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Direct Magnitude Estimation of Articulation Rate in Boys with Fragile X Syndrome

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[†] Posthumous

Abstract

Purpose—This study a) compared perceived articulation rate of boys with fragile X syndrome (FXS) to chronologically age-matched boys (CA), and b) determined segmental and/or prosodic factors which account for perceived rate.

Method—Ten listeners used direct magnitude estimation (DME) procedures to judge the articulation rates of 7 boys with FXS only, 5 boys with FXS and a diagnosis of autism spectrum disorder (ASD), and 12 CA boys during sentence repetition. Sentences had similar articulation rates in syllables per second as determined acoustically. Four segmental/prosodic factors were used to predict perceived rate: a) percent consonants correct, b) overall fundamental frequency (F_0) level, c) sentence-final F_0 drop, and d) acoustically-determined articulation rate with the final word of the sentence excluded.

Results—Boys with FXS-ASD were judged to talk faster than CA controls. Multiple linear regression indicated that articulation rate with the final word of the sentence excluded and sentence-final F_0 drop accounted for 91% of the variance for perceived rate.

Conclusions—The findings suggest that descriptions of speakers with FXS as having fast and/ or fluctuating articulation rates may be influenced by autism status. Also, atypical sentence-final prosody may be related to perceived rate in boys with FXS-ASD. Clinical implications are discussed.

Keywords

fragile X syndrome; speaking rate; articulation rate; direct magnitude estimation; prosody

Fragile X syndrome (FXS) is the most common inherited cause of intellectual disability in males, with an estimated prevalence of about 1 in 4,000 (Crawford, Acuna, & Sherman, 2001; Turner, Webb, Wake, & Robinson, 1996). Most boys with FXS have moderate to severe delays in speech and language with speech intelligibility affected to various degrees. In addition, many boys with FXS have difficulties in adaptive and social skills and often exhibit behaviors characteristic of autism: poor eye contact, social withdrawal, stereotypic/repetitive behaviors, and hyperactivity (Cohen et al., 1988; Dykens, Hodapp, Ort, & Leckman, 1993; Kerby & Dawson, 1994; Reiss & Freund, 1992). Indeed, there is general

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agreement that 15–25% of males with FXS meet diagnostic criteria for autism (Bailey et al., 1998; Dykens & Volkmar, 1997; Hagerman, 2002; Turk & Graham, 1997); recent figures are as high as 30–50% (Hagerman, 2006; Hall, Lightbody, & Reiss, 2008; Rogers et al., 2001).

One of the more commonly reported speech characteristics among boys with FXS is a fast speaking rate (Borghgraef, Fryns, Dielkens, Pyck, & Van den Bergh, 1987; Brun-Gasca & Antigas-Dallares, 2001; Hanson, Jackson, & Hagerman, 1986; Reis & Freund, 1992). Hanson et al. (1986) described a "fast and fluctuating rate of speech" in 9 of 10 boys (ages 3 to 8 years) with FXS which they believed was characteristic of "cluttering." Borghgraef et al. (1987) surveyed the parents, speech-language pathologists, and teachers of 23 boys with FXS (ages 2 to 12 years) on various speech characteristics and found that 85% of the boys were reported to use a "rapid speech rhythm." Reis and Freund (1992) found that parents reported that 34 boys with FXS (3 to 18 years old) had an unusual rate of speech as compared with 32 boys with developmental delays. More recently, Brun-Gasca and Antigas-Dallares (2001) noted in a review article that "unequal rhythm" was a primary speech characteristic of children with FXS.

