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Narrative Analysis in Adolescents with Fragile X Syndrome

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Abstract

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Introduction

Fragile x syndrome (FXS) is caused by a trinucleotide repetitive sequence on $(CGG)_n$ of the fragile X mental retardation 1 gene (*FMR1*) (Verkerk et al., 1991). Individuals with more than 200 CGG repeats are considered to have full mutation FXS (Imbert & Mandel, 1995; Loesch, Huggins, & Hagerman, 2004). FXS is the leading known inherited cause of intellectual disability (Crawford, Acuña, & Sherman, 2001). Delays in language development can first be observed in male children with FXS at 9 months of age and continue through adolescence (Roberts et al., 2009).

While individuals with FXS are generally delayed in their development, there is still wide variety in the presentation of language and social skills in this population. This variability is due to both differences in sex (Imbert & Mandel, 1995) and methylation status of the *FMR1* gene (Wright-Talamante et al., 1996). As this is an X-chromosome linked syndrome, males tend to have more significant deficits compared to females (de Vries et al., 1996).

Males with FXS generally have cognitive deficits that can be severe and increase overtime (Freund, Peebles, Aylward, & Reiss, 1995). However, cognitive ability can range in males with FXS depending on the degree of methylation of the *FMR1* gene. Average IQs range from 46.9-80.5 in males (Wright-Talamante et al., 1996). Behavioral differences frequently observed in males with FXS include increased sensitivity to sensory stimuli, a reduction in attention and increased hyperactivity. A secondary diagnosis of autism spectrum disorders occurs in a substantial proportion of males, and even those without a diagnosis tend to display some symptoms of autism such as stereotypic behavior and perseveration (Cohen et al., 1991; Demark, Feldman, & Holden, 2003; Roberts et al., 2016).

Females with FXS typically have one X chromosome with the full genetic mutation and an unaffected X chromosome that acts as a protective factor and lessens the impact of the *FMR1* gene (Imbert & Mandel, 1995). A large group of females with FXS present with IQs in the normal to borderline range. Deficits are still present in this population in the form of specific learning difficulties and deficits in executive functioning, including reduced attention and increased impulsivity (Mazzocco, Pennington, & Hagerman, 1993). Social difficulties and elevated levels of anxiety are also often present, possibly leading to limited use of social language (Lesniak-Karpiak, Mazzocco, & Ross, 2003). This study analyzed the impact of structural language, cognition, and autism symptomatology on the narrative skills of male and female adolescents with FXS and the association between narratives and literacy in this population.

Narrative Association with Language and Cognition

Narratives can be analyzed on two aspects of language, the narrative macrostructure and the narrative microstructure (Kintsch & van Dijk, 1978). Macrostructure analysis measures the overall organization of the story and the inclusion and frequency of necessary story elements that create a narrative. Microstructure analysis measures the language structure used within the story including the diversity of vocabulary used and the syntactic structures employed (Liles, Duffy, Merritt, & Purcell, 1995). In the current investigation, we analyzed the macrostructure because we were interested in the children's ability to organize events systematically through a narrative structure.

Many typically developing children begin to tell simple stories as early as 2 years old (Applebee, 1978). Children start by labeling and describing a series of unrelated actions (Stadler & Ward, 2005). During the preschool years, stories progress to statements around a central topic

and events are linked together into story episodes at ages 3-4 (Khan et al., 2016). As children's narrative skills advance, they connect story events with perceptual links followed by temporally-linked statements (Stadler & Ward, 2005). In later childhood, overall plots and character goals are used along with character motivations and thoughts (Khan et al., 2016).

The production of a coherent and complex narrative requires the use of more complex language than conversational speech (Petersen, 2011). The narrator must put the story in context, use descriptive language to depict the events of the story and connect the events with casual language. While advanced semantic and morphosyntactic skills aid in the development of complex narrative macrostructure (Stadler & Ward, 2005), the macrostructure and microstructure of the story are different constructs (Liles et al., 1995). In analyzing children with learning disabilities Feagans & Appelbaum (1986) found distinct groups of children based on language scores. Some groups' narrative macrostructure exceeded their semantic and syntactic skills while others had the opposite profile (Feagans & Appelbaum, 1986). This demonstrates that while macrostructure and microstructure are both measures of oral language use, there are other factors that contribute to narrative development beyond morphosyntactic and semantic skills.

Cognitive skills such as executive functioning (Friend & Bates, 2014), theory of mind (Kim, 2016), and working memory (Montgomery, Polunenko, & Marinellie, 2009; Nordberg, Dahlgren, & Miniscalco, 2015) have been associated with children's abilities to understand and produce narratives. Executive functioning skills and narrative abilities support each other's development between ages 4 and 5 as the child's narratives are becoming more complex and connected (Friend & Bates, 2014). The narrator needs to hold multiple events in their mind and plan the order in which to tell the story so the story is logical and events are connected (Westby, 1999). Theory of mind, defined as the ability to understand that a person has their own thoughts

and emotions that differ from one's own (Losh et al., 2012), is needed to decide what information the listener needs to hear to understand the story. Theory of mind can also help when a narrator is generating the thoughts and feelings of characters in the story (Applebee, 1978).

Narratives and Literacy

The ability to understand and tell a cohesive story serves as a key transition between oral and written language (Westby, 1999). Strong narrative skills have predicted literacy outcomes in typically developing children (Reese, Suggate, Long, & Schaughency, 2010), children with learning disabilities (Feagans & Appelbaum, 1986), reading disabilities (Feagans & Short, 1984; Roth & Speece, 1996) and co-occurring speech sound and language disorders (Wellman et al., 2011).

