Speech disfluencies in individuals with Tourette syndrome

Luc F. De Nila,b,* Jayanthi Sasisekaran,a Pascal H.H.M. Van Lieshouta, Paul Sandorb

aGraduate Department of Speech–Language Pathology, University of Toronto, Room 160, 500 University Avenue, Toronto, ON, Canada M5G 1V7
bToronto Western Research Institute, University Health Network, Canada

Received 2 December 2003; accepted 8 June 2004

Abstract

Objective: The purpose of the present study was to analyze the frequency and type of speech disfluencies in a relatively large group of individuals with Tourette’s syndrome (TS) and to compare their results with similar speech data from a control group of unselected individuals. Method: Self-report data, as well as conversation and reading samples, were obtained from 69 children diagnosed with TS (mean age = 12.49) and 27 control participants (mean age = 10.9). Results: Self-report data on fluency difficulties did not reveal significant group differences; however, detailed analysis of fluency during reading and spontaneous speech revealed an overall higher level of more typical (normal) disfluencies in the TS group. No overall differences in less typical (stuttering) disfluencies were observed between the two groups of children. Conclusion: Results are discussed in light of previous studies proposing a common aetiology and neuropathological link between TS and developmental stuttering.

© 2005 Elsevier Inc. All rights reserved.

Keywords: Tourette’s syndrome; Stuttering; Children; Speech disfluencies

Introduction

Tourette’s syndrome (TS) is a neuropsychiatric disorder characterized by a wide range of neuromotor and behavioural symptoms, including speech and language difficulties and learning disabilities (LD) [1–3]. Previous research has reported that the speech of individuals with TS is characterized by word repetitions, hesitations, interjections, and prolongations [4,5]. The presence of such atypical speech features has led some to suggest that speech in individuals with TS is “stutter-like” [6,7]. Similarly, the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV; [8]) lists stuttering as one of the initial symptoms of the Tourette’s complex.

In addition to the presence of speech disfluencies, TS and developmental stuttering seem to share a number of other characteristics, which have been interpreted as suggesting possible shared etiological factors [9–12]. Both have a strong genetic component, are most prevalent in males, are prone to developmental changes in behavioural and psychomotor functions, and symptom severity is influenced by communication situations and emotional stress [1–3,13]. Children diagnosed with developmental stuttering also have been reported to exhibit behaviours frequently observed in the TS population, including obsessive compulsion, motor tics, and hyperactivity [12,15].

While the presence of speech disfluencies in individual patients with TS seems to be well documented, the prevalence of such symptoms is not well understood, and reported data vary widely. A number of studies have surveyed the prevalence of stuttering in TS. For instance, Comings and Comings [1] and Singer et al. [7] investigated the family history of 325 TS individuals ranging in age from 2 to 70 years using a family study method and reported a
prevalence rate of 31.3%. In contrast, Pauls et al. [10] using a family study method reported a prevalence of 15.3% in 85 TS individuals ranging in age from 7 to 62 years. Such differences in reported prevalence may indicate methodological differences in the studies or variations in the definition of stuttering. More recently, as part of the Canadian-American TS database project, data were collected on 3500 patients from 22 different countries [16]. The results from this multinational study indicated an average prevalence of stuttering in the TS population of 8% (range 2–17%), which is much lower than previously reported. The reported prevalence in TS-only individuals, who did not demonstrate comorbid conditions, was lower, still at 4%. However, the authors of this study caution that “relatively few individuals were directly evaluated by a speech–language pathologist. Therefore, it is likely that only the individuals with more severe stutters were identified” (p. 443).

In one of the few studies where speech disfluencies were directly analyzed, Van Borsel and Vanryckeghem [17] investigated disfluent and phonemic tics in an 18-year-old male diagnosed with TS. The participant exhibited interjections, whole word and phrase repetitions, rapid speech rate, disorganised and confused wordings, poor grammar, and slurred articulation. They concluded that the speech pattern of the participant did not conform to the classical pattern of stuttering but did bear more resemblance to cluttering [18].

Given the paucity of direct studies of disfluencies in individuals with TS, the primary purpose of the present study was to use direct analysis of frequency and type of speech disfluencies in a large group of individuals with TS to investigate the presence of stuttering and other disfluencies.

