Journal of Intellectual Disability Research doi: 10.1111/j.1365-2788.2006.00888.x
VOLUME 51 PART 5 pp 387–400 MAY 2007

Gender differences in repetitive language in fragile X syndrome

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Abstract

Background Verbal perseveration (i.e. excessive self-repetition) is a characteristic of male individuals with fragile X syndrome; however, little is known about its occurrence among females or its underlying causes. This project examined the relationship between perseveration and (1) gender, (2) cognitive and linguistic ability, and (3) language sampling context, among youth with fragile X syndrome.
Method Language transcripts were obtained from adolescent male ($n = 16$) and female participants ($n = 8$) with fragile X syndrome in two language contexts (i.e. narration and conversation) designed to elicit spontaneous language samples. Transcripts were coded for utterance-level repetition (i.e. repetition of words, phrases, dependent clauses or whole utterances), topic repetition and conversational device repetition (i.e. repetition of rote phrases or expressions).
Results Male participants produced more conversational device repetition than did female participants. Gender differences in conversational device repetition were not explained by differences in non-verbal cognitive or expressive language ability. Context influenced the type of repetition observed; for example, more topic repetition occurred in conversation than in narration regardless of gender.
Conclusions The observed gender differences in conversational device repetition among adolescents with fragile X syndrome suggest that, relative to females, male participants may rely more heavily on rote phrases or expressions in their expressive language. Further, results suggest that this gender difference is not simply the result of the correlation between gender and cognitive or linguistic ability in fragile X syndrome; rather, gender may make an independent contribution to conversational device repetition. Repetition type also varied as a function of expressive language context, suggesting the importance of assessing language characteristics in multiple contexts.

Keywords communication, fragile X syndrome, gender differences, language, perseveration, self-repetition

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Introduction

Fragile X syndrome is the most common known inherited cause of intellectual disability, occurring in approximately 1 in 4000 males and 1 in 8000 females (Hagerman 1999). The syndrome occurs as the result of an excessive lengthening of a repetitive sequence of trinucleotides (CGG) in a gene (FMR1) on the long arm of the X chromosome (Yu *et al.* 1991). When this expansion consists of 200 or more repetitions (also known as a *full mutation*), it leads to a reduction in the protein (FMRP) that is produced by the gene. In most cases, males with the full mutation have cognitive impairments ranging from moderate to severe (Hagerman & Sobesky 1989; Bennetto & Pennington 1996). In contrast, cognitive ability among females who have the full mutation typically ranges from mild intellectual disabilities to no noticeable impairments (Hagerman & Sobesky 1989; Bennetto & Pennington 1996). However, it is unclear whether females are simply affected in the same way but to a lesser degree than males, or whether there are also qualitative differences in the patterns of impairments characterizing males and females (as reviewed by Murphy & Abbeduto 2003). This study was designed to examine gender differences among individuals with fragile X syndrome in the domain of language.

Language deficits are characteristic of males with fragile X syndrome (as reviewed by Murphy & Abbeduto 2003). Most notably, verbal perseveration, or excessive self-repetition, is frequent among males with fragile X syndrome (Sudhalter *et al.* 1990, 1991, 1992; Ferrier *et al.* 1991; Abbeduto & Hagerman 1997) and may distinguish them from males with other developmental disabilities, such as Down syndrome or autism (Sudhalter *et al.* 1990). For example, Sudhalter *et al.* (1990) found that males with fragile X syndrome, Down syndrome, and autism who were matched on chronological age, Vineland Communication domain age-equivalent scores and overall Adaptive Behavior Score could be distinguished on the basis of self-repetition. Both males with fragile X syndrome and males with autism produced significantly more repetitive language than did males with Down syndrome. However, 86% of the repetitive language produced by those with fragile X syndrome was perseverative (i.e. a repetition of the self) and 10% was echolalic (i.e. a repetition of others), whereas only 2% of repetition in autism was perseverative and 79%, echolalic. Consistent with these findings, Ferrier *et al.* (1991) found that males with fragile X syndrome produced proportionally more self-repetitions than did males with Down syndrome or autism, who were matched to them on age and IQ.

In contrast to the numerous studies of language among males with fragile X syndrome, few studies have examined the language of females with the syndrome, and fewer still the occurrence of verbal perseveration (Dykens *et al.* 1994; Hagerman 1996; Abbeduto & Hagerman 1997). Madison *et al.* (1986) examined communication skills in a single family of individuals with fragile X syndrome. With respect to the language characteristics of the female members in the family, they observed 'a detailed, run-on narrative style' (p. 139) in conversational speech and repetition of 'automatic phrases such as "course", "well" or "special" ' (p. 139). Although the Madison *et al.* results provide evidence of repetitive language, the fact that the participants were from a single family limits the generalizability of the findings. Moreover, Madison *et al.* did not include a comparison group, which makes it difficult to assess the extent or seriousness of the repetition observed. The present study was designed to contribute to our understanding of perseveration in fragile X syndrome by examining excessive repetition in both males and females with the syndrome.

