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Phonological Awareness and Reading in Boys with Fragile X Syndrome

Suzanne M. Adlof, **Jessica Klusek**, **Svetlana V. Shinkareva**, **Marissa L. Robinson**, and **Jane E. Roberts**
Department of Psychology, University of South Carolina, Columbia, SC, USA

Abstract

Background—Reading delays are well documented in children with fragile X syndrome (FXS), but few studies have examined linguistic precursors of reading in this population. This study examined the longitudinal development of phonological awareness and its relationship to basic reading in boys with FXS. Individual differences in genetic, social-behavioral and environmental factors were also investigated as predictors of phonological awareness.

Methods—Participants included 54 boys with FXS and 53 typically developing (TD) mental age-matched peers who completed assessments of phonological awareness, nonverbal intelligence, and reading annually for up to four years. FMRP level and autism symptomatology were also measured within the FXS group. Hierarchical linear modeling was used to examine change in phonological awareness over time and its predictors. Linear regression was used to examine phonological awareness as a predictor of word reading.

Results—Boys with FXS exhibited slower growth than TD peers in phonological awareness only when nonverbal cognitive abilities were not controlled. The rate of change in phonological awareness decreased significantly after age 10 in boys with FXS. Phonological awareness accounted for 18% unique variance in basic reading ability after controlling for nonverbal cognition, with similar relationships across groups.

Conclusion—Phonological awareness skills in the boys with FXS were commensurate with their nonverbal cognitive abilities, with similar relationships between phonological awareness and reading as observed in the TD mental age-matched peers. More research is needed to examine potential causal relationships between phonological awareness, other language skills, and reading abilities in individuals with FXS and other neurodevelopmental disorders.

Introduction

Fragile X syndrome (FXS) is the leading known genetic cause of autism and inherited intellectual abilities. FXS results from excessive CGG repeats (>200) on the *FMR1* gene, which regulates the production of *FMR1* protein (FMRP), which is necessary for normal

Correspondence Suzanne M. Adlof, University of South Carolina, Williams Brice Nursing Building, 6th Floor, 1621 Greene St. Columbia, SC 29208, USA; sadlof@mailbox.sc.edu.

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brain development and function (Loesch, Huggins, & Hagerman, 2004). Prevalence estimates suggest that 1 in 2500 males display the full mutation (Hagerman, 2008). Because FXS is an X-linked syndrome, males tend to be more severely affected than females (Hagerman & Hagerman, 2002). Behavioral symptoms include language, cognitive, and social impairments, which in turn, limit functional skills including literacy, an important indicator of future employment and quality of life. To date, little research has investigated the linguistic skills that underlie emerging reading skills in this population or in other neurodevelopmental disorders. This study aims to address this gap in the literature by examining the development of phonological awareness and its relationship with basic word reading skills in males with FXS.

Language and Cognitive Development in Children with FXS

As a group, children with FXS exhibit delays in acquiring first words (Brady, Skinner, Roberts, & Hennon, 2006), and continue to display language deficits into adulthood (cf. Finestack, Richmond, & Abbeduto, 2009, for review). Receptive and expressive language weaknesses include delays in vocabulary and morphosyntax as well as higher-level processes; whereas delays relative to chronological age-peers are consistent across studies, mixed findings have been reported regarding language performance relative to mental-aged peers (Abbeduto et al., 2003; Price et al., 2008; Price, Roberts, Vandergrift, & Martin, 2007; Roberts et al., 2007).

Studies examining within-syndrome variability have found that after controlling for sex, higher levels of FMRP are associated with stronger cognitive abilities (Hall, Burns, Lightbody, & Reiss, 2008; Loesch et al., 2004). Language and cognitive delays also tend to be more pronounced in individuals with comorbid diagnoses of autism and FXS (Martin, Losh, Estigarribia, Sideris, & Roberts, 2013; Skinner et al., 2005). In addition to biological variation, environmental factors including maternal education level (Roberts et al., 2005) and maternal responsivity (Warren, Brady, Sterling, Fleming, & Marquis, 2009) have been associated with developmental outcomes.

Development of Reading Skills in Children with FXS

Individuals with FXS exhibit delays in reading skills relative to typically developing (TD) peers (Hodapp, Dykens, Ort, Zelinsky, & Leckman, 1991; Kemper, Hagerman, & Altshul-Stark, 1988). There is some evidence that growth in reading skills plateaus after approximately age 10, similar to reports of a developmental plateau for other more general cognitive abilities in children with FXS (e.g., Fisch et al., 1996; Wright-Talamante et al., 1996). Roberts and colleagues (2005) examined growth in basic reading and other academic disciplines (math, science, social studies, and the humanities) in males with FXS, recruited between the ages of 4 and 13 years and followed for an average of 4.5 years. Hierarchical linear models revealed rapid early growth in reading skills followed by a plateau at approximately 10 years of age.

