



Differentiating tic-related from non-tic-related impairment in children with persistent tic disorders



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ABSTRACT

Children with persistent (chronic) tic disorders (PTDs) experience impairment across multiple domains of functioning, but given high rates of other non-tic-related conditions, it is often difficult to differentiate the extent to which such impairment is related to tics or to other problems. The current study used the Child Tourette's Syndrome Impairment Scale - Parent Report (CTIM-P) to examine parents' attributions of their child's impairment in home, school, and social domains in a sample of 58 children with PTD. Each domain was rated on the extent to which the parents perceived that impairment was related to tics versus non-tic-related concerns. In addition, the Yale Global Tic Severity Scale (YGTSS) was used to explore the relationship between tic-related impairment and tic severity. Results showed impairment in school and social activities was not differentially attributed to tics versus non-tic-related impairment, but impairment in home activities was attributed more to non-tic-related concerns than tics themselves. Moreover, tic severity was significantly correlated with tic-related impairment in home, school, and social activities, and when the dimensions of tic severity were explored, impairment correlated most strongly with motor tic complexity. Results suggest that differentiating tic-related from non-tic-related impairment may be clinically beneficial and could lead to treatments that more effectively target problems experienced by children with PTDs.

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1. Introduction

Persistent tic disorders (PTDs), including Tourette's disorder (TD) and persistent (chronic) motor or vocal tic disorder, are a class of childhood-onset neurobiological conditions defined by the production of sudden, rapid, recurrent, nonrhythmic movements (motor tics) and/or sounds (vocal tics) that persist for at least one year [1]. Although tics are fairly common in school-age children, PTDs are much less common; occurring in 0.8%–1.9% of youth [2–4].

Prior research has shown that children with PTDs experience impairment across a variety of functional domains [5–11]. Several studies have shown that frequent and intense tics are associated with academic problems, such as difficulties concentrating in class, writing, reading, and completing tests and homework [5,10]. In addition, youth with tics often struggle to maintain strong social relationships, are often victimized by peers, and are rated by their peers as being more withdrawn, aggressive, and less popular than children without PTD [6,9]. Further,

Robinson et al. [11] and Espil et al. [5] demonstrated that tics had some influence on children arguing with their parents and their ability to do chores. Likewise, Storch et al. [12] found that 24% of parents in their study reported that tics caused at least one significant problem across home activities (e.g., difficulty going places, doing chores), and Ramanujam et al. [13] found that the number and intensity of a child's tics was associated with increased objective caregiver strain (e.g., missing work, disruption of family routines, etc.).

In addition to tics, an estimated 78% to 90% of individuals with PTDs experience one or more comorbid psychiatric problem(s) such as attention-deficit hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD), anxiety and mood disorders, and impulse control disorders [11,14–17], and many of these disorders are impairing in their own right [18–20]. Other behavioral and emotional symptoms that do not fall under any particular diagnosis or cut across multiple diagnoses, but are also common, include elevated levels of psychosocial stress, hypersensitivity to sensory stimuli, difficulties with emotion regulation, general impulsivity, difficulties with visual motor integration, and procedural learning difficulties [21,22]. However, limited research has focused on the degree to which non-tic-related issues versus tics themselves contribute to functional impairment.

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Several studies have examined quality of life (QoL) and psychosocial functioning in children with TD-only and TD + comorbidities (TD+). For instance, O'Hare et al. [23] found strong associations between comorbidity and decreased global QoL, impaired emotional and school functioning, and increased emotional symptomatology in youth with TD. Further, Debes, Hjalgrim, and Skov [24] demonstrated that children with TD and other comorbidities such as OCD and ADHD have higher rates of psychosocial and educational problems. Similarly, Sukhodolsky et al. [25] and Stephens and Sandor [26] found that children with TD+ have more disruptive behaviors than children with TD-only and unaffected controls. Finally, Ramanujam et al. [13] found that parents of children with PTD and one or more comorbid internalizing or externalizing conditions reported higher levels of objective and subjective caregiver strain compared to parents of children with PTD without comorbidity.

Although the aforementioned studies suggest that non-tic-related problems (i.e., comorbidities/elevated levels of psychological difficulties), as opposed to the tics themselves, lead to greater functional impairment, parents and those affected by PTDs have rarely been asked to make the attribution themselves. Differentiating tic-related from non-tic-related impairment may be clinically beneficial, as such information could lead to treatments or treatment sequencing that more effectively target problems experienced by children with PTDs. To answer this question, Storch et al. [12] developed the Child Tourette's Syndrome Impairment Scale - Parent Report (CTIM-P), which assesses the degree to which parents of children with PTDs report impairment across 37 activities (home, school, social), as well as the extent to which that impairment is attributed to tic versus non-tic-related problems.