For example, Feagans & Short (1984) found that children with reading disorders demonstrated poorer performance on measures of narrative content and complexity in generated narratives compared to their peers who read at grade level. This relationship between narratives and reading is also present at the level of word decoding. Narrative macrostructure predicted real word decoding in children with speech sound disorders (Wellman et al., 2011). In a group of typically developing school-age children, oral narrative quality predicted later reading fluency, even after controlling for early literacy skills (Reese et al., 2010). Wellman and colleagues (2011) found the same relationship in children with co-occurring speech sound disorders and language impairment.

Narratives in Fragile X Syndrome

Language is highly variable in individuals with FXS, with some individuals remaining nonverbal and others showing advanced language skills (Abbeduto, Brady, & Kover, 2007). Narrative analysis may help us to better understand the functional language use and literacy

abilities of individuals with FXS. Previous studies of narrative macrostructure analysis in individuals with FXS have included children and adolescents with mean ages ranging from 9.73 years (Hogan-Brown, Losh, Martin, & Mueffelmann, 2013) to 16.68 years (Keller-Bell & Abbeduto, 2007). Despite this range in age, narrative macrostructure scores have been found to match typically developing peers matched on mental age in most studies (Hogan-Brown et al., 2013; Keller-Bell & Abbeduto, 2007). Furthermore, only one study has included females with FXS and no significant difference was found based on sex. The authors attributed this to the small sample size and called for further investigation (Keller-Bell & Abbeduto, 2007).

Based on the narrative story structure (NSS) scoring rubric from SALT, Finestack, Palmer & Abbeduto (2012) found individuals with FXS macrostructure scores most closely matched those of their typically developing peers when matched on MLU than on mental age. However other studies used detailed frequency-based scoring system and found that the narrative macrostructure of individuals with FXS closely matched the macrostructure of typically developing children with the same mental age (Hogan-Brown et al., 2013; Keller-Bell & Abbeduto, 2007).

These discrepant findings are thought provoking. Language and cognition have been found to be closely linked (Price, Roberts, Vandergrift, & Martin, 2007; Rice, Warren, & Betz, 2005; Roberts, Mirrett, & Burchinal, 2001); hence one would expect narrative skills to be linked to both cognition and language measures. Most individuals with FXS have delays in both cognitive and language skills but identifying the degree to which narratives relate to measured cognitive and language skills could provide guidance for intervention. For example, if language skills appear to be driving narrative performance, interventions that target underlying language structures could benefit narratives. In contrast, finding that cognitive skills appear to be more

related to narrative outcomes would suggest alternative interventions such as those focused on sequencing of events or theory of mind may have more impact on narratives.

Impact of ASD

Individuals with ASD also have difficulties in generating organized narratives with appropriate referents and use of mental state words (Baixauli, Colomer, Rosello, & Miranda, 2016). Therefore, it is likely that individuals with FXS and many characteristics of ASD (FXS-ASD) may be additionally disadvantaged compared to their peers with FXS only (FXS-O). In a previous study of narrative retell abilities of individuals with FXS-O and FXS-ASD, no significant differences were found between the two groups. However, boys with FXS-ASD demonstrated significantly poorer performance than typically developing peers matched on mental age whereas boys with FXS-O did not (Estigarribia et al., 2011). Further investigation is needed into the impact of ASD symptoms on narrative performance by individuals with FXS.

Current Study

The purpose of the current study is to describe the narrative macrostructure abilities of a sample of adolescent boys and girls with FXS who vary according to language, cognitive and autism symptoms. Specifically, we addressed three primary research questions.

1. What are the macrostructure abilities of adolescent boys and girls with FXS in a narrative generation task and does performance differ based on sex?
2. How is variability in language, cognition and autism symptoms related to narrative scores in adolescents with FXS after controlling for age?
3. Is there a relationship between literacy skills and narrative macrostructure skills in adolescents with FXS?

Methods

Participants

Narratives were collected from 32 adolescents with full mutation FXS between the ages of 12 years, 5 months and 18 years (mean age of 15 years, 10 months) at the time of assessment. Participants were part of a larger longitudinal study investigating the development of individuals with FXS (n=45 at the first adolescent visit) (BLINDED FOR PEER REVIEW). The current analysis focuses on narratives produced at the first adolescent timepoint of this longitudinal study. A narrative sample was collected from all participants who communicated with speech and verbalized at least one utterance per page of the book--32 participants met these criteria (8 females, 24 males). 87.5% were Caucasian, 6.3% African American, 6.3% multiple races. Two participants were Hispanic, and the rest of the sample identified as Non-Hispanic. This is a sample of convenience that was recruited from list-serves, registries and support groups

Assessment Measures

All procedures were approved by the Human Subjects Committee at the [BLINDED FOR PEER REVIEW] (HSC #17261, TITLE BLINDED FOR REVIEW). The following measures were completed by a trained graduate research assistant during data collection visits in the participants' homes and will be used for analyses. The total assessment time was approximately an hour and a half.

Peabody Picture Vocabulary Test- Fourth Edition (PPVT-4). The PPVT-4 (Dunn & Dunn, 2007) is a standardized measure of receptive vocabulary. Participants were asked to select the picture that represented a given vocabulary item out of a field of four images.

Expressive Vocabulary Test- Second Edition (EVT-2). The EVT-2 (Williams, 2007) is a standardized measure of expressive vocabulary and word retrieval. The participant answers the question asked by the examiner regarding the picture depicted on the test easel. Three types of

responses are elicited including labeling the image, answering a question and providing a synonym.