Bailey and colleagues (2009) also examined literacy attainment in a cross-sectional survey of functional skills of individuals with FXS from birth through adulthood. Similar to Roberts et al. (2005), results indicated rapid growth in literacy skills between the ages of five and 10 years, followed by a plateau beyond age 10. By adulthood, most individuals with FXS could recognize letters and letter-sounds, and most could read some words, with more females than males showing mastery. In terms of text reading, 44% of males were able to read basic books and 19% of males were able to read books containing new words or concepts.

Phonological Awareness in Children with FXS

Whereas delays in the acquisition of reading skills have been documented in children with FXS, less is known about the development of precursor linguistic skills such as phonological awareness. Phonological awareness is defined as sensitivity to the sound structure of words, e.g., the awareness that words rhyme or begin with similar or different sounds (Scarborough & Brady, 2002). In typically developing children, early phonological awareness is predictive and causal of later word reading skills (Ehri et al., 2001). Phonological awareness helps children notice the ways letters represent sounds, enabling phonological decoding to "sound out" words (Al Otaiba, Kosanivich, & Torgesen, 2012). Explicit phonological awareness instruction is recommended as an evidence-based best practice for teaching reading to TD children (National Reading Panel, 2000). In contrast, educational recommendations for children with FXS have tended to emphasize whole-word or "logographic" reading, at least during the early stages of reading instruction (Braden, 2002; National Fragile X Education Project, 2004).

Two previous studies have examined phonological awareness in individuals with FXS. Buckley and Johnson-Glenberg (2008) found that young adult males with FXS exhibited severe deficits in phonological awareness, performing between the 2nd and the 8th percentiles relative to chronological age expectations. Williams (2004) examined relationships between phonological processing measures, including phonological awareness, phonological memory, and rapid naming, and reading abilities in 7-13 year old boys with FXS and TD mental age matched peers. As a group, the boys with FXS performed significantly lower than chronological age expectations on all measures, and significantly worse than mental age peers on measures of nonword decoding and two of the five measures of phonological processing. However, substantial variability was evident, with some members of the FXS group performing within chronological age-based expectations. These studies measured phonological awareness at a single point in time, and much remains to be learned about the developmental trajectory of phonological awareness and its relationship to subsequent literacy achievement in FXS.

Study Rationale and Research Questions

As a group, individuals with FXS exhibit reading difficulties relative to TD peers. However, there is variability in the level of literacy attainment, and little is known about potentially associated cognitive processes. Therefore, this study examined the development of phonological awareness skills in boys with FXS longitudinally. Delays relative to typical chronological age norms were expected, but we were interested in whether phonological awareness growth was commensurate with nonverbal cognitive development, as well as how individual differences in biological and environmental factors were related. Finally, given our interest in informing knowledge of reading development in FXS, we investigated the relationship between phonological awareness and basic reading abilities. The research questions were:

1. Does growth in phonological awareness skills in boys with FXS differ from mental age-matched TD boys?
2. Does phonological awareness growth in boys with FXS plateau at 10 years of age?
3. To what extent do autism severity and FMRP level account for variability in phonological awareness growth among boys with FXS?
4. Does phonological awareness explain individual differences in basic reading in boys with FXS?

Method

Participants

Data for this study were collected as part of a longitudinal study of cognitive development in children with FXS conducted at the University of North Carolina. Informed consent was obtained from participants' parents in accordance with university IRB regulations. Participants were assessed annually and included 54 males with FXS assessed one to five times (127 total observations), and 53 TD mental aged-matched males assessed one to three times (115 total observations), with most assessed twice. The majority of participants across both the FXS and TD groups were Caucasian (82% and 83%, respectively). At study entry, the participants with FXS were matched in nonverbal mental age to the TD group. Phonological awareness data were available for all participants, although the number of observations varied slightly. Subsets of the FXS and TD groups for whom data on FMRP, autistic behavior and word reading were available were used for selected analyses. See Table 1 for an overview of the participant characteristics at study entry and basic descriptive statistics.

Measures

Phonological Awareness—Phonological awareness was measured using the Phonemic Awareness composite score of the Woodcock-Johnson III Test of Cognitive Abilities (WJ-III; Woodcock, McGrew, & Mather, 2001). The Phonemic Awareness score is comprised of two subtests: Sound Blending (synthesizing sounds to form words), and Incomplete Words (identifying complete words from words presented with missing phonemes). Sound Blending measures the participant's ability to synthesize sounds to form words. For example, the participant is presented with a prompt such as "What word do these sounds make when you put them together? /k/ /a/ /t/?" Incomplete Words requires participants to identify a complete word from a word presented with missing phonemes. For example, the examiner may present the following prompt "What word am I trying to say? Alli_a_or?" purposefully omitting the /g/ and /t/ sounds from the word *alligator*. Phonological awareness skills were assessed at each time point for both the FXS and TD samples using W scores, which are Rasch-model scores that include item difficulty as a parameter in estimating a person's ability, and are thus ideal for measuring change over time. The W scale is an equal-interval scale centered on a value of 500 for fifth graders.

