorrect responses than children with SLI (t = −7.202, p < .001). Performance did not differ between children with FXS and TD (t = 0.304, p = .762; see Table 3).

Figure 1. Group performance on the third-person singular probe and past tense probe.

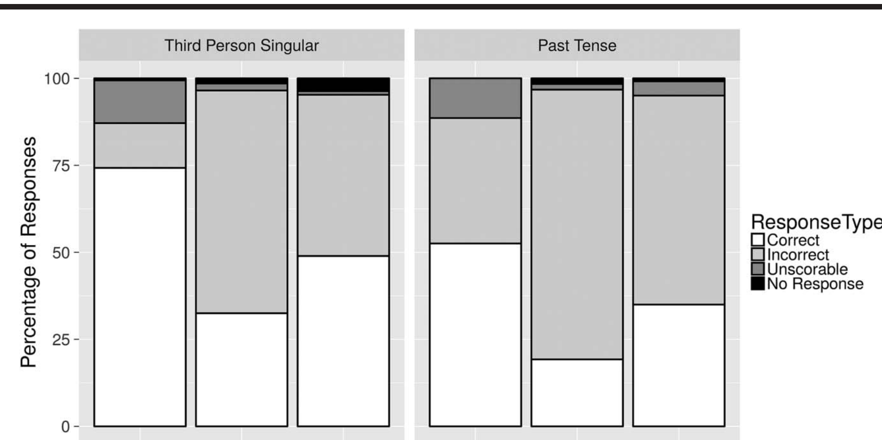
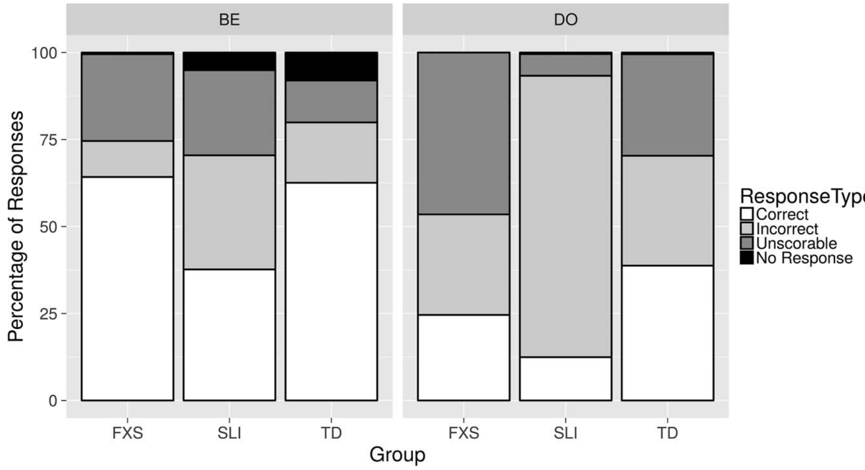


Figure 2. Group performance on the be/do probe.



Unscorable Responses

Group Comparisons of Unscorable Responses

The second research question asked if there were group differences in unscorable responses. Regression models tested for group differences in the mean-centered percentage of unscorable responses within each TEGI subtest. Planned orthogonal contrasts tested for group differences according to the hypotheses. The first contrast tested if the FXS group produced a significantly higher percentage of unscorable responses than the TD and SLI groups. The second contrast compared the percentage of unscorable responses between the TD and SLI groups.

The percentages of unscorable responses were first compared in the third-person singular probe. Children with TD and SLI produced significantly lower percentages of unscorable responses than children with FXS ($t = -3.135$, $p = .003$), and children with TD and SLI did not differ ($t = -0.250$, $p = .804$; total $R^2 = 15.7\%$). In the past tense probe, children with TD and SLI produced significantly lower percentages of unscorable responses than children with FXS ($t = -2.770$, $p = .008$). In addition, there were no significant differences between children with TD and SLI in the percentage of unscorable responses in the past tense probe (total $R^2 = 13.3\%$).

The percentages of unscorable responses on *be* verb items on the be/do probe were examined next. It was surprising that children with FXS did not statistically differ from children with TD and SLI. However, children with SLI produced a significantly higher percentage of unscorable responses than children with TD. To better understand group performance, we observed the group mean percentage of unscorable responses on *be* items. We gathered that children with FXS and SLI produced more unscorable responses, on average, than children with TD. We confirmed this observation by conducting a follow-up analysis with orthogonal comparisons testing if children with FXS and SLI produced significantly higher percentages of unscorable responses than children with TD and if children with FXS and SLI produced similar percentages of unscorable responses. Indeed, children with FXS and SLI produced significantly more unscorable responses than children with TD ($t = 2.631$, $p = .011$), and there were no significant differences between the children with FXS and SLI ($t = 0.055$, $p = .956$; total $R^2 = 11.6\%$). This pattern held after p values were corrected for this follow-up analysis.

