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Jordan T. Stiede Marquette University

Jennifer R, Alexander Marquette University, jenny.alexander@marquette.edu

Brianna Wellen University of Utah

Christopher C. Bauer Marquette University

Michael B. Himle University of Utah

See next page for additional authors

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Authors Jordan T. Stiede; Jennifer R, Alexander; Brianna Wellen; Christopher C. Bauer; Michael B. Himle Mouton-Odum; and Douglas W. Woods	; Suzanne

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Differentiating Tic-Related from Non-Tic-Related Impairment in Children with Persistent Tic Disorders

Jordan T. Stiede
Marquette University, Milwaukee, WI
Jennifer R. Alexander
Marquette University, Milwaukee, WI
Brianna Wellen
University of Utah, Salt Lake City, UT
Christopher C. Bauer
Marquette University, Milwaukee, WI
Michael B. Himle
University of Utah, Salt Lake City, UT
Suzanne Mouton-Odum
Psychology Houston, PC, Houston, TX
Douglas W. Woods
Marquette University, Milwaukee, WI

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.

Keywords

Impairment, Tourette's disorder, Tic severity, Children

1. Introduction

Persistent <u>tic disorders</u> (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], [3], [4]].

Prior research has shown that children with PTDs experience impairment across a variety of functional domains [[5], [6], [7], [8], [9], [10], [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], [15], [16], [17]], and many of these disorders are impairing in their own right [[18], [19], [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 <u>impulsivity</u>, difficulties with visual motor integration, and procedural <u>learning difficulties</u> [21,22]. However, limited research has focused on the degree to which non-tic-related issues versus tics themselves contribute to functional impairment.

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 (<u>Table 1</u>). Consistent with the male-biased gender distribution in PTD [<u>17,23</u>], 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. <u>Table 1</u> shows the percentage of children meeting diagnostic criteria for each disorder.

Table 1. Participant Demographics and Clinical Characteristics.

Variables	% (n)
Sex	
Male	75.9 (44)
Female	24.1 (14)
Race	
White	82.8 (48)
African American	5.2 (3)
Asian	3.4 (2)
Mixed	8.6 (5)
Current medication use	32.8 (19)
Tic diagnosis	

Dorsistant motor tip disorder	C O (4)
Persistent motor tic disorder	6.9 (4)
Persistent vocal tic disorder	3.4 (2)
Tourette's disorder	89.7 (52)
Comorbid diagnoses (according to MINI-KID)	
Major depressive disorder	1.7 (1)
Major depressive episode	17.2 (10)
Suicidality	5.2 (3)
Dysthymic disorder	1.7 (1)
Panic disorder	1.7 (1)
Agoraphobia	1.7 (1)
Separation anxiety disorder	5.2 (3)
Social phobia/social anxiety disorder	8.6 (5)
Specific phobia	8.6 (5)
Obsessive-compulsive disorder	15.5 (9)
Attention deficit hyperactivity disorder	22.4 (13)
Oppositional defiant disorder	6.9 (4)
Generalized anxiety disorder	10.3 (6)
Pervasive developmental disorder	3.4 (2)
Age M (SD)	11.90 (2.745)

Note. MINI-Kid = Mini International Neuropsychiatric Interview for Children and Adolescents.

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) not currently being managed or managed ineffectively. Nine persons with YGTSS scores > 30 were allowed into the RCT after considering overall appropriateness of the participants. The study was reviewed and approved by the Institutional Review Boards at the respective sites, and this work was supported by a grant from the National Institute of Health (NIH; R44MH09634402). All participants were compensated for their time.

2.2. Measures

2.2.1. Child Tourette's syndrome impairment scale – Parent report about child (CTIM-P)

The CTIM-P [12] is a 37-item parent-rated instrument that examines impairment over the past month in home (11 items; e.g., getting dressed in the morning, bathing or grooming, doing household chores, etc.), school (11 items; e.g., getting to school on time, missing school, giving oral reports/reading out loud, etc.), and social (15 items; e.g., making new friends, keeping friends, spending time with friends, etc.) activities. For each of the activities listed in the CTIM-P, parents rated the extent to which they believed their children's impairment in the

activity was tic-related and the extent to which their children's impairment in the activity was due to non-tic-related concerns (e.g., anxiety, depression, a comorbid condition, etc.). These impairment ratings were made on a 4-point scale anchored by 0 ("not at all") and 3 ("very much"). Items that parents perceived to not be relevant for their child were rated as "not applicable" (e.g., having a boyfriend/girlfriend for a 9-year-old).