Leiter International Performance Scale-Revised (Leiter-R). The Leiter-R (Roid & Miller, 1997) is a standardized measure of cognitive function for individuals aged 2-20 years, 11 months. It is individually administered and involves the use of manipulatives. Four subtests were administered that make up the Brief IQ scale: figure ground, form completion, sequential order and repeated patterns.

Vineland Adaptive Behavior Scales- Second Edition (VABS-2). The VABS-2 (Sparrow, Cicchettim, & Balla, 2006) is a measure of adaptive behavior across multiple areas of development. The survey interview form of the VABS-2 was administered to the mother of each participant. Questions about the child's development were asked and assigned a value of 0-2 (i.e. never to usually). The written language subtest was used for this analysis, as a measure of literacy skills. Questions range from a child's knowledge of and ability to write the alphabet, to the activities in which the child uses reading and writing skills at school and in life.

Childhood Autism Rating Scale- Second Edition (CARS-2). The CARS-2 (Schopler, Van Bourgondien, Wellman, & Love, 2010) was used to determine the degree of autism symptomatology for each participant assessed. The examiner rates the presence of various social communication abilities and restrictive and repetitive behaviors witnessed during an interaction resulting in a total score that captures the degree of autism symptoms displayed. In adolescents, a score below a 28 indicates minimal to no symptoms of autism, a score at or above 28 indicates mild symptoms and a score of 34 or higher is categorized as severe symptoms of ASD.

Table 1 below outlines the participants' scores on the measures administered at the visit. Raw scores were used for analyses because floor effects on standardized assessments are

common in this population and result in insufficient variability in scores for analysis. Standard scores are also reported for interpretation. Overall, the participants presented with a range of abilities as represented by the large standard deviations.

Story Generation Protocol

This study was modeled after Finestack et al. (2012) by using the same administration protocol and NSS scoring rubric. Finestack et al. (2012)'s sample included 12 boys with FXS who had a mean age of 14.95 years and a mean nonverbal IQ of 39.50. Their mean length of utterance in morphemes (MLU-M) was 5.97, which was higher than the males of our sample.

We completed a narrative macrostructure analysis for each participant based on stories generated from the picture book *Frog Goes to Dinner* (Mayer, 1974). The task was administered through the narrative protocol developed by Abbeduto et al. (1995). The protocol has been verified as an effective measure of expressive language skills in a typically developing population (Channell et al., 2018). All narratives were collected in the child's home environment in a quiet room using audio and video recorders. The examiner sat next to the participant and turned the pages of the book to control the pace of the task. A previewing of the book was done first. The participant looked at each page for ten seconds and was instructed to not to tell the story yet.

The sequence of instruction for narrative elicitation is shown in Figure 1. In addition to the prompts in Figure 1, if the participant started talking about an unrelated topic, the examiner would prompt the child to return to telling the story. The examiner remained engaged with the participant during the entire activity through eye contact and facial expressions; however, no statements were made other than those indicated by the series of prompts. Procedural fidelity was assessed through a checklist (available from the first author upon request) on 25% of the files, chosen randomly. The average procedural fidelity was 94.15%.

Transcription

Each narrative was transcribed from the audio recording using Express Scribe software (NCH© Software). The utterances were segmented by c-unit. The video recording was used to clarify any unintelligible words. A second trained transcriber reviewed each transcript for accuracy and segmentation of c-units. Finally, the examiner that administered the task was asked to review the transcript to clarify any unintelligible words. 25% of files were randomly selected to be transcribed by an independent coder for reliability. The narratives included 6 males and 2 females. Point-by-point agreement for c-unit segmentation ranged from 85.1% to 97.8% (M = 90.9%) and 76.9% to 97.3% (M = 85.4%) for bound morphemes.

C-units are made up of an independent clause and its corresponding modifiers. For example, “and he walks out the door with the rest of his family” contains an independent clause and a modifier and is considered one c-unit. The conventions from the Systematic Analysis of Language Transcripts (SALT; Miller & Iglesias, 2016) were used to divide words into morphemes (e.g. dog/s) and indicate mazes within utterances. Mazes are repetitions and revisions at the word and phrase levels. All transcripts were analyzed using SALT software to derive the mean length of MLU-M as a measure of utterance length and complexity.

Macrostructure Coding

The narrative transcripts were scored using the narrative story structure rubric (NSS) (Heilmann, Miller, & Nockerts, 2010; Heilmann, Miller, Nockerts, & Dunaway, 2010; Malone, 2010). The NSS allows for the measurement of specific story elements as well as the general organization of the story. A Likert-type scale is used to score each narrative macrostructure aspect on a range of 0-5 (SALT Software, LLC, 2017). The seven narrative macrostructure aspects include: introduction, character development, mental state, conflict/resolution, referencing, cohesion, and conclusion. The NSS was initially adapted by Finestack et al. (2012)

for the *Frog Goes to Dinner* by defining each score (0-5) for each narrative aspect and providing examples related to the book. We further refined the scoring definitions to increase reliability. Every use of a given macrostructure aspect is considered when it is scored. For example, when scoring the aspect of conflict/resolution the coder looks for the presence of each conflict/resolution pair listed on the coding sheet and a score is given based on the number of complete and partial conflict/resolution pairs the individual included in the story. These individual scores are then added up to generate a total narrative macrostructure score, which was used for our analyses. The full scoring rubric is available from the first author upon request. Example quotes from narratives produced by children with FXS and their assigned scores are shown in Table 2.