In contrast to the above literature, Zajac et al. (2006) reported similar articulation rates as determined by acoustic analysis between young boys with FXS and typically developing boys matched on chronologic age (CA). Zajac et al. suggested that articulation rate might not be a primary factor contributing to perceptual impressions of increased speaking rate in boys with FXS. They speculated that prosodic factors such as utterance duration, number and duration of pauses within an utterance, and intonation contours might better account for the perceived fast speaking rate. Prosodic differences could be contributing to the fast rate in some males with FXS given disordered prosody is a frequently reported characteristic of many individuals with autism (McCann, Peppe, Gibbon, O'Hara, & Rutherford, 2007; Shriberg et al., 2001) and the high occurrence of autism as a co-diagnosis in boys with FXS. Zajac et al. (2006), however, determined mean articulation rates and did not attempt to evaluate fluctuations in rate which may have occurred within an utterance. Furthermore, Zajac et al. did not identify boys with FXS who may have been diagnosed with autism.

The purpose of the present study was to further analyze a subset of speakers previously investigated by Zajac et al. (2006) relative to perceived speaking rates. Two specific research questions were proposed: 1) Do listeners perceive boys with FXS as speaking faster than CA controls even when utterances are matched on overall acoustically-determined articulation rate? 2) If so, what factors within the utterances account for perceived articulation rate? To answer these questions, we first selected boys with FXS and CA controls who had similar acoustically-determined articulation rates during a sentence imitation task. Approximately half of the boys with FXS were also diagnosed with "autism" or "autism spectrum" disorder (ASD). Next, we used direct magnitude estimation procedures to determine perceived articulation rates. To account for perceived rate, we then determined a) the accuracy of the sounds in the sentences, b) overall fundamental frequency (F_0) levels of the sentences, c) sentence-final intonation contours, and d) acousticallydetermined articulation rate of the sentence minus the final word. The latter measure was used to account for rate fluctuations that might have occurred within a sentence. We selected a sentence imitation task because it largely controlled for the effects of utterance duration and pausing within the utterance. We focused on F₀ and sentence-final intonation as prosodic factors because previous research has suggested that overall pitch level and changes in pitch influence perception of articulation rate (e.g., Ainsworth & Lindsay, 1986; Kohler, 1986; Rietveld & Gussenhoven, 1987).

Method

Participants: Speakers and Listeners

Speakers were a subset of 12 young boys with FXS and 12 typically developing boys matched on CA previously investigated by Zajac et al. (2006). Additional details about the larger sample of boys have been reported by Roberts et al. (2005) and Zajac et al. (2006). Five of the boys with FXS also met the criteria for ASD according to the Autism Diagnostic Observation Schedule (ADOS) (Lord, Rutter, DiLavore, & Risi, 2002). All boys selected for the present study were between the ages of 10 to 15 years. Mean age of boys with FXS only (FXS-O) was 11.6 years (SD=1.5); mean age of boys with FXS and a co-diagnosis of ASD (FXS-ASD) was 12.4 years (SD=1.7); mean age of CA controls was 12.6 years (SD=1.7). These boys were selected because they imitated sentences at similar articulation rates as described below.

Thirteen adult listeners (11 females, 2 males) participated in two direct magnitude estimation (DME) procedures described below. They were between the ages of 20 to 25 years. All listeners were undergraduate students recruited from the University of North Carolina at Chapel Hill campus who were naïve to the purpose of the study. Each listener passed a pure-tone hearing screening at 25 dB HL in the better ear at 500, 1000, 2000, and 4000 Hz. In addition, all listeners reported 1) English as a first language, 2) no history of speech or hearing disorders, and 3) being unfamiliar with speech characteristics of FXS.

Speech Sample

As part of the larger project, the boys repeated a set of 15 sentences modeled by an examiner (see appendix). Seven of the 15 sentences were first used by Gordon-Brannan and Hodson (2000). The 15 sentences were all simple declarative statements that varied in length from 5 to 7 syllables and were modeled with a falling intonation contour. Perceptual and acoustic analysis using F_0 tracking of the recorded examiner models confirmed that all sentences were produced with a falling intonation.