An important factor that could influence the occurrence of perseveration in both males and females with fragile X syndrome is the individual's cognitive ability (e.g. IQ) or language ability. Evidence suggests that among both males and females, those who exhibit more of the physical and behavioural features of fragile X syndrome tend to be more cognitively impaired (Dykens *et al.* 1994; Bennetto & Pennington 1996; Hagerman 1996). If this relationship holds true for perseveration, males should perseverate more than females. When comparing the language characteristics of males and females, however, gender *per se* might also account for differences in perseveration. Gender differences in language competence and performance exist among the population of individuals who are typically developing (Ely 1997) and among individuals with other types of intellectual disabilities even when controlling for differences in IQ (Wilkinson 1999; Wilkinson *et al.* 1999). The present study was designed to determine whether gender differences in perseveration were fully explained by differences in cognitive and linguistic ability or whether gender itself also made a unique contribution over and above the contribution of cognitive and linguistic ability.

Previous efforts to understand the nature of perseveration in fragile X syndrome have been complicated by lack of a consistent and precise definition of what constitutes perseveration. For example, the repetition measured by Paul *et al.* (1987) was described as 'topics on which the subject perseverated' (p. 462), whereas Ferrier *et al.* (1991) coded repetition as 'complete self-repetition', 'complete repetition of the examiner' (i.e. other repetition), 'partial self-repetition', or 'partial repetition of other', but for 'major syntactic constituents only, i.e. nouns, verbs, adjectives, and adverbs' (p. 780). An even more inclusive definition of perseveration was adopted by Sudhalter *et al.* (1990), who defined perseveration as: 'excessive repetition of a word, phrase, sentence, or topic' (Sudhalter *et al.* 1990) and combined it for purposes of analysis with other forms of 'deviant repetitive language', which included echolalia, jargoning, and affirming by repetition of other. Consequently, the diagnostic group differences observed by Sudhalter *et al.* (1990) may not only reflect differences in perseveration.

More generally, these varying definitions of perseveration are problematic because the repetition of different types of linguistic units may reflect different underlying problems. For example, repetitions that occur at the utterance-level (i.e. of syllables, words, phrases, or whole sentences) may reflect word-finding difficulties, false starts, or self-corrections (Miller 1991), whereas topic repetition may reflect overall ability to organize, execute and maintain the continuity of discourse (Brinton & Fujiki 1984). In the present study, we distinguished repetition at the *utterance-level* from *topic* and *conversational device* repetition (i.e. routinized social phrases, such as 'That's a wrap').

Also unclear from previous studies is whether repetitions occur within utterances (e.g. 'Maybe I <u>Maybe I</u> just can't read'; repetition is underlined) or between utterances (e.g. 'He went to the store. <u>The store</u>'; repetition is underlined). Again, different mechanisms might underlie these two types of self-repetition. Among typically developing children, repetitions that occur within an utterance are often associated with sentence-formulation difficulties (Miller 1991), whereas repetitions between utterances are thought to reflect a strategy for holding the turn while preparing for the next utterance (Brinton & Fujiki 1984). In the present study, we distinguished between repetitions occurring *within* and *between* utterances.

Another factor that has been neglected in previous research, but that may be important for understanding perseveration in fragile X syndrome, is the context in which the language sample for measuring perseveration is taken. For example, work with typically developing individuals (Dollaghan *et al.* 1990; Evans & Craig 1992) and youth with intellectual disabilities of varying aetiology (Abbeduto *et al.* 1995), has demonstrated that story telling, or narrative, contexts elicit more syntactically complex utterances on average than do conversation. In contrast, conversation leads to the production of a greater number of utterances (Dollaghan *et al.* 1990). As a result, both narration and conversation language samples are need to provide a full characterization of expressive language ability (Dollaghan *et al.* 1990). However, previous research on fragile X syndrome has relied on conversation primarily. In the present study, language samples were collected in standardized narrative and conversational tasks.

In summary, the purpose of this study was to examine the relationship between the occurrence of repetitive language and (1) gender, (2) cognitive and linguistic ability, and (3) language sampling context. More specifically, we determined whether there were differences in the extent and nature of repetitive language based on gender or language sampling context, and examined the contribution of gender to the extent of repetition in fragile X syndrome relative to cognitive and linguistic ability. We also distinguished between utterance-level, topic and conversational device repetition, as well as repetitions that occurred within or between utterances.

Method

Participants

Twenty-four adolescents (8 females, 16 males) were drawn from a group of 51 individuals with fragile X who were participating in a larger study on language and communication (see below for exclusionary criteria and Table 1 for participant characteristics). The larger study primarily recruited families from the midwestern USA, although families came from other regions in the USA, including Florida, Colorado, Nebraska and Texas. Recruitment was done through Internet postings, newspaper advertisements, flyer distribution at genetics clinics and mailings to special

Table 1 Participant characteristics: mean (and standard deviation)

	Females $n = 8$	Males $n = 16$
Chronological age (years)	15.35 (2.53)	17.03 (3.01)
Non-verbal IQ*	65.5 (16.67)	40.12 (7.35)
OWLS-OES standard score†	78.12 (11.42)	53.81 (14.79)

* Calculated based on three subtests of the Stanford Binet, 4th Edition: Bead Memory, Copying, Pattern Analysis (Thorndike *et al.* 1986).