Nonverbal Intelligence—The Brief IQ composite score from the Leiter International Performance Scales-Revised (Leiter-R; Roid & Miller, 1997) was used to measure nonverbal cognition. The Leiter-R provides an estimate of nonverbal mental age, which is particularly well-suited for use with children with cognitive and language impairments, and it has been used widely in studies of children with FXS, e.g., (Hooper, Hatton, Baranek, Roberts, & Bailey, 2000; Skinner et al., 2005). The Leiter-R was measured concurrently with the WJ-III.

Autistic behavior—Autistic behavior in the FXS group was measured using the Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Renner, 1988), an examiner rating scale evaluating symptoms of autism in 15 areas including social behaviors, activity level, adaptation, and communication. Nineteen of the 54 participants (35%) had a CARS score consistent with a diagnosis of autism, which is consistent with previous reports (Bailey et al., 1998).

FMRP—Consistent with previous studies (Bailey, Hatton, Tassone, Skinner, & Taylor, 2001; Roberts et al., 2005), FMRP was determined by scoring 200 lymphocytes for the absence or presence of FMRP resulting in the percent producing FMRP (Willemsen et al., 1995). Procedures to collect and analyze FMRP are described in detail elsewhere (Bailey et al., 2001; Hatton et al., 2003). FMRP was available for 42 of the 54 participants with FXS. Inspection of means for all outcome measures suggested no significant performance differences between children for contributing or not contributing FMRP data.

Maternal Education—Maternal education level was collected through a demographic survey. Education level was analyzed as a continuous variable corresponding to the cumulative years of formal education, ranging from less than high school (11 years) to professional or advanced degree (20 years).

Basic Reading—Basic letter- and word- identification skills were measured using the Letter-Word Identification subtest of the Woodcock-Johnson Test of Academic Achievement-Revised (WJ-R; Woodcock & Johnson, 1990). Initial items measure the ability to recognize symbols, and subsequent items measure the ability to name alphabet letters and read single words of increasing complexity. At Time 1, W-scores were available for 52 of the participants with FXS and 39 of the TD controls (see Table 1 for group descriptives). Mean scores for both groups corresponded to an early kindergarten grade-level; however the range included preschool through second-grade performance. Predictive analyses were limited to the FXS group given little data for the TD group. The earliest phonological awareness score and the latest reading score were selected for predictive analyses; the time between these assessments varied with a mean of 1.89 years (SD = 0.96). The average age of the FXS groups at the last reading assessment point was 12.16 (SD = 1.75), and the average W-score at that age was 410.03 (SD = 32.81; range = 350.00-479.00), which is approximately equal to a middle-kindergarten grade level.

Procedures

Annual assessments were administered to each participant over the course of two consecutive days of testing. Upon completion of the assessment, raw protocol scores were calculated with 100% of the protocols verified followed by double entry into the database followed by 20% of the database checked for accuracy. If 20% of the dataset did not meet a pre-established accuracy rate of > 80%, then 100% of the database was checked to ensure accuracy.

Data Analysis

Hierarchical linear models (HLMs) were used to examine questions regarding within-and between- group differences in initial status and rate of change over time in phonological awareness skills. For the first model addressing the first research question, chronological age was nested within participant as the marker of change over time. Group was the primary predictor, and the interaction between group and chronological age tested group differences in the rate of phonological awareness growth over time. Next, nonverbal mental age and maternal education were tested as covariates, along with their interaction with time. Given that the nonverbal mental age of the participants varied over time, this variable was treated as a time-varying predictor. Chronological age and an intercept were included as random effects; all other effects were fixed. Following the addition of each variable to the model, changes in deviance statistics were tested using the χ^2 distribution to confirm a significant improvement in model fit at $p < .05$. Covariates that did not have a significant impact on the outcome were dropped from the final model. To reduce collinearity and facilitate interpretation of the main effects, chronological age, mental age, and maternal education were centered at the grand mean at initial status.

Given prior research indicating a plateau in literacy acquisition starting at 10 years in FXS (e.g., Bailey et al., 2009; Roberts et al., 2005), an alternative nonlinear change model was conducted to test for discontinuity in the rate of phonological awareness growth after 10 years of age in the FXS group (research question #2). First, a linear HLM model was fit to describe phonological awareness change only within the FXS group, employing the same approach and covariates as for the model described above, minus the “group” variable. After a final model was specified, a second time-varying temporal predictor representing the epochs before and after 10 years was added as a random effect. This additional temporal predictor allowed each individual trajectory to have two distinct slopes: one representing the rate of change before 10 years and the other representing change after 10 years (Singer & Willett, 2003).