Audio Recording Procedures

The boys were audiotape recorded using a portable digital audio tape (DAT) recorder (TASCAM, DA-P1) at a sampling rate of 44.1 kHz. A Shure WBH headset microphone was positioned approximately 3 inches from the child's mouth. Location of the recording sessions varied among the boys. They were recorded in quiet rooms either at the laboratory, in their school, or in their home.

Determination of Articulation Rate

The 24 boys included in the present study (7 FXS-O, 5 FXS-ASD, 12 CA controls) were selected solely on the basis of acoustically-determined articulation rate. Specifically, a boy was included if he produced at least 3 sentences that contained at least 6 syllables with an articulation rate within a range of 3.5 to 4.5 syllables/s. In addition, all sentences selected had no acoustically-determined pauses that exceeded 200 ms.

To determine overall articulation rate, the 72 sentences (24 boys times 3 sentences) were digitally transferred to the Computerized Speech Lab (CSL, Kay Elemetrics, Inc, Model 4400), low-pass filtered, and down sampled to 22.05 kHz. The second author (AH) determined the duration of the utterance in seconds and counted the number of syllables to calculate articulation rate in syllables/s. Conventional criteria were used to identify segment onsets and offsets of the sentences (Kent & Read, 2001; Klatt, 1976). Mean articulation rate was calculated for each speaker based upon the 3 sentences.

We targeted an articulation rate of approximately 4.0 syllables/s because it was similar to the mean rate of the larger sample of males with FXS and CA controls. Also, because we were interested in the effects of articulation, sentences were not excluded if produced with speech errors (e.g., sound omissions or substitutions). By using these criteria, we were able to match boys on overall acoustically-determined articulation rate and eliminate length of utterance and pauses within the utterance as possible confounders to perceived rate. As described below, although overall articulation rate was controlled, boys with FXS often exhibited atypical sentence-final intonation which caused rate fluctuations within the sentences. This occurred most often for boys with FXS-ASD.

DME Procedures

DME procedures were used to scale perceived rate of articulation. These procedures were similar to those reported in the literature for scaling hypernasality (Zraick & Liss, 2000; Whitehill, Lee, & Chun, 2002), intelligibility (Yunusova, Weismer, Kent, & Rusche, 2005), and speaking rate (Turner & Weismer, 1993). All 72 sentences were transferred to a perceptual rating program (Ratewav, PERCI-SARS, Version 1.1). A standard sentence representing an articulation rate of 4.0 syllables/s was assigned a modulus value of 100. The standard sentence was produced by a CA boy who was not part of the present study. Although the standard sentence was selected primarily on the basis of acoustically-determined articulation rate, it was also judged to have a perceptually normal rate and a natural sentence-final falling intonation. The standard sentence was presented at the beginning of the rating session and after every 6 sentences.

Ten listeners were instructed to use the standard sentence as a referent when estimating the rate of the sentences. They were instructed, for example, to assign a sentence the number 50 if they judged the rate to be half as fast as the standard or the number 200 if they judged the rate to be twice as fast. Sentences were played via a loudspeaker to individual listeners in a sound-attenuated booth at a distance of approximately 3 feet and a sound pressure level of approximately 70 dB(C). Before judging the actual sentences, the listeners were presented with five practice sentences representing a range of articulation rates produced by boys not in the present study. For the actual ratings, the Ratewav program randomized the order of the 72 sentences for each listener. The listeners recorded their responses on pre-numbered forms.

The above DME procedures used a standard sentence spoken by a CA boy who exhibited perceptually normal rate and intonation. Because of this, the listeners may have experienced difficulty when attempting to judge the rate of boys with FXS who used atypical prosody and/or fluctuating rate. To determine if the use of a standard sentence spoken by a CA boy affected the validity of the DME procedures, three additional listeners participated in a second DME task that used a sentence produced by a boy with FXS as a standard. This sentence was also produced with an overall acoustically-determined articulation rate of 4.0 syllables/s but included a perceptually salient rise-fall intonation during the final word. The three additional listeners judged the original 72 sentences for perceived rate using the same DME procedures as described above. In addition, 15 of the sentences were randomly repeated.

