

Receptive Language Skills of Adolescents and Young Adults With Down or Fragile X Syndrome

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Abstract

We investigated the receptive language of adolescents and young adults with Down syndrome ($n = 25$) or fragile X syndrome ($n = 19$). We were interested in syndrome differences and gender differences within fragile X. Comparison of the syndromes and MA-matched typically developing children ($n = 24$) revealed that individuals with the syndromes differed in relative achievements across the domains of receptive vocabulary, receptive syntax, and nonverbal cognition as well as in the organization of their linguistic knowledge. Comparison of males and females with fragile X revealed that each displayed synchronous development across the three domains, despite the fact that the receptive language levels of females surpassed that of males.

Down syndrome and fragile X syndrome are the two most common genetic causes of mental retardation (Dykens, Hodapp, & Finucane, 2000). Each syndrome is associated with a range of physical and behavioral sequelae. Most individuals with Down syndrome function in the mild to moderate range of mental retardation (Chapman & Hesketh, 2000). The range of affectedness is broader in fragile X syndrome. Males with the full mutation of the fragile X gene (FMR1) typically display IQs in the mild to moderate range of mental retardation (Hagerman, 1999). Females with the full fragile X mutation can have mental retardation, a learning disability, or social adjustment difficulties without cognitive effects (Mazzocco, 2000). Language problems are almost invariably associated with Down syndrome (Chapman & Hesketh, 2000). Although less well-studied, language problems have also been documented in fragile X (Schopmeyer, 1992). In this article, we report on a study of the receptive language skills of adolescents and young adults with Down syndrome or fragile X syndrome. We focused on receptive language because it is understood less well than the expressive aspects of language, particu-

larly in relation to fragile X syndrome (Abbeduto & Hagerman, 1997). Our goals were (a) to determine whether the two syndromes are characterized by different receptive language profiles and (b) to characterize within-syndrome variation in fragile X. Pursuance of these goals will contribute to our knowledge of the behavioral phenotypes of the two syndromes and provide guidelines for intervention.

Speech and language problems may be among the most salient and limiting challenges facing individuals with Down syndrome (Chapman & Hesketh, 2000). These individuals are substantially delayed in the age at which they produce their first spoken words (Berglund, 2001). Even after they begin producing words, they make very slow progress in acquiring new skills in virtually all domains of expressive language (Fabbretti, Pizzuto, Vicari, & Volterra, 1997). Early in development, receptive language appears to be less problematic than expressive language. Indeed, most young children with Down syndrome display levels of receptive language commensurate with measures of their nonlinguistic cognitive growth, such as nonverbal MA (Miller, 1999). By

adolescence, however, comprehension of syntax lags behind nonverbal MA (Chapman, Schwartz, & Kay-Raining Bird, 1991; Rosin, Swift, Bless, & Vetter, 1988). The comprehension of vocabulary, however, continues to keep pace with, or even exceed, nonverbal MA during adolescence and young adulthood (Chapman et al., 1991; Rosin et al., 1988). Receptive syntax and vocabulary are also predicted by partly different sets of variables during adolescence and young adulthood (Chapman et al., 1991), which suggests that the syntactic and lexical difficulties of individuals with Down syndrome may result in part from different mechanisms. As a result, the nature of the interventions targeting these domains might need to be different as well. In this study, we were interested in determining whether the pattern of relationships among receptive syntax, receptive vocabulary, and nonverbal MA that characterizes adolescents and young adults with Down syndrome also characterizes those with fragile X syndrome.

Males with fragile X are delayed in many domains of speech and language (Pennington, O'Connor, & Sudhalter, 1991; Schopmeyer, 1992). These delays increase in magnitude relative to age-matched typically developing peers during late childhood and adolescence (Fisch et al., 1999; Freund, Peebles, Aylward, & Reiss, 1995; Prouty et al., 1988; Roberts, Mirrett, & Burchinal, 2001). Most studies of males have been focused on impairments in language expression, particularly impairments thought to reflect the influence of social anxiety and hyperarousal (e.g., Belser & Sudhalter, 1995, 2001; Ferrier, Bashir, Meryash, Johnston, & Wolff, 1991; Sudhalter, Cohen, Silverman, & Wolf-Schein, 1990). There are, however, few data on the receptive language skills of males with fragile X and fewer still on the relation between receptive language and nonverbal cognition or between the lexical and syntactic domains of receptive language (Abbeduto & Hagerman, 1997). Such data are critical for developing interventions for fragile X that can target the areas of greatest challenge or exploit areas of relative strength when trying to impart new skills (Hodapp & Dykens, 2001; Hodapp & Fidler, 1999).

