



HHS Public Access

Author manuscript

Am J Ment Retard. Author manuscript; available in PMC 2017 September 25.

Published in final edited form as:

Am J Ment Retard. 2008 May ; 113(3): 214–230. doi:
10.1352/0895-8017(2008)113[214:SNOLAC]2.0.CO;2.

Signaling Noncomprehension of Language: A Comparison of Fragile X Syndrome and Down Syndrome

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Abstract

Signaling noncomprehension of the spoken messages of others was examined for youth with fragile X or Down syndrome in comparison with each other and nonverbal MA-matched typically developing children. A direction-following task was used in which some of the directions were inadequate. Both syndrome groups signaled noncomprehension less often than did the typically developing children. The ability to signal noncomprehension appropriately was related to a measure of receptive vocabulary and syntax. Preliminary analyses indicated that males with fragile

X syndrome signaled noncomprehension less often than did their female peers, even after controlling for differences in nonverbal MA.

For a discourse to be successful, the participants must fulfill the obligations associated with their roles as speaker and listener (Clark, 1996). In the role of listener, a participant must use all available sources of information to construct the speaker's intended meaning. Moreover, the listener must signal when comprehension is not possible so that the speaker can provide clarification. If the listener fails to signal noncomprehension, he or she will find it increasingly difficult to construct an accurate representation of the talk and to make meaningful contributions (Clark & Schaefer, 1989). Individuals with intellectual disabilities often fail to signal noncomprehension (Abbeduto, Davies, Solesby, & Furman, 1991; Abbeduto, Short-Meyerson, Benson, & Dolish, 1997; Abbeduto, Short-Meyerson, Benson, Dolish, & Weissman, 1998; Ezell & Goldstein, 1991; Fujiki & Brinton, 1993). There is, however, considerable within-group variability in most domains of language use, including noncomprehension signaling (Abbeduto & Rosenberg, 1980; Bedrosian & Prutting, 1978). The causes and correlates of such variability are poorly understood (Abbeduto & Hesketh, 1997). In this study, we examined the possibility that the nature and extent of problems in noncomprehension signaling vary with etiology by focusing on Down syndrome and fragile X syndrome, the two most common genetic causes of intellectual disabilities (Dykens, Hodapp, & Finucane, 2000). We also examined the sources of between- and within-syndrome differences in non-comprehension signaling. Such data can provide the foundation for language interventions designed to meet the unique needs of the individual with intellectual disabilities (Dykens et al., 2000; Hodapp & Fidler, 1999; Murphy & Abbeduto, 2005).

Signaling noncomprehension requires that the listener continuously monitor his or her understanding and formulate linguistic responses that make clear to the speaker what aspects of the utterance are problematic and, thereby, the nature of the clarification that the speaker must provide (Clark, 1996). In turn, this ability presupposes the development of a number of linguistic, cognitive, and social-cognitive skills (Abbeduto et al., 1997, 1998; Abbeduto & Short-Meyerson, 2002; Ackerman, 1993; Golinkoff, 1986; van der Meij, 1988; Robinson & Whittaker, 1985; Tager-Flusberg, 2001; Whitehurst & Sonnenschein, 1985), virtually all of which are delayed or impaired in Down syndrome and fragile X syndrome.

Most individuals with Down syndrome have impairments in numerous cognitive skills, as reflected by IQs in the range of mild to moderate intellectual disabilities (Chapman & Hesketh, 2000). The range of cognitive impairments is broader in fragile X syndrome, with virtually all males and half of females with the full mutation having a diagnosis of intellectual disabilities (Hagerman, 1999). Below age-level mastery of the linguistic system is almost always found in individuals with Down syndrome (Chapman, 2003) and is characteristic of most males and many females with fragile X syndrome (Murphy & Abbeduto, 2003). Performance in the social-cognitive domain (e.g., in reasoning about mental states) is also delayed relative to typically developing age-matched peers in Down syndrome (Zelazo, Burack, Benedetto, & Frye, 1996) and fragile X syndrome (Cornish et al., 2005; Garner, Callias, & Turk, 1999; Grant, Apperly, & Oliver, 2007; Lewis et al., 2006).

Thus, the impairments that define the behavioral phenotypes of Down and fragile X syndromes are likely to lead to substantial delays in noncomprehension signaling.

Despite the commonalities, there are differences in the behavioral phenotypes of Down syndrome and fragile X syndrome. First, the development of the linguistic system, especially its syntactic aspects, is more delayed in Down syndrome than in fragile X syndrome (Abbeduto et al., 2003). Second, individuals with Down syndrome display deficits in theory of mind (i.e., reasoning about mental states) that are more severe than their deficits in other areas of cognitive functioning (Zelazo et al., 1996), whereas individuals with fragile X syndrome reason as accurately about mental states as do their MA-matched typically developing peers (Garner et al., 1999; Lewis et al., 2006). Third, auditory memory is especially impaired (i.e., relative to other facets of memory) in Down syndrome (Marcell & Weeks, 1988; Seung & Chapman, 2000), whereas no such asynchrony has been documented for fragile X syndrome (Dykens et al., 2000). Fourth, maladaptive behavior occurs at relatively low rates in Down syndrome, but is frequent in fragile X syndrome, with the latter being troubled by, for example, social anxiety (Bregman, Leckman, & Ort, 1988; Mazzocco, Baumgardner, Freund, & Reiss, 1998) and attentional difficulties (e.g., Bregman et al., 1988; Cornish, Sudhalter, & Turk, 2004; Dykens, Hodapp, & Leckman, 1989; Mazzocco, Pennington, & Hagerman, 1993). Although the relative contributions of language, theory of mind, auditory memory, and maladaptive behavior to the typical development of noncomprehension signaling have yet to be determined (Abbeduto et al., 1997), it is reasonable to suppose that phenotypic differences in these domains could lead to within- and between-syndrome differences in noncomprehension signaling. Moreover, comparisons of Down syndrome and fragile X syndrome should be especially useful in clarifying the contributions of these behavioral domains to the development of mature noncomprehension signaling.