Reliability. The first author coded all narratives used for analysis and a trained research assistant coded files for reliability. The research assistant was trained to a minimum Krippendorff's alpha of .80 for total narrative score and .67 for individual narrative aspects. Thirty-one percent of files were randomly selected for reliability including narratives from 3 females and 7 males. These files were independently coded by a trained research assistant and scores were compared using Krippendorff's alpha (Krippendorff, 2004)-- a coefficient of agreement where 0 is perfect disagreement and 1 is perfect agreement. Krippendorff's alpha was chosen because it can be used to assess reliability among any level of measurement and can be used with small sample sizes (Hayes & Krippendorff, 2007). Values above .67 are considered to reflect tentative agreement and values above .80 are considered acceptable agreement (Hayes & Krippendorff, 2007; Krippendorff, 2004). The alpha values for the NSS are as follows: introduction α=.94, character development α=.68, mental state α=.89, referencing α=.78, conflict/resolution α=.90, cohesion α=.83, conclusion α=.72, total score α=.89, all aspects

combined α=.89. All aspect scores were used as they were all above the .67 cut-off used in a similar study (Finestack et al., 2012).

Analysis Plan

Differences in performance among females and males with FXS was anticipated and analyzed in the first research question. Due to this anticipated difference multiple analyses were conducted for each research question. To answer the first and third questions, analyses were conducted on the whole sample and then the males and females separately. Due to the small number of females in the sample, confidence intervals are provided for correlations. Both the total NSS score and the subscale scores were used to describe narrative performance of the two groups in research question 1 and to analyze the relationship between narratives and literacy skills for question 3.

The second research question was answered by conducting two sets of multiple regression analyses—one for the whole sample and one with males only. The small sample size of the female group did not allow for a separate analysis to be conducted. Tentative conclusions can be drawn regarding the female narratives by comparing the results of the whole sample to the results of the male-only sample. Multiple regression analyses were used to determine how scores on language, cognitive and autism symptoms predicted NSS scores after controlling for age. The dependent variable was the total narrative macrostructure score from the NSS. A language composite *Z* score was generated by first creating individual *Z* scores for the raw scores from the PPVT-4, EVT-2, and the MLU-M and then averaging these scores into one composite language *Z* score. Total raw scores were used from each of the other measures. Age was entered into the first block of the regression and then the variables of language composite *Z* score, Leiter raw and CARS scores were entered into the model together in the second block to determine the

combined variance explained by the model. The part correlation value was squared to acquire the R^2 change value for each variable and its unique contribution.

Results

Research Question 1 *What are the macrostructure abilities of adolescent boys and girls in a narrative generation task and does performance differ based on sex?*

The total macrostructure score varied widely, with a range of 2-31 and the average score was below half of the total 35 possible points *M*=14.31 *SD*= 7.61. The aspects that the participants preformed best on were *introduction M*=2.91 *SD*= 1.53 and *mental state use M*=2.25 *SD*=1.69. For all other aspects, participants had a mean score close to 2 on a 5-point scale. The full range of scores were only used for *mental states* and *conclusions.* On average, participants produced 82.69 utterances in their narratives *SD*= 36.98. The average MLU-M was 5.05 for the narratives segmented by c-unit, *SD*=2.18. The descriptive statistics for the narrative macrostructure and MLU are presented in Table 4.

Sex differences. There was a statistically significant difference on the total narrative macrostructure score between males (*M*=11.96) and females (*M*=21.38) after controlling for age, $F(1, 29)= 12.05$, $p=.002$, $d=1.46$. However, there was no statistically significant difference between males and females on the total narrative macrostructure score after controlling for cognitive abilities through the Leiter raw scores, $F(1, 29)=.07$, $p>.05$.

Narratives from males. On average, the male participants scored between a 2 (minor details included) and a 1 (immature or minimal) for most macrostructure aspects, indicating that they were including some story structure aspects, but at a minimal level. Overall, the males provided little elaboration of story details and the stories lacked cohesion with no connections between events within the story. Many males' narratives primarily consisted of off-topic

comments and included only basic story elements such as the main characters and a few actions. This resulted in low scores across all narrative aspects with some aspects scored as 0.

Narratives from females. Like males, females scored best on introduction and mental state use. The lowest score for females was character development illustrating a potential weakness in the inclusion of details of main characters. Females also scored in the middle of the rubric on referencing, which combined with the poor score in character development could indicate a lack of labeling of characters and using pronouns with no reference.

Research Question 2 *How is variability in language, cognition and autism symptoms related to narrative scores after controlling for age?*

Whole sample. The combination of language composite score, Leiter Brief raw score and CARS-2 total score explained a significant proportion of the variance in narrative macrostructure scores after accounting for age, R^2_{Δ}= .75, p<.001. The part and partial correlations reveal that the language composite score alone explains a significant unique proportion of the variance above and beyond Leiter Brief raw score and CARS-2 score, R^2_{Δ}=.16, p<.001. Neither the Leiter Brief raw score, R^2_{Δ}=.001, p=.74 nor the CARS-2 score, R^2_{Δ}=.005, p=.48 explained a significant proportion of variance above and beyond the other two variables.

Males. Together, the language composite score, Leiter Brief raw score and CARS-2 total score explain a significant proportion of the variance in narrative macrostructure scores after accounting for age, R^2_{Δ}= .67, p= <.001. The part and partial correlations reveal the language composite score alone explains a significant proportion of the variance above and beyond Leiter Brief raw score and CARS-2 score, R^2_{Δ}=.30, p=.001. The amount of variance explained by the language composite score alone for the male-only sample is almost double that of the whole sample. However, the total variance explained by all three variables combined is less in the

male-only sample (67%) than in the whole sample (75%). Again, neither the Leiter Brief raw score, $R^2_{\triangle}$=.00006, p=.95 nor the CARS-2 score, $R^2_{\triangle}$=.0001, p=.94 explained a significant proportion of variance above and beyond the other two variables.