**Method**

**Participants**

Children diagnosed with TS [8] were recruited from the TS clinic at the Toronto Western Hospital. Control participants were recruited from the General Medical Clinic at the same hospital. Patients were invited to participate when they were first assessed at the clinic. The use of therapeutic drugs was not controlled for in our study, but information about current use of medication was collected. Informed consent to participate was obtained from all participants and from their parents according to the guidelines of the Human Subject Review Committee at the hospital.

Sixty-nine children (8 females and 61 males) diagnosed with TS were recruited. Their age ranged from 4 to 18 years (mean age = 12.49, S.D. = 3.32). Eight of the children (10%) had TS only, 9 (12%) were diagnosed with TS and obsessive-compulsive disorder (OCD), 20 (28%) had TS and attention deficit hyperactivity disorder (ADHD), and 32 (45%) had TS, OCD, and ADHD. The findings in the current study of a ratio of 1:8.5 of TS-only to TS + comorbidity, as well as a gender ratio of 7.6:1 (male/female), are similar with those reported by Freeman et al. [16]. Twenty-seven control participants (14 females, 13 males) were recruited from the General Medical Clinic at the same hospital. Their age ranged from 6 to 17 years (mean age = 10.9; S.D. = 3.08). To analyze changes in fluency with age, the participants were divided into three age groups: 4–10 years (TS 26; Control 11), 11–14 years (TS 22; Control 9), and 15–18 years (TS 21; Control 7).

**Procedures**

**Clinical assessment**

Prior to participation in the study, all TS participants underwent a routine clinical medical assessment as used in the Tourette’s clinic at the hospital. This assessment consisted of a semistructured, self-administered questionnaire complemented by a clinical interview and examination by an experienced neuropsychiatrist. The primary objective of the interview was to diagnose the presence of TS and associated conditions, such as OCD, ADHD, and LD. DSM-III-R criteria were used in the diagnosis of TS and DSM-IV in the diagnosis of OCD and ADHD [19]. For the control group, self, and parent reports were reviewed by the first author, a certified speech–language pathologist, and the fourth author, a psychiatrist who also is the director of the Tourette’s Clinic, to establish the absence of comorbid conditions.

**Speech and language questionnaire**

All participants, or their parents in case of younger children, were administered a speech and language questionnaire designed specifically for this study. This questionnaire was intended to collect data on the self-reported presence, nature, and familial incidence of speech and language difficulties. The *Speech–Language Section* of the questionnaire included questions on self-observed occurrences of speech and language problems, including learning difficulties, stuttering, voice problems, and articulation disorders. Participants were presented with 20 questions probing for speech, language, hearing, and reading difficulties. They rated each question on a four-point scale (1 = never, 2 = sometimes, 3 = often, and 4 = very often). The *Family History Section* probed for the presence of a family history of speech and language problems in immediate (parents and siblings) and distant family members (grandparents, cousins, aunts, uncles, nieces, and nephews). Typically, for children younger than 16 years, their parent(s) was present in the room, and the questionnaire was completed with their input.

**Speech sample**

Each participant also was videotaped using an AG-190 model Panasonic VHS video/audio recorder during two speech tasks: (1) reading a standard, age-appropriate text of approximately 250 words and (2) a free flowing conversation (approximately 500 words) with a trained
research assistant on a variety of age-appropriate topics. For children who were nonreaders, the reading task was replaced by a story narration task using standardized pictures. The video recording was done in a quite room in the hospital with the research assistant and the participant (and his or her parent(s) for younger children) seated across a table.

Disfluency analysis

Two trained research assistants performed the disfluency analysis on the reading and conversation samples, respectively. Overall frequency of speech disfluencies and the relative frequency of various types of disfluencies were calculated off-line using the system proposed by Yaruss [20]. In this system, disfluencies are differentiated between less typical and more typical. Less typical disfluencies include sound and syllable prolongations, repetitions, and speech blocks and are more characteristic of developmental stuttering. More typical disfluencies include word repetitions, filled and unfilled pauses, hesitations, and interjections that are observed in the speech of stuttering, as well as nonstuttering, individuals. To calculate the intra- and interjudge reliability of the disfluency scores, a randomly selected sample of 10% of the reading and conversation speech samples was reanalyzed by one of the two original research assistants (intra) and by a third trained assistant (inter). The mean difference in the disfluency frequency score was 2.01 disfluencies (S.D. = 1.88, \( r = .91 \)) for intra-judge and 0.12 disfluencies (S.D. = 0.72, \( r = .98 \)) for interjudge reliability measures.