In light of the importance of noncomprehension signaling to successful discourse, it is surprising that there are no published studies on the ability of individuals with Down or fragile X syndrome to engage in noncomprehension signaling. It is interesting to note, however, that very young children with Down syndrome show delays in learning to respond as speakers to requests for clarification from other people (Coggins & Stoel-Gammon, 1982; Scherer & Owings, 1984), although the extent of the delay (e.g., whether excessive relative to other domains of language) is not clear (Rosenberg & Abbeduto, 1993). In addition, there is some evidence that the ability to evaluate the fit of a spoken utterance to the discourse context is impaired in individuals with fragile X syndrome, even in adult females with typical-range IQs (Simon, Keenan, Pennington, Taylor, & Hagerman, 2001). These studies reinforce the notion that noncomprehension signaling may pose a special challenge for individuals with Down or fragile X syndrome; however, the extent of that challenge for either syndrome remains to be determined.

In examining noncomprehension signaling, it is important to recognize that the type of problematic message and its context can have dramatic effects on young listeners (Shatz, 1983). In terms of message type, incompatible messages (i.e., those for which there are no reasonable available referents) are highly salient and associated with minimal information-processing constraints and, thus, typically developing children as young as 2 or 3 signal

noncomprehension of such messages (Lempers & Elrod, 1983; Revelle, Wellman, & Karabenick, 1985). In contrast, it is not until the age of 4 and up that typically developing children respond to ambiguous messages (i.e., messages for which there are multiple, plausible referents) and other more subtle types of problems (Ackerman, Szymanski, & Silver, 1990; Lempers & Elrod, 1983; Revelle et al., 1985). In terms of context, young typically developing children are less likely to fully analyze the potential referent array, which is necessary for identifying instances of noncomprehension, the larger that array (Glucksberg, Krauss, & Higgins, 1975; Roberts & Patterson, 1983). The noncomprehension signaling of adolescents and adults with intellectual disabilities has also been found to be influenced by such variables, although studies to date have included only samples that were heterogeneous with respect to etiology and etiology was not a variable of interest (Abbeduto et al., 1997, 1998; Fujiki & Brinton, 1993).

In the present study, we had three goals. The first was to determine the extent and nature of the delay in noncomprehension signaling for individuals with Down syndrome or fragile X syndrome. We were interested in documenting the delay in this domain of language use relative to typically developing children at similar developmental levels as well as the possibility of syndrome differences in the extent or nature of the delay. In addressing this goal, we examined noncomprehension signaling as a function of type of problematic message and size of the potential referent array. The second goal was to determine how the noncomprehension signaling of individuals with Down or fragile X syndrome is shaped by their levels of cognition, language, social cognition, and maladaptive behavior. Our third goal was to explore possible differences between males and females with fragile X syndrome in the extent and pattern of their noncomprehension signaling. Few studies of language have included both males and females assessed under comparable task conditions; thus, whether they differ quantitatively or qualitatively in language is not clear (Murphy & Abbeduto, 2003). Because only a small number of females with fragile X syndrome participated in this study, the results are only preliminary in nature.

Method

Participants

The participants were 18 adolescents and young adults with fragile X syndrome, 22 adolescents and young adults with Down syndrome, and 17 typically developing 3- to 6-year-olds. Participants in the syndrome groups were recruited by advertisements in local newspapers, mailings to local educators and administrators of genetics clinics, and notices to families enrolled in a university research registry. The syndrome groups were also recruited nationally through postings on Internet websites and listservs and in the newsletters of national organizations focused on developmental disabilities. Typically developing children were recruited locally through university research registries, community postings, and preschools. A few of the participants were also included in analyses reported in Lewis et al. (2006).

Sample characteristics are summarized in Table 1. Participants were selected to achieve a group-wise match across the three groups on non-verbal mental age (MA), which, as described below, was measured using three subtests from the Stanford-Binet Intelligence

Scale, 4th edition (Thorndike, Hagen, & Sattler 1986). Although the groups did not differ significantly in nonverbal MA, $F(2, 54) = 2.03, p = .14$, the match achieved was less close than suggested by Mervis and Robinson (1999) and, thus, nonverbal MA was used as a covariate in the primary analyses (see *Results* section). The syndrome groups also did not differ in nonverbal IQ on the Stanford-Binet subtests, $t(38) = .10, p = .92$, or chronological age (CA), $t(38) = .01, p = 1.00$. Although the number of males was greatest in the fragile X syndrome group, the groups did not differ significantly in gender composition, $\chi^2(2, N = 57) = 2.44, p = .30$.

No participant had more than a mild hearing loss (i.e., a mean pure tone threshold of 30 dB or worse in the better ear across the frequencies of 500, 1000, and 2000 Hz) at the time of testing. No participant met criteria for autism (see Lewis et al., 2006, for details).

Physician or hospital reports providing DNA confirmation of the full mutation were available for all participants with fragile X syndrome. Two males with fragile X syndrome were mosaic. Participants with Down syndrome all had trisomy 21 according to parental report, which was confirmed by physician/hospital records of the karyotype results for most of them. Three families had more than one child with fragile X syndrome participate.

There was no difference across the groups in racial composition, $\chi^2(2, N = 57) = 3.91, p = .14$, with 52 of the 57 participants self-identifying as White. The groups also did not differ in maternal education, $\chi^2(2, N = 56) = 2.17, p = .34$, with 43 of the 56 mothers who provided such data indicating that they had a 4-year college degree or higher.

Characteristics of the participants with fragile X syndrome are presented by gender in Table 2. The males and females differed on nonverbal IQ, $t(16) = 4.48, p < .0005$, and nonverbal MA, $t(16) = 5.08, p < .0005$, but not age, $t(16) = .00, p = 1.00$. These differences are consistent with previous findings of greater affectedness in males than females (Hagerman, 1999). Note, however, that because of our interest in making comparisons between nonverbal MA-matched syndrome groups, the females with fragile X syndrome all had nonverbal IQs in the intellectual disabilities range. Moreover, the males and females with fragile X syndrome overlapped considerably in the range of nonverbal IQs (37 to 57 and 44 to 56, respectively). Thus, the females in this sample should be seen as representing the more affected end of the continuum of females with fragile X syndrome; that is, the half who meet diagnostic criteria for intellectual disabilities (Hagerman, 1999).