The CTIM-P yields 8 different impairment scores: (1) tic-related impairment in home activities (possible range = 0–33), (2) non-tic-related impairment in home activities (possible range = 0–33), (3) tic-related impairment in school activities (possible range = 0–33), (4) non-tic-related impairment in school activities (possible range = 0–33), (5) tic-related impairment in social activities (possible range = 0–45), (6) non-tic-related impairment in social activities (possible range = 0–45), (7) total tic-related impairment (i.e. sum of home, school, and social activities; possible range = 0–111), and (8) total non-tic-related impairment (possible range = 0–111). For all 8 impairment scores, higher scores indicate greater perceived impairment attributed to either tics or non-tic problems. The CTIM-P has demonstrated excellent internal consistency and acceptable convergent and discriminant validity [12]. The results from this study showed excellent internal consistency for CTIM-P tic-related total scores (α = 0.94 and 0.96, respectively) and good to excellent internal consistency for CTIM-P tic-related (α s = 0.85, 0.89, 0.96) and non-tic-related (α s = 0.90, 0.89, 0.96) home, school, and social subscale scores.

2.2.2. Yale global tic severity scale (YGTSS)

The YGTSS [28] is a clinician-rated instrument designed to assess current motor and vocal tic severity according to five different dimensions (i.e., number, frequency, intensity, complexity, and interference), as well as current tic-related <u>distress</u> and impairment in interpersonal, academic, and occupational situations. The YGTSS yields three severity scores: motor tic severity score (ranging from 0 to 25), vocal tic severity score (ranging from 0 to 25), and total tic severity score (ranging from 0 to 50). For each of these severity scores, higher scores indicate greater tic severity. The YGTSS also yields a tic impairment scale score, which ranges from 0 to 50, with higher scores indicating greater tic-related impairment. The YGTSS has demonstrated good interrater agreement, acceptable internal consistency and convergent and divergent validity [28,31]. In the current sample, the YGTSS showed good internal consistency for YGTSS total ($\alpha = 0.80$) and vocal tic severity ($\alpha = 0.83$) scores and acceptable internal consistency for YGTSS motor scores ($\alpha = 74$).

2.2.3. Mini international neuropsychiatric interview for children and adolescents (MINI-KID)

The MINI-KID [32] is a structured diagnostic interview designed to assess children from 6 to 17 years old based for 23 axis 1 psychiatric disorders as defined by the <u>DSM-IV</u> and <u>ICD-10</u>. The MINI-KID has demonstrated reliable and valid psychiatric diagnoses for children and adolescents [32].

2.2.4. Clinical global impression – severity scale (CGI-S)

The CGI-S [30] is a clinician-rated instrument designed to assess the severity of the patient's illness at the time of assessment. This study used a modified version of this scale to assess global tic-related severity. Clinicians rate the perceived patient global tic-related severity according to the following 7-point scale: Normal (1), Not at all ill (2), Mildly ill (3), Moderately ill (4), Markedly ill (5), Severely ill (6), Among the most extremely ill patients (7).

2.2.5. Wechsler abbreviated scale of intelligence-II (WASI-II)

The WASI-II [29] is an abbreviated measure of cognitive intelligence used for individuals 6 to 90 years old. The Full-Scale IQ-2 (FSIQ-2) consists of two subtests and was used to estimate IQ in this study. The FSIQ-2 scoring of the WASI-II has demonstrated good reliability and test-retest stability and acceptable validity.

2.3. Procedures

Interested persons first participated in a phone <u>screening</u> to determine preliminary study eligibility. Those deemed likely eligible to participate in the RCT were invited to complete a face-to-face screening conducted by an independent evaluator (IE). During this visit, an IE administered a series of clinician-rated, parent-report, and self-report measures, including the CTIM-P, YGTSS, MINI-KID, CGI-S, and WASI-II. Eight participants at the University of Utah were recruited and assessed via videoconferencing. Methods for recruiting these participants were very similar to those noted above, with the following procedural differences: remote participants were emailed the consent and assent forms, and then engaged in consent procedures over the phone before signing the assent and consent electronically. These participants were also mailed copyrighted forms to fill out and completed all other forms via REDCap, a confidential online survey platform. Further, these participants engaged in the clinical interviews through VSee, a confidential videoconferencing program.