The few studies of males with fragile X that have been focused on the relation between receptive language and cognition or between different domains of receptive language (Madison, George, & Moeschler, 1986; Paul, Dykens, Satson, Breg, & Cohen, 1987) have yielded findings that are contradictory or difficult to interpret. In part,

these ambiguous findings are due to the use of small and restricted samples (i.e., a single family in the Madison et al. study and 12 institutionalized adults in the Paul et al. study). Interpretive difficulties also arise, however, because of the failure to include a typically developing comparison group. Such a comparison is needed when each domain of interest is assessed using a different standardized test (Mervis & Robinson, 1999), as was the case in the Madison et al. and Paul et al. studies. The individuals on which different tests have been normed can differ in potentially important (and unknown) ways, including in average level of ability. This makes it impossible to know what scores would be achieved on the different tests by the "average" child. As a result, it is not clear what magnitude of difference between scores on the tests is needed to conclude that there is an asynchrony (i.e., a difference in relative level of difficulty) across domains for the syndrome group. In the present study, therefore, comparisons were made not only between fragile X and Down syndrome but also between the syndrome groups and a group of developmental-level-matched typically developing children.

Although there is much we do not know about the language problems of males with fragile X, even less is known about the language of females with fragile X. Moreover, in existing studies researchers have focused on expressive language (e.g., Benetto & Pennington, 1996; Mazzocco, Pennington, & Hagerman, 1993; Sobesky et al., 1996), with two exceptions. First, Madison et al. (1986) reported on the receptive language skills of several females from a single family; however, the small sample size and wide variability in age and level of functioning across the participants make generalization impossible. Second, Simon, Keenan, Pennington, Taylor, and Hagerman (2001) investigated discourse comprehension in high-functioning females with fragile X. These investigators found that females with the full mutation had difficulty selecting appropriate humorous endings for stories that they read, which suggested that they had problems computing coherent representations for the stories. This finding is particularly noteworthy because the women studied were functioning in the normal range of intelligence. It is reasonable to expect more serious comprehension problems, and with more "basic" facets of language (e.g., vocabulary and syntax), in females with fragile X who are also cognitively challenged. Unfortunately, there are no data to evaluate this

expectation. As a result, clinicians and educators who provide services to females with fragile X cannot turn to empirical data for the decisions they make about assessment or intervention but must rely, instead, on data from affected males. In the present study, therefore, we included females as well as males with fragile X in order to determine whether females differed from males in the extent of their receptive language impairments or in the pattern of relations among domains. However, because only a small number of females with fragile X participated, the results of these comparisons should be viewed as preliminary and in need of replication.

In summary, the present study was designed to answer two questions. The first question was: Are Down syndrome and fragile X syndrome characterized by similar relationships among receptive syntax, receptive vocabulary, and nonverbal MA during the adolescent and young adult years? We addressed this question by examining (a) the relative developmental levels achieved on measures of these domains by adolescents and young adults with Down syndrome, adolescents and young adults with fragile X syndrome, and cognitively matched typically developing children and (b) the correlations among these measures within each group. The second question was: Are there gender differences in receptive language among individuals with fragile X syndrome? We addressed this question by comparing males and females with fragile X in terms of (a) the extent of their delays in receptive language, (b) their relative delays in receptive language and nonverbal cognition, and (c) their relative delays in the lexical and syntactic domains of receptive language.

Method

Participants

Three groups were included: adolescents and young adults with Down syndrome ($n = 25$), adolescents and young adults with fragile X ($n = 19$), and typically developing 3- to 6-year-olds ($n = 24$). Participants with Down syndrome or fragile X were recruited through advertisements in local newspapers, announcements in the newsletters of regional and national organizations focused on developmental disabilities, postings on the Internet, a university-based registry of families having a son or daughter with a disability, and mailings to local special educators and genetics clinics. Because of differences in prevalence, participants

with fragile X were recruited from a wider geographical region than were those with Down syndrome. All participants with Down syndrome or fragile X lived at home with their parents or legal guardians. Typically developing children were recruited locally through preschools, notices posted in public places, and a university-based registry. Parents of typically developing children indicated that the child had no diagnosed disability and that he or she was not receiving special education services other than speech therapy at the time of the study.