Measures Providing Putative Predictors of Noncomprehension Signaling

The following measures assessed domains important in recognizing and resolving comprehension failures. The measures were administered to each participant as part of a more comprehensive battery. The measures were administered over several sessions, typically on the same day as, or within a few days of, the noncomprehension signaling task (described below).

Nonverbal cognition—The Bead Memory, Pattern Analysis, and Copying subtests of the Stanford-Binet, 4th edition were administered. These subtests, which require a minimum of verbal instructions and only nonverbal responses, have been used in previous studies to

create cognitively matched comparisons of the three groups included in the present study (e.g., Abbeduto et al., 2003; Chapman et al., 1991). Nonverbal MA and IQ were computed from the three subtests (see Table 1). Nonverbal MA was the basis of the group-wise matching and was used to evaluate the contribution of nonverbal cognitive ability to noncomprehension signaling.

Auditory short-term memory—The Digit Span subtest from the Wechsler Scales of Intelligence for Children, 3rd edition (Wechsler, 1991) was administered. In this subtest, the participant must immediately repeat verbatim digit sequences spoken by the examiner at the rate of one digit per second. The number of correctly repeated sequences (see Table 1) was used to evaluate the contribution of auditory short-term memory to noncomprehension signaling.

Language—We used the Oral Expression Scale of the Oral and Written Language Scales (Carrow-Woolfolk, 1995) to measure expressive skills. This scale measures numerous dimensions of expressive language, from vocabulary to syntax to discourse-level rules. Each item requires the participant to produce a word, phrase, or sentence in response to a verbal prompt from the examiner. The prompts are accompanied by drawings. The age-equivalent score (see Table 1) was used to evaluate the contribution of expressive language ability to noncomprehension signaling.

Receptive language was assessed using the Test for Auditory Comprehension of Language-3 TACL-3 (Carrow-Woolfolk, 1999). This test includes items tapping understanding of vocabulary, grammatical morphemes, and syntactic rules and relations. Each item requires the participant to point to the one picture from among several alternatives that correctly conveys the meaning of the word, phrase, or sentence spoken by the examiner. The total test age-equivalent score (see Table 1) was used to evaluate the contribution of receptive language to noncomprehension signaling.

Theory of mind—We administered a false belief task similar to those commonly used to study theory of mind (Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998). In this task, the participant is asked a series of questions about the beliefs of various story characters after listening to and watching a story told and enacted with miniature figures and props. The test questions assess whether the participant differentiates between his or her own true beliefs and the story characters' false beliefs. Some questions require reasoning about a character's beliefs about an object's location (first-order reasoning), whereas others require reasoning about a character's beliefs about yet another character's beliefs about an object's location (second-order reasoning). Typically developing children succeed at such tasks near the age of 4, which is when they recognize that the human mind represents rather than copies the world (Tager-Flusberg, 2001). The proportion of test questions answered correctly (see Table 1) was used to evaluate the contribution of theory of mind to noncomprehension signaling. This task is described in detail elsewhere (Lewis et al., 2006).

Maladaptive behavior—The Child Behavior Checklist/4-18 (Achenbach, 1991) was completed for each participant with fragile X syndrome or Down syndrome by his or her parent. This informant-report measure has been widely used for decades, including for

individuals with developmental disabilities. We computed a Total Problems T score (see Table 1) as well as T scores for each of the five subscales expected to distinguish the fragile X and Down syndrome groups: Withdrawn, Anxious/Depressed, Attention Problems, Thought Problems, and Social Problems. We used T scores to evaluate the contribution of maladaptive behavior to the noncomprehension signaling task.

Noncomprehension Signaling Task

Task overview—Participants, who were tested individually, played the role of listener, and a second researcher played the role of speaker. The participant and speaker sat at a table facing each other. The participant had an easel book; each page contained a colorful scene (e.g., a seascape). Moveable magnetic pieces, each with a colored drawing of an object (e.g., a seashell), were situated at the bottom of the page. A magnetic strip in the scene could hold one of the pieces. The speaker also had an easel book containing the scenes, but with one of the pieces already printed on the scene. For each page, the speaker produced a one-sentence direction indicating which piece the participant should move into the scene (e.g., “Put the seashell on the beach”). The goal was for the participant to make each page identical to the speaker’s page. The participant could not see the speaker’s page.

The speaker’s direction allowed the intended referent to be unambiguously identified on the pages of the informative condition (e.g., the potential referents on one page were four crayons of different colors, including a red one, and the direction was “Put the red crayon on the box”). The directions were less than fully informative for other pages. In some cases, the direction referred to a piece that was not available (i.e., the incompatible condition). On one page, for example, the scene was of a dinner plate and place setting, and the magnetic pieces were drawings of forks of different colors. The speaker said, “Put the black fork on the plate,” but none of the forks was black. In other cases, the direction did not contain an adjective to indicate which piece was the one intended (i.e., the ambiguous condition). On one page, for example, the scene was of a painting on an easel; the magnetic pieces contained drawings of paint brushes, each dipped in a different color of paint. The speaker’s direction was “Put the brush on the painting.” Finally, the speaker’s direction sometimes contained an adjective whose meaning was highly unlikely to be known by the participant (i.e., the unfamiliar condition). On one page, for example, the scene was a sky, and one magnetic piece depicted a blue hot air balloon and another piece depicted a yellow hot air balloon. The speaker’s direction for this page was “Place the azure balloon in the sky.” Thus, a participant could immediately select a referent and move it into the scene for the informative directions; however, he or she had to signal noncomprehension and, thereby, solicit more information to be sure of making a correct referent choice for the three types of inadequate directions. In addition to direction type, we also manipulated the number of potential referents available per page. Each page included two or four potential referents.