3. Results

3.1. Handling of missing data

For CTIM-P tic-related impairment, 6 participants were missing one item, 1 participant was missing two items, and 2 participants were missing three items. For non-tic-related impairment, 6 participants were missing one item, 3 participants were missing two items, 2 participants were missing three items, 1 participant was missing 6 items, and 8 participants were missing over seven items. Item scores were imputed if participants had 20% or less (i.e., 7 items) of their data missing. Specifically, item scores were replaced with the subject's own subscale average item score.

3.2. Statistical analyses

Scores on the YGTSS and CTIM-P did not follow a normal distribution; therefore, nonparametric tests (i.e., Wilcoxon signed rank test and Spearman rho correlation coefficients) were calculated when examining tic-related versus non-tic-related impairment at the domain and activity level and when examining correlations between tic-related impairment and tic severity. Also, when examining correlations between tic-related impairment at the CTIM-P domain level and dimensions of tic severity, a Bonferroni correction for multiple comparisons was used. Although the test was exploratory, we still addressed the concern of multiple comparisons. However, when exploring activity differences in tic-related and non-tic-related impairment, the concern of multiple comparisons was not addressed as it would be difficult to observe significant results using a correction for 37 separate comparisons.

3.3. Domain differences in tic-related versus non-tic-related impairment

It was hypothesized that problems during home, school, and social activities would be attributed more strongly to non-tic than tic-related symptoms. Results of Wilcoxon signed-rank tests partially supported this hypothesis. Results showed that parents attributed their children's levels of impairment in home activities more to non-tic-related symptoms (M = 4.62, SD = 5.65) than tics themselves (M = 3.45, SD = 4.91). However, tic-related impairment in school (M = 5.03, SD = 4.94) and social activities (M = 5.57, SD = 8.80) did not differ from non-tic-related impairment in these same activities (M = 5.13, SD = 6.10; M = 5.58, SD = 9.21). Also, it was found that overall tic-related impairment scores (M = 14.07, SD = 14.99) did not significantly differ from overall non-tic-related impairment scores (M = 15.60, SD = 18.81). See <u>Table 2</u> for statistics associated with each of the Wilcoxon signed rank tests associated with the comparisons described in this section.

Table 2. Differences in Tic-Related and Non-Tic-Related Impairment.