Research Question 3 *Is there a relationship between the literacy skills and narrative macrostructure skills in adolescents with FXS?*

Our third research question asked about the relationship between literacy skills and narrative skills in adolescents with FXS. This question was addressed by separate analyses for the whole sample, males only and females only. Our female sample was large enough to conduct a separate correlational analysis (Aggarwal & Ranganathan, 2016), but not a multiple regression analysis. The mean raw score of the VABS-2 written subtest in the whole sample was slightly higher M=25.28 SD=11.17 than that of the male-only sample M=23 SD=9.41 and as expected the females achieved the highest scores, M= 32.13 SD=13.79. After controlling for age, raw scores on the written language scale of the VABS-2 were significantly correlated with the total narrative macrostructure scores and all subscale scores for the whole sample. For the male-only sample, significant correlations were reported for the total NSS score and the subscale scores of introduction, referencing and cohesion after controlling for age (see Table 6). The sample with the highest correlations between the VABS-2 and the narrative macrostructure score was the female only sample. High, significant correlations were found between VABS-2 written raw scores and the total macrostructure score (.94) along with many of the subscales in the female sample after controlling for age.

The same correlations were run but with MLU-M from the narratives as the control variable (see Table 7). After controlling for MLU-M, significant correlations were found in the between the total macrostructure score and the subscales of character development, mental state

& conclusion in the female-only sample. Figure 2 shows a scatterplot depicting the relationship between scores on the NSS and VABS-2 written language subscale.

Discussion

This study sought to investigate the narrative macrostructure abilities of both male and female adolescents with FXS syndrome. The participants demonstrated a wide variety of narrative skills. Most participants included at least basic story elements and a general story structure. Scores on the introduction and mental state subscales were consistently higher than the other subscales. The average total NSS score for the whole sample was 14.31, which is less than half the total possible score of 35.

Sex Differences

As expected, we observed significant differences in narrative quality for males and females in our sample when controlling for age. The narratives of the male group scored significantly lower than those of the females. However, when controlling for cognitive ability through Leiter raw scores there was no significant group difference in narrative scores. This is to be expected as females with FXS generally have higher IQs than males (Rinehart, Cornish, & Tonge, 2011). Thus, differences in narrative scores between males and females with FXS are likely due to difference in cognition between the two groups.

The adolescent males told narratives that, for the most part, lacked structure, cohesion and important story details. The majority of the male narratives introduced characters and described a string of actions by those character without linking the events together. They often lacked an overarching plot to the story. This description matches the narratives of a typically developing preschooler (Applebee, 1978). These low narrative scores reflect difficulties many males with FXS have with using language in a functional context. Though we didn't assess

academic or personal narratives, our findings suggest that many will have difficulties with generating narratives that are necessary for academic success (Feagans & Appelbaum, 1986) and social connections (Nelson, 1991).

The females provided structure and more details in their narratives than the males. They also attempted to connect events to create cohesion in the story. However, their stories lacked specific details about characters and were missing elements such as linking words and a detailed conclusion. Females with FXS with stronger language skills also had strong narrative skills. Further research is needed to determine how females with FXS apply narrative skills in academic and social contexts.

There was a range of performance in narrative generation in both the male and female groups. A subset of males were high performing and produced narratives that more closely matched the females' narratives. Additionally, a few of the females produced lower scoring narratives that more resembled the narratives of the male group. These differences may be accounted for by differences in cognition within each group. To better understand narrative performance in this population multiple types of narratives should be collected including story retell narratives and personal narratives.

Language, Cognition, Autism Symptomatology and Narratives

Our second aim was to determine how variability in language, cognition and autism symptomatology predicted narrative scores. Overall, we found language scores explained a significant proportion of the variance in total narrative macrostructure scores above and beyond the cognitive and autism measures combined, after controlling for age, for both the whole and male-only samples. The amount of variance explained by the language composite score alone is reduced by almost half when the females are added to the analyses (16%) compared to the male-

only sample (29%). This reduction in variance explained is not due to a lack of variance in the female language scores, as the SDs on the female language measures were larger than the males'. The reduction in variance may indicate language has less impact on the development of narrative skills in females compared to males in adolescents with FXS. Further investigation is needed with a larger group of females to determine how language impacts narratives.

These findings align with those of Finestack et al. (2012) who found that narratives of adolescents with FXS more closely matched typically developing peers' narratives when matched on MLU rather than on mental age. By language explaining a significant proportion of the variance above and beyond the Leiter and CARS scores, it appears language has a stronger impact on narrative skills. However, cognition likely still plays a role. The narrative macrostructure scores were highly correlated with both language and the Leiter Brief IQ measures, reflecting relationships between all three variables. This is to be expected due to the tight relationship of cognition and language (Komesidou et al., 2017; Roberts et al., 2001). Sex differences in narrative performance within this sample can be partially explained by cognitive differences between the sexes given that group differences were reduced, but not eliminated, by the inclusion of a covariate for cognitive ability.