Materials—The participant’s book contained 32 pages (2 practice and 30 experimental items). Each 28 cm × 21.5 cm page depicted a background scene into which a potential referent could be placed. Each potential referent was drawn on a separate 5 cm × 5 cm card. Each card had a magnet on its back. A magnet large enough to hold a single card was located within each scene. The potential referents were arrayed in a line at the bottom of the

page in a single random order. The scenes and potential referents were drawn with a standard clip art computer program. The pages and cards were laminated and combined into a single spiral-bound book that stood up on an easel so that each page could be easily viewed by the participant but was out of view of the speaker.

Half of the pages for the experimental items contained two potential referents and half contained four potential referents. Six two-referent and six four-referent pages were selected at random and assigned to the informative direction condition. The 18 remaining pages were assigned at random to the incompatible, ambiguous, and unfamiliar direction condition, with an equal number of pages per condition and an equal number of two- and four-referent pages per condition.

The speaker's directions were scripted for each experimental item. Each direction was a single-clause imperative directing the participant to move a specific potential referent into the scene. Each imperative began with a verb such as *put*, *place*, or *move*. The basic structure of the imperative was Verb + Noun Phrase + Prepositional Phrase. The directions were six to eight words in length.

A script of speaker responses to possible participant verbal responses was also created for each speaker direction. The script specified a pragmatically appropriate speaker response for any possible signal of noncomprehension from the participant (see Table 3). The script also specified the nature of the speaker's response should the participant respond to the direction by doing something other than signaling noncomprehension or moving a potential referent into the scene. Note that nonspecific questions such as "huh?" or "what?" were treated as requests for repetition of the original direction rather than signals of non-comprehension based on previous studies demonstrating that such questions most often elicit a repetition of the original utterance rather than new information from speakers in natural conversation (e.g., Garvey, 1977).

Two versions of the easel book were created, each comprised of a different random order of the 30 experimental items. Participants were randomly assigned to one or the other order.

Procedure—In explaining the task, the examiner stressed the need to listen carefully and achieve an exact match with the speaker. The examiner also explained that "you can talk with ___ [speaker name], ask him/her questions, or say anything to him/her. You need to make sure your pictures match." This latter instruction was designed to assure the participant that there were no prohibitions against talking, an instruction that has been found to be important in ensuring the validity of this task (Abbeduto et al., 1997).

The two practice trials, each of which involved an informative direction, immediately followed the reading of the task instructions by the examiner. For the practice items, the participant and speaker were allowed to compare their pages after each direction with either positive or corrective feedback provided as necessary. No corrective feedback or opportunity to compare pages was provided after the practice trials, although non-contingent general praise (e.g., "I like how you're listening") was delivered according to a script throughout the task. The examiner's participation in the remainder of the task was minimal.

The speaker looked at his or her own book when producing each direction and maintained that focus until the participant had either signaled noncomprehension or moved a potential referent into the scene. Eye contact between participant and speaker was thereby avoided so as not to convey the impression that the speaker was necessarily expecting a verbal response from the participant. The speaker responded verbally to signals of noncomprehension according to the script. Responses in which the participant simply moved a potential referent into the scene received no response from the speaker.

The examiner scored the participant's responses to the speaker's directions as they occurred, noting which potential referent was selected and whether a signal of noncomprehension was produced and transcribing any signal produced. The entire session was also audio- and videotaped so that the accuracy of the examiner's notations with regard to the occurrence and transcription of noncomprehension signaling could be checked for accuracy.

Scoring noncomprehension signals—Each page of the book was scored for the presence or absence of a signal of noncomprehension by the participant. The types of participant responses that were scored as signals of noncomprehension are illustrated in Table 3. We did not distinguish between responses containing multiple versus single signals of noncomprehension. Interrater agreement, calculated for 9 participants (3 per diagnostic group), was found to be 100% for the occurrence of a signal of noncomprehension.

Checks on task materials/manipulations—Additional data were collected either from the participants or from different participants in the developmental range of interest in pilot studies to ensure that (a) participants understood the nouns used in the directions, (b) the meanings of the adjectives used in the unfamiliar directions were not known by the participants, (c) the shorter length of the ambiguous directions (i.e., in contrast to the other inadequate directions, they lacked an adjective) had no impact on the non-comprehension signaling of the participants, and (d) all participants understood the color adjectives used and could discriminate among the colors used. (Details are available from the first author.)

Results

Correct Referent Selections for Informative Directions

We analyzed the number of correct (i.e., intended) referent selections in response to the 12 informative directions in a 3 (diagnostic group) \times 2 (number of potential referents) ANCOVA. Number of potential referents was a within-participant variable and nonverbal MA was the covariate. Only the main effect of diagnostic group was significant, $F(2, 53) = 6.46$, $p = .003$, $\eta^2 = .20$; however, even the participants with Down syndrome, who were the lowest performing group, did well, selecting the correct referent on a covariate-adjusted mean of 5.55 across the two- and four-referent informative directions. The covariate-adjusted means for the fragile X syndrome and typically developing groups were 5.99 and 6.01, respectively. Thus, the participants understood the task and could process the linguistic forms used for the directions, although those with Down syndrome were somewhat less capable in this regard.

Diagnostic Group Comparisons of Noncomprehension Signaling

The number of trials on which a signal of noncomprehension was produced was analyzed in a 3 (diagnostic group) \times 4 (direction type) \times 2 (number of potential referents) ANCOVA. Direction type and number of potential referents were within-participant variables, and the covariates were nonverbal MA and number of correct referent selections on informative directions. We employed the Greenhouse-Geisser adjustment for all significance tests involving the within-participant variables to control for violations of the sphericity assumption. Because there were twice as many informative directions as any other direction type, the number of signals produced in response to informative signals was divided by two to ensure that the scale was constant across conditions. Note that covariate-adjusted means are presented throughout. The adjusted means and standard errors for each condition for each diagnostic group are presented in Table 4.

We found a main effect of diagnostic group, $F(2, 52) = 6.60, p = .003$. Using Fisher's LSD (Levin, Serlin, & Seaman, 1994), we found that post-hoc comparisons of the diagnostic groups indicated that the typically developing participants produced significantly more signals of noncomprehension than did participants in either of the syndrome groups, who did not differ from each other. The covariate-adjusted mean number of signals (elicited by the three directions per each combination of direction type and number of referents) was .52, .81, and 1.64 for the Down syndrome, fragile X syndrome, and typically developing groups, respectively. The effect of diagnostic group was moderate to large, $\eta^2 = .20$, according to the guidelines proposed by Cohen, Cohen, West, and Aiken (2003).