Table 2. Group comparisons of correct finiteness marking.

Independent Variables	Third-person singular probe				Past tense probe				Be: Be/do probe				Do: Be/do probe			
	β	*SE*	*t*	*p*	β	*SE*	*t*	*p*	β	*SE*	*t*	*p*	β	*SE*	*t*	*p*
Intercept	.00	2.98	0.39	.701	.00	3.04	0.22	.782	.00	2.97	0.22	.806	.00	3.70	0.06	.957
FXS and TD versus SLI	.51	6.20	4.70	< .001	.49	6.34	4.21	< .001	.50	6.19	4.17	< .001	.37	7.71	2.50	.016
FXS versus TD	–.37	7.42	–3.43	.001	–.18	7.58	–1.57	.123	–.03	7.41	–0.25	.808	.20	9.22	1.56	.126
		$R^2 = .383$				$R^2 = .271$				$R^2 = .248$				$R^2 = .144$		

Note. FXS = fragile X syndrome; TD = typical development; SLI = specific language impairment.

Table 3. Group comparisons of incorrect finiteness marking.

Independent Variables	Third-person singular probe				Past tense probe				Be: Be/do probe				Do: Be/do probe			
	β	*SE*	*t*	*p*	β	*SE*	*t*	*p*	β	*SE*	*t*	*p*	β	*SE*	*t*	*p*
Intercept	.00	2.56	−0.55	.582	.00	2.55	−0.44	.660	.00	1.79	−.32	.749	.00	3.38	−0.37	.712
FXS and TD versus SLI	−.59	5.34	−6.46	< .001	−.62	5.31	−6.16	< .001	−.56	3.72	−5.06	< .001	−.70	7.03	−7.20	< .001
FXS versus TD	.48	6.39	5.25	< .001	.31	6.36	3.09	.003	.18	4.45	1.60	.116	.03	8.41	0.30	.762
	R^2 = .559				R^2 = .466				R^2 = .343				R^2 = .495			

Note. FXS = fragile X syndrome; TD = typical development; SLI = specific language impairment.

Last, the percentages of unscorable responses on *do* items on the be/do probe were assessed. Children with FXS had significantly higher percentage of unscorable responses than children with SLI and TD ($t = -2.917$, $p = .005$). Also, children with TD produced a significantly greater percentage of unscorable responses than children with SLI ($t = 2.101$, $p = .040$). Observation of group means led us to understand that children with TD produced a fairly large percentage of unscorable responses for *do* items, but it was unclear if the children with FXS produced a significantly higher percentage of unscorable responses. To test this, we conducted a follow-up analysis with orthogonal comparisons testing if children with FXS and TD produced significantly higher percentages of unscorable responses than children with SLI and if children with FXS and TD produced similar percentages of unscorable responses. As expected, children with FXS and TD produced significantly higher percentages of unscorable responses than children with SLI ($t = 3.493$, $p = .002$). However, differences between children with FXS and TD did not reach significance ($t = -1.531$, $p = .132$; total R^2 = 19.8%). This pattern persisted after correcting *p* values for multiple comparisons.

Unscorable Characterizations

To address our third research question, we completed descriptive comparisons and characterized the unscorable responses produced by each group. Table 4 provides a complete list of the types of unscorable responses the groups produced. In the third-person singular probe, the FXS group produced two categories of unscorable responses. They primarily produced responses that contained verb tenses other than the third-person singular form; the most common nontarget grammatical forms were present progressive *do* and *be* verb forms. Children with SLI produced unscorable responses with other verb forms, including primarily present progressive verb forms. Children with TD produced other verb forms in unscorable responses that contained *do* verb forms.