The participants for this study were drawn from a larger pool of 87 individuals recruited into the project. Excluded from the present study were individuals who were unable to complete the tests of interest, individuals with Down syndrome or fragile X whose nonverbal IQs on the test we administered (described subsequently), were above 70, and typically developing children whose nonverbal IQs on the test we administered were below 80. We also excluded any individual whose nonverbal MA on the test we administered was outside the range of nonverbal MAs for each of the other groups, thereby ensuring that we could arrive at a groupwise nonverbal MA match. In addition, all participants were screened using the Autism Behavior Checklist (Krug, Arick, & Almond, 1980). Those who met screening criteria for autism were referred to a clinical psychologist for a follow-up evaluation. The psychologist judged that two of the five individuals referred to her met DSM-IV criteria for autism. These two individuals, both of whom had fragile X, were excluded. Together these exclusionary criteria resulted in the final sample of 68 participants.

Characteristics of the participants are provided in Table 1. The participants in the three groups were selected such that they were matched groupwise on nonverbal MA, which was determined by administering subtests (described subsequently) from the Stanford-Binet, 4th edition (Thorndike, Hagen, & Sattler, 1986), $F(2, 65) = .02$, $p = .98$. The participants with Down syndrome and those with fragile X were also selected such that they were matched groupwise on both nonverbal IQ (determined from the Stanford-Binet), $t(42) = 1.23$, $p = .23$, and chronological age, $t(42) = .70$, $p = .49$. There was no difference between the Down syndrome and fragile X groups with regard to the percentage of participants who had mothers (or female guardians) with college or advanced degrees, $\chi^2(1, N = 68) = .33$, $p = .57$. In contrast,

Table 1. Participant Characteristics by Group

Characteristic	Group ^a					
	DS (n = 25)		FXS (n = 19)		TD (n = 24)	
	Mean ^b	SD	Mean	SD	Mean	SD
Nonverbal MA (in years) ^c	4.8	1.1	4.9	1.6	4.8	1.0
Nonverbal IQ ^c	40.7	6.2	43.7	10.0	97.7	10.3
CA (in years)	16.6	3.1	16.0	3.3	4.9	0.7
No. of mothers with college degrees	12	—	11	—	23	—
No. of Caucasians	21	—	17	—	24	—
No. of males	12	—	13	—	6	—

^aDS = Down syndrome, FXS = fragile X syndrome, TD = typically developing. ^bOr number, where indicated. ^cBased on administration of the Pattern Analysis, Copying, and Bead Memory subtests of the Stanford-Binet Intelligence Scale, 4th ed.

all but one of the mothers of the typically developing children had earned a college degree. Maternal education within the syndrome groups, however, was not significantly related to any of the dependent measures, and, thus, this variable was not considered further. Ninety-four percent of the participants were Caucasian. The proportion of males varied across groups, $\chi^2(2, N = 41) = 8.15, p = .017$. This variation in gender was due in large part to the higher proportion of males in the group with fragile X and is consistent with prevalence estimates for the syndrome.

Hearing was evaluated for each participant by determining pure tone air-conduction thresholds across the "speech" frequencies of 500 Hz, 1000 Hz, and 2000 Hz. Consistent with the exclusionary criterion of Chapman et al. (1991), no participant had more than a mild hearing loss. Mean thresholds, however, differed significantly across groups, $F(2, 63) = 17.04, p \leq .0005$, reflecting the fact that the group with Down syndrome had a higher threshold than did either of the other groups, who did not differ. Thresholds were not correlated with the dependent measures for any group, so this variable is not considered further.

The parents/guardians of all participants with Down syndrome reported the etiology as being trisomy 21 (rather than mosaic or translocation). We were able to obtain reports confirming the karyotype for 19 of these 25 participants. DNA confirmation of the full mutation was available for all but one of the participants with fragile X, with cytogenetic confirmation available for him. Three of the males with fragile X were described

in the confirming reports as mosaic. Within the fragile X group, two families each had 2 affected children who participated, and one family had 3 affected children who participated.