Predictor Variables

One segmental and three prosodic variables were determined from the sentences as predictors of perceived rate: a) percent consonants correct (PCC), b) overall F_0 level, c) sentence-final intonation contour, and d) acoustically-determined articulation rate minus the final word of the sentence (hereafter rate_mfw).

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PCC—Because poor consonant articulation may affect perceived rate of speaking (Kent & Rosenbek, 1982), we determined percentage of consonants correct (PCC) for all boys. For each sentence, PCC was calculated to assess accuracy of consonant production (Shriberg & Kwiatkowski, 1982). PCC is the number of correct consonants divided by the number of consonant targets, and was computed using Computerized Profiling (Long, Fey, & Channel, 2003).

F₀ and Sentence-Final Intonation Contour—The first author (DZ) used the pitch trace routine of TF32 (Milenkovic, 2001) to extract the fundamental frequency (F₀) contours of the sentences produced by 10 of the boys with FXS and 8 of the CA boys. Six boys (2 FXS, 4 CA controls) were excluded because the pitch trace failed to track F₀ completely during one or more of the sentences. Use of additional acoustic programs (e.g., CSL, Praat) could not resolve all F₀ contours. The sentences of the remaining boys were relatively free of F₀ tracking errors. Small tracking errors less than 10 to 20 Hz such as those illustrated in Figure 2 below were not corrected. Mean F₀ during the entire sentence was determined for each boy using the summary statistics provided by the TF32 software.

Because most of the boys with FXS tended to use a relatively extreme rise-fall and/or falling intonation pattern at the end of their sentences (see below), we determined the difference in F_0 from the beginning to the end of the final word of the sentence. When final words consisted of a single syllable, this measure usually reflected the maximum F_0 fall. When final words consisted of two syllables, this measure did not always reflect the maximum F_0 fall (see Figure 2). To control for differences in overall F_0 level, final F_0 drops were converted from Hertz to semitones.

Rate_mfw—Because atypical sentence-final intonation contours such as a rise-fall pattern will prolong segment durations (see Figure 2), we also determined articulation rate for each sentence with the final word excluded. The first author used TF32 to determine the duration of the sentence minus the final word.

Data Analysis

Geometric means of perceived articulation rate were calculated for each boy based on the scores from all 10 listeners in the first DME task and from all three listeners in the second DME task. Arithmetic means of PCC, F_0 level, final F_0 drop, and rate_mfw were calculated based upon the 3 sentences produced by each boy.

Reliability

Acoustically-determined articulation rate—The second author (AH) repeated rate measurements on 25 randomly selected sentences to determine intra-examiner reliability for overall articulation rate. A trained research assistant repeated the rate measurements for all 72 sentences to determine inter-examiner reliability. A correlation coefficient of .87 (p<. 001) was obtained between the measurements repeated by the second author. The mean difference between measurements (0.02 syllables/s) was not significant (t[24]=-0.403, p>. 05). A correlation coefficient of .92 (p<.001) was obtained between the articulation rate measurements made by the second author and the trained research assistant. The mean difference between measurements (0.04 syllables/s) was not significant (t[71]=1.548, p>. 05).

DME of articulation rate—Intra-listener reliability of the DME ratings was assessed by calculating intraclass correlation coefficients for the three listeners in the second DME task who repeated ratings for 15 sentences. These correlations were .74, .91, and .47, respectively. Although the correlation for one listener was relatively low, the mean

difference for all three listeners between repeated ratings (9 scale points) was not significant (t[44]=-1.671, p>.05).