CTIM-P	Tic related mean	Non-tic related	Z Value	Significance	Effect size
	(SD)	mean (<i>SD</i>)		level	(r)
Home activities					
Total home activities	3.45 (4.91)	4.62 (5.65)	-2.14	0.03*	0.28
Getting dressed in the morning	0.18 (0.50)	0.37 (0.65)	-2.14	0.03 <u>*</u>	0.28
Bathing or grooming	0.18 (0.50)	0.41 (0.74)	-2.36	0.02*	0.31
Doing household chores	0.28 (0.68)	0.65 (0.91)	-2.30	0.02*	0.30
Eating meals at home	0.23 (0.57)	0.28 (0.71)	-0.63	0.53	0.08
Getting ready for bed at night	0.23 (0.47)	0.44 (0.72)	-1.64	0.10	0.22
Sleeping at night	0.58 (84)	0.51 (0.91)	-0.06	0.95	0.01
Getting along with siblings	0.48 (0.79)	0.62 (0.87)	-1.82	0.07	0.24
Getting along with parents	0.36 (0.67)	0.58 (0.71)	-2.56	0.01*	0.34
Visiting relatives	0.22 (0.50)	0.33 (0.67)	-1.61	0.11	0.21
Going on family vacation	0.21 (0.64)	0.31 (0.72)	-1.44	0.15	0.19
Going to religious services	0.41 (0.77)	0.31 (0.70)	-0.30	0.76	0.04
School activities					
Total school activities	5.03 (4.94)	5.13(6.10)	-0.43	0.67	0.06
Getting to school on time	0.14 (0.40)	0.38 (0.68)	-2.70	0.01*	0.35
Missing school	0.19 (0.55)	0.22 (0.57)	-0.07	0.94	0.01
Giving oral reports/reading out loud	0.60 (0.77)	0.42 (0.69)	-2.20	0.03*	0.29
Being prepared for class	0.41 (0.73)	0.70 (0.87)	-2.57	0.01*	0.34
Writing in class	0.81 (0.96)	0.49 (0.81)	-2.35	0.02*	0.31
Taking tests or exams	0.67 (0.80)	0.63 (0.96)	0	1	0
Doing homework	0.74 (0.91)	0.78 (0.98)	-0.55	0.58	0.07
Participating in gym	0.17 (0.38)	0.19 (0.59)	-0.35	0.73	0.05
Doing fun things during	0.17 (0.46)	0.31 (0.75)	-1.61	0.11	0.21
recess/free time					
Concentrating on his/her work	0.90 (0.95)	0.80 (0.86)	-0.45	0.66	0.06
Eating meals with other kids	0.22 (0.50)	0.22 (0.60)	-0.19	0.85	0.02
Social activities					
Total social activities	5.57 (8.80)	5.58 (9.21)	-0.69	0.67	0.09
Making new friends	0.54 (0.80)	0.62 (1.0)	-0.86	0.39	0.11
Keeping friends	0.40 (0.73)	0.36 (0.74)	-0.78	0.44	0.10
Spending time with friends	0.33 (0.63)	0.41 (0.74)	-0.87	0.38	0.11
Having conversations with other kids	0.56 (0.73)	0.55 (0.89)	-0.12	0.90	0.02
Being teased by peers	0.77 (0.82)	0.34 (0.78)	-3.36	< 0.01 <u>*</u>	0.45
Leaving the house	0.28 (0.64)	0.20 (0.60)	-1.89	0.06	0.25
Being with a group of strangers	0.52 (0.88)	0.67 (0.95)	-1.29	0.20	0.17
Going to a friend's house during the day	0.24 (0.66)	0.33 (0.67)	-0.83	0.41	0.11
Having a friend at the house during the day	0.19 (0.58)	0.29 (0.66)	-1.51	0.13	0.20
Spending the night at a friend's house	0.31 (0.75)	0.53 (1.0)	-2.07	0.04*	0.27
Having a friend spend the night	0.22 (0.65)	0.35 (0.82)	-1.40	0.16	0.18
Having a boyfriend/girlfriend	0.22 (0.65)	0.31 (0.72)	-1.19	0.23	0.16
Going shopping	0.35 (0.77)	0.29 (0.71)	-0.70	0.48	0.09
Eating in public places	0.26 (0.69)	0.26 (0.71)	0	1	0
Going to the movies	0.36 (0.74)	0.19 (0.59)	-2.23	0.03*	0.29
Overall impairment scores	14.07(14.99)	15.6 (18.81)	-0.22	0.83	0.03

Note. The statistics presented in this table correspond with <u>Wilcoxon signed rank tests</u> calculated to examine difference between tic-related versus non-tic-related impairment across home, school, and social domains and activities. CTIM-P = Child <u>Tourette's Syndrome</u> Impairment Scale - Parent Report. *p < .05.

3.4. Activity differences in tic-related and non-tic-related impairment

Although parents did not report that impairment across the domains of home, school, and social activities was more related to tics than to non-tic-related problems, it is possible that, for particular items, impairment was more attributed to tics than non-tic-related concerns. As a result, we conducted several exploratory Wilcoxon signed-rank tests to examine tic-related versus non-tic-related impairment on individual CTIM-P items. See <u>Table</u> 2 for the statistics associated with each of the calculated Wilcoxon signed-rank tests.

For home activities, having trouble with getting dressed in the morning, bathing or grooming, doing household chores, and getting along with parents were more strongly attributed to non-tic-related factors than tic-related factors. For school activities, children's difficulties with giving oral reports/reading out loud and writing in class were more attributed to tics than non-tic problems. However, having trouble getting to school on time and being prepared for class were attributed to other problems more than tics. For social activities, being teased by peers and difficulty going to the movies were attributed more to tics than non-tic-related issues, but difficulties in spending the night at a friend's house was more attributed to non-tic-related factors.