† OWLS-OES, Oral and Written Language Scales – Oral Expression Scale (Carrow-Woolfolk 1995).

education directors. All participants lived at home with parents or guardians.

Adolescents were excluded if no DNA results were available to confirm the fragile X diagnosis ($n = 2$), they had a sibling participating in the study (so as not to violate the assumption of statistical independence associated with ANOVA; $n = 8$), they had a diagnosis of autism (discussed subsequently; $n = 9$), were younger than 12 years of age ($n = 4$), or were unable to complete the study protocol ($n = 2$). Two participants were also excluded because no transcripts were available at the time of the coding and analysis conducted for this study. We set age 12 as a lower bound because findings in the literature on fragile X syndrome suggest that many of the physical and linguistic characteristics are exacerbated during adolescence, at least among males (Dykens *et al.* 1994). Moreover, in adolescence, compared with childhood, language demands change and increase as language and thought become more abstract (as reviewed by Nippold 1998). As seen in Table 1, male and female participants did not differ in chronological age, $t(22) = 1.35$, $P = 0.19$.

The diagnosis of fragile X syndrome was obtained via DNA testing. Seven of the male participants were mosaic (i.e. they had both the full mutation and a premutation). This rate of mosaicism is consistent with other estimates of its prevalence among male with fragile X syndrome (Nolin *et al.* 1994). Previous studies of perseveration in fragile X have not distinguished between individuals with the full mutation and individuals who are mosaic. Thus, to allow replication of previous findings regarding males and consistent with our focus on gender differences, we did not distinguish among male participants according to mosaic status.

No participant with fragile X syndrome in the present sample met the DSM-IV criteria for autistic disorder. All participants were screened for autism using the Autism Behavior Checklist (Krug *et al.* 1980) followed by an evaluation by a clinical psychologist for those who met screening criteria (see Abbeduto *et al.* 2003 for details). Screening criteria consisted of an Autism Behavior Checklist score of 44 or greater (Volkmar *et al.* 1988) from two of three informants (i.e. mother, father, teacher) in two-parent families, or one of two informants (e.g. teacher and mother) in single-parent families. Any participants that the clinical psychologist determined meet criteria for autism were excluded from the present study.

Materials

Non-verbal cognitive ability

Participants were administered the Bead Memory, Pattern Analysis and Copying subtests of the Stanford–Binet Intelligence Scale, 4th edition (Thorndike *et al.* 1986). The standard scores for the three subtests were used to determine a partial composite IQ score, which was used to evaluate the contribution of cognitive ability to the occurrence of repetition. The internal-consistency reliabilities for the three subtests over the developmental levels represented in the present study have been found in previous research to be above 0.80 and mean test–retest reliabilities are generally above 0.60 (Thorndike *et al.* 1986). Moreover, factor analyses suggest that the three subtests load highly (at 0.60 or more) on *g* (Thorndike *et al.* 1986). As expected, females in the present study had higher partial composite IQ scores than males, $t(22) = -5.24$, $P < 0.001$ (see Table 1).

Language ability

Participants were administered the *Oral Expression Scale* of the Oral and Written Language Scales (OWLS; Carrow-Woolfolk 1995). The standard score was computed for each participant and used to evaluate the contribution of expressive language ability to the occurrence of repetition. Previous research has shown that the split-half reliabilities for the total score range from 0.76 to 0.91 across ages, whereas the average test–retest reliability is 0.81 across ages (Carrow-Woolfolk 1995). As expected, females in the present study had higher OWLS standard scores than males, $t(22) = -4.07$, $P = 0.001$ (see Table 1).

Language samples

Each participant completed two structured tasks designed to elicit spontaneous language samples, which were then analysed for the occurrence of repetitive language. The first language task, narration, required the participant to tell a story based on pictures in Mercer Mayer's wordless picture book, *A Frog Goes to Dinner*. Procedures for eliciting narratives from this book were standardized (see Abbeduto *et al.* 1995 for details). The second language task, conversation required the participant to talk to an experimenter for 10 min. In order to elicit 10 min of speech, the experimenter introduced a predetermined set of topics developed by Abbeduto *et al.* 1995 and slightly modified for the current project. The topics included school, favourite teacher, after-school activities, pets, favourite sports and hobbies. The procedures for introducing and maintaining each topic were standardized, although the participant largely determined the flow of the interaction within each topic (see Abbeduto *et al.* 1995 for details).

Language transcription

All speech during the narration and conversation tasks was audio-taped then transcribed and analysed using Systematic Analysis of Language Transcripts (SALT; Miller & Chapman 1990). SALT is a software program that specifies a set of conventions for transcribing language samples. The conventions are based on well-established procedures in child language research. SALT also provides preset and user-defined summaries of transcripts prepared according to its conventions.

A single transcriber produced an initial transcript of each language sample. A second transcriber then checked the transcript by listening to the audio-taped language sample and marking any disagreements on the transcript. The first transcriber listened to the sample again and incorporated the feedback from the second transcriber, as he or she deemed necessary, to produce a final version of the transcript for coding. The transcribers were unaware of the hypotheses of the study or the adolescents' level of cognitive or language ability. In order to evaluate the reliability of the transcripts used for coding perseveration, inter-transcriber reliability was calculated for a randomly selected subset of transcripts (16%). The mean percent agreement across the dimensions relevant to coding (i.e. utterance segmentation, intelligibility, mazes, number of morphemes, number of words and word identification) was 0.90 across narration and conversation.

