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Do individuals with fragile X syndrome show developmental stuttering or not? Comment on “Speech fluency in fragile X syndrome” by van Borsel, Dor and Rondal

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Abstract

Van Borsel, Dor, and Rondal (2007) examined the speech of seven boys and two young male adults with fragile X syndrome and considered whether their speech was comparable to that reported in the developmental stuttering literature. They listed five criteria which led them to conclude that the speech patterns of speakers with fragile X syndrome differed from those observed in developmental stuttering. The differences noted were: 1) distribution of type of dysfluency; 2) the class of word on which dysfluency occurred; 3) whether word length affected dysfluency; 4) number of times words and phrases were repeated; and 5) whether there were influences of material type on fluency (spontaneous speech, repeated material etc.). They concluded that the speech of speakers with fragile X syndrome differed from developmental stuttering. The comparisons that van Borsel et al. (2007) made between participant groups were not for speakers of comparable ages. Comparisons with groups of corresponding ages support the opposite conclusion, namely the young speakers with fragile X syndrome show patterns similar to developmental stuttering.

Keywords

Fragile X syndrome; developmental stuttering; dysfluency

Introduction

van Borsel, Dor, and Rondal (2007) reported analyses of fluency patterns in the speech of nine males with fragile X syndrome. The nine participants were aged between 10 and 22 years, two of whom were young adults (older than 18 years) and the sample had modal ages of 11 and 14 years. They examined a range of material types, and they reported gross speech rate measures for each participant.

Comparison of their results with previously published ones on developmental stuttering, led the authors to conclude that “individuals with fragile X syndrome show a pattern of dysfluency that is not identical to developmental stuttering.” They based this conclusion on five features in the speech patterns where the two types of speakers differed. These were:

1. Distribution of types of dysfluency. van Borsel et al. adapted the scheme used by Johnson and Associates (1959) to categorize dysfluencies. van Borsel et al. divided dysfluent events into those that occurred between, and those that occurred within, words. The between-word category included events “such as interjections, revisions and incomplete phrases”. The within-word category included events “such as part-

word repetitions, whole-word repetitions, blocks, prolongations and broken words". For these dysfluency categories, between-word dysfluencies made up over 70% of the dysfluencies evident in the group with fragile X syndrome. They claimed that this contrasted with Conture's (1990) report about developmental stuttering where within-word dysfluencies occurred more frequently than between-word dysfluencies.

2. Dysfluency and word class. In the fragile X syndrome group, more dysfluencies occurred on function words than content words whereas they claimed that the opposite happens in developmental stuttering. For stuttering, they cited Brown (1937) and Wingate (1979) as support.
3. Effect of word length on fluency. Word length did not lead to more dysfluency in fragile X syndrome individuals whereas they maintain that Brown and Moren (1942) and Wingate (1967) showed that word length does lead to more dysfluency in developmental stuttering.
4. Number of elements repeated in word and phrase repetitions. They noted that most word and phrase repetitions in speakers with fragile X syndrome involved a single repetition. They cited Yairi (1981) as evidence that this differed from the situation in developmental stuttering.
5. Effect of different types of material on dysfluency. No difference in frequency of dysfluencies was found between different speech material types for the participants with fragile X syndrome. The material types were: 1) automatic series, as when saying the days of the week in order, 2) repetition of 50 words of different length, and repetition of six phrases that increased in word length, 3) monologue which involved picture naming and 4) spontaneous speech based on responses to 12 probe questions. In developmental stuttering, reviewers of the literature often consider that certain types of material are affected more than others (van Borsel et al. cited Bloodstein, 1995) although there is little empirical evidence for this.

They also noted three ways in which the speech of males with fragile X syndrome was affected in a similar way to developmental stuttering. These were the position the dysfluent part takes in: 1) a word; and 2) a sentence; and 3) whether the word started with a consonant or vowel (dysfluency is more likely for both participant groups when a word starts with a consonant).

As has been said, van Borsel et al. rely heavily on literature to make their comparisons. This is alright as long as precautions are taken to ensure the comparison groups are matched on dimensions that could affect dysfluency patterns: Developmental stuttering refers to the form that starts in childhood and which can continue throughout life. The factors that affect incidence and type of dysfluency in developmental stuttering have different influences over ages. van Borsel et al. (2007) often compared across disorders using groups with very different ages. When comparison is made across groups that are similar in age, this leads to the opposite conclusion to that drawn by van Borsel et al. (2007). Namely, as documented below, the participants with fragile X syndrome appear to have a similar pattern of dysfluencies to that seen in developmental stuttering. Each of the five distinguishing features is considered in turn.