In an initial study of the CTIM-P, Storch et al. [12] examined parent responses in a sample of 59 parents of children with PTDs and found that impairment due to tics occurred mostly in school and social activities. Parents reported that tics "pretty much" or "very much" interfered with activities such as writing in class (24.6%), doing homework (21.9%), concentrating on work (21.8%), and being prepared for class (18.5%). Parents also perceived that being teased by peers (17.5%) and difficulties making new friends (15.8%) were "pretty much" or "very much" related to tics. Regarding non-tic-related impairment, parents perceived impairment as "pretty much" or "very much" related to non-tic-related problems in the following school activities: concentrating on work (38.9%), doing homework (37.1%), being prepared for class (27.0%), and taking tests or exams (25.9%). Additionally, parents perceived impairment as "pretty much" or "very much" related to non-tic concerns in activities such as making new friends (21.3%), doing household chores (17.6%), sleeping at night (17.3%), and being with a group of strangers (15.3%). It was much more common for impairment in these activities to be rated as being "pretty much" or "very much" related to non-tic problems rather than to tics.

In another study, Cloes et al. [27] examined tic-related and non-tic-related impairment using both the parent and child versions of the CTIM (CTIM-P and CTIM-C, respectively). In contrast to the methodology used by Storch et al. [12], this study compared CTIM scores of children with PTDs to healthy controls. Not surprisingly, children with PTDs experienced higher levels of tic-related and non-tic-related impairment compared to healthy controls. Consistent with Storch et al. [12], school impairment was strongly attributed to both tic and non-tic problems for individuals with PTDs. The highest tic-related impairment scores for children involved impairment in concentrating on work, doing oral reports/reading out loud, and taking tests. Interestingly, parents of these children attributed impairment in these school activities more to non-tic-related concerns than to tics. Parent ratings of their children's non-tic-related impairments were consistently higher than child's ratings of such impairment; however, the study did not directly compare tic versus non-tic-related impairment across the domains.

In addition to reporting the extent to which impairment was attributed to tics versus non-tic behaviors, Cloes et al. [27] and Storch et al.

[12] examined the relationship between tic-related impairment and tic severity. Results showed that motor, vocal, and total tic severity scores on the Yale Global Tic Severity Scale (YGTSS; [28]) positively correlated with parent ratings of tic impairment on the CTIM-P. However, the relationship between specific dimensions of tic severity (i.e., number, frequency, intensity, complexity, interference) and CTIM-P scores were not explored. Such analyses may be important, given research demonstrating that different tic dimensions may differentially relate to aspects of impairment. For instance, Espil et al. [5] found that, after controlling for anxiety and ADHD, tic intensity was a stronger predictor of impairment in close relationships with friends and family, ability to do home chores, and school productivity than was tic frequency. Better understanding of how specific dimensions of tic severity contribute to functional impairment may be helpful in allowing therapists to prioritize and target those dimensions when treating tics.

The current study had three aims. First, we sought to partially replicate the studies by Storch et al. [12] and Cloes et al. [27]. This was done by administering the CTIM-P to a sample of treatment-seeking families and examining parental attributions of functional impairment associated with tics versus non-tic-related problems across three broad domains (i.e. home, school, social). It was hypothesized that impairment across school, home, and social activities would more likely be attributed to non-tic concerns than tics. Second, we examined the degree to which tic-related impairment correlated with tic severity. It was hypothesized that YGTSS motor, vocal, and total tic severity scores would positively correlate with total tic-related impairment on the CTIM-P. Third, we performed exploratory analyses to examine whether tic-related impairment was correlated with the various dimensions of motor and vocal tic severity (i.e. number, frequency, intensity, complexity, and interference).

2. Method

2.1. Participants

Participants were 58 children and adolescents between 8 and 17 years old who had been diagnosed with a PTD (Table 1). Consistent with the male-biased gender distribution in PTD [17,23], the sample was predominantly male ($n = 44$). Of the 58 children, 48% ($n = 28$) had at least one comorbidity, and the average number of comorbid diagnoses for those 28 children was 2.5. Table 1 shows the percentage of children meeting diagnostic criteria for each disorder.

Participants were recruited through regular clinic flow at three university-based tic disorder specialty clinics. In addition, participants were recruited via the Tourette Association of America (TAA) website and membership emails, local TAA support groups, and local TD treatment providers. This study was part of a larger, multi-site randomized control trial (RCT) testing the efficacy of an internet-based treatment for children with PTDs. Inclusion criteria included English fluency, age 8–18 years, Diagnostic and Statistical Manual of Mental Disorder-5 (DSM-5) diagnostic criteria for TD or PTD, FSIQ > 70 on the Wechsler Abbreviated Scale of Intelligence – 2 subtest (WASI-II; [29]), current display of at least one motor and/or vocal tic multiple times per day, Clinical Global Impressions – Severity (CGI-S; [30]) score ≥ 3 (mildly ill or worse), unmedicated or on stable psychotropic medication (i.e., 6 weeks with no changes or planned changes in dosage), and availability of a personal computer with internet access. Exclusion criteria included a YGTSS score > 30; WASI-II score FSIQ < 70; DSM-5-defined diagnosis of substance abuse, substance dependence, or conduct disorder within the past 3 months; current or past (i.e., >4 sessions) non-pharmacological treatment for tics; lifetime DSM-5 diagnosis of mania, or psychotic disorder; and any serious psychiatric or neurological condition (e.g., obsessive-compulsive disorder (OCD), attention deficit hyperactivity disorder (ADHD), major depressive disorder (MDD), severe aggression, childhood disintegrative disorder)

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