Perseveration coding

For each task, all speech was segmented into utterances. For this study, an utterance was equivalent to Loban's (1976) *Communication Unit* (*C-unit*); namely, 'an independent clause and any of its modifiers, which could include dependent clauses' (Abbeduto *et al.* 1995). For example, the sentence 'The boy went home with the girl who went to school' would count as one utterance, whereas the sentence 'The boy went home and the girl went to school' would count as two utterances. Each utterance was then coded for the occurrence of repetition. The mean (and range) for the number of utterances in narration were 70.19 (23–424) and 52.37 (25–94), for male and female participants, respectively. In conversation, the mean (and range) was 138.44 (25–203) and 123.13 (59–173) for male and female participants, respectively. No significant differences were found between males and females in either the narration or conversation context as regards the total number of utterances ($P = 0.61$ and $P = 0.39$, respectively).

Three types of self-repetition were assessed. Detailed examples of each type of repetition are provided in the Appendix.

First, *utterance-level repetition* captured repetition occurring within the structural level of the utterance, including repetition of words, phrases, dependent clauses and whole utterances. To count as a repetition in these categories, the repeated segments had to be said more than one time in immediate succession, preserve the basic structure of the original segment and add no new semantic content. Each repetition was further classified according to the location of the repetition: each repetition could either occur *within* an utterance (e.g. 'Maybe I <u>Maybe I</u> just can't read.'), or *between* utterances (e.g. 'He went to the store. <u>The store</u>.'). Note that in the preceding examples, the repetition is underlined. Also, note that the within-between distinction was not meaningful for the remaining repetition types.

Second, *topic repetition*, captured repetition that occurred at the broader level of meaning abstracted from the utterances. A topic was defined as 'a proposition (or set of propositions) expressing a concern (or set of concerns) that the speaker is addressing' (Keenan & Schieffelin 1976; p. 343). In order to determine whether the topic of the utterance was repetitious, the coder was instructed to decide whether the topic of each utterance reintroduced the topic from a pervious utterance without providing new information about the topic (i.e. expanding or elaborating on the topic). An example of topic repetition is as follows.

> The experimenter says, 'You can't think of any time you went on a vacation?' To which the participant responds, 'Out of [state name], no . . . I never went out of [state name] . . . I have never gone out of home, out of [state name] . . . Everything's in [state name] . . .'.

In some instances, as in the previous example, topic repetitions occurred as a series of consecutive repetitive utterances on a particular topic (i.e. once an individual started talking, the topic did not change, but rather multiple repetitive utterances were devoted to the topic). In other instances, the same topic or topics were repeatedly reintroduced at periodic intervals throughout the talk even though they were not directly related to the topic at hand. An example of this type of repetition is as follows:

> The experimenter says, 'Tell me about what you like to read.' The participant responds, 'I liketa read books about box car children . . . I have that at home because my mom wants to (utterance abandoned). Well, I don't like the [Team A name]. I like the [Team B name] . . .'

In this example, despite being asked about reading, the participant introduces a different topic, the Team A football team. The topic of the Team A had been discussed previously and was unrelated to the current topic (reading), so this reintroduction would count as a repeated topic.

Third, *conversational device repetition*, captured repetition that controlled the flow of the interaction without adding content to the topic. More specifically, any utterance that did the mechanics of the interaction, but did not necessarily have any semantic content was considered a conversational device (e.g. the same utterance repeated through the transcript). An example of a conversational device would be a phrase like 'That's about it', 'All that kind of stuff', or 'That's a wrap' repeated throughout the transcript. In the following example, the participant repeatedly answers the experimenter's questions then adds 'How about you?' Even though this question carries semantic meaning, it is used indiscriminately in this exact form through out the language sample.

> Experimenter: 'Are there any sports that you like other than football?'
> Participant: 'My favourite sport is baseball. And my favourite team is the [team name]. Those are cool teams. How about you?' . . .
> Experimenter: 'How do you play that [hockey]?'
> Participant: 'Well, my Dad likes the [team name]. and my team is famous. The [team name]. How about you?' . . .
> Experimenter: 'Tell me about some of their [music group] other songs you like?'
> Participant: 'Well, my favourite one is [song name]. Yeah. How about you?'

Perseveration coding reliability

Reliability of repetition coding was computed for a random subset (44%) of the transcripts. Two independent coders coded the transcripts separately. Overall, kappa agreement, which corrects for chance agreement (Cohen 1960), for utterance-level repetition was 0.82 in narration and 0.89 in conversation, which is 'almost perfect' (Landis & Koch 1977, p. 165). The kappa value for topic and conversational device repetition was 0.77 in narration, which is 'substantial'.