Inter-listener reliability of the DME ratings was estimated in two ways. First, Cronbach's alpha was calculated for all listeners in the first DME task. As noted by Whitehill et al. (2002), this measure is equivalent to intraclass correlation type (3,k) described by Shrout and Fleiss (1979). The reliability of the DME ratings was .92. Second, split-half reliability of the listeners in the first DME task was also determined. This was done by randomly assigning the 10 listeners to two groups of five listeners and calculating the mean DME ratings of the 24 boys for both groups of listeners. A correlation coefficient of .87 (p<.001) was obtained and there was no significant difference in mean ratings between the groups (t[23]=0.808, p>.05).

PCC—All sentences were glossed (orthographically transcribed) by two speech-language pathologists (SLP). Gloss reliability, calculated using word-by-word comparison, was 97.5%. A third SLP then phonetically transcribed speech productions that differed from the target production using transcription guidelines described by Shriberg and Kent (2003). Inter-observer agreement was calculated on approximately 17% of the sample (2 boys with FXS and 2 TD boys), and point-by-point comparison of consonant segments yielded an agreement of 89.5%.

Sentence-Final Intonation Contour—The first author repeated the measurement of sentence-final F_0 drop on 25 randomly selected sentences. A trained research assistant repeated the measurements on all 54 sentences (18 speakers times 3 sentences). A correlation coefficient of .95 (p<.001) was obtained between the F_0 measurements repeated by the first author. The mean difference between measurements (5.4 Hz) was not significant (t[24]=1.135, p>.05). A correlation coefficient of .94 (p<.001) was obtained between the measurements made by the first author and the trained research assistant. The mean difference between measurements (5.1 Hz) was not significant (t[53]=1.478, p>.05).

Rate_mfw—Intra-examiner reliability of the first author's measurements of rate_mfw was determined by randomly selecting 20 sentences and repeating the measurements. A correlation coefficient of .99 (p<.001) was obtained between the repeated measurements. The mean difference between measurements (0.04 syllables/s) was not significant (t[19]=-1.111, p>.05). Inter-examiner reliability was determined by having the second author repeat the measurements for 20 randomly selected sentences. A correlation coefficient of .96 (p<.001) was obtained between the first and second authors' measurements. The mean difference between measurement (0.04 syllables/s) was not significant (t[19]=-1.011) was obtained between the first and second authors' measurements. The mean difference between measurement (0.04 syllables/s) was not significant (t[19]=-0.021).

Validity of DME Ratings

Table 1 lists the geometric means (GM), standard deviations (SD), minimum (Min), and maximum (Max) of perceived articulation rate for the two DME tasks. As shown, regardless of the standard sentence used as a modulus, boys with FXS were judged to use a faster rate than CA boys. While the choice of a standard sentence influenced absolute DME ratings, the relative difference in perceived rate between boys with FXS and CA boys was essentially identical regardless of the standard sentence. That is, boys with FXS were judged to speak 21% faster than CA boys when a standard sentence from a CA boy was used as a modulus; they were judged to speak 20% faster when a standard sentence from a boy with FXS was used as a modulus. These findings are similar to Weismer and Laures (2002) who reported that relative differences in scaled intelligibility across dysarthric speakers were maintained despite the use of standards that varied in severity as a modulus.

Statistical Analysis

Question 1—To determine if listeners perceived boys with FXS as speaking faster than CA controls, an analysis of variance (ANOVA) was performed using perceived articulation rate obtained from the first DME task as the dependent variable and diagnostic category of boys as a factor with three levels (i.e., FXS-O, FXS-ASD, and CA).

Question 2—To determine what factors accounted for perceived articulation rate, multiple linear regression was performed. Perceived articulation rate obtained from the first DME task was used as the dependent variable. Five predictors of perceived rate were evaluated: 1) diagnostic category of the boys (FXS-O, FXS-ASD, and CA), 2) PCC of the sentences, 3) overall Fo level of the sentences, 4) final Fo drop of the sentences, and 5) rate_mfw of the sentences. Multiple linear regression was performed using stepwise, automatic forward entry of variables using SYSTAT 11 (2004).