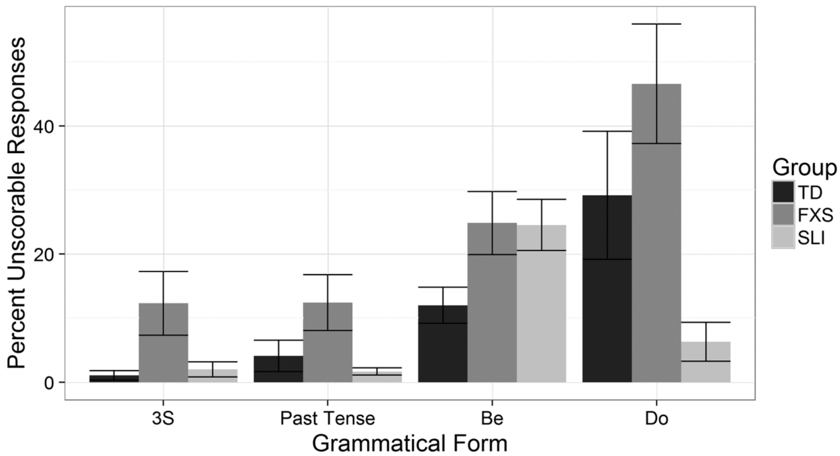
In the past tense probe, children with FXS primarily produced other verb forms, such as third-person singular and *be* verbs. In addition, the FXS group omitted verbs

Table 4. Percentage of unscorable response types.

Unscorable code	Example	FXS	SLI	TD
			Third-person singular probe	
Other verb tense	*He is playing.*	61.9	100	100
Omission of verb	*She hat. He running.*	38.1		
			Past tense probe	
Other verb tense	*She digs. He will eat.*	81.6	66.7	100
Omission of verb	*He going. He fast.*	15.8	16.7	
Subject error	**He ran.*		16.7	
Unintelligible	*He X cookie. XXX.*	2.6		
			Be/do probe	
Other verb tense	*He cried. She jumps.*	17.1	13.5	60.2
Omission of verb	**Do they want some?*	21.3	39.1	14.4
Transpose	*They are tired?*	9.8	29.7	19.5
Subject error	*Are you hungry?*	39.4	6.5	0.6
Yes/no response	*No. Yes.*	2.6	6.5	2.5
Echo	*Ask the puppet.*	7.3	1.5	0.9
Unintelligible	*They X hungry.*	2.1	0.7	1.7
Wh-question word	*Why they like milk?*	0.5		

Note. FXS = fragile X syndrome; SLI = specific language impairment; TD = typical development; * = omitted word; X = unintelligible word or set of words.

Figure 3. Percentage of unscorable responses across grammatical forms. Error bars depict standard error from the mean.



and produced unintelligible responses. The unscorable responses produced in the SLI group primarily contained other verb forms, such as *be* verbs. Unscorable responses also contained omissions of a verb and subject errors in the SLI group. Last, the TD group produced other verb tense forms such as third-person singular.

The greatest variety of unscorable responses was produced in the be/do probe. The most frequent unscorable responses produced by the children with FXS included responses with subject errors, omitted verbs, and other verb tenses. The SLI group most often produced responses with an omitted verb. In addition, many children with SLI produced transposed responses. For example, instead of asking, "Is the bug hungry?" a child said, "The bug is hungry?" Children with TD also produced several transposed responses as well as other verb tenses, such as present tense and modal forms.

As depicted in Figure 3, the percentage of unscorable responses generally increased across the grammatical forms. An exception was seen in the SLI group, who produced relatively few unscorable responses for *do* items. An additional approach to examining child productions of unscorable responses across the grammatical forms is to identify the number of children who produced at least one unscorable response within each subtest. As predicted, an increasing number of children who produced at least one unscorable response was observed across the three subtests. Within the FXS group, the following percentages of children produced at least one unscorable response: 41% third-person singular probe, 65% past tense probe, 100% be/do probe. The same pattern was true for the TD group: 11% third-person singular probe, 21% past tense probe, 89% be/do probe. Last, more children with SLI produced unscorable responses in the be/do probe (95%) than the first two probes (30%).

In addition to characterizing the types of unscorable responses, we coded the grammaticality of unscorable responses. The TD group produced the highest percentage of grammatical unscorable responses across the three TEGI subtests. More than half of the unscorable responses were grammatically correct (100% third-person singular probe, 85.7% past tense probe, 61.9% be/do probe). Greater variability in the FXS group was seen with percentage of grammatical correctness of 16.7% in the third-person singular probe, 73% in the past tense probe, and 45.1% in the be/do probe. Last, the SLI group produced grammatical responses 50% of the time or less (45.5% in the third-person singular probe, 50% in the past tense probe, and 20.9% in the be/do probe).

Discussion

In the current study, we directly compared finiteness marking in children with FXS, SLI, and typical development who were matched on MLU. To our knowledge, this comparison has not yet been examined in the literature despite the fact that children with FXS have deficits in finiteness marking. In addition, we examined nontarget responses that were categorized as unscorable according to the TEGI manual.