Twelve of the participants with fragile X and 5 of those with Down syndrome were regularly receiving medications to control problem behavior, especially hyperactivity and anxiety (e.g., Ritalin, Prozac). If parents asked, we instructed them to administer medications on the day(s) of the visits as though it were a typical school day. This decision was made so that we could evaluate the participants under conditions that would allow generalization to their typical behavior in school and other important settings in their lives. Medication status (i.e., on/off) was not related to any of the dependent measures.

Characteristics of the participants with fragile X are presented as a function of gender in Table 2. The 13 males and 6 females differed significantly on nonverbal MA, $t(17) = 5.62, p < .0005$, and nonverbal IQ, $t(17) = 6.01, p < .0005$, but not age. These differences are consistent with previous findings that males are affected more than females (Hagerman, 1999).

Materials

Measure of receptive language. The Test for Auditory Comprehension of Language-Revised (Carroll-Woolfolk, 1985) was administered. In this test, the participant responds by pointing to the one drawing of three that matches the meaning of a word, phrase, or sentence spoken by the examiner. The 120 items are organized into three

Table 2. Characteristics of Participants With Fragile X by Gender

Characteristic	Males (n = 13)		Females (n = 6)	
	Mean	SD	Mean	SD
Nonverbal MA (in years) ^a	4.0	1.1	6.8	0.7
Nonverbal IQ ^a	38.2	5.7	55.5	6.1
CA (in years)	16.4	3.7	15.1	2.1

^aBased on administration of the Pattern Analysis, Copying, and Bead Memory subtests of the Stanford-Binet Intelligence Scale, 4th ed.

subtests: Word Classes & Relations measures vocabulary comprehension, whereas Grammatical Morphemes and Elaborated Sentences are syntactically oriented. The Grammatical Morphemes subtest measures comprehension of inflectional and derivational morphology (e.g., the plural *-s* and past tense *-ed*). The Elaborated Sentences subtest measures comprehension of basic clause and multiclausal patterns (e.g., passive sentences, such as *The boy is chased by the girl*). The Test for Auditory Comprehension of Language was normed on 1,003 children ages 3 to 9.92 years, but is appropriate for adolescents with language problems, including those with mental retardation (Chapman et al., 1991). In the standardization sample, split-half reliabilities for the total score ranged from .88 to .97 across ages, and test-retest reliability was .95 (Carrow-Woolfolk, 1985). The test also discriminates children with and without diagnosed comprehension problems (Carrow-Woolfolk, 1985).

Age-equivalent scores for the test overall (total scores) and for each subtest (subtest scores) served as the primary dependent variables. Standard scores were also computed, but these have a floor of 65, which is only 2 standard deviations (*SDs*) below the mean. The majority of individuals with mental retardation would be expected to score more than two *SDs* below the mean for their age (Rosenberg & Abbeduto, 1993) and, thus, standard scores have limited sensitivity for the populations of interest here.

Measure of nonverbal cognition. We derived the nonverbal IQs and MAs described previously by administering the three subtests from the Stanford-Binet, 4th edition (Thorndike et al., 1986): Bead Memory, Pattern Analysis, and Copying. Administration requires minimal verbal instruc-

tions and the participant responds nonverbally. There are several advantages of these subtests. First, they yield measures of cognition that are minimally influenced by language ability, which is critical for estimating the developmental synchrony of the two domains (Rosenberg & Abbeduto, 1993). Second, they include, but are not limited to, an assessment of short-term memory (i.e., Bead Memory). Chapman et al. (1991) have argued that short-term memory is important for language comprehension and, therefore, should be represented in any measure used for matching in studies of receptive language, although it is not included in many popular tests of nonverbal intelligence. Third, although the subtests used in the present study require the participant to manipulate blocks and beads, the majority of the items are un-timed and manageable for the level of fine-motor skill of the participants in this study. More generally, there is no evidence to suggest that the relative difficulty of the three subtests would differ between the two syndrome groups (see Batshaw, 1997, for a review of the relevant literatures). Indeed, the fragile X and Down syndrome groups did not differ in their mean age-equivalent scores on any of the subtests. According to Thorndike et al. (1986), internal-consistency reliabilities for three subtests over the developmental levels represented in the present study are above .80, and mean test-retest reliabilities are generally above .60. Previous factor analyses have also demonstrated that the three subtests load highly (at .60 or greater) on *g* (Thorndike et al., 1986).