Results

Question 1

Figure 1 shows geometric means (GM) and standard deviations (SD) of perceived articulation rate for the three groups of boys. Listeners perceived CA boys as speaking the slowest (GM=94, SD=23), boys with FXS-O as speaking faster (GM=107, SD=16), and boys with FXS-ASD as speaking the fastest (GM=125, SD=23). An ANOVA indicated a significant main effect of diagnostic group (F[2, 21]=7.750, p<.01). Post hoc Tukey tests indicated a significant difference between boys with FXS-ASD and CA boys (p<.01). Overall, boys with FXS-ASD were judged to speak approximately 33% faster than CA boys.

Question 2

Figure 2 illustrates examples of the acoustic waveforms, F₀ traces, and broadband spectrograms for the sentence The bus driver is smiling produced by a boy with FXS-ASD (top) and CA control (bottom). By design, the overall acoustically-determined sentence durations and articulation rates were similar between the boys (4.5 syllables/s for the boy with FXS-ASD and 4.3 syllables/s for the CA control). Perceived articulation rates, however, were 129 for the boy with FXS-ASD and 92 for the CA control. Clear differences are apparent in overall F₀ level and final F₀ drop between the boys. While the CA boy exhibited a relatively low F_0 level throughout the sentence and a slightly falling F_0 during smiling, the boy with FXS-ASD exhibited a relatively high F₀ with a sharp rise-fall contour during the word *smiling*. Figure 2 also reveals that acoustically-determined articulation rate with smiling excluded (rate_mfw) was much faster for the boy with FXS-ASD (6.0 syllables/s compared to 4.7 syllables/s for the CA boy). Because the boy with FXS-ASD used an atypical sentence-final intonation pattern, this prolonged segments of *smiling* and caused overall articulation rates to be similar for both boys. Among the 5 boys with FXS-ASD, 4 boys used a sentence-final F₀ pattern characterized by a rise-fall. Conversely, among the 5 boys with FXS-O, one used a rise-fall F_0 pattern.

Table 2 shows group descriptive statistics of the predictor variables for CA boys, boys with FXS-O, and boys with FXS-ASD. As shown, boys with FXS-ASD exhibited faster rate_mfw than boys with FXS-O and CA boys. Boys with FXS-O and FXS-ASD, however, exhibited higher F_0 levels and larger F_0 drops than CA boys. Table 3 shows the correlation matrix of the predictor variables with perceived articulation rate across all boys. Three variables were significantly correlated with perceived rate: a) rate_mfw (r=.82), b) final F_0 drop (r=.61), and overall F_0 level (r=.56).

Multiple regression analysis revealed that only rate_mfw and final F_0 drop were significant predictors of perceived rate accounting for 91% of the total variance (F[2, 15]=75.890, p<. 01). Rate_mfw accounted for 75% of the variance while final F_0 drop accounted for 16% of the variance.

Discussion

The purpose of this study was to determine perceived articulation rates in a subset of boys with FXS and CA controls previously investigated by Zajac et al. (2006). Two specific research questions were proposed: 1) Do listeners perceive boys with FXS as speaking faster than CA controls even when utterances are matched on overall acoustically-determined articulation rate? 2) If so, what factors within the utterances account for perceived articulation rate?

Relative to the first question, we found that when overall articulation rate in syllables per second was matched among groups, boys with FXS-ASD were judged to sound faster than CA boys. Although boys with FXS-O were also judged to sound faster than CA boys, the difference was not significant. These findings are somewhat at odds with previous research which suggests that FXS speakers are judged to exhibit a fast and fluctuating rate. While acoustic analysis of boys with FXS-ASD in our study revealed rate fluctuations, boys with FXS-O did not exhibit rate fluctuations compared to CA boys. We must emphasize, however, that previous studies have not reported the autism status of FXS speakers. Given that ASD may co-occur with FXS in up to 50% of cases (Hall et al., 2008), it is possible that previous reports may have focused on speakers who had both diagnoses.

