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Spatio-temporal gait characteristics in children with Tourette syndrome: A preliminary study

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ABSTRACT

Earlier studies had suggested that variability of stride length in gait is a pathological sign of basal ganglia disease. Some evidence implicates the involvement of the basal ganglia and related thalamocortical circuitry in Tourette syndrome (TS). To date, the gait of subjects with TS has only discussed in case reports. This investigation compared the spatial and temporal gait characteristics of a sample of children with TS ($N = 8$) with those of healthy controls (HC; $N = 8$). All children were instructed to walk under two speed conditions: "preferred" and "fastest." Gait parameters were measured using an electronic walkway. Spatial and temporal gait parameters were compared using a two-way (group) - × (conditions) repeated measures ANOVA. The preliminary results suggested that similar to HC children, children with TS were capable of regulating temporal characteristics of gait based on walking speed. They also exhibited subtle gait anomalies such as irregular step length, as evidenced by significant differences in step length differential ($p = 0.003$), detectable despite the small sample size. These findings warrant further investigation into the gait control of children with TS.

1. Introduction

Tourette syndrome (TS) is a childhood-onset neurodevelopmental disorder defined by the presence of phonic and motor tics. Tics are involuntary, brief, rapid, and nonrhythmic muscle contractions causing purposeless, stereotyped motor actions (motor tics) and sounds (phonic tics). TS is no longer considered a rare disease—Robertson, Eapen, and Cavanna (2009) estimated that the overall international incidence of TS is 1% of the population—yet the pathophysiology of tics is not fully understood. Some evidence suggests that the basal ganglia and related thalamocortical circuitry are involved in the disorder (Baym, Corbett, Wright, & Bunge, 2008; Mink, 2001; Peterson et al., 1993, 2003; Raz et al., 2009; Singer et al., 1993).

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The basal ganglia play an important role in the ability of humans to maintain balance and walk (Iseki & Hanakawa, 2010; Morris, Iansek, Matyas, & Summers, 1994a; Takakusaki, Tomita, & Yano, 2008). Several balance problems have been identified in children with TS (Lemay, Le, & Richer, 2010; Lemay et al., 2007). Gait, which is also an indicator of deficits in motor control performance, has not been fully explored in children with TS, with only limited case studies describing the unusual gait characteristics of subjects with TS and comorbid disorders (Demirkol, Erdem, Inan, Yigit, & Guney, 1999; Fasano, Ruzicka, & Bloem, 2012). Demirkol et al. reported an adult with TS and comorbid obsessive-compulsive disorder, attention deficit-hyperactivity disorder (ADHD), stuttering and gait disturbance who exhibited unusual gait characteristics, such as walking on the balls of his feet. Fasano et al. reported tic-induced gait disturbance for four subjects with tic disorders, which included three subjects with TS. These studies raised important questions about gait characteristics in children with TS without tic episodes.

Severe gait disturbances have been noted in other conditions involving deficits in the basal ganglia and related thalamocortical circuitry, such as Parkinson's disease (Blin, Ferrandez, & Serratrice, 1990; Morris et al., 1994a; Morris, Iansek, Matyas, & Summers, 1994b), ADHD (Buderath et al., 2009; Leitner et al., 2007), and autism (Rinehart et al., 2006). Of notes, subjects with Parkinson's disease exhibit marked gait hypokinesia (Morris et al., 1994a, 1994b); children with ADHD tend to walk with higher variability in stride time (Leitner et al., 2007); and children with autism show higher variability both in stride length and duration in preferred walking speeds (Rinehart et al., 2006).

Walking speed influences the elements of gait, such as cadence, stride length, ground reaction forces, and stride time (Diop et al., 2005). Gait deviations in children with various developmental disorders might be highly apparent when children walk fast. Fast walking speed is associated with greater gait deficits, such as stride length variability and inability to increase stepping frequency, because the duration of double limb support becomes shorter. Earlier studies have also shown that stride length variability may be a pathological sign of basal ganglia disease (Blin et al., 1990). Subjects with Parkinson's disease have difficulties in the regulation of stride length and exhibit a relative increase in cadence as a compensatory mechanism when walking at faster speeds (Morris et al., 1994b). Young adults with Williams syndrome—a developmental disorder accompanied by basal ganglia deficits, subtle cerebellar signs, and mild extrapyramidal signs-disproportionately increase their cadence (stepping frequency) with increased walking speeds (Hocking, Rinehart, McGinley, & Bradshaw, 2009). Since those young adults were able to compensate for reduced stride length by increasing cadence when walking at a fast speed, it is safe to assume their fundamental regulation mechanisms governing stepping frequency to be unaffected; rather, this observation suggests that individuals with William syndrome have problems in regulating their stride length according to walking speed.

The above results suggest that investigating gait at fast walking speeds for children with TS may be worthwhile to elucidate the underlying mechanism of gait problems. The aims of the present study were to explore the gait characteristics of children with TS under two conditions—walking at their preferred speed and walking at their fastest possible speed—and to conduct a comparative analysis against a control group of healthy children.

2. Methods

2.1. Participants

The Institutional Review Boards for Human Studies at the Chang Gung Memorial Hospital approved this protocol. Written informed consents for participation of the children in the study were obtained from all children and their parents. Inclusion criteria for this study were that the children should be between 5 and 12 years old, attend regular classes at an elementary school, and have normal or corrected-to-normal vision, and (for the TS group) no diagnosed with any other condition. For inclusion in the TS group, children were required to have been diagnosed by a pediatric neurologist according to the DSM-IV-TR (American Psychiatric Association, 2000) criteria for TS. Children were excluded from this study based on their parental reports and medical records if they met any of the following conditions: known pervasive developmental disorders or ADHD, inability to follow verbal orders, a history of head injury, any other neurological disorder, or musculoskeletal disorders; or on any medication within one month of the start of this study. All children with TS had been diagnosed with TS more than three months before their participation in this study, and underwent a complete neurological evaluation before testing. Tic severity was assessed using the Yale Global Tic Severity Scale (YGTSS) (Leckman et al., 1989), a clinician-rated instrument of motor and phonic tic severity based on a semi-structured interview with the parent(s) and child and on behavioral observations. Out of eight children in the TS group, 37.5% had moderate tic severity (YGTSS scores 20 to $<$30), and 62.5% had mild tic severity (YGTSS scores $<$20). Boys in the healthy control (HC) group were recruited from acquaintances of the researchers and age-matched to boys in the TS group. Thus, eight boys with TS (mean age 9.6 $\pm$ 1.7 years, range 7–12 years) form the TS group and eight typical developing boys (mean age 9.3 $\pm$ 1.6 years, 7–12 years) formed the HC groups. Table 1 shows that there were no significant differences between the TS and the HC groups in term of their anthropometric information.

2.2. Procedure

First, the children were instructed to walk in the preferred walking speed condition, where they received the following verbal instructions: "Walk as you walk in your daily activities. You can swing your hands as you like. Just feel relaxed and walk." Then, the children were instructed as follows for the fastest walking speed condition: "Walk as if a dog is chasing you.