Each subtest yields a standard score and an age-equivalent score. The mean standard score for the three subtests provided the estimate of nonverbal IQ and the mean of the age-equivalents from the subtests provided the estimate of nonverbal MA.

Procedure

Participants were tested individually in a quiet laboratory room at a university research center. The Test for Auditory Comprehension of Language and Stanford-Binet, 4th edition, subtests were part of a longer research protocol designed to investigate the linguistic and cognitive profile of individuals with Down syndrome or fragile X. Subtests for both instruments were administered in separate sessions, with the former test administered first. The sessions were conducted on different days whenever possible. If parents felt it appropriate, the sessions were administered on

one day with a 1- to 2-hour break between them. The mean number of days between administration of the Test for Auditory Comprehension of Language and Stanford-Binet was 6.8 days. Hearing was tested on the same day that the Test for Auditory Comprehension of Language was administered. For any given participant, the same examiner administered the entire protocol. Across the entire sample, five examiners were involved.

Results

Diagnostic Group Analyses

Age-equivalent total scores from the Test for Auditory Comprehension of Language are presented for each group in Figure 1. These scores were analyzed in a one-way ANOVA, with group (Down syndrome, fragile X syndrome, typically developing) as a between-participants factor. The effect of group was significant, $F(2, 65) = 3.08, p = .05$. Post-hoc contrasts (using Fisher's least significant difference technique to maintain familywise alpha at .05—Levin, Serlin & Seaman, 1994) indicated that the group effect was due to the fact that the mean age-equivalent score for the participants with Down syndrome was significantly lower than that for the participants with fragile X. The difference between the Down syndrome and typically developing groups just failed to reach sig-

nificance. The effect of group, $f = .25$, was of medium size (Kirk, 1995). Despite the relatively limited variability in standard scores from the Test for Auditory Comprehension of Language, the trends for those scores were consistent with those observed for the age-equivalent scores with regard to the difference between the participants with Down syndrome and those with fragile X.

Age-equivalent scores from each subtest of the Test for Auditory Comprehension of Language are presented by group in Figure 2. These scores were analyzed in a Group (Down syndrome, fragile X syndrome, typically developing) \times Subtest (Word Classes & Relations, Grammatical Morphemes, Elaborated Sentences) ANOVA, with repeated measures on the last factor. This analysis yielded a main effect of subtest, $F(2, 130) = 8.91, p \leq .0005$, and a Group \times Subtest interaction, $F(4, 130) = 2.56, p = .04$. Simple effects tests (with .05 familywise alpha) indicated that the age-equivalent scores varied significantly across subtests, but only for the participants with Down syndrome. Post-hoc contrasts (with .05 familywise alpha) indicated that, for the participants with Down syndrome, age-equivalent scores were significantly higher for the Word Classes & Relations subtest than for either of the other subtests. The Group \times Subtest effect, $f = .30$, was of medium size (Kirk, 1995). Despite relatively limited variability, the trends for the standard scores from the Test for Auditory Comprehension of Language were consistent with those observed for the age-

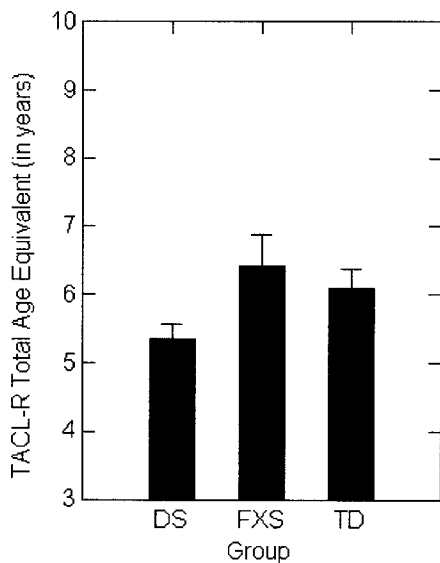


Figure 1. Test for Auditory Comprehension of Language-Revised. Total test age-equivalent scores by group. DS = Down syndrome, FXS = fragile X syndrome, TD = typically developing.