To address the third research question, another series of HLMs was conducted to test autism severity (CARS) and FMRP as predictors of phonological awareness growth within the FXS group over time. CARS scores were centered at the group mean at the initial wave of data collection and treated as a time-varying predictor. A log transformation was applied to the FMRP levels to correct for positive skewing; FMRP was treated as a time-invariant. Age, maternal education, and their interactions with time were included as covariates. Age and an intercept were included as random effects; all other effects were fixed. Deviance statistics were evaluated to determine whether more complex models provided significantly better fit to the data than models with fewer predictors.

To address the fourth research question, linear regression was used to test phonological awareness skills as a predictor of concurrent reading skills in both groups then as a predictor of later reading skills in the FXS group. The initial model included the control variables of group and mental age. Next, phonological awareness and its interaction with group were entered to determine the degree to which phonological awareness uniquely predicted reading skill after accounting for control variables and if these differed between groups.

Results

Phonological Awareness Growth in FXS and TD

The first HLM estimated growth in phonological awareness as a function of chronological age, group, and their interaction. There was a significant effect of group and a significant interaction between group and chronological age, with the FXS group exhibiting slower annual growth in phonological awareness than the TD group (see Figure 1). Next, the effects of nonverbal mental age and maternal education and their interactions with time were examined. There was a significant effect for mental age, but the interaction between group and chronological age was reduced and no longer significant (p = .095) after controlling for mental age (see Figure 1). These results indicated that boys with FXS and TD boys had similar rates of phonological awareness growth over time with mental age in the model, highlighting the significant effect of mental age in these analyses. The effects of maternal education, the interaction of maternal education and time, and the interaction of mental age and time were not significant and were dropped from the final model (see Table 2).

Nonlinear Phonological Awareness Growth in FXS

To test for nonlinear change, an HLM model was fit to describe linear change within the FXS group (estimates are presented in Table 3). Then, a second time-varying predictor was added to the model to test for discontinuity in slope. The estimate for the time-varying predictor was significant (p = .034), indicating a significant discontinuity in the rate phonological awareness growth before and after 10 years of age. Deviance statistics indicated that the discontinuous change model was a better fit to the data than the linear change model (p <.05. These results indicate that the rate of phonological awareness growth slowed after 10 years of age in the FXS group; see Figure 2.

Predictors of Phonological Awareness Growth in FXS

We examined the effect of autistic behavior (i.e., CARS scores) and FMRP on the growth of phonological awareness in boys with FXS. Mental age had a significant effect on the rate of phonological growth in FXS (p = .015). Although deviance statistics indicated that a model including CARS and FMRP was significantly better fitting (p < .05), the estimates for both CARS and FMRP were non-significant within the model (both p >.10). Estimates from the final model are provided in Table 4.

Phonological Awareness Associated with Literacy

Finally, we examined phonological awareness in relation to concurrent and future basic reading abilities in both groups. Zero-order correlations are displayed in Table 5. Table 6 reports the results of hierarchical linear regression models predicting basic reading abilities at Time 1 in both groups. In the first step, nonverbal cognitive abilities and group accounted for 22% of the variance in basic reading skills; the addition of phonological awareness accounted for an additional 18% of unique variance. In Step 3, the phonological awareness by group interaction term was not significant. Thus, phonological awareness skills predicted concurrent letter-word identification skills in a similar manner across groups. Next, we examined the longitudinal relationship between phonological awareness at Time 1 and the basic reading abilities at the latest assessment point for the FXS group alone. Phonological awareness accounted for 16% of the unique variance in later reading after controlling for nonverbal mental age. However, when reading at Time 1 was entered as the autoregressor, it accounted for the majority (64%) of the variance, and phonological awareness was no longer a significant contributor to the model (see Table 7).

Discussion

This study examined growth in phonological awareness and its relationship to basic reading skills in males with FXS as compared with TD peers matched for nonverbal cognitive abilities. Analyses examining growth across chronological age indicated that the boys with FXS evidenced slower growth in phonological awareness than the TD group; however, the difference in growth rates was reduced and not significant after controlling for nonverbal cognitive abilities. These findings suggest that although males with FXS displayed a general weakness in phonological awareness skills relative to chronologically same-aged TD peers, phonological awareness growth was commensurate with nonverbal cognitive development.