Although the kappa value for topic and conversational device repetition in conversation was considered 'moderate' (kappa = 0.51), it was noticeably lower than for the other coding categories. The challenge of reliably coding topic repetition is not limited to this study; others investigating this topic have found a similarly broad range of percent agreement (Brinton & Fujiki 1984; Dorval & Eckerman 1984). Dorval & Eckerman (1984), for example, report a range of percent agreement for topic coding between 73% and 96% for a more developmentally mature sample of participants than those in this study (i.e.

typically developing school-age children and adolescents). The percent agreement for topic calculated for the present study is therefore consistent with the reliability obtained in previous studies. Regardless, the somewhat lower agreement for topic coding in the present study suggests using caution when interpreting the results for topic repetition in conversation.

Procedure

The measures for this study were part of a more comprehensive protocol, which was individually administered. A single experimenter administered the entire protocol to any given participant. There were five experimenters (all women) across participants. The protocol was administered during the course of two visits to the research centre. The measures reported here were administered during the second visit beginning with the Stanford–Binet, 4th edition (Bead Memory, Pattern Analysis, then Copying) followed by the narration or the conversation task, a short break, and then the remaining language task. The order of the presentation of the narration and conversation was counterbalanced across participants.

Dependent measures

Four dependent measures were calculated separately for each language sampling context.

Proportion of utterance-level repetition: the sum of all utterance-level repetition divided by the total number of utterances. Utterance-level repetition was calculated by adding together the number of word, phrase, dependent clause and whole utterance repetitions.

Proportion of topic repetition: the number of repeated topics divided by the total number of utterances. This measured the extent to which repetition reflected repetition of themes or ideas occurring throughout the discourse as opposed to repetition of rote phrases.

Proportion of conversational device repetition: the number of repeated conversational devices divided by the total number of utterances. This measured the extent to which repetition reflected repetition of phrases to control the flow of interaction without adding content to the discourse.

Proportion of within-utterance repetition was calculated to investigate repetition within the broad category of utterance-level repetition, and consisted of the sum of all utterance-level repetition that was coded as within-utterance repetition divided by the total number of utterances. This measure reflected the extent to which repetition at the utterance-level occurred within rather than between utterances. Note that together the within- and between-utterance variables were mutually exclusive and exhaustive, so only the former was used in the analysis.

Results

Descriptive analyses

Regardless of context, 99% of all utterance-level repetitions were self-repetitions, which is consistent with other findings in the literature on fragile X syndrome (Sudhalter *et al.* 1990; Ferrier *et al.* 1991). No additional analyses were conducted on self repetitions. In addition, the majority (82%) of these utterance-level repetitions occurred within utterances. Independent sample *t*-tests revealed no gender differences on the proportion of within-utterance repetition in narration or conversation ($Ps > 0.93$) (see Table 2 for means and standard deviations).

Repetition analyses

All proportions were arcsine transformed, which is a standard method for dealing with the fact that the mean and variance are correlated for proportions in violation of the homogeneity of variance assumption of analysis of variance. Inferential statistics were conducted on the transformed proportions; however, for ease of interpretation, descriptive statistics are reported for the untransformed proportions. A separate repeated-measures ANOVA was conducted for each dependent variable. Alpha levels were not adjusted despite having multiple dependent measures because the exploratory nature of the study made a less conservative alpha appropriate. In addition, because the amount of repetition in males should be greater than or equal to the amount in females, the gender comparisons were conducted using one-tailed tests. The proportions of the various types of repetition are presented in Table 2.

The influence of gender and sampling context were addressed in a Gender (male, female) X Context (narration, conversation) repeated-measures ANOVA, with a separate analysis for (1) the proportion of utterance-level repetition, (2) the proportion of topic repetition, and (3) the proportion of conversational device repetition.

Table 2 Proportion of utterance-level, topic and conversational device repetition by gender and context: mean (standard deviation)

	Narration		Conversation	
Variables	**Females**	**Males**	**Females**	**Males**
Number of utterances	52.37 (20.39)	70.19 (96.05)	123.13 (36.43)	138.44 (42.07)
Utterance-level repetition*	0.06 (0.10)	0.08 (0.05)	0.08 (0.06)	0.10 (0.07)
Within-utterance repetition†	0.78 (0.44)	0.78 (0.36)	0.85 (0.35)	0.88 (0.17)
Topic repetition	0.05 (0.06)	0.11 (0.13)	0.12 (0.11)	0.16 (0.15)
Conversational device repetition	0.002 (0.006)	0.07 (0.12)	0.002 (0.006)	0.04 (0.03)

Note. Unless otherwise indicated, values based on $n = 8$ females and $n = 16$ males.
* During narration three females and one male did not produce any utterance-level repetition. During conversation, 1 male did not produce any utterance-level repetition.
† Distinction is only applicable to utterance-level repetition, and is only meaningful when utterance-level repetition is produced. Thus, values reflect the proportion of within-utterance repetition only for individuals who produced utterance-level repetition ($n = 5$ females and 15 males in narration; $n = 8$ females and $n = 15$ males in conversation).

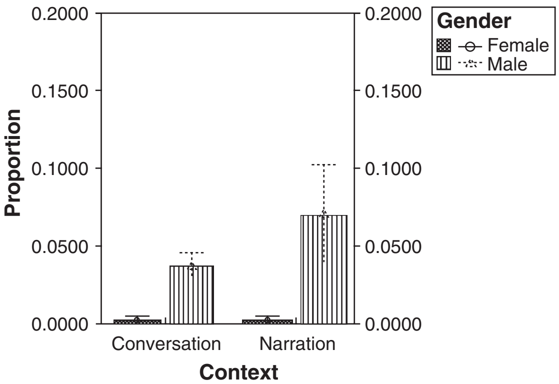

Figure 1 Conversational device repetition as a function of gender and language sampling context.

