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Speech disfluencies in individuals with Tourette syndrome

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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.

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

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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 phonic 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