Our second research question examined whether children with FXS exhibited a plateau in phonological awareness growth after age 10, consistent with previous studies pinpointing a plateau in literacy-related skills at that time (e.g., Bailey et al., 2009; Roberts et al., 2005). Our results indicated that there was a significant discontinuity in the rate of change in the FXS group, such that phonological awareness grew at a slower rate after age 10 than before age 10. Future studies should examine what is the mechanism that explains this potential plateau. For example, one hypothesis is that biological mechanisms lead to a general cognitive slowing as children with FXS approach adolescence. Another hypothesis is that as children with FXS move into adolescence, instructional goals and methods change, perhaps due to a focus on other “functional” skills, such that basic literacy skills do not grow as quickly. Our data cannot address these questions, but we believe the finding of a significantly different rate of growth on measures that are psychometrically appropriate for measuring growth (i.e., W scores from WRMT) is an important finding that should be followed up in future studies.

The third research question examined whether autism symptomatology and FMRP level showed associations with phonological awareness development within FXS. These two features have been established as significant predictors of other aspects of development, including nonverbal cognitive ability (Skinner et al., 2005) and social communication skills

(Losh, Martin, Klusek, Hogan-Brown, & Sideris, 2012). Results of our models were somewhat difficult to interpret. Although the model fit was better when autism symptomatology and FMRP level were included, their estimates within the model were non-significant, suggesting that once nonverbal cognitive skills are controlled, autism symptoms and FMRP do not significantly predict phonological awareness growth. However, because our data may not have been sufficiently powered to detect such predictors, these findings should be reexamined in future studies.

The fourth research question asked whether phonological awareness shows a similar relationship with basic reading in boys with FXS as in TD boys. We found that phonological awareness uniquely explained individual differences in basic reading skills after controlling for group and mental age. Furthermore, the relationship between phonological awareness and basic reading did not significantly differ between groups. Although this study involves correlational data, such findings raise the question of whether phonological awareness might have a similar causal relationship with word reading in children with FXS as it does for TD children. Results of predictive analyses involving only the FXS group indicated that phonological awareness maintains a similarly strong relationship with basic word reading skills over time. However, early phonological awareness did not add to the prediction of later reading skills after controlling for initial reading level.

We are unaware of any published studies comparing approaches for teaching reading or pre-literacy skills to children with FXS. Thus far, clinical experts have recommended visual “sight word” or whole-language approaches over phonics-based approaches, as a means of working around working memory and sequential processing deficits in this population (Braden, 2002; Braden, 2004). Sight word approaches can be effective for a specific set of target words; however, one limitation of these approaches is that students are not able to generalize the knowledge to decode new words. For TD students, this limitation is best overcome using a phonics-based approach; such programs have been found to be highly effective for most (though not all) TD children (Ehri et al., 2001). Significant effects of phonological awareness and phonics-based reading instruction have also been found for children with Down syndrome and other moderate to severe intellectual disabilities, although effect sizes have tended to be more modest (Browder, Wakeman, Spooner, Ahlgrim-Delzell, & Algozzine, 2006; Goetz et al., 2008; Lemons & Fuchs, 2010). It remains to be determined whether such approaches will prove to be effective for children with FXS. However, the significant relationship between phonological awareness and basic reading skills in this study suggests that tests of the feasibility and effectiveness of phonological awareness interventions in children with FXS are warranted.

This study is the first to link linguistic skills to literacy outcomes in FXS, but phonological awareness is but one. More research is needed to investigate relationships between other factors known to impact literacy in typical development, including vocabulary, orthographic awareness, and higher-level language skills in children with FXS and other neurodevelopmental disorders. For example, while the initial relationship between phonological awareness and reading development may be similar for TD children and children with Down syndrome, the longitudinal relationship appears to differ in subtle, yet important ways (Hulme et al., 2012; Steele, Scerif, Cornish, & Karmiloff-Smith, 2013).

Understanding the similarities and differences between emergent literacy skills and reading development in children with FXS is important for the development of valid assessments and effective instructional methods for this population, as well as other children with inherited intellectual disabilities.

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Key Points

- Reading delays are well documented in children with fragile X syndrome (FXS), but few studies have examined linguistic precursors of reading in this population.
- We examined the development of phonological awareness longitudinally, and its relationship with reading performance in boys with FXS and typically developing (TD) peers who were matched on nonverbal mental age at study outset.
- Although phonological awareness in boys with FXS was delayed compared to chronological age norms, it was commensurate with their nonverbal cognitive development.
- The rate of change in phonological awareness in boys with FXS slowed significantly after age 10.
- Phonological awareness was significantly associated with basic reading skills in both FXS and TD groups.