Results

The data did not reveal any statistically significant differences in speech disfluencies between the children with TS-only and children with TS+comorbid conditions. Therefore, the data from these two groups were pooled for analysis. Of the children diagnosed with TS, 81.5% were taking neuroleptics, antidepressants, stimulants, anxio-lytics, or anticonvulsants at the time of the study. None of the participants in the control group were taking any of these drugs.

Self-report data

Speech and language problems

The total score obtained from the Speech–Language Section of the questionnaire was analyzed using univariate ANOVA (\( \alpha = .05 \)), with group, age, and gender as the fixed factors. None of the main or interaction effects were statistically significant. Descriptively, participants with TS were more likely (17.76%) than the control participants (11.1%) to respond “often” or “very often” to questions concerning speech or language problems (see Table 1). The largest difference between the TS and the control participants was observed on questions related to reading difficulties (TS: 17.45%; control: 3.7%).

Table 1

Percentage responses to questionnaire items for TS and control participants

<table>
<thead>
<tr>
<th>Questionnaire items</th>
<th>TS (n = 69)</th>
<th>Control (n = 27)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Never</td>
<td>Sometimes</td>
</tr>
<tr>
<td>Difficulty speaking</td>
<td>34.8</td>
<td>26.1</td>
</tr>
<tr>
<td>Trouble thinking of common</td>
<td>31.9</td>
<td>39.1</td>
</tr>
<tr>
<td>words while speaking</td>
<td>31.9</td>
<td>39.1</td>
</tr>
<tr>
<td>Hard to express thoughts</td>
<td>24.6</td>
<td>42.0</td>
</tr>
<tr>
<td>Others find speech difficult to</td>
<td>37.7</td>
<td>29.0</td>
</tr>
<tr>
<td>understand</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Voice loss</td>
<td>40.2</td>
<td>29.9</td>
</tr>
<tr>
<td>Soft voice</td>
<td>31.9</td>
<td>39.1</td>
</tr>
<tr>
<td>Loud voice</td>
<td>63.8</td>
<td>17.4</td>
</tr>
<tr>
<td>Throat ache</td>
<td>68.1</td>
<td>15.9</td>
</tr>
<tr>
<td>Reading difficulties</td>
<td>33.3</td>
<td>17.4</td>
</tr>
<tr>
<td>Difficulty reading magazines</td>
<td>36.2</td>
<td>15.9</td>
</tr>
<tr>
<td>Mix up letters when talking</td>
<td>68.1</td>
<td>15.9</td>
</tr>
<tr>
<td>Average</td>
<td>45.7</td>
<td>23.3</td>
</tr>
<tr>
<td>Stuttering related</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Asked to repeat frequently</td>
<td>33.3</td>
<td>36.2</td>
</tr>
<tr>
<td>Stutter</td>
<td>55.1</td>
<td>29.0</td>
</tr>
<tr>
<td>Words stuck in the mouth/throat</td>
<td>50.7</td>
<td>20.3</td>
</tr>
<tr>
<td>Repeat words or letters while talking</td>
<td>47.8</td>
<td>21.7</td>
</tr>
<tr>
<td>Certain letters hard to say</td>
<td>19.6</td>
<td>11.6</td>
</tr>
<tr>
<td>Avoid words with certain letters</td>
<td>75.4</td>
<td>13.0</td>
</tr>
<tr>
<td>Hard to say name</td>
<td>73.9</td>
<td>8.70</td>
</tr>
<tr>
<td>Average</td>
<td>57.9</td>
<td>15.7</td>
</tr>
</tbody>
</table>
average, 6.20% of the TS children reported that they experienced these symptoms often or very often, which was very comparable with the frequency observed in the control group (6.05%). Of interest, however, was the fact that on one of the questions in which the children (or their parents) were asked specifically whether they stutter, 29% of the TS respondents marked “sometimes” compared with only 7.4% for the control group.