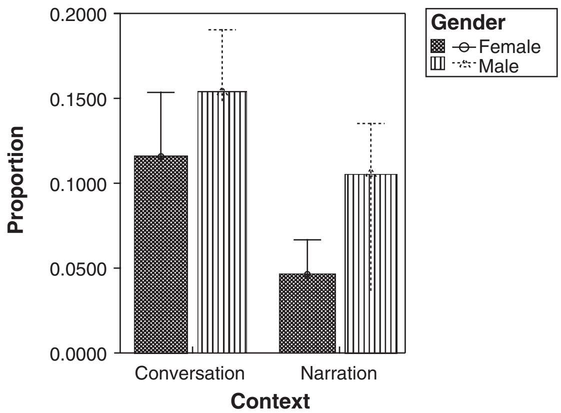

Figure 2 Topic repetition as a function of gender and language sampling context.

No differences were found as a function of gender for utterance-level repetition ($P = 0.13$, one-tailed) or topic repetition ($P = 0.13$, one-tailed). However, a main effect of gender indicated that males produced a higher proportion of conversational device repetition than did females regardless of context, $F_{1,22} = 8.13$, $P = 0.004$ (one-tailed), partial $\eta^2 = 0.27$ (see Fig. 1).

A main effect of context for topic repetition, indicated that proportionally more topic repetition occurred in conversation than in narration regardless of gender, $F_{1,22} = 10.59$, $P = 0.004$, partial $\eta^2 = 0.32$ (see Fig. 2). In addition, although not significant ($P = 0.07$), proportionally more utterance-level repetition occurred during conversation than during narration, $F_{1,22} = 3.60$, partial $\eta^2 = 0.14$. No significant difference in conversational device repetition according to context was observed ($P = 0.87$).

Regression analyses were conducted to examine the contribution of gender and cognitive or linguistic ability to the prediction of repetition. Before performing the regressions, a principle components analysis (with Varimax rotation) was conducted on the arcsine-transformed proportions to reduce the number of dependent variables for the regression. Two factors emerged. Factor 1 was defined by high positive loadings of four variables: (1) utterance-level repetition in conversation, (2) utterance-level repetition in narration, (3) topic-level repetition in conversation, and (4) topic-level repetition in narration. Factor 1 accounted for 43.6% of the total variance. Factor 2 was defined by high positive loadings of conversational device repetition in conversation and in narration. Factor 2 accounted for 22.5% of the total variance. Standardized score for these two factors were then used in the regressions.

The relationship between Factor 1 (utterance-level and topic repetition) and gender was examined in two sets of regression analyses. In the first set, ability was represented by non-verbal IQ score (NVIQ), calculated from the three Stanford–Binet subtests. In the second set, NVIQ was replaced with the standard score obtained from the Oral Expression scale of the OWLS. In both sets of analyses, gender was entered into the equation at the first step and then the ability measure was added, with interest in whether a significant relationship between gender and the dependent measure remained after ability was simultaneously considered. Based on these analyses, the variation in Factor 1 was not accounted for by gender or by the combined effect of gender and non-verbal cognitive ability or language ability ($Ps > 0.17$, one-tailed).

The relationship between Factor 2 (conversational device repetition) and gender was examined following the same procedure used to analyse Factor 1. Gender alone accounted for 27% of the variance in conversational device repetition, $F_{1,22} = 9.51$, $P = 0.002$, one-tailed, partial $\eta^2 = 0.30$. When both gender and NVIQ were included in the model, a significant proportion of variance in the dependent measure was explained, $F_{2,21} = 4.54$, $P = 0.01$, one-tailed. However, NIVQ did not contribute significantly to the prediction of Factor 2 scores over and above the effects of gender ($P = 0.50$, one-tailed), whereas the effect of gender remained significant ($\beta = -0.55$, $P = 0.029$, one-tailed, partial $\eta^2 = 0.16$). A similar pattern was observed when the OWLS standard score was used. When both gender and OWLS standard score were included, the overall model was significant, $F_{2,21} = 4.67$, $P = 0.01$, one-tailed. However, OWLS standard score did not contribute significantly to the prediction of Factor 2 scores over and above the effects of gender ($P = 0.33$, one-tailed), but the effect of gender remained significant ($\beta = -0.48$, $P = 0.029$, one-tailed, partial $\eta^2 = 0.16$). Taken together, these results suggest that gender differences in Factor 2 were not explained simply by differences in non-verbal cognitive ability or expressive language ability.

Discussion

This study was designed to investigate gender differences in repetitive language among individuals with fragile X syndrome. Towards that end, we examined the occurrence of utterance-level repetition, topic repetition and conversational device repetition during narration and conversation. The relative contributions of cognitive and expressive language ability and gender to the occurrence of repetitive language were also investigated.