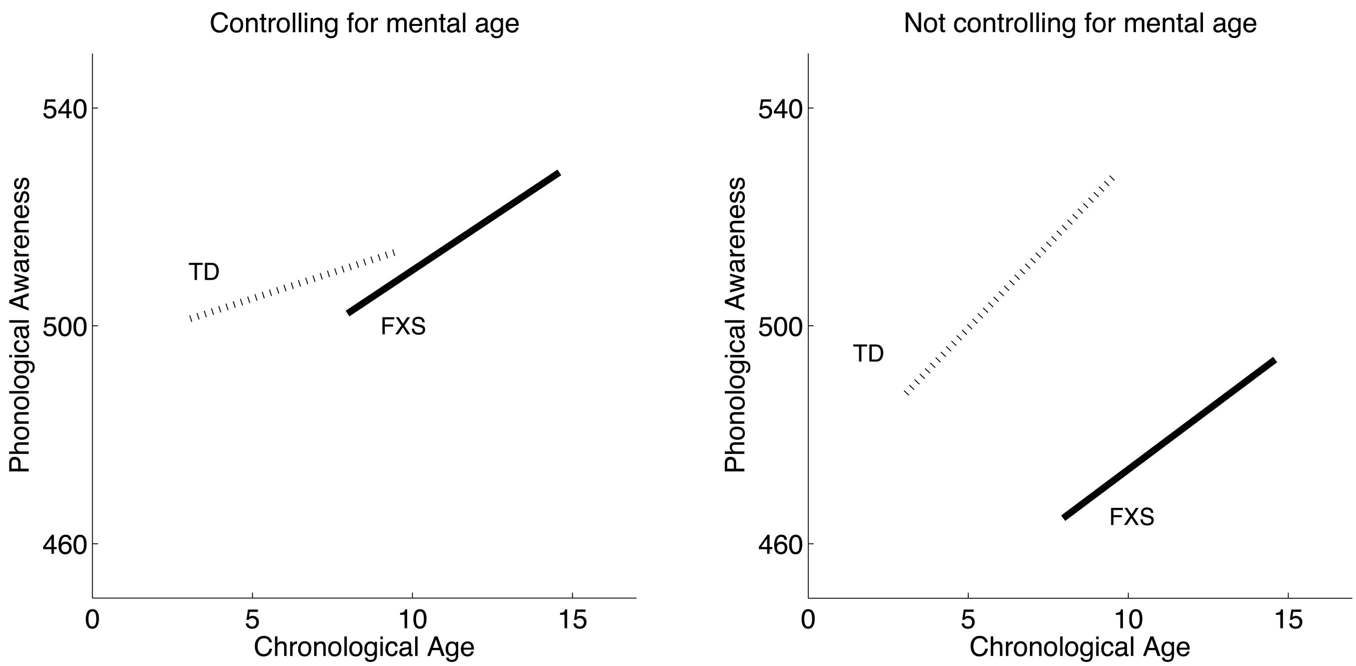

Figure 1.
Growth in Phonological Awareness by Group

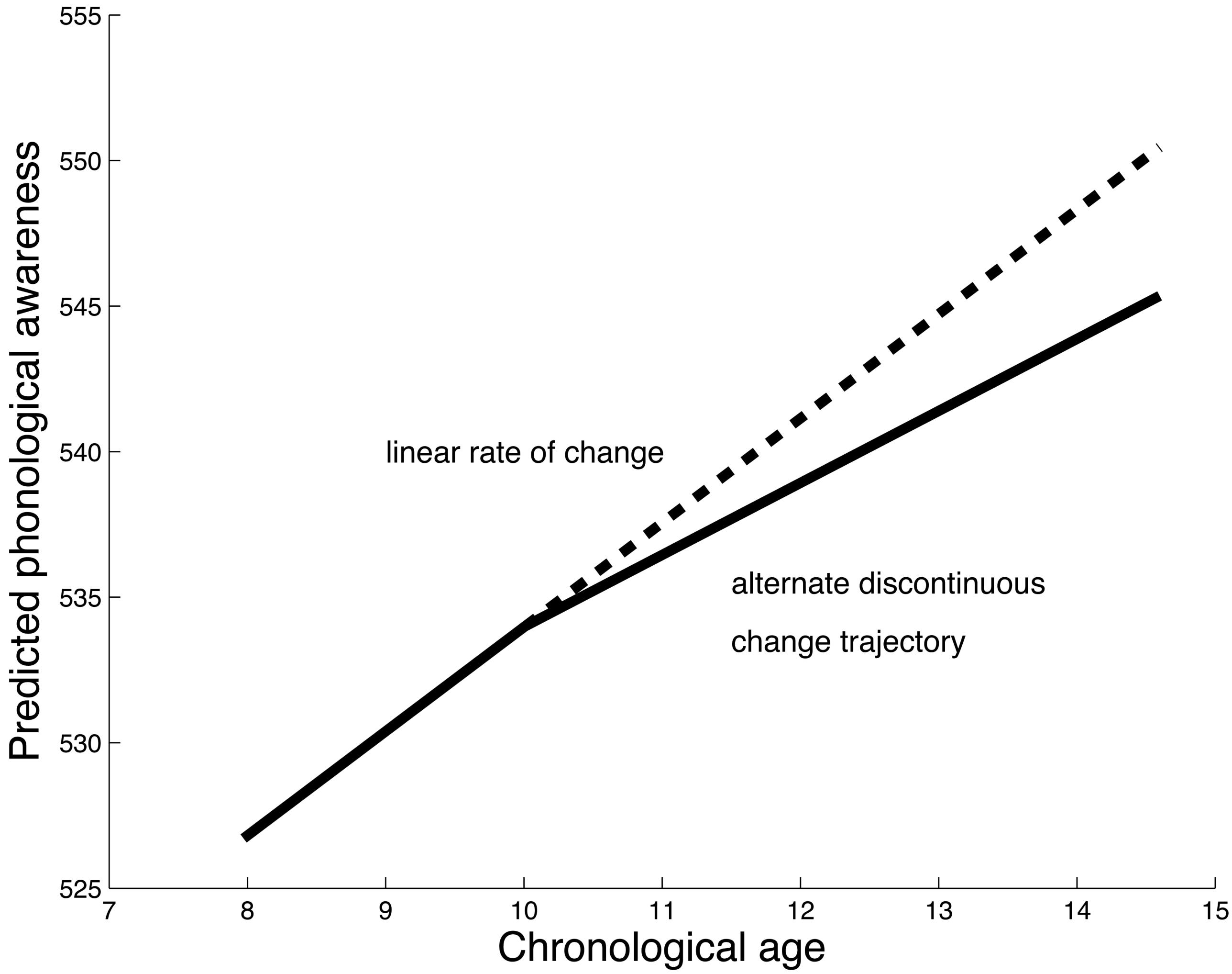

Figure 2.
Alternate Nonlinear Change Trajectories for Boys with FXS

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Table 1

Demographic and Descriptive Data for First Assessment

Variable	FXS			TD		
	n	***M (SD)***	**Range**	***n***	***M (SD)***	**Range**
Chronological Age	54	10.26 (1.67)	7.97-14.04	53	5.13 (0.88)	3.06-7.48
Maternal Education	47	14.28 (1.41)	13.00-17.00	48	15.15 (1.44)	13.00-17.00
Nonverbal Mental Age	52	5.31 (0.64)	4.00-6.67	53	5.23 (0.92)	2.83-7.50
Phonological Awareness	53	478.92 (16.91)	446.00-507.00	53	488.15 (12.21)	437.00-511.00
Basic Reading	52	398.65 (30.90)	335.00-450.00	39	388.97 (21.17)	350.00-450.00
FMRP[1]	42	9.11 (8.24)	1.50-40.00			
Autistic Behavior[2]	50	27.55 (4.80)	17.00-38.50			

Note. Descriptive statistics represent values at Time 1.

[1]Fragile X mental retardation 1 protein

[2]Childhood Autism Rating Scale.

Table 2

Longitudinal Analysis Testing Group Differences in Initial Status and Growth in Phonological Awareness over Time