The large amount of variance explained by language may also be due to the type of narrative scoring metric used in this and Finestack et al. (2012)'s study. The NSS is a macrostructure measure; however, it is influenced by vocabulary and morphosyntactic skills. For example, the inclusion of more descriptive terms about the characters leads to higher scores on *Introduction*. The ability to use transitional words between events increases scores on *Cohesion*. The studies that found a clear developmental trajectory of narrative macrostructure often used more frequency-based measures that count the number of times a specific narrative aspect is used

rather than the rating scale of the NSS (Khan et al., 2016; Stadler & Ward, 2005). This demonstrates how the narrative macrostructure measure used may influence results. Frequency based measures of story elements may better capture small variations in narrative performance better than a rating scale measure of the NSS.

The CARS-2 measure also did not explain any additional variance above and beyond the language and cognitive measure. This may be due to the small sample of participants who received a high CARS-2 score. Only 6 children received a CARS score at or above 28 and would be classified as having symptoms of ASD. Many of the participants with high CARS-2 scores were unable to complete the narrative generation task and hence excluded.

Narratives and Literacy

As expected, narrative skills and literacy skills were related in this study after controlling for age. Individuals with stronger narrative skills had higher literacy skills. The VABS-2 written language subtests were significantly correlated with all individual narrative aspect scores when the whole sample was included. The narratives of the female only group correlated strongly with the VABS-2 written language subtest when controlling for age. When the same correlations were run and MLU-M was entered as the control variable, neither the whole nor male-only samples were correlated with the VABS-2 written language scores. However, the female narratives continued to be significantly related to literacy skills on both the total narrative score and several subscales.

These results indicate a strong relationship between some narrative and literacy skills in females with FXS. Previous studies have shown a relationship between narrative skills to reading fluency (Reese et al., 2010) and word decoding (Feagans & Short, 1984). The VABS-2, however, is a gross measure based on parent report and not reflective of subtle changes in

fluency and decoding. Further investigation is needed to determine the nature of the relationship between narratives and specific literacy skills in the population with FXS. The presence of a relationship between narratives and literacy suggests that targeting narrative skills in intervention may have spillover effects on literacy and thereby increase academic success.

Strengths and Limitations

A strength of this study was the inclusion of females in our sample. The inclusion of females allowed for a better understanding of the full scope of narrative skills in adolescents with FXS. However, the number of females included was too small to analyze specific predictors for females. Future narrative studies in this population should obtain a larger sample of females to better assess their strengths and weaknesses in narration.

Another strength was our rigorous measure of narratives. We used an adapted scoring rubric based on Finestack et al. (2012) and improved it by further operationally defining the categories of character development, referencing and cohesion. Our modifications of the NSS increased reliability so that all narrative aspects could be used for analysis.

We also extended the study of narratives to include the relationship between narratives and literacy skills in this population. However, the VABS-2 written language measure is based on caregiver report and is not a comprehensive measure of literacy. Future research may delve into specific areas of literacy that relate to narratives such as reading comprehension, reading fluency or word decoding.

We used Brief scores from the Leiter International Performance Scale-Revised as our measure of nonverbal cognitive skills. Cognitive skills such as theory of mind, working memory and executive functioning that relate to narrative skills are not reflected in this measure. Each of these areas are theoretically linked to narrative skills and are also areas that could be directly

addressed through interventions. Future investigation of how theory of mind and memory skills relate to narrative skills of individuals with FXS may be informative.

We did not gather narratives from a matched comparison sample of typically developing participants for this investigation. This prohibited us from demonstrating how the performance of our sample of adolescents compared to typically developing children of the same mental age or language level. An ADOS was collected for this longitudinal study but at a later timepoint than this data set. Therefore, the CARS scores collected at the concurrent timepoint were used for this analysis as it provided the best picture of the participant's autism symptomatology at that time. Finally, narratives differ greatly across culture. We assessed narratives within the cultural-specific expectations of Western European storytelling which differs from other cultures.

Clinical Implications

Our findings suggest two main points regarding the importance of narratives for clinical work with children who have FXS. First, narrative analysis is an important measure because it provides insight into an individual's strengths and weaknesses in expressive language. Narratives can also elicit language skills required for successful academic and social communication.

Secondly, the skill of narration is an important target of language intervention for some individuals with FXS. A narrative intervention for individuals with FXS has the potential to not only increase language skills, but also literacy skills, social skills and academic achievements. A narrative intervention my impact overall academic success as evidenced by the predictive relationship seen in other populations (Feagans & Appelbaum, 1986); however further research is needed with individuals with FXS. Narrative intervention has the potential to improve both skills in structuring stories and the vocabulary and syntax used in their stories (Petersen, 2011).

Hence, narrative interventions may provide a way to address language skills within a functional context.

In conclusion, our results suggest that narratives yield valuable diagnostic information, and may be potential targets for intervention for individuals with FXS. The skill of narration can be taught and improved with intervention as evidenced in other populations (Petersen, 2011). Further investigation is needed to determine an effective narrative intervention in this population and to determine the effects of the intervention on other areas of language, social and academic achievement.