We also found a significant Diagnostic Group \times Direction Type interaction, $F(4.1, 106.7) = 4.24, p = .003$, with an η^2 of .14, indicating a small to moderate effect size. Simple effects tests were conducted to examine differences among diagnostic groups separately for each type of direction using the Holm sequential procedure to prevent inflation of Type I error (Holm, 1979; Levin, Serlin, & Seaman, 1994). This procedure required the largest F for diagnostic group to reach an alpha of .013 for significance (i.e., $\alpha/4$); the next largest F , an alpha of .017 ($\alpha/3$); the next largest, an alpha of .025 ($\alpha/2$); and the smallest, an alpha of .05. The effect of diagnostic group was significant for unfamiliar directions, $F(2, 52) = 7.62, p = .001$, and ambiguous directions, $F(2, 52) = 6.39, p = .003$, but just failed to reach significance for incompatible directions, $F(2, 52) = 3.93, p = .026$. The effect of diagnostic group was not significant for the informative directions. Post-hoc comparisons (using Fisher's LSD technique) indicated that the participants with fragile X syndrome and those with Down syndrome produced fewer signals of noncomprehension than did the typically developing children on both the unfamiliar and the ambiguous directions. In the case of incompatible directions, the marginally significant finding reflected the fact that the participants with Down syndrome produced fewer signals of noncomprehension on the incompatible directions than did the typically developing participants. None of the comparisons between the two syndrome groups were significant.

The interaction of diagnostic group and number of referents was also significant, $F(2, 53) = 3.46, p = .04, \eta^2 = .12$. Simple effects tests were conducted to examine differences among diagnostic groups separately for the two-referent and four-referent condition using the Holm sequential procedure. This procedure required the largest F for diagnostic group to reach an

alpha of .025 for significance (i.e., $\alpha/2$) and the smallest to reach an alpha of .05. The effect of diagnostic group was significant for both the four-referent, $F(2, 52) = 7.88, p = .001$, and two-referent, $F(2, 52) = 5.24, p = .008$, conditions. Post-hoc comparisons (using Fisher's LSD technique) indicated that the participants with fragile X syndrome and those with Down syndrome produced fewer signals of non-comprehension than did the typically developing children on both the two- and four-referent conditions, although the magnitude of the difference between groups was greater on the four- than the two-referent arrays. None of the comparisons between the two syndrome groups were significant.

Comparisons of Males and Females With Fragile X Syndrome

As noted previously, the participants with fragile X syndrome were virtually perfect in selecting the correct referent in response to informative directions, with a covariate-adjusted mean of 5.99 across the two- and four-referent arrays. Thus, we did not perform any statistical analyses involving gender and this dependent measure.

The number of signals of noncomprehension produced by the participants with fragile X syndrome was analyzed in a 2 (gender) \times 4 (direction type) \times 2 (number of potential referents) AN-COVA. Direction type and number of potential referents were within-participant variables, and nonverbal MA was the covariate. (Number of correct referent selections on informative directions was not included as a covariate because there was no variability on this measure.) Again, the number of signals produced for informative directions was divided by 2.

We found that males with fragile X syndrome signaled noncomprehension less often than did females with the syndrome, $F(1, 15) = 3.46, p = .04$ (one-tailed), $\eta^2 = .19$ (i.e., a moderate effect size). The (covariate-adjusted) mean frequencies for males and females were .51 and 1.72, respectively. No other main or interaction effects were significant.

We also reran the Diagnostic Group \times Direction Type \times Number of Potential Referents ANCOVA described in the previous section, excluding females with fragile X syndrome, but including the females with Down syndrome or typical development because there was no reason to expect gender differences in the latter two groups. Our aim was to determine whether the diagnostic group differences in noncomprehension signaling and the interactions of diagnostic group with direction type and with number of potential referents had somehow been distorted by the inclusion of the females with fragile X syndrome in that analysis. Although the means of the fragile X and Down syndrome groups were even more similar to each other in the re-analysis than in the primary analysis that included females with fragile X syndrome, the pattern of statistical significance was unchanged. (The results of this re-analysis are available from the first author.)

Examination of Putative Predictors of Noncomprehension Signaling

Multiple regression was used to examine the relationship between a measure of "appropriate" noncomprehension signaling and the various predictors. The dependent measure was created by first computing the proportion of inadequate (i.e., incompatible, ambiguous, and unfamiliar) directions that elicited signals of noncomprehension from a participant and then subtracting the proportion of informative directions that elicited such

signals from the participant, thereby controlling for any indiscriminate signaling. This proportional dependent variable was subjected to an arcsine transformation prior to analysis. Because the number of predictors was large relative to the sample size, we adopted a conservative approach to model building, limiting the number of predictors while still testing whether the relationships among variables differed across diagnostic groups (see Abbeduto et al., 2006, for a similar approach).

The regression analysis proceeded in three steps. In Step 1, we entered two dummy variables to represent the three diagnostic groups (Cohen et al., 2003). The first dummy variable indexed whether the participant had Down syndrome and the second, whether the participant had fragile X syndrome. A significant coefficient for a dummy variable indicated that the indexed group and the typically developing group differed on the dependent variable. In Step 2, nonverbal MA, TACL-3 age-equivalent score, Oral Expression Scale age-equivalent score, proportion correct on the false belief task, and number of correctly recalled sequences on the digit span task were entered simultaneously. We also entered gender at Step 2 because of the gender differences in the frequency of noncomprehension signaling among the fragile X syndrome participants described in the preceding section. In Step 3, we entered interactions between the predictors that were significant at Step 2 and the dummy variables indexing the diagnostic groups. Each interaction was represented by the product of the dummy variable and predictor variable. A significant coefficient for an interaction term indicated that the relationship between the significant Step 2 predictor and the dependent variable differed between the diagnostic group indexed by the dummy variable and the typically developing group. This approach to testing interactions reflected the assumption that the interactions would be ordinal (i.e., the relationships would vary in strength but not direction across groups), which means that significant interactions were likely to be associated with significant main effects. We expected the predictors to be related positively (or not at all) to the dependent variable and, thus, main effects were evaluated with one-tailed tests. We used two-tailed tests to evaluate all interactions. This analytical strategy allowed us to assess potential sources of both within- and between-group differences in appropriate noncomprehension signaling.