	Effect	**Estimate**	**SE**	**DF**	***t***	***p***	**Deviance Statistics (−2 log likelihood)**	**Pseudo R^2 Statistic**	
Model 1	Intercept	487.98	1.57	105	310.87	<.001*	1915.5		
	Time	2.08	0.39	83	5.38	<.001*			
Model 2	Intercept	429.63	4.99	104	86.15	<.001*	1795.8	.37	.34
	Time	2.56	1.39	82	1.84	.069			
	Group (TD)	39.21	3.03	50	12.96	<.001*			
	Group*Time	1.84	0.90	50	2.05	.045*			
Model 3	Intercept	445.77	5.59	102	79.80	<.001*	1747.9	.46	.46
	Time	3.91	0.59	81	6.65	<.001*			
	Group (TD)	24.36	4.03	49	6.04	<.001*			
	Group*Time	−2.01	1.18	49	−1.70	.095			
	Mental Age	4.81	0.92	49	5.14	<.001*			

Note. Model 1 is the unconditional growth model. Model 2 is the initial model of differences in growth between groups with no covariates. Model 3 includes estimates from the final model, which considered all covariates. Maternal education, maternal education*time, and mental age*time did not have a significant impact on the outcome and were dropped from this final model. R^2_0 is the proportion of between-person variance in initial status explained by the model. R^2_1 is the proportion of between-person variance in rate of change explained by the model.