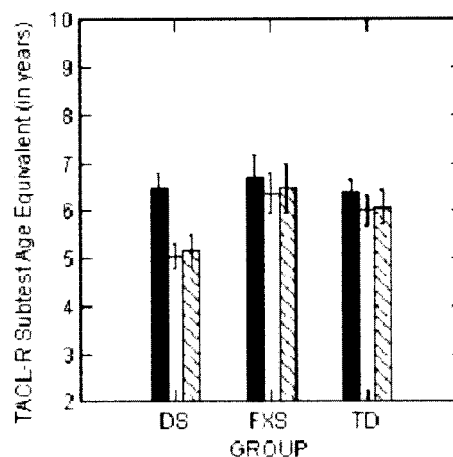


Figure 2. Test for Auditory Comprehension of Language-Revised: subtest age-equivalent scores by group. Black bars = Word Classes & Relations, white bars = Grammatical Morphemes, hatched bars = Elaborated Sentences.

equivalent scores with regard to group differences in the pattern of performance across subtests.

In addition to examining group differences in the levels of performance achieved on the various subtests of the Test for Auditory Comprehension of Language, we also investigated the patterns of within-group correlations among the various measures (see Table 3). The simple (zero-order) correlations among measures are presented above the diagonal in each panel of the table. The correlations below the diagonal are the correlations among the Test for Auditory Comprehension of Language subtests with the contribution of nonverbal MA partialled out of the relationship. In evaluating the zero-order correlations, we used an alpha level of $\leq .008$ for the significance of any individual correlation in order to maintain a .05 familywise alpha level. As can be seen from Table 3, all of the zero-order correlations involving nonverbal MA and the Test for Auditory Comprehension of Language subtests were significant for the fragile X and the typically developing groups. In the case of the Down syndrome group, the zero-order correlations between nonverbal MA and each of the Test for Auditory Comprehension of Language subtests were significant, but only

one of the zero-order correlations among these subtests was significant. In evaluating the partial correlations, we required an alpha level of .017 for the significance of any individual correlation in order to maintain a .05 familywise alpha level. As can be seen from Table 3, none of the partial correlations was significant for the Down syndrome group. In contrast, the partial correlation between the Word Classes & Relations and the Elaborated Sentences subtests was significant for the fragile X group and the partial correlations between Elaborated Sentences and each of the other subtests were significant for the typically developing group.

Comparisons of Males and Females With Fragile X

The age-equivalent scores from the Test for Auditory Comprehension of Language for the participants with fragile X were examined according to gender in three analyses. In the first, females were found to have higher total age-equivalent scores than did males ($M_s = 8.69$ and 5.37 , respectively, $t(17) = 6.23$, $p \leq .0005$). In the second analysis, a difference score was computed by

Table 3. Simple and Partial Correlations Among TACL-R Subtests and Nonverbal MA by Group

Group/Measure	Nonverbal MA	WC & R	Grammatical morphemes	Elaborated sentences
Down syndrome				
Nonverbal MA	—	.63*	.47*	.56*
WC & R		—	.40	.43
Grammatical morphemes		.14	—	.47*
Elaborated sentences		.12	.29	—
Fragile X				
Nonverbal MA	—	.84*	.70*	.83*
WC & R		—	.62*	.86*
Grammatical morphemes		.09	—	.61*
Elaborated sentences		.53**	.08	
Typically developing				
Nonverbal MA	—	.68*	.54*	.69*
WC & R		—	.54*	.72*
Grammatical morphemes		.29	—	.79*
Elaborated sentences		.48**	.68**	—

Note. Simple (zero-order) correlations appear above the diagonal. Partial correlations (with nonverbal MA partialled out) appear below the diagonal. TACL-R = Test for Auditory Comprehension of Language-Revised, WC & R = Word Classes & Relations.

* $p < .008$, one-tailed. ** $p < .017$.

subtracting the nonverbal MA for each participant from his or her age-equivalent total score on the Test for Auditory Comprehension of Language. We found that females and males did not differ significantly on their mean difference scores ($M_s = 1.9$ and 1.3 , respectively). In the third analysis, a Subgroup (males, females) \times Test for Auditory Comprehension of Language Subtest repeated-measures ANOVA yielded only an effect of subgroup, $F(1, 17) = 28.41$, $p \leq .0005$. The means for Word Classes & Relations, Grammatical Morphemes, and Elaborated Sentences were, respectively, 8.6, 8.0, and 9.1 years for females and 5.8, 5.6, and 5.3 years for males.