The Step 1 model indicated that the two dummy variables indexing the group contrasts were significant, $F(2, 51) = 6.78, p = .002$. The addition of the six predictors (main effects) at Step 2 was associated with a significant change in the R^2 , $F(6, 45) = 2.47, p = .04$; however, the only predictor of the six that yielded a significant beta at Step 2 was the TACL age-equivalent score, $\beta = .47, t = 1.93, p = .03$ (one-tailed). The betas for the dummy variables representing the two group contrasts remained significant at Step 2. At Step 3, we entered the two interaction terms for group and TACL age-equivalent score. The addition of the interaction terms did not lead to a significant change in the R^2 , although the Step 3 model yielded an adjusted R^2 of .28, $F(10, 43) = 3.03, p = .005$. The only predictor associated with a significant beta at Step 3 was the TACL age-equivalent score, $\beta = .64, t = 1.81, p = .04$ (one-tailed). Neither group contrast was significant in the Step 3 model.

We also conducted regression analyses examining the relationship between Child Behavior Checklist scores and the arcsine-transformed measure of appropriate noncomprehension signaling. These analyses involved only the two syndrome groups and were conducted in the

same way as the regression described previously, with one analysis including the Child Behavior Checklist Total score as predictor and the other analysis including the five subscale scores as predictors. Scores on the Child Behavior Checklist did not contribute to prediction in either analysis.

Discussion

Diagnostic Group Differences and Similarities in Noncomprehension Signaling

Our first goal was to determine the extent and nature of the delay in noncomprehension signaling for individuals with Down syndrome or fragile X syndrome. We found that both syndrome groups were less likely to signal noncomprehension of inadequate directions than were the MA-matched typically developing children. In fact, on average individuals with Down syndrome or fragile X syndrome signaled noncomprehension on only 30% of the inadequate directions they heard compared to 70% for the typically developing comparison children. Moreover, the advantage of the typically developing children over the two syndrome groups in noncomprehension signaling was evident for both the two- and four-referent arrays. At the same time, both syndrome groups were virtually perfect in selecting the intended referent for informative directions, suggesting that their low frequencies of noncomprehension signaling did not arise solely from limitations in their linguistic knowledge or ability to process the specific language forms used in the noncomprehension signaling task. This claim is further supported by the fact that the typically developing children did not differ from either syndrome group in their scores on the TACL-3, which is a standardized test of receptive language. Thus, the findings suggest that youth with fragile X syndrome or Down syndrome are (a) poor at monitoring their comprehension, thereby failing to recognize when they do not understand; and/or (b) unable to create and execute a plan for soliciting corrective information from the speaker, thereby allowing detected problems to go uncorrected. It may also be that these youth view the noncomprehension signal as a sign of *their failure* rather than *the failure of the message* and thus refrain from signaling non-comprehension so that they, like the adults with mild intellectual disabilities studied by Edgerton (1993), can assume a “cloak of competence.” In any event, given the likely negative consequences for everyday comprehension in a host of settings, from school to informal conversations with peers and care providers, it is important that noncomprehension signaling be a target of intervention. Unfortunately, there have been few attempts to develop effective interventions in this area (see Ezell & Goldstein, 1991, for an exception).

We also found that differences in noncomprehension signaling between the typically developing children and the syndrome groups were less pronounced for incompatible directions than for ambiguous or unfamiliar directions. Of the three types of inadequate directions, the problem exemplified in the incompatible directions is the most salient and least difficult to resolve and, thus, is the first to be dealt with successfully by young typically developing children (e.g., Lempers & Elrod, 1983; Revelle et al., 1985). Thus, although noncomprehension signaling is quite delayed in fragile X and Down syndrome, it is not qualitatively different compared to typically developing children.

The two syndrome groups did not differ significantly in their rates of noncomprehension signaling in any comparison. The only marginal difference occurred for incompatible directions, on which only the Down syndrome participants tended to differ from the typically developing participants. Despite the generally similar levels of performance exhibited by the two syndrome groups, it is unlikely that their low rates of noncomprehension signaling are simply a manifestation of their having intellectual disabilities. This conclusion is suggested by the fact that several researchers using a variety of methodologies have not found differences in noncomprehension signaling rates between youth with mental retardation who are heterogeneous with respect to etiology and MA-matched typically developing children (Abbeduto et al., 1997; Abbeduto et al., 1998; Ezell & Goldstein, 1991; Fujiki & Brinton, 1993; Rueda & Chan, 1980; however, see Abbeduto et al., 1991, for an exception). Additional studies in which investigators include a nonspecific intellectual disabilities comparison in addition to the two syndrome groups (Dykens et al., 2000), however, are needed to verify this claim. If differential impairments in noncomprehension signaling are demonstrated for individuals with fragile X syndrome or Down syndrome, on the one hand, and individuals with other forms of intellectual disabilities, on the other hand, it would be important to determine the causes of those differences, with the possibilities including not only the genetic and psychological characteristics of the individuals, but their environmental histories as well. From a clinical perspective, such a finding would suggest the need to develop different types of language interventions, or at least interventions with different targets, for individuals with these two syndromes compared to individuals with intellectual disabilities of other origins.

Putative Predictors of Noncomprehension Signaling

Our second goal was to determine how non-comprehension signaling is shaped by an individual's levels of nonverbal cognition, language, theory of mind, auditory memory, and maladaptive behavior and whether there are differences between the syndromes and MA-matched typically developing children in this regard. We found that only scores on the TACL-3, which was designed to measure the ability to understand a range of lexical items and grammatical elements, patterns, and rules, made a unique contribution to the dependent measure, with higher TACL-3 scores being related to more frequent noncomprehension signaling. Moreover, the relation between the TACL-3 and noncomprehension signaling was similar across the two syndrome groups.