Finiteness Marking

Analyses of the percentage of correct responses produced in the TEGI subtests revealed that children with FXS and TD produced more correct responses than children with SLI. Previous studies have found that children with SLI often perform more poorly on finiteness marking when compared with children with TD who are matched on MLU (e.g., Hoover et al., 2012; Leonard, Eyer, Bedore, & Grela, 1997). It is interesting to note that children with FXS performed similarly to children with TD and even produced a significantly higher percentage of correct third-person singular productions. This is not to say that children with FXS do not have deficits in finiteness marking. In fact, an observation-based comparison of normative scores in the TEGI manual revealed that the boys with FXS, on average, performed below nonverbal mental age expectations on all finiteness markers (Rice & Wexler, 2001, p. 65). Previous studies also have found that boys with FXS have delayed expressive language, including grammatical complexity, relative to nonverbal mental age expectations (Estigarribia et al., 2011; Price et al., 2008; J. E. Roberts et al., 2007). However, relative to MLU expectations, boys with FXS performed similarly to younger children with TD and consistently outperformed the MLU-matched children with SLI. Our data indicate that boys with FXS may not demonstrate the delay-within-a-delay profile described in the literature on children with SLI (Rice, 2003) but instead demonstrate a pattern of strengths and weaknesses depending on the measurement tool. As such, especially in children with FXS, the description of language abilities may differ according to both the language variable (e.g., vocabulary, syntax) and assessment method (e.g., language sample, standardized assessment) used to benchmark language skills.

We chose MLU for the group matching measure because it is a highly accepted linguistic benchmark in the SLI literature (e.g., Leonard et al., 1997; Rice, Wexler, & Hershberger, 1998). It is interesting to note that we did not find deficits in finiteness marking beyond MLU expectations in our group of boys with FXS. This may be because MLU is significantly delayed in boys with FXS (Finestack et al., 2013; Sterling et al., 2012). MLU may be particularly low because it is typically obtained through spontaneous language samples acquired through interpersonal tasks and therefore influenced by social linguistic deficits (Bailey et al., 2001; Finestack et al., 2013). It is noteworthy though that the boys with FXS performed similarly to the group of children with TD on the norm-referenced measure of finiteness marking. The results highlight the importance of using multiple contexts to study language, particularly in disorders with complex behavioral phenotypes, such as FXS. The reliance of measures derived solely from language samples, such as MLU may underestimate the grammatical profile seen in FXS. Performance on finiteness marking may differ according to the assessment tool (e.g., language sample vs. standardized assessment). This is not the case in SLI, with which MLU and performance on the norm-referenced tasks are often corroborative in identifying language impairment.

We also analyzed the percentage of incorrect responses produced on the basis of the full range of response types noted in the TEGI: correct, incorrect, unscorable, and no response. Given our interest in understanding the types of responses and the potential compensatory strategies children might be using, we examined the full range of response types and identified patterns across groups. For example, children might produce similar percentages of correct productions but differ in incorrect or unscorable responses. This was the case for the past tense probe. Children with FXS and TD produced similar percentages of correct responses; however, children with FXS produced fewer incorrect responses and more unscorable responses than children with TD. Beyond this example, patterns of correct and incorrect productions were similar, suggesting that finiteness marking abilities may be similar in FXS and MLU-matched children with TD.

Unscorable Responses

The current study expanded upon previous literature by examining unscorable responses. In general, children produced responses that were correct or incorrect, and the no response code was almost never assigned. Given the number of children in each group producing unscorable responses, we were interested in examining the type, frequency, and grammaticality of each response. We hypothesized that boys with FXS would produce more unscorable responses than children in the TD and SLI groups, and this was confirmed with the data. Why though, did children with FXS produce more unscorable responses? One possibility is that it reflects a lack of task understanding. This is unlikely though given that the majority of responses to task items were correct or incorrect, demonstrating that children with FXS attempted targeted finiteness markers the majority of the time. As an alternative, unscorable responses may reflect attentional deficits, which are often noted in children with FXS (Bailey, Raspa, Olmsted, & Holiday, 2008). Difficulty sustaining attention could have resulted in children with FXS inconsistently providing responses with the target subject and finiteness form.

Another potential explanation is that unscorable responses may be an outcome of pragmatic deficits (see J. A. Roberts et al., 2004, for a similar hypothesis for ASD). The pragmatic deficits in FXS (Klusek, Martin, & Losh, 2014) may have hindered their ability to flexibly adapt to the linguistic contexts unique to each subtest on the TEGI and, as a result, to produce more tangential responses. The boys with FXS produced a significant percentage of unscorable responses for *do* items in the be/do probe. *Do* items consisted of targeted questions (e.g., *Does the bug want milk?*); several *be* items were intended to be questions, but others were statements. Many boys with FXS produced subject errors (e.g., *Do you want milk?*). It is possible that the boys with FXS struggled to understand the social nature of the task, resulting in responses that contained an incorrect subject and possibly a nontargeted verb form.