Discussion

Although the development of receptive language in individuals with Down syndrome has been reasonably well-characterized, little is known about the extent and nature of receptive language problems in fragile X. In the present study, we were interested in determining whether different patterns of relationships among receptive syntax, receptive vocabulary, and cognition characterize (a) Down syndrome and fragile X and (b) males and females with fragile X. The comparison of Down syndrome and fragile X can yield important data on the behavioral consequences of the genetic anomalies underlying the two syndromes, thereby providing a foundation for assessment and intervention strategies that are more closely tailored to the needs of affected individuals (Hodapp & Dykens, 2001; Hodapp & Fidler, 1999). The comparison of males and females with fragile X can elucidate the extent, nature, and causes of within-syndrome variability in receptive language for fragile X, thereby providing clinically useful information (Abbeduto & Hagerman, 1997).

The first question we addressed was: Are Down syndrome and fragile X syndrome characterized by different relationships among receptive syntax, receptive vocabulary, and nonverbal MA during the adolescent and young adult years? In addressing this question, we examined mean levels of performance across the three nonverbal MA-matched groups and within-group correlations among the domains. In terms of mean levels of performance, we found that individuals with Down syndrome achieved total scores on the Test for Auditory Comprehension of Language that were significantly lower than those of the MA-matched participants with fragile X and margin-

ally lower than those of the typically developing comparison children. This result is consistent with those of previous studies (Chapman et al., 1991; Rosin et al., 1988) in suggesting that by adolescence, individuals with Down syndrome find receptive language more challenging than would be expected based on their levels of nonverbal cognitive development. We also found that the participants with Down syndrome achieved scores on the Test for Auditory Comprehension of Language that were higher for the Word Classes & Relations subtest than for either the Grammatical Morphemes or the Elaborated Sentences subtest. This result is consistent with the findings of Chapman et al. (1991) in suggesting that the comprehension of syntax is more challenging than the comprehension of vocabulary for adolescents and young adults with Down syndrome. Taken together, these results suggest that language intervention for adolescents and young adults with Down syndrome must devote considerable attention to increasing the syntactic capabilities of affected individuals and that such interventions may need to be more intense than those targeting lexical skills.

In contrast to individuals with Down syndrome, those with fragile X (a) did not differ from the typically developing comparison group in their age-equivalent total scores on the Test for Auditory Comprehension of Language and (b) displayed similar age-equivalent scores across the three subtests of this instrument. These results suggest that individuals with fragile X achieve equivalent levels of development in receptive syntax, receptive vocabulary, and nonverbal cognition during the adolescent and young adult years. The straightforward interpretation afforded by the present results relative to previous studies (e.g., Madison et al., 1986; Paul et al., 1987) illustrates the advantage of including an appropriately matched typically developing comparison group in studies of the language development of individuals with fragile X. The present results also suggest that language interventions for youth with fragile X should focus on all areas of receptive language (e.g., syntactic and lexical) rather than singling out one particular facet for more intensive treatment.

In terms of the within-group correlations among measures, we found that both receptive syntax and receptive vocabulary were strongly related to cognitive development (as indexed by nonverbal MA). This was true even for the partic-

ipants with Down syndrome, who displayed an asynchrony between the mean levels of achievement in receptive syntax and vocabulary. Although the direction of causation cannot be unambiguously determined from these correlations, the results are consistent with theories positing that the acquisition and use of language depends in important ways on the maturation of a broader conceptual and information-processing system (Abbeduto, Evans, & Dolan, 2001). At the same time, the existence of substantial correlations between receptive language and nonverbal MA across all three groups, despite variability in their relative achievements in these domains, are difficult to reconcile with Chomskyan claims of the modularity (i.e., independence) of language and cognition (Smith, 1999). The present findings replicate those of previous studies on Down syndrome (Chapman et al., 1991) and provide the first evidence of a strong relationship between receptive language and cognitive development in people with fragile X. More importantly, the results suggest that planners of language intervention for youths with Down syndrome or fragile X may need to identify and treat cognitive limitations that are barriers to the acquisition and use of targeted language skills.