It is not surprising that knowledge of word meanings and syntactic structure would have an impact on noncomprehension signaling; for example, the ability to parse the syntactic structure of a sentence is likely to facilitate recognition of the source of noncomprehension and creation of a plan for soliciting precisely the sort of linguistic information needed to resolve the problem. Variation in noncomprehension signaling, however, cannot be reduced simply to differences in receptive lexical and syntactic ability. The syntax and vocabulary of the directions in the noncomprehension signaling task were quite simple and chosen so that they would be within the competence of the participants. In fact, all three groups displayed a high level of skill in processing informative directions, which were syntactically identical to the inadequate directions. Moreover, the participants with fragile X syndrome were poor at signaling noncomprehension despite TACL-3 scores that exceeded those of the participants

with Down syndrome and that were not significantly different from those of the typically developing children. Thus, noncomprehension signaling requires skills that extend beyond linguistic knowledge. The implication of this conclusion is that traditional language intervention, which is focused largely on teaching new language targets (Brady & Warren, 2003), is likely to have rather minimal impacts on comprehension monitoring and the other skills needed to engage in effective noncomprehension signaling. Future researchers should focus on evaluating more broadly conceived language interventions.

Contrary to expectations, individual differences in nonverbal cognition, social cognition, auditory memory, and maladaptive behavior did not make unique contributions to noncomprehension signaling. This may reflect the small sample size or limitations of the measures; however, we have also failed to find contributions from these domains to other aspects of social communication in these syndromes or in typically developing children (Abbeduto et al., 2006), suggesting that, at least in the developmental range of our participants (approximately 3 to 7 years), nonverbal cognition, social cognition, auditory memory, and maladaptive behavior do not constrain acquisition or use of the skills underlying noncomprehension signaling. All four domains, however, have been found to impact other aspects of language at other points in development for individuals with developmental disabilities as well as for typically developing children (e.g., Belser & Sudhalter, 1995; McDuffie, Chapman, & Abbeduto, in press; Tager-Flusberg, 2001). Together, such findings underscore the importance of expanding language assessment to include domains such as auditory memory while tailoring the focus of the assessment of nonlinguistic domains to the child's developmental level and the profile of language problems. In terms of typical development, the findings suggest that claims about the pervasive role played by theory of mind and auditory memory in communication will need to be more constrained and nuanced, recognizing that the role of skills in these domains will vary across developmental periods and different facets of communication.

We note, however, that we evaluated non-comprehension signaling under “ideal” conditions; that is, testing occurred in a quiet room with a skilled adult partner, with few distractions, and without other people involved or available to participate in the interaction. Of course, this is a very different context than is true of most of the daily experience of a youth with Down syndrome or fragile X syndrome (e.g., in school). The domains of nonverbal cognition, social cognition, auditory memory, and maladaptive behavior might make more important contributions to noncomprehension signaling in these everyday contexts than was observed in this study. Future researchers, therefore, should examine noncomprehension signaling and its determinants in a range of contexts, including laboratory-based tasks in which the demands on nonverbal cognition, social cognition, auditory memory, and the constraints on the occurrence of maladaptive behavior are systematically manipulated. Moreover, we included only very broad summary measures of nonverbal cognition and language ability and measures of rather narrow “slices” of theory of mind and auditory memory. Measurement of a different set of skills from each of these domains might lead to different conclusions and should be evaluated.

Differences in Noncomprehension Signaling by Males and Females With Fragile X Syndrome

Our third goal in the study was to explore possible gender differences in noncomprehension signaling among those with fragile X syndrome. Despite including only a small number of females with this syndrome, we found that they were more likely to signal noncomprehension than were their male counterparts. This difference emerged despite statistical equation of males and females on nonverbal MA through ANCOVA and despite the fact that we included only those females whose nonverbal IQs placed them among the lower half of females with fragile X syndrome (i.e., those meeting criteria for an intellectual disability). Moreover, even the males with fragile X syndrome were virtually perfect in selecting the intended referent for informative directions. Thus, the difference in noncomprehension signaling between males and females was not due simply to the within-syndrome variations in cognitive and linguistic ability that are inherently correlated with gender. This is consistent with recent findings concerning various aspects of expressive language, including the verbal perseveration that is so characteristic of individuals with fragile X syndrome (Murphy & Abbeduto, 2007). Such findings suggest that differences in the behavioral phenotypes of males and females with fragile X syndrome are not simply quantitative in nature, reflecting differences in severity; instead, different profiles of relative strengths and weaknesses may characterize males and females with the syndrome as well. Indeed, the rate of noncomprehension signaling in females with fragile X syndrome approached that observed for the typically developing matches, suggesting that below-MA rates of noncomprehension signaling characterize largely males with the syndrome. It will be an important task for future researchers to more completely characterize the language phenotypes of males and females, which will require studies using tasks and measures that allow direct comparison of performance across the two (Murphy & Abbeduto, 2003).

Conclusion

We must acknowledge three limitations of the study. First, the sample size, especially with regard to the comparison of males and females with fragile X syndrome, was small; thus, there is a need to replicate the findings and to do so with larger samples. Second, although we examined noncomprehension signaling in relation to other dimensions of behavioral functioning, we assessed only narrowly within some domains of interest and with only a single method of measurement per domain. Moreover, other aspects of the behavioral phenotypes of the two syndromes distinguish them and may impact the signaling of noncomprehension (e.g., visual search). Third, we included only individuals who did not also have an autism diagnosis, which means that we omitted as many as one fourth of the population of persons with fragile X syndrome (Demark, Feldman, & Holden, 2003). There is evidence that these two subgroups differ in both their degree of impairment, with the co-morbid subgroup being lower functioning on average, and in their profiles of behavioral strengths and weaknesses (Hepburn, Hayes, Hagerman, & Rogers, 2004; Lewis et al., 2006). It will be interesting to determine whether variations in noncomprehension signaling also distinguish between individuals with fragile X syndrome with and without a co-morbid diagnosis of autism.