The use of unscorable responses may also reflect a compensatory strategy for children who have not yet mastered the targeted structures. If this hypothesis is true, we may expect unscorable responses to increase in frequency for later acquired finiteness markers. In addition, unscorable responses would most likely be grammatically correct. This generally was seen in unscorable responses from the TD group. The children with FXS and SLI were using more unscorable responses in the be/do probe compared with the third-person singular and past tense probes. However, the grammaticality of the responses varied. Unscorable responses for the past tense probe in the FXS group were mostly grammatically correct. The majority of these unscorable responses contained third-person singular finiteness forms.

It is possible that this particular pattern may indicate a unique compensatory strategy with the children using perseverative language. The past tense probe directly follows the third-person singular probe, and so it is not surprising that children defaulted to a third-person singular tense response. This seems to be most likely in the FXS group because previous literature has documented perseverative language in children with FXS, similar to patterns in ASD (J. A. Roberts et al., 2004). It is also possible that the children with FXS frequently produced *you* as the subject within the be/do probe as a compensatory strategy to manage the complex demands of switching subjects (*he* vs. *they*) throughout the subtest.

Although less common, children with TD and SLI also produced unscorable responses. Children with SLI produced a particularly high percentage of unscorable responses for *be* items on the be/do probe. Most often, they omitted verbs or produced transposed question forms (e.g., *He is eating? He is hungry?*), which indicates that most problems stemmed from question forms. In contrast, a large number of unscorable responses occurred for *do* questions in the TD group. Unscorable responses most often contained another verb tense or transposed forms. Given that questions are acquired later than statements, it is possible that children had insufficient knowledge of question forms, leading to transposed forms. Also, the high percentage of incorrect responses and unscorable responses with omitted verbs is likely to at least partially reflect the extended optional infinitive (Rice et al., 1995) and the optional infinitive stages (Wexler, 1994). In line with extended optional infinitive/optional infinitive theory, children may have treated *be* and *do* verbs in question forms as optional, leading to responses such as *He hungry?* In cases in which *be* and *do* were omitted, children often used rising intonations to mark question forms, indicating emerging understanding of question forms.

Study Limitations

The present study was limited in several respects. First, the participants were not gender matched. Girls with FXS present differently than boys with FXS (Bailey et al., 2008; Finestack & Abbeduto, 2010; Hagerman & Hagerman, 2002). To avoid mixed findings on the basis of genetic reasons, we chose to only include boys with FXS. In contrast, SLI is not an X-linked disorder, and gender differences are not as extreme. Although boys tend to have higher rates of SLI when using direct assessment (Whitehouse, 2010), most studies include boys and girls with SLI (e.g., Leonard et al., 1992; Rice et al., 2009, 1998). Taken together with gender differences in TD, it will be important for future studies to more evenly match participants on gender. Another limitation was difference in language sampling tasks. The language sampling technique was selected on the basis of the appropriateness for chronological age: play-based for TD and SLI and conversation-based for FXS. It is important to note that the language samples followed a conversation style in both the play- and conversation-based language sampling tasks. Although clinically recommended language elicitation strategies were used with all participants, it is possible that differences between a conversation-based and play-based context may have influenced expressive language production. For example, there may be more opportunities in conversation compared with play for advanced syntax (Evans & Craig, 1992). Future studies should use the same language sampling method and may wish to base MLU off of the same number of utterances or length of language sample across participants to avoid potential problems with generalizability of findings.

Clinical Implications

Findings from this study indicate that clinicians should be aware that assessment context can influence child language performance. Also, observation of nontarget or unscorable responses may provide clinically meaningful information about expressive language skills and compensatory strategies.

Conclusion

The current study builds upon previous work by Sterling et al. (2012) by examining *be* and *do* forms in FXS and, to our knowledge, is the first comparison of SLI and FXS in terms of finiteness marking abilities. We found that boys with FXS do not demonstrate deficits in finiteness marking relative to MLU benchmarks. This profile differs from the delay-within-a-delay profile observed in children with SLI. Furthermore, although it occurred infrequently relative to correct responses, on average, boys with FXS produced significantly more unscorable responses than children with typical development and SLI, possibly reflecting the effect of pragmatic language deficits on the validity of assessment measures. Future work should continue to examine areas of overlap and distinction in language phenotypes in children with language impairment.

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