1. Dysfluency types. Conture (1990) was emphatic that there is no clearcut criterion for distinguishing people who stutter from those who are fluent though there are some general guidelines that he noted. Other respected clinicians have noted how the pattern of dysfluencies in developmental stuttering changes over age (Bloodstein, 1960a; 1960b; 1961, Froeschels, 1921, van Riper, 1954). All these authors point to changes from simplex forms involving word repetition to more complex forms involving dysfluencies on parts of words (i.e. a change from between-word dysfluencies at younger ages to within-word dysfluencies at later ages). A further factor that might operate in the fragile X syndrome group that would affect stuttering is that cognitive

delay could lead to speech delay, which would be manifest as earlier ontogenetic dysfluency forms. This would operate to extend between-word dysfluency types to older ages. If so, speakers with fragile X syndrome should be compared with younger IQ matched developmental stutterers (the latter do not have major cognitive delays). Whether or not the groups are matched as just indicated, van Borsel et al.'s report of high rates of between-word dysfluencies in young speakers with fragile X syndrome would be consistent with what has been reported for young speakers with developmental stuttering (which most their sample were). More empirical evidence is needed about differences in dysfluency types over ages for speakers in this age range for developmental stuttering and fragile X syndrome.

2. **Word type.** Systematic differences have been reported between the types of word affected by dysfluency over age groups. Bloodstein and Gantwerk (1967), and Bloodstein and Grossman (1981) showed that young speakers stuttered predominantly on function words. Brown (1937) showed adults have more problems on word types that commentators have classed as content words (Brown himself did not refer to function and content word classes in his series of studies). Howell's group has reported a trade off between dysfluencies on function words (young speakers) and on content words (older speakers) for English (Howell, Au-Yeung & Sackin, 1999), German (Dworzynski, Howell, Au-Yeung & Rommel, 2004) and Spanish (Au-Yeung, Vallejo Gomez & Howell, 2003). In all cases young speakers showed a predominance of function word dysfluencies rather than content word dysfluencies. Function word rate dropped off progressively with age whilst content word rate gradually increased. For present purposes, the main detail these results show is that function words are commonly involved in dysfluency in younger speakers and dysfluency on these words continues to predominate at least to adolescence. Based on the pattern in developmental stuttering, it would not be surprising to see a high rate of function word dysfluencies in the fragile X syndrome speakers as the majority were adolescent and teenage. Cognitive delay might again make more juvenile forms persist to later ages.
3. **Word length.** van Borsel et al. reported an analysis of dysfluency rate for words with different numbers of syllables. For words with one, two or three syllables, the percentage of dysfluent words out of all words for the respective number of syllables and the absolute numbers of dysfluent and fluent words were reported. They found no effect of word length on occurrence of dysfluency (presumably on the percentage scores) for the speakers with fragile X syndrome. In contrast, word length has been reported to affect dysfluency rate in developmental stuttering (Brown & Moren, 1942). The first thing to warn about is drawing conclusions from insignificant findings which is not acceptable. Second, Brown and Moren (1942) used the same 32 speakers as in the earlier Brown (1937) study. As already discussed, Brown's speakers who stuttered had a higher number of content word dysfluencies than the fragile X syndrome group, so they would also have had more long words. Short words are likely to be function words and long words are likely to be content words (Howell, Au-Yeung, Yaruss & Eldridge, 2006). The lack of a word length effect in van Borsel et al.'s fragile X group could be because there are fewer of content words which tend to be longer in this group compared to Brown's group of speakers who stutter. That is, van Borsel et al. may have failed to find an effect of word length whereas Brown did because of the confound between word length and function/content word type and the different proportions of content words in the two groups. Thus finding a word length effect for the speakers who stuttered but no such effect for fragile X syndrome might have been expected. Ideally what is necessary is for an analysis in which word length and word types are un-confounded by analyzing for a word length effect separately for function and content words.

4. Once-only repetitions. Rate of word repetition can be estimated from van Borsel et al.'s study. 85.5% involved one word. If the remaining 14.5% had just two repetitions, the rate over all repetitions would be 1.145. Contrary to what van Borsel et al., say, this is almost exactly the same as Yairi (1981) who reported 1.13 (in a near-contemporary study Wexler, 1982 reported 1.10). Here, in contrast to earlier, it is of note that the speakers Yairi (1981) used were probably too young to compare with van Borsel et al.'s participants.
5. Material type. There were differences between material types (e.g. 13.98% in the spontaneous task and 7.17% for repetition) in van Borsel et al.'s study, although this is reported as not significant. The lack of significance is probably because the speakers were variable, in their response to different material types so a study with a small N has insufficient statistical power. In addition, for a comparison to be appropriate, it would need to have speakers of the same age and gender, and control for factors like IQ, sub-culture, and to use the same forms of material. Until more extensive data for developmental stutterers are available, the effect of material type is probably best considered as “unproven” (I could not locate any data in Bloodstein (1995) about different rate of dysfluency across material type in developmental stuttering). A further point about material is that Brown's whole series of studies that is relied on heavily (see Brown, 1945 for a review) was conducted on recordings where participants read written prose. In contrast, many of the analyses van Borsel report were on spontaneous material. If van Borsel et al. are right that developmental stuttering and the speech of fragile X speakers differ across material types, such comparisons are not valid.

There is no clearcut evidence for differences between speakers with fragile X syndrome and developmental stuttering on the five criteria van Borsel et al. discuss. There may be other characteristics of the speech of children with fragile X syndrome that lead the dysfluencies in this group of speakers to be distinct from those seen in developmental stuttering. For instance, the prominent mandible and high-arched palate that van Borsel et al. mention might conceivably lead to certain articulatory gestures being difficult for these children to achieve, that causes a disproportionate number of dysfluencies on speech sounds that are specific to these speakers.

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