When the contribution of nonverbal MA was statistically removed from the correlations among the Test for Auditory Comprehension of Language subtests, different patterns of relationships emerged across the groups. We hypothesize that these differences reflect variations in the extent to which different forms of knowledge (i.e., the lexical and the syntactic) have been organized into a single system. Increased organization is an important dimension of developmental change (Karmiloff-Smith, 1986). In the typically developing children, scores on the Elaborated Sentences subtest were significantly correlated (after removing the contribution of nonverbal MA) with scores on Word Classes & Relations and on Grammatical Morphemes. This pattern suggests that typically developing 3- to 6-year-olds, as a group, have organized their linguistic knowledge into a coherent system in which achievements in one component (e.g., the lexical) inform and motivate achievements in other components (e.g., the syntactic) and vice versa. Among the adolescents and young adults with fragile X syndrome, only the partial correlation between the Word Classes & Relations subtest and the Elaborated Sentences subtest was significant. This pattern of results suggests that the

fragile X group has also achieved some integration of the different components of language, although they have been less successful in this regard than their typically developing MA-matched peers. In contrast, none of the partial correlations was significant for the participants with Down syndrome, which raises the possibility that their linguistic knowledge may be comprised of poorly organized sets of representations that are only loosely linked. Poorly organized knowledge can be difficult to access under the time constraints of natural language comprehension. Thus, the present results suggest that language intervention for fragile X and especially Down syndrome may need to foster integration and coordination of existing linguistic knowledge as well as the acquisition of new knowledge.

In addition to addressing the question of syndrome differences, we asked: Are there differences between males and females with fragile X in (a) the extent of their delays in receptive language, (b) the relative delays in receptive language and nonverbal cognition, or (c) the relative delays in the lexical and syntactic facets of receptive language? The results suggest that females are less severely challenged in the area of receptive language than are males. This finding is consistent with the results of previous studies of cognitive and behavioral functioning in suggesting that females may be buffered at the level of the phenotype from the full effects of the FMR1 gene (Hagerman, 1999). Despite the gender differences in mean level of performance, the magnitude of the difference between age-equivalent scores on the Test for Auditory Comprehension of Language and nonverbal MAs was not significantly different for males and females with fragile X; moreover, scores did not vary across the different subtests for either males or females. Thus, despite the difference in the severity of their receptive challenges, both males and females with fragile X display a profile characterized by synchrony in their levels of achievement in the language and cognitive domains assessed, at least during the adolescent and young adult years. This, too, is consistent with the results of previous studies in suggesting that the fragile X phenotype is not substantially different across males and females in terms of their profiles of strength and weakness (Dykens et al., 2000). More importantly, this pattern of results suggests that similar types of language interventions may be effective for both genders, despite the differences in the severity of their impairments.

Several directions for future research are suggested by results of the present study. First, these results should be replicated with a new sample of participants. Although the sample size in this study was larger and more diverse than those of previous studies of receptive language in individuals with fragile X, it was still small, especially with regard to the within-syndrome comparison of males and females. Replication of small-sample studies is particularly important in light of the wide range of affectedness in fragile X. Second, future researchers should replicate these results using multiple measures of receptive language rather than relying on a single measure of receptive language as we have here. This is particularly important because task factors are likely to play an important role in the language-processing of individuals with fragile X, perhaps even more than is true for typically developing children because of the problems with attention and social anxiety observed among those with fragile X (Hagerman, 1999). Third, it is important that future investigators measure numerous facets of cognitive ability and examine the relationships among specific cognitive and linguistic achievements. Here we dissected receptive language into its major components, but we relied on only a single summary measure of a rather narrow set of nonverbal cognitive abilities. Fourth, future researchers should focus on exploring the variations in receptive language that are associated with the autism status of individuals with fragile X. In doing so, it will be possible to extend the generalizability of the results to a wider portion of the population of persons with fragile X. Finally, the relatively straightforward interpretation of the results of this study illustrates the advantage of including an appropriately matched typically developing comparison group in studies of the language development of individuals with genetic syndromes.

In conclusion, the present study has yielded new, clinically important, data about differences in receptive language ability between Down syndrome and fragile X and about within-syndrome variability in receptive language among individuals with fragile X. It also has demonstrated the utility of including both another syndrome group and a typically developing comparison group when evaluating behavioral aspects of syndrome phenotypes and of comparing males and females with fragile X directly in studies of language functioning. In addition, we have outlined a series of questions for future researchers to consider when

evaluating language characteristics among groups of individuals with mental retardation.

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