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Figure 1

Prompting Hierarchy for Narrative Elicitation Protocol

<table>
<tr><td colspan="2">First page: “Tell me how the story starts”</td></tr>
<tr><td>Adequate Response
→ 5 second wait time
→ “Anything else?”</td><td>Inadequate Response
→ “That’s a good start. Tell me a little more”
→Inadequate Response
→ “What about the frog, what is he doing?” (continue with each character until an adequate response is reached)</td></tr>
<tr><td colspan="2">Subsequent pages: No prompt</td></tr>
<tr><td>Adequate Response
→ 5 second wait time
→ “Anything else?” (Used if the examiner thinks the child has more to say)
OR
→ “Okay next page”</td><td>Inadequate Response
→ “Tell me everything that is happening in this part of the story”
→Inadequate Response
→ “Anything else?”</td></tr>
<tr><td colspan="2">Last page: “How does the story end?”</td></tr>
</table>

Figure 2

Plot of VABS-2 Written Scale and Total Narrative Macrostructure

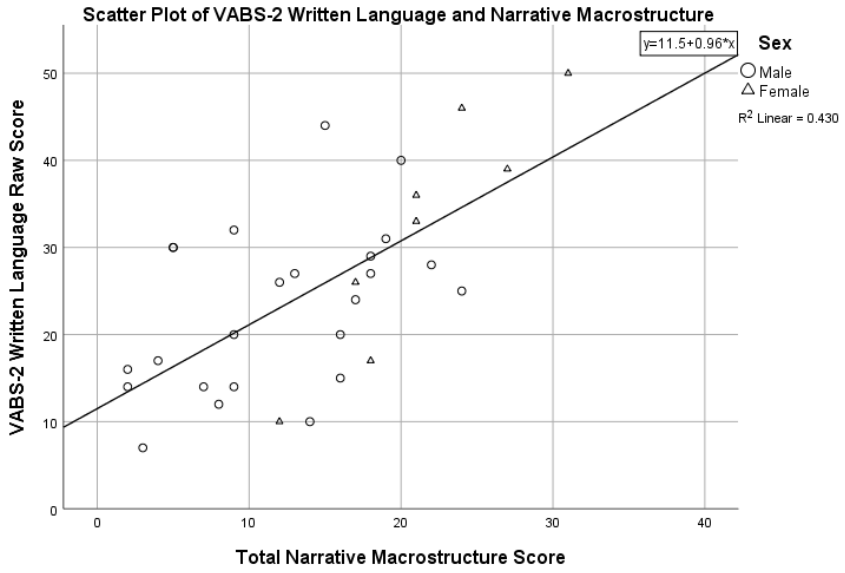

Table 1

Descriptive Statistics for Study Variables

Variable	Males (n=24)			Females (n=8)		
	M	*SD*	Range	*M*	*SD*	Range
Age (years;months)	16;0	1;2	12;5- 18;0	15;5	1;0	14;2- 16;11
PPVT-4 Raw Scores	92.17	32.73	29-147	149.25	47.86	60-189
PPVT-4 Standard Scores	41.38	16.79	20-71	76.00	28.39	25-103
EVT-2 Raw Scores	69.96	23.61	20-114	111.38	33.48	70-148
EVT-2 Standard Scores	47.42	17.30	20-78	79.13	24.20	48-105
Leiter Brief Raw Score	42.17	13.61	17-68	80.38	24.46	49-106
Leiter Brief Standard Scores	37.67	3.05	36-48	57.63	18.64	36-79
VABS-2 Written Raw Score	23.00	9.41	7-44	32.13	13.79	10-50
VABS-2 Written V Scale	7.38	1.50	5-11	9.63	3.62	5-17
MLU-M	4.32	1.50	1.79-6.88	7.25	2.52	3.24-10.63
CARS-2 Total Score	23.75	5.43	18-34.5	20.19	2.09	17-24
NSS Total Score	11.96	6.63	2-24	21.38	5.97	12-31

Table 2

High, Middle and Low Scoring Narrative Examples for Each NSS Aspect

High Score (4-5)	Middle Score (2-3)	Low Score (0-1)
Introduction- The inclusion of main characters and description of the settings throughout the story.		
"A boy getting dressed with a dog, turtle and frog looking at him."	"He put on his favorite tie. Then goodbye. He's going out to eat."	"The frog. He grab it. He goes frog."
Character Development- The inclusion and description of the characters in the story.		
"Okay so Jimmy and his family are gonna go out. He's got Kyle the dog, Tee the turtle and Trevor the frog"	"A boy getting dressed with a dog, turtle and frog looking at him" "There's a guy in the background laughing" "The waiter serves the lady her salad"	"The boy" "Hey frog in mirror" "'Hey' yell the woman" (continues to repeatedly use 'the woman' throughout the story)
Mental State- The inclusion of mental state words that describe the mental state of the characters to display emotion and/or thought process.		
"There's a guy being all mad." "The lady gets shocked… and starts crying" "His wife is perturbed by the fact that there's a frog there" "And he's just sad because he wanted to make sure the frog was okay. And he felt bad for the frog. And the frog just sat there in shock"	"She's scared" "He's laughing" "He's angry"	"He got mad"
Referencing- The use of clear pronouns and antecedents throughout the story.		
"The boy says goodbye to the dog and he walks out the door with the rest of his family"	"and the lady's just yelling at *them* like 'I want a refund'…And then *they* just sat." "and the dad looks like he's mad. And *he*'s like 'wait, wait that's my frog'"	"*He* got mad. *He* watching him. *He* go to *him*"
Conflict Resolution- The presence of conflict and resolution pairs throughout the story. The pairs represent actions and reactions between the characters; 8 pairs were chosen prior to scoring the narratives.		
"The frog then jumps into a salad. The waiter serves the lady her salad and as she starts eating it the frog comes out. The lady gets shocked and falls over."	Does not complete most conflict resolution pairs. "He grab the frog. He felt angrier. That his frog."	"When the man was a happy man for he saw the guy. I saw the man for two years. They guy say 'come over and play'"