Despite these limitations, the present study has yielded new, clinically important data about the serious challenges facing youth with fragile X syndrome or Down syndrome as listeners. Indeed, the failure to signal noncomprehension can have pervasive negative effects in an interaction, as misunderstandings that are not resolved are sure to be compounded as the interaction progresses. Moreover, when coupled with, as displayed in this study, a tendency to select a referent despite inadequacies in the messages heard, listeners with these syndromes are likely to “guess” wrong and say or do things that are inappropriate to the speaker’s intent, leading to further breakdowns in the interaction. This study has also demonstrated the advantage 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 syndrome directly using the same measures.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

Acknowledgments

Preparation of this manuscript was supported by Grants R01 HD024356 and P30 HD003352 from the National Institutes of Health and by the Graduate School Research Committee of the University of Wisconsin-Madison as well as by funds from the Waisman Center and Friends of the Waisman Center. The authors are deeply grateful to the participating families for their time, patience, and enthusiastic support. Thanks also to Randi Hagerman and Marcia Braden for their advice on recruitment and testing accommodations, Robert Nellis and Jill Roxberg for conducting the audiological evaluations, Pamela Lewis for assistance with the autism diagnostic evaluations, and Susan Kirkpatrick for assistance with genetic testing and interpretation. Brief summaries of preliminary analyses of some of the data reported here have appeared in several chapters in edited volumes, including Abbeduto and Murphy (2004) and Murphy and Abbeduto (2003), and were presented at the 2006 meeting of the Society for the Study of Behavioral Phenotypes, Dublin, Ireland.

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Table 1

Participant Characteristics by Diagnostic Group

Characteristic	Group ^d					
	DS (<i>n</i> = 22)		FX (<i>n</i> = 18)		TD (<i>n</i> = 17)	
	Mean	SD	Mean	SD	Mean	SD
CA (in years)	17.59**	3.68	17.58**	3.35	4.54	.58
S-B ^b Nonverbal MA (in years)	5.22	1.06	5.03	1.52	4.50	.70
S-B ^b Nonverbal IQ	42.32**	6.38	42.56**	8.42	98.00	6.90
OES ^c age-equivalent (in years)	4.60*	1.78	8.09	4.34	5.62*	.89
TACL-3 ^d age-equivalent (in years)	5.62*	1.47	6.83	1.69	5.95	1.06
Proportion correct false belief	.29	.28	.47	.35	.50	.39
No. correct digit sequences ^e	3.14	2.42	3.33	2.03	4.35	1.93
Child Behavior Checklist Total T score ^f	55.09	8.29	57.06	9.67	—	—
No. Caucasians	22	—	16	—	14	—
No. mothers with college degree ^g	15	—	13	—	15	—
No. males	12	—	13	—	8	—

^aDS = Down syndrome, FX = fragile X syndrome, TD = typically developing.

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

^cOral Expression Scale of the Oral and Written Language Scales. Age-equivalents are missing for 1 participant with DS and 2 TD participants.

^dTest of Auditory Comprehension of Language-3.

^eBased on the forward recall portion of the Digit Span subtest of the Wechsler Scales of Intelligence for Children, 3rd edition.

^fChild Behavior Checklist/4-18.

^gLevel of maternal education was not available for 1 participant with FX.

* Significantly different, *p* < .05, from FX.

** Significantly different, *p* < .05, from TD.

Table 2

Characteristics of Participants With Fragile X Syndrome by Gender

Characteristic	Males (<i>n</i> = 13)		Females (<i>n</i> = 5)	
	Mean	<i>SD</i>	Mean	<i>SD</i>
CA (in years)	17.58	3.64	17.58	2.79
Nonverbal MA ^a (in years)	4.31	1.05	6.90	.65
Nonverbal IQ ^a	38.77	6.07	52.40	4.83

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

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Table 3

Types of Noncomprehension Signals and the Speaker's Responses to Them

Type of signal	Example	Examples of responses elicited from speaker
Request for confirmation	The blue hat? This one? (plus holds up card for speaker to see)	Yes, I meant the blue hat. Actually, I meant the red one.
Request for definition	What's <i>russet</i> mean? What's <i>tawny</i> ? What's a _____? (uttered with an intonation suggesting that completion by the speaker is expected)	<i>Russet</i> is a kind of red. <i>Tawny</i> is another word for orange.
Request for specific information	Which one? Which fork do you mean?	Sorry, I meant the yellow duck. I'm sorry . . . I meant the yellow fork. The red jar . . . <i>russet</i> is another word for red (for unfamiliar directions only).
Statement of nonexistence	There is no brown book. There's not one like that. I can't find that one.	Actually, I meant the yellow duck. Sorry, I meant the blue hat.
Statement of existence	There are four forks. There are lots of those you know.	Actually, I meant the yellow one. Sorry, I meant the green letter.
Other	For example, participant holds up a potential referent to show the examiner while looking expectantly.	Yes, I meant the blue hat. Actually, I meant the yellow duck.

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Frequency of Noncomprehension Signaling: Covariate-Adjusted Means and Standard Errors (SEs)

Table 4

Condition	Group ^a					
	DS (n = 22)		FX (n = 18)		TD (n = 17)	
	Mean	SE	Mean	SE	Mean	SE
Informative directions						
Two-referent arrays	.04	.05	.00	.05	.16	.05
Four-referent arrays	.06	.06	.06	.06	.15	.06
Incompatible directions						
Two-referent arrays	.93	.31	1.59	.32	2.23	.34
Four-referent arrays	.99	.31	1.60	.32	2.32	.34
Ambiguous directions						
Two-referent arrays	.54	.31	1.00	.32	2.00	.34
Four-referent arrays	.60	.29	.78	.30	2.23	.32
Unfamiliar directions						
Two-referent arrays	.56	.27	.70	.28	1.77	.30
Four-referent arrays	.45	.28	.74	.29	2.22	.31

^aDS = Down syndrome, FX = fragile X syndrome, TD = typically developing.