The overall rate of self-repetition found in the present study is consistent with findings from previous studies of fragile X syndrome (e.g. Ferrier *et al.* 1991; Sudhalter *et al.* 1991). Sudhalter *et al.* (1991), for example, found that approximately 19% of the utterances produced by males with fragile X syndrome in conversation with an adult contained repetitions. Their work collapsed across utterance-level, and topic repetition. If we do the same, our percentage is similar (i.e. 26%).

Gender differences in repetitive language emerged only for conversational device repetition. Conversational devices control the flow of the interaction without contributing substantively to the topic, and include rote phrases or sayings, such as 'that's a wrap', 'right on', 'that is interesting', 'that's it'. Compared with females, males in the present sample were more likely to repeat conversational devices. This finding suggests that males may rely more heavily than females on rote phrases or expressions in their expressive language regardless of whether they are telling a story or carrying on a conversation. In addition, the results of the regression analyses suggest that gender may make an independent contribution to the occurrence of conversational device repetition, such that the gender difference is not simply the result of correlated differences in non-verbal cognitive ability or expressive language ability.

At present, it is unclear what factor or factors underlie the observed gender difference in conversational device repetition. However, possible mechanisms may arise from gender differences in aspects of cognitive ability, such as working memory or executive function that were not fully captured by non-verbal IQ in the present study. For example, the language tasks used in the present study require synthesizing visual and verbal cues, as well as formulating verbal responses and tracking the progression of the conversation, or story. Simultaneously remembering and manipulating the cognitive, linguistic and social aspects of these tasks may place demands on working memory that lead to an increased reliance on rote phrases or expressions as a means by which to facilitate the discourse. Although females with fragile X have difficulty with aspects of executive function and working memory (e.g. Cornish *et al.* 2004; Kirk *et al.* 2005), they may be less impaired than males, and so may rely less on conversational devices.

Alternatively, physiological characteristics, such as differences in arousal in response to social stimulation, may contribute to the observed gender difference in conversational device repetition in addition to, or instead of, executive function. Although both males and females experience elevated arousal in social situations, difficulty with arousal is especially pronounced among males (Hessl *et al.* 2002). Elevated arousal is associated with other behaviours, such as gaze aversion (Hall *et al.* 2006; Hessl *et al.* 2006), and may also be related to language characteristics, including tangential and perseverative language, at least in males with fragile X syndrome (Belser & Sudhalter 1995).

In contrast to the results for conversational device repetition, no significant gender differences were found for utterance-level or topic repetition. One interpretation is that these latter two types of repetition result from different underlying mechanisms than do conversational device repetitions. Indeed, the principal components analyses indicated that utterance-level and topic repetition were more closely correlated with each other than with conversational device repetition. Further, conversational device repetition displayed different patterns of correlation with gender, cognitive ability and expressive language ability than did utterance-level and topic repetition. Thus, the pattern of results for utterance-level, topic and conversational device repetition together suggests that different mechanisms may account for the occurrence of different types of repetition and these mechanisms may be differentially affected in males and females.

It should be noted at this point that one limitation of the present study is that we did not distinguish between males with only the full mutation and those who are mosaic in this study. Differences in repetition between males and females may thus have been attenuated by the inclusion of males with mosaicism compared to those with the full mutation-only. That is, men who are mosaic may experience fewer effects of the syndrome owing to the production of the protein associated with fragile X (FMRP) in unaffected cells (Tassone *et al.* 1999). Moreover, it is possible that the gender differences observed should be reinterpreted simply as an effect of FMRP levels. In other words, it is possible that differences in repetition arise as a function of protein level rather than gender or genetic status *per se*. Although the limited sample size in the present study precluded the inclusion of genetic status in the analyses, as a group, the male participants with mosaic fragile X syndrome displayed higher non-verbal IQ and less utterance-level repetition than did males with the full mutation. Future research aimed at exploring the differences within males with fragile X syndrome based on mosaic status, as well as the relationship between FMRP levels and repetition in both males and females will help to clarify the factors contributing to perseveration in fragile X syndrome.

In addition to differences in repetition as a function of gender, differences were also observed based on the context in which the language sample was taken. In particular, topic repetition occurred more frequently in conversation than in narration. This pattern of results is consistent with the notion that language characteristics can vary as a function of the processing and social demands associated with the sampling context (Abbeduto *et al.* 1995). Thus, context differences in topic repetition may reflect the different demands of narration vs. conversation. For example, narrating a story from a book compared with conversing about topics may require less interpersonal interaction, such as eye contact, which is associated with elevated arousal and avoidance among males with fragile X (Belser & Sudhalter 1995).

An alternative explanation of the context effect is that the presence of a book from which to narrate reduces information processing demands by providing visual cues absent from conversation. Contextual differences in repetition may then be related to the extent to which the situation provides structure or cues to aid performance. It is also possible that narrating a story provides sufficient structure to minimize the influence of the executive function deficits associated with fragile X syndrome relative to conversation, which is more influenced by executive function deficits because it has less inherent structure.

Future research can test these hypotheses by manipulating the interpersonal or processing demands of the language-sampling context. Examination of the source of contextual differences in repetition may inform our understanding of the underlying factors in perseveration as well as routes for intervention. For example, it may be helpful for clinicians and others who work with individuals with fragile X syndrome to know that context can influence the types of language characteristics observed, particularly topic repetition. Reliance on only one context for language sampling may result in a misrepresentation of repetitiveness. Therefore, evaluating language characteristics, such as excessive self-repetition, in multiple contexts may be informative.