Relative to the second question, we found that both boys with FXS-ASD and FXS-O exhibited higher F_0 levels and greater final F_0 drops than CA boys. In addition, boys with FXS-ASD exhibited faster articulation rates minus the final word of the sentence (rate_mfw) than the other groups of boys. Multiple regression analysis, however, indicated that only rate_mfw and final F_0 drop predicted approximately 75% and 16% of the variance in perceived articulation rate, respectively. While the large effect of rate_mfw was not unexpected, it highlights the perceptual influence of rate fluctuations within an utterance on perceived rate. The lack of diagnostic group as a significant predictor in the regression model was somewhat unexpected given that group was a significant effect in the ANOVA. The loss of group suggests that this variable was highly collinear with rate_mfw and final F_0 drop. Also, we must note that power was reduced for the multiple regression analysis compared to the ANOVA given that 6 boys were omitted due to missing F_0 data.

The above findings relative to F_0 are generally consistent with previous research which has shown that overall pitch level and changes in pitch influence perception of articulation rate (e.g., Ainsworth & Lindsay, 1986; Kohler, 1986; Rietveld & Gussenhoven, 1987). Kohler (1986), for example, conducted experiments using German speakers and listeners to show that the perception of speech rate is "multidimensional" and depends upon "various aspects of duration and F_0 " (p. 135). Specifically, Kohler reported that listeners perceived faster speech when overall F_0 levels were high and when utterance-final F_0 was rising; listeners perceived slower speech when utterance-final F_0 was falling. Kohler further noted that a) "sentence-internal F_0 structure" involving pitch movement was dominant over global pitch level, and b) falling F_0 was a "stronger" perceptual cue than rising F_0 .

Given the perceptual importance of falling F_0 suggested by Kohler (1986), one might question the relatively fast articulation rate estimated by listeners in the present study for boys with FXS-ASD who showed the largest final F_0 drop. This apparent discrepancy may be explained by a combination of a) faster articulation rates at the beginning of the sentence,

and b) the use of an extreme rise-fall F_0 pattern at the end of the sentence. Relative to the latter, it may be that an extreme F_0 rise is perceptually more salient than a F_0 fall when both occur within a sentence. Regardless of the actual perceptual mechanisms, the present findings provide additional evidence of the role of sentence-final intonation contours on the perception of rate.

Clinical Implications

In the present study, boys with FXS-ASD used a fast articulation rate during the initial part of a sentence and a perceptually distinctive rise/fall F_0 pattern at the end of a sentence. Atypical stress patterns have been reported to be characteristic of speakers with ASD. Indeed, Shriberg et al. (2001) state, "... the prosody characteristics of a person with autism constitute one of the most significant obstacles to his or her social integration and vocational acceptance" (p. 1099). We believe that targeting rate and/or atypical prosodic patterns of boys with FXS-ASD in speech intervention could possibly result in reduced social stigma associated with perceptually "unusual" or "peculiar" speech. Obviously, research is needed to explore the effectiveness of such intervention in boys with FXS-ASD.

Limitations

Several limitations of this study exist. First, some listeners find DME procedures difficult to perform even following adequate instructions and practice. As reported above, one listener in the second DME task showed relatively low intra-listener reliability. Although interlistener reliability in the main DME task was high, we did not obtain repeated judgments for intra-listener reliability in this task. Second, because we separated boys with FXS into groups based upon autism status, the numbers in each group were small. Power was additionally reduced for the multiple regression analysis given the omission of some boys due to missing F_0 data. This may have contributed to the loss of diagnostic group as a significant predictor. Last, because we used a sentence imitation task, the present findings may not be characteristic of boys with FXS during conversational speech.