Family history

The reported frequency of family history of speech or language problems was not significantly different between TS and control participants ($\chi^2 = 1.145, P > .05$). When the participants were asked specifically to identify stuttering in their immediate and distant family members, 50% of the TS individuals with a positive family history of speech and language problems reported stuttering among relatives compared with only 20% of the control participants, a difference that approached statistical significance ($\chi^2 = 3.1, P = .06$).

Speech fluency data

Conversation

The mean and SD of percentage disfluencies for the TS and control participant groups are presented in Table 2. Using analysis of variance, no significant differences for less typical disfluencies were found between the TS and control participants. The TS individuals, however, showed a statistically significant higher frequency of more typical disfluencies during conversation compared with the control group [$F(1,83) = 7.02, P < .05, \text{power} = 0.74$; see Fig. 1]. Descriptively, a higher percentage of more typical disfluencies was observed among female (8%) than among male participants (4.03%), but only for the TS participants. In the control participants, males (3.03%) and females (2.9%) did not show the same difference.

Reading

The overall frequency of less typical disfluencies during reading was comparable between the TS and the control groups, and no statistically significant between-group differences were found. Descriptively, the data suggested that the children with TS consistently showed a higher percentage of less typical disfluencies compared with the control group in the two older age groups, while the opposite was true for the youngest age group. In addition, two other observations were made: (1) Females in the TS group tended to have a greater percentage of less typical disfluencies than the males did. This was not the case in the control group, with the exception of the youngest children, who showed an overall higher level of disfluencies during reading, possibly related to emerging reading skills in some. (2) The frequency of less typical disfluencies decreased with age in the control participants but not in the TS group. This Group $\times$ Age $\times$ Gender interaction was statistically significant [$F(2,83) = 3.32, P = .04, \text{power} = 0.61$].

The total percentage of more typical disfluencies in the TS children was comparable with that for the control group (see Fig. 1), and none of the main or interaction effects were statistically significant. Descriptively, however, a number of interesting trends could be observed. In the TS group, the frequency of more typical disfluencies during reading (4%) was similar to that in the control speakers (3.2%). The percentage of more typical disfluencies in the TS group was relatively constant with age (4–10 years: 3.9%; 11–14 years: 3.9%; 15–18 years: 4.2%). In contrast, in the control group, the youngest age group showed a higher frequency of more typical disfluencies compared with the two older age groups. In both subject groups, a somewhat higher percentage of disfluency was observed in male than in female participants.

![Fig. 1. More typical and less typical disfluencies in conversational and reading samples of TS and control participants (*$P \leq .05$).](image-url)
Discussion

In contrast to previous reports of higher prevalence of stuttering in persons with TS, the results of the present study did not support such a straightforward conclusion. Instead, a more complex picture emerged.

The self-report data did not result in any significant differences between the two groups of children, although descriptively, there were a number of interesting observations. In particular, children with TS tended to report more self-perceived reading difficulties, as well as a higher likelihood of stuttering among relatives. Similarly, more TS respondents indicated that they “sometimes” stuttered compared with the control group. No attempt was made in the present study to define stuttering for the participants, and as such, this observation needs to be interpreted carefully, given that people may not always differentiate between the various types of speech disfluencies or between less typical and more typical disfluencies. Nevertheless, these self-report data are intriguing and warrant further study.

The fact that the self-report data did not show significant differences in self-perceived stuttering between the two subject groups is supported by the results of our direct analysis of speech disfluencies during spontaneous speech and reading. Direct analysis of speech did not show a higher frequency of less typical disfluencies, commonly identified as the core features of developmental stuttering [21], in the TS participants. In contrast, the two subject groups clearly differed with respect to the frequency of more typical disfluencies, which are commonly observed in the speech of stuttering as well as nonstuttering speakers. During our conversational speech task, the children with TS showed a significantly higher frequency of more typical disfluencies compared with the control group, a difference that was not affected by either age or gender. This observation, coupled with the lack of between-group differences for less typical disfluencies, suggests that the previously reported higher prevalence of stuttering in TS may simply reflect a generally more disfluent conversational speech pattern.