* $p < .05$

Table 3

Longitudinal Analysis Testing Nonlinear Change in in Phonological Awareness over Time

	Effect	Estimate	SE	DF	*t*	*p*	Deviance Statistics (−2 log likelihood)	Pseudo R^2 Statistic	
Model 1	Intercept	479.74	1.94	52	246.79	<.001*	956.2		
	Time	4.51	0.68	39	6.64	<.001*			
Model 2	Intercept	483.56	1.87	50	258.77	<.001*	925.6	.40	.40
	Time	3.58	0.70	39	5.13	<.001*			
	Mental Age	8.02	1.83	31	4.38	0.001*			
Model 3	Intercept	488.06	2.46	49	198.21	<.001*	915.5	.35	.34
	Time	9.74	2.47	39	3.95	<.001*			
	Mental Age	7.03	1.75	24	4.02	<.001			
	Temporal predictor of change before/ after 10 years	−7.26	2.77	7	−2.63	0.034*			

Note. Model 1 is the unconditional growth model. Model 2 includes estimates from the final linear model, which considered all covariates. Maternal education, maternal education*time, and mental age*time did not have a significant impact on the outcome and were dropped from this final model. Model 3 adds a second temporal predictor to the final model, testing discontinuous change before and after 10 years of age. $R^2{}_0$ is the proportion of between-person variance in initial status explained by the model. $R^2{}_1$ is the proportion of between-person variance in rate of change explained by the model.

* $p < .05$

Table 4

Predictors of Phonological Awareness Change in FXS

	Effect	Estimate	*SE*	*DF*	*t*	*p*	Deviance statistics (−2 log likelihood)	Pseudo R^2 Statistic R^2_0	R^2_1
Model 1	Intercept	479.74	1.94	52	246.79	<.001*	694.2		
	Time	4.51	0.68	39	6.64	<.001*			
Model 2	Intercept	474.59	5.13	36	92.58	<.001*	522.8	.48	.50
	Time)	3..76	0.79	25	4.74	<.001*			
	FMRP	8.19	5.41	8	1.51	.169			
	CARS	0.54	0.31	8	1.77	0.114			
	Mental Age	7.23	2.36	8	3.06	.016			

Note. Model 1 is the unconditional growth model. Model 2 includes estimates from the final model. Maternal education, maternal education*time, mental age*time, and CARS*time did not have a significant impact on the outcome and were dropped from the final model. R^2_0 is the proportion of between-person variance in initial status explained by the model. R^2_1 is the proportion of between-person variance in rate of change explained by the model.

* $p < .05$

Table 5

Correlations between Nonverbal Mental Age, Phonological Awareness, and Letter-Word Identification Skills

	1	2	3
1. WJ PA	1.00	.582**	.677**
2. Mental Age	.54**	1.00	.625**
3. WJ LWID (Concurrent)	.568**	.371*	1.00
4. WJ LWID (Predictive)	.568**	.473*	.935**

Note. Shaded cells list correlations for the FXS group; non-shaded cells list correlations for the TD group. WJ PA = Phonemic Awareness composite score of the Woodcock-Johnson III Test of Cognitive Abilities; WJ LWID = Letter-Word Identification subtest of the Woodcock-Johnson Test of Academic Achievement- Revised.

* $p < .01$

** $p < .001$

Table 6

Regression Coefficients Depicting Initial Phonological Awareness as a Predictor of Concurrent Basic Word Reading

	Effect	*B (SE)*	*t*	*p*	R^2
Step 1	Intercept	323.75 (20.25)	15.99	<.001*	.22
	Group	−8.55 (5.29)	−1.62	.110	
	Mental Age	1.31 (0.29)	4.59	<.001*	
Step 2	Intercept	−59.34 (77.60)	−.77	.447	.40
	Group	−18.53 (5.06)	−3.66	<.001*	
	Mental Age	0.55 (0.29)	1.86	.066	
	Phonological Awareness	0.92 (0.18)	5.07	<.001*	
Step 3	Intercept	−57.79 (230.76)	−0.25	.803	.40
	Group	−19.78 (175.67)	−0.11	.911	
	Mental Age	0.55 (0.30)	1.80	.075	
	Phonological Awareness	0.92 (0.48)	1.94	.056	
	Phonological Awareness*Group	0.00 (0.36)	0.02	.994	

* $p < .05$

Table 7

Regression Coefficients Depicting Initial Phonological Awareness as a Predictor of Later Basic Word Reading in FXS

	Effect	*B (SE)*	*t*	*p*	R^2
Step 1	Intercept	275.92 (42.84)	66.44	<.001*	.21
	Mental Age	2.09 (0.66)	33.17	.003*	
Step 2	Intercept	−105.36 (134.16)	−0.79	.437	.37
	Mental Age	1.11 (0.69)	1.61	.115	
	Phonological Awareness	0.92 (0.31)	2.97	.005*	
Step 2	Intercept	30.38 (24.82)	1.22	.229	.88
	Mental Age	0.15 (0.30)	0.50	.623	
	Time 1 Word Reading	0.92 (0.07)	13.40	<.001*	
Step 3	Intercept	55.25 (61.05)	0.91	.372	.88
	Mental Age	0.19 (0.32)	0.60	.553	
	Time 1 Word Reading	0.94 (0.08)	11.90	<.001*	
	Phonological Awareness	−0.07 (0.16)	−0.45	.658	

*$p < .05$