Summary

A fast and/or fluctuating rate of speaking is a perceptual characteristic often reported for boys with FXS. Zajac et al. (2006) reported no significant difference between boys with FXS and CA controls during conversation when overall articulation rate was calculated in syllables per second across utterances. In that study, however, boys with FXS used shorter utterances and a tendency to pause less often. In the present study, we used a sentence imitation task to control for utterance duration and pausing while we determined perceived articulation rates. The findings confirm previous descriptions of a perceptually fast and fluctuating articulation rate only among boys with FXS and a co-diagnosis of ASD. The findings also suggest that atypical sentence-final intonation may contribute to perceived articulation rate in boys with FXS-ASD.

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Appendix

Sentences used during an imitation task as part of a larger study. Participants in the present study were selected if they produced any 3 of the sentences containing 6 or 7 syllables (indicated with asterisks) using an articulation rate of approximately 4.0 syllables per second without pausing.

- 1. The bus driver is smiling.*
- 2. He wears a scary mask.*
- 3. A baby bird is chirping.*
- **4.** Smoke comes out the chimney.*
- 5. The cowboy rides a horse.*
- 6. The rabbit eats a carrot.*
- 7. The clown is really funny.*
- **8.** The telephone rings.
- 9. I like potato chips.*
- 10. The sun is yellow.
- **11.** The baby bird hops away.*

- **12.** The boy's hair is brown.
- **13.** The books are on the shelf.*
- **14.** The new shoes are black.
- **15.** The cat chased the mouse.



Figure 1.

Geometric means (MG) and standard deviations (error bars) of perceived articulation rate for CA boys, boys with FXS-O, and boys with FXS-ASD.

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Figure 2.

Examples of the acoustic waveforms, F_0 traces, and broadband spectrograms for the sentence *The bus driver is smiling* produced by a boy with FXS-ASD (top) and CA control (bottom). Vertical cursors indicate the final F_0 contour.

Table 1

Geometric means (GM), standard deviations (SD), minimum (Min), and maximum (Max) of perceived articulation rate using two different standard sentences as a modulus of 100.

	GM	SD	Min	Max
CA Standard				
CA Boys	94	9	81	117
FXS Boys	114	21	84	159
FXS Standard				
CA Boys	65	15	31	89
FXS Boys	78	34	21	151

Table 2

Means, standard deviations (SD), minimum (Min), maximum (Max), and number of boys (N) for percent consonants correct (PCC), F₀ level (F₀), final F₀ drop (F_0 Drop), and rate_mfw.

CA PCC					
PCC					
	0.66	2.3	92.8	100.0	12
F_0	175.0	52.1	98.3	245.0	×
F_0 Drop	1.5	3.2	-2.9	8.1	8
Rate_mfw	4.4	0.6	3.3	5.3	12
FXS-0					
PCC	84.4	12.8	65.6	100.0	٢
F_0	239.7	24.0	207.0	272.3	5
F_0 Drop	6.2	1.7	4.8	8.9	5
Rate_mfw	4.4	0.7	3.5	5.1	٢
FXS-ASD					
PCC	85.3	12.6	70.0	97.0	5
F_0	254.0	45.3	213.3	327.7	5
F_0 Drop	8.8	3.8	5.7	14.4	5
Rate_mfw	5.0	0.9	3.9	5.8	5

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Table 3

Correlation matrix of perceived articulation rate (DME Rate), percent consonants correct (PCC), sentence F₀ level (F₀), final F₀ drop (F₀ Drop), and rate_mfw.

	DME Rate	PCC	\mathbf{F}_{0}	$F_0 \ Drop$	Rate
DME Rate	1.00				
PCC	-0.34	1.00			
F_0	0.56^*	-0.40	1.00		
F_0 Drop	0.67^{**}	-0.63	0.74***	1.00	
Rate_mfw	0.82^{***}	0.03	0.24	0.33	1.00
*** p<.001,					
** p<.01,					
* p<.05					