Cohesion- The telling of the story in a logical order and the use of transitional words.		
"The frog jumped into the saxophone. *Then* the man couldn't play." "*Next* the frog jumped into the salad. The lady screamed *after* she saw the frog."	"*And then* the other guy's angry. He's like, 'why'*?* *And then* the other guy's just laughing and smiling because it's funny".	"The frog" "He grab it" "And then his friends" "The house" "They're hungry"
Conclusion- The inclusion of a final event and the inclusion of an overall wrap-up of the story		
"And then Trevor, Timmy, Kyle and Tee are all talking about the night and they just find it hilarious. Trevor's like 'we should do that again'. And then they all go 'no'!"	"He's laughing"	"The end"

Table 3

Correlations for Study Variables with the Whole Sample (n= 32;above diagonal) and Males Only Sample (n= 24;below diagonal)

Variable	1	2	3	4	5	6	7
1. Total Narrative Macrostructure Score	—	.73**	.81*	.82**	.84**	-.57**	.67**
2. Leiter Brief Raw Score	.50*	—	.85**	.85**	.77**	-.50*	.71**
3. EVT-2 Raw Score	.70**	.60*	—	.94**	.83**	-.59**	.75**
4. PPVT-4 Raw Score	.69**	.69**	.91**	—	.80**	-.55**	.75**
5. MLU-M	.80**	.42*	.63**	.61**	—	-.58**	.67**
6. CARS-2 Total Score	-.55*	-.55*	-.63**	-.56*	.63**	—	-.42*
7. VABS-2 Written Expression Raw Score	.47*	.49*	.59*	.57*	.52*	-.39	—

* $\alpha \leq .05$ ** $\alpha \leq .001$

Table 4

Descriptive Statistics for the NSS Aspects

Narrative Macrostructure Aspect	FXS Whole Sample (n=32)		FXS Male Sample (n=24)		FXS Female Sample (n=8)	
	M	*SD*	*M*	*SD*	*M*	*SD*
Introduction	2.91	1.53	2.42	1.38	4.38	.92
Character Development	1.75	.88	1.50	.59	2.50	1.19
Mental States	2.25	1.69	1.92	1.61	3.25	1.58
Referencing	1.97	1.06	1.75	1.03	2.63	.92
Conflict/Resolution	1.88	1.26	1.46	1.10	3.13	.83
Cohesion	1.97	1.09	1.71	1.12	2.75	.46
Conclusion	1.59	1.16	1.21	.83	2.75	1.28
Total	14.31	7.61	11.96	6.63	21.38	5.97
MLU-M	5.05	2.18	4.32	1.50	7.25	2.52

Table 5

Summary of Multiple Regression Analysis for Variables Predicting Narrative Macrostructure Performance for Whole Sample (n=32) and Male Only Sample (n=24)

	Whole Sample				Male Only			
	Age	Language Composite Score	Leiter Brief Raw Score	CARS-2 Total Score	Age	Language Composite Score	Leiter Brief Raw Score	CARS-2 Total Score
B	-.04	6.92	-.02	-.13	-.06	8.35	-.005	-.02
SE B	.05	1.63	.06	.18	.06	2.02	.09	.23
Partial Correlation	-.16	.63	-.06	-.14	-.23	.69	-.01	-.02
Part Correlation	-.08	.40	-.03	-.07	-.14	.55	-.01	-.01
R^2 Change	.01	.16	.0008	.004	.02	.29	.0001	.0001
Sig.	.40	<.001	.74	.48	.32	.001	.95	.94

Table 6

Correlations of VABS-2 Written Scale and Narrative Macrostructure Aspects Controlling for Age

Narrative Macrostructure Aspect	FXS Whole Sample (n=32)		FXS Male Sample (n=24)		FXS Female Sample (n=8)	
	r	95% CI	*r*	95% CI	*r*	95% CI
Introduction	.58**	.29-.77	.47*	.08-.73	.77*	.13-.96
Character Development	.51*	.20-.73	.20	-.22-.56	.59	-.19-.92
Mental State	.59**	.30-.78	.37	-.04-.67	.92*	.60-.99
Referencing	.55**	.25-.76	.50*	.11-.75	.80*	.22-.96
Conflict/Resolution	.60**	.31-.78	.41	0-.70	.90*	.53-.98
Cohesion	.52*	.21-.74	.46*	.07-.73	.74	.07-.95
Conclusion	.68**	.44-.83	.40	0-.69	.90*	.55-.98
Total Macrostructure Score	.66**	.40-.82	.48*	.09-.74	.94*	.70-.99

* $\alpha \leq .05$ ** $\alpha \leq .001$

Table 7

Correlations of VABS-2 Written Scale and Narrative Macrostructure Aspects Controlling for MLU-M

Narrative Macrostructure Aspect	FXS Whole Sample (n=32)		FXS Male Sample (n=24)		FXS Female Sample (n=8)	
	r	95% CI	*r*	95% CI	*r*	95% CI
Introduction	.11	-.24-.45	.18	-.24-.54	.18	-.60-.79
Character Development	.15	-.21-.47	-.26	-.16-.60	.68*	-.04-.94
Mental State	.23	-.13-.54	.06	-.35-.45	.85*	.36-.97
Referencing	.10	-.26-.43	.09	-.33-.48	.28	-.53-.82
Conflict/Resolution	.19	-.17-.51	.20	-.22-.56	.58	-.21-.91
Cohesion	.06	-.30-.40	.11	-.30-.49	.22	-.57-.80
Conclusion	.30	-.06-.59	.05	-.36-.45	.84*	.32-.97
Total Macrostructure Score	.23	-.13-.54	.12	-.30-.50	.82*	.26-.97

* $\alpha \leq .05$