Our results for the reading task yielded a more complex picture. While no overall group differences were observed for more typical disfluencies, the frequency of less typical disfluencies was affected by the age and gender of the participants. As expected, the control group showed a decreasing level of disfluency during reading with age [22]. Such changes have been attributed to maturation of the neuromotor and the cognitive–linguistic subsystems [23]. The children with TS, on the other hand, did not show such a decrease with age, an observation not unlike that in children with developmental stuttering who typically also show increased frequency of disfluency with age [24,25]. In contrast to our findings with TS children, however, even very young children with developmental stuttering show an elevated level of disfluency compared with their normally fluent peers [26,27]. Instead, the youngest children with TS in our study showed a reduced frequency of less typical disfluencies compared with the control participants. This observation may suggest that between-group differences in speech fluency observed in our study are due primarily to an age-related decrease in both less typical and more typical disfluencies in our control participants, while the speech fluency in children with TS remains essentially stable with age.

The data in the current study revealed higher levels of less typical disfluencies among female participants with TS compared with the control group. This gender effect was statistically significant only for the reading task, but a similar nonsignificant trend was observed for spontaneous speech. Such a gender effect was not observed among the control participants, with the exception of the youngest children, during reading. Given that both TS and developmental stuttering are more likely to occur in males [2,3,14], one could speculate that the apparent gender effect in our study reflects a higher genetic or biological loading in females, resulting in a more severe disease complex. Clearly, the relatively low number of female participants in our subject groups, especially among the children with TS, should qualify such an interpretation. Nevertheless, the observed gender effects clearly call for further study using larger sample sizes and more balanced gender representation.

In contrast to the differences in stuttering prevalence in children with TS with or without comorbid conditions reported by Freeman et al. [16], the data in our study did not reveal any significant differences between these two groups. It is possible that the difference between our study and the Freeman et al. study [16] is related to the fact that the evaluation of speech fluency in the Freeman study was not based on detailed fluency analysis and, therefore, may represent a broader range of disfluency types. The relatively small sample size in each of the diagnostic subgroups in the present study, however, prevents any conclusive discussion of this finding at this time, and the influence of comorbid conditions on speech fluency needs to be investigated further. Furthermore, no attempt was made in the current study to control for the use of drugs in the TS group. Eighty-seven percent of the TS participants were taking medication for their symptoms at the time of data collection. It cannot be excluded that some of the prescribed drugs may have influenced speech fluency and the effects of specific classes of therapeutic drugs on speech fluency in TS deserves further study.

In contrast to previous studies, speech disfluencies in our TS and non-TS participants were analyzed in-depth during both a spontaneous speech and a reading task. In addition, self-report data were collected. The data obtained from such direct observations failed to confirm the presence of a generally higher prevalence of stuttering disfluencies in children with TS. Instead, the speech of children with TS was characterized by a higher frequency of more typical disfluencies. This finding suggests that the
reported higher prevalence of stuttering associated with TS in the literature may, at least in part, be the result of a failure to differentiate between so-called more typical (normal) and less typical (stuttering) types of disfluencies. Our finding of a higher frequency of less typical disfluencies during reading in the older children with TS, especially females, also points to the need to differentiate various types of disfluencies. Such a differentiation is important in light of the potential differences in aetiology between these disfluencies [28,29] and also may be relevant in further investigations of common pathophysiology between the two disorders [3,9,15,30].

In conclusion, our data show that the relationship between stuttering and TS is more complex than previously reported. If what is shared between children with TS and children with developmental stuttering is a higher frequency of disfluent speech itself, rather than stuttered speech, this may suggest that the link between the two disorders, if any, is not a common set of etiological factors but rather the effect that these conditions have on the overall stability of the speech fluency system.

Acknowledgments

The authors would like to thanks Jessica Steinman, Katie Sharpe, and Sophie Lafaille for their assistance in data collection and analysis. This research was supported by grants from the Natural Sciences and Engineering Research Council of Canada and the Bluma Appel TS Research Fund awarded to the first and fourth authors, respectively.

References