It is important to acknowledge that another limitation of the present study is that we defined topic as a linear, turn-by-turn process at the level of the utterance, which may not sufficiently reflect or adequately capture how topics are introduced and maintained throughout discourse. As a result, it was occasionally difficult to determine the topic of a given utterance. This difficulty is reflected in the acceptable, but somewhat lower, reliability scores for topic repetition compared with utterance-level repetition. Alternative ways of measuring topic that focus on the multifaceted nature of topic may better capture how topics are introduced, maintained, or repeated in the discourse of individuals with fragile X syndrome.

In addition, lack of additional gender differences may be the result of limited statistical power to detect differences because of the small sample size, and especially the limited number of females in the present study. Replicating these findings among larger samples will contribute towards understanding the mechanisms underlying different types of repetition and the relative contribution of gender to their occurrence. Moreover, although not included in the present study, comparisons between typical development and fragile X syndrome can inform the extent to which repetition is commensurate with developmental age and language ability or is, instead, an especially problematic domain. For example, studies of conversation in typical development suggest that repetition is a characteristic of discourse that functions as a strategy for maintaining engagement (Dorval & Eckerman 1984; Brinton & Fujiki 1984). It may also reflect difficulty with sentence formulation or the demands of real-time communication.

Despite consistency across studies in rate of repetition, neither previous studies nor our study have addressed the point at which repetition 'becomes' perseveration at a clinical level. As a result, the conclusions drawn reflect the nature and extent of repetition among individuals with fragile X, not a clinical assessment of perseveration. It is important that future research address the validity and clinical significance of coding systems like the one applied here. Regardless, improved understanding of repetition in fragile X syndrome is necessary before it is possible to thoroughly study perseveration and its causes. Addressing the extent and nature of repetition contributes to the overall conceptualization of the fragile X syndrome's impact on language and the factors that mediate and moderate these effects, while providing information about the etiology of language challenges critical for informing intervention strategies.

Acknowledgements

This research was supported in part by NIH grants R01 HD24356 and P30 HD03352. The work was conducted as part of the first author's masters thesis at the University of Wisconsin-Madison. The authors gratefully acknowledge Selma Karadottir, Susen Schroeder, Elissa Jones, and Katherine Verberkmoes for their assistance. We also wish to thank the young people and their families who so kindly participated in this research.

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Accepted 25 May 2006

Appendix

Examples of repetition categories. Note that in each example, the repetition is *underlined*. All repetition coded was *self-repetition*.

1. *Utterance-Level Repetition*.

A. A repetitive word:
  a. Within utterance: Or, <u>or, or</u> I think he's trying to . . .
  b. Between utterances: Happy! <u>Happy.</u>

B. A repetitive phrase:
  a. Within utterance: Maybe I <u>Maybe I</u> just can't read.
  b. Between utterances: He went to the store. <u>The store</u>.

C. A repetitive dependent clause:
  a. Within utterance: After the movie <u>After the movie</u>, we went for ice cream.
  b. Between utterances: The frog who jumped into the glass. <u>Who jumped into the glass.</u>

D. A *repetitive utterance*: Because of the definition of utterance, all repetitions of independent clauses had to be complete utterances. As a result, these repetitions always occurred between utterances, for example, 'They must have fainted too. <u>They must have fainted.</u>'

2. *Topic repetition*: Topic repetitions could be (a) concentrated (i.e. occur in successive utterances) or (b) interspersed through out the language sample. In the following examples, 'E' indicates and examiner's utterance, whereas 'C' indicates the child's utterance.

a. Consecutive Topic Repetitions:
  i. Conversation: Here C3 and C5 both repeat the topic introduced in C2
    E. Tell me how you play [baseball].
    C1. Well, all the rules are simple.
    C2. Well, I don't know any rules about it.
    C3. <u>But I don't know any rules about</u> (utterance abandoned).
    C4. I mostly don't want it.
    C5. <u>I really don't know much of the rules . . .</u>
  ii. Narration: Here C3 repeats the topic introduced in C1.
    C1. And the frog is going in the boys pocket.
    C2. No [not] in pants pocket, but his coat pocket.
    C3. <u>The frog just jumps in the pocket.</u>

b. Interspersed Topic Repetitions:
  i. Conversation: Here C3 repeats the topic introduced in C1.
    E. Tell me why you like doing that [reading and math].
    C1. . . . I've got to get used to this new school I've been going to.
    Later in the conversation.
    E. You don't want to have to mention the name [of your favourite teacher].
    C2. <u>. . . So I haven't gotten used to this new place [school].</u>

ii. Narration: Here C3 repeats the topic introduced in C1.
Page 2: C1. The frog has to come in to the coat and go to the store.
Page 2: C2. He is going to the store.
Page 5: C3. I think they are going to the store to buy groceries.

3. *Conversational Devices*: Any utterance that does the mechanics of the interaction, but does not have semantic content (e.g. the same utterance repeated through the transcript). Examples include: I don't know, that's it, interesting, that's a wrap, next page.