3.5. Correlations between tic-related impairment on the CTIM-P and tic severity

To explore the relationship between tic severity and <u>functional impairment</u>, Spearman rho <u>correlation coefficients</u> were calculated between total tic-related impairment on the CTIM-P and YGTSS motor, vocal, and total tic severity. Results are presented in <u>Table 3</u> and show that all three of these correlation coefficients were significant and positive. Thus, subsequent relationships between the three CTIM-P domains and YGTSS total, motor, and vocal tic severity scores were calculated. Results indicated that (a) YGTSS total tic severity and motor tic severity were significantly positively correlated with CTIM-P tic-related impairment in home, school, and social activities and (b) vocal tic severity was significantly positively correlated only with tic-related impairment in social activities. Correlation coefficients for these analyses are presented in <u>Table 3</u>.

	Clinician-rated Tic Severity.

		1	
CTIM-P	YGTSS total	Motor total	Vocal total
Total tic-related impairment	0.55*	0.57*	0.37*
Home activities	0.36 <u>*</u>	0.37 <u>*</u>	0.25
School activities	0.41*	0.51 <u>*</u>	0.20
Social activities	0.47≛	0.40*	0.41*

Note. CTIM-P = Child <u>Tourette's Syndrome</u> Impairment Scale - Parent Report; YGTSS = Yale Global Tic Severity Scale.

*p < .05.

Additional Spearman rho correlation coefficients (with Bonferroni corrections for multiple comparisons) were calculated to examine the relationships between dimensions of motor severity (i.e., number, frequency, intensity, complexity, and interference) and tic-related impairment in all three CTIM-P domains. As shown in Table 4, motor tic intensity, complexity, and interference scores showed a significant correlation with tic-related impairment in school activities. Only motor tic complexity correlated with tic-related impairment in home and social activities.

Table 4. Correlations between tic-related impairment at the CTIM-P domain level and clinician-rated dimensions of tic severity.

CTIM-P	YGTSS	YGTSS	YGTSS	YGTSS	YGTSS
	number	frequency	intensity	complexity	interference
Motor tics					
Home activities	0.34*	-0.01	0.19	0.40**	0.29*
School activities	0.25	0.12	0.50**	0.49**	0.46**
Social activities	0.33*	0.14	0.30 <u>*</u>	0.36**	0.32*
Vocal tics					
Social activities	0.38**	0.30 <u>*</u>	0.37**	0.21	0.39**

Note. Correlations between the dimensions of YGTSS vocal tic severity and CTIM-P home and school activities are not reported because YGTSS total vocal tic severity was not significantly correlated with CTIM-P school and home activities. CTIM-P = Child <u>Tourette's Syndrome</u> Impairment Scale - Parent Report; YGTSS = Yale Global Tic Severity Scale.

Because social activities were the only domain to significantly correlate with vocal tic severity, Spearman rho correlations were calculated to explore the relationships between dimensions of vocal tic severity (i.e., number, frequency, intensity, complexity, and interference) and social activities. As shown in <u>Table 4</u>, vocal tic number, intensity, and interference significantly correlated with tic-related impairment in social activities after a Bonferroni correction.

4. Discussion

Although several studies have confirmed that children with PTDs experience impairment across several functional domains [5], [6], [7], [8], [9], [10], [11]], the relative contribution of tics versus non-tic-related problems remains unclear. Accordingly, the current study replicated earlier studies by Storch et al. [12] and Cloes et al. [27] by examining tic and non-tic-related impairment across broad functional domains (i.e., home, school, and social) using an established measure of tic- and non-tic-related interference (the CTIM-P). In addition, for particular items on the CTIM-P, we examined whether impairment was more strongly attributed to tics or non-tic-related problems. Finally, the association between tic severity and CTIM-P tic-related impairment was examined, and the relationships between tic-related impairment and the five dimensions contributing to YGTSS motor and vocal tic severity scores were explored.

4.1. Differences in tic-related and non-tic-related impairment at the domain level

It was hypothesized that parents of children with PTDs would attribute more impairment in home, school, and social activities to non-tic problems than tics. This hypothesis was partially supported. Impairment in home activities was attributed more to non-tic-related problems than tics, but there was no difference between tic-related and non-tic-related impairment in school or social activities. Reasons for these findings are not clear. Perhaps the home setting gives parents a greater opportunity to observe their children's behaviors relative to school or social settings and as a result parents can make attributions more definitively at home, whereas in school or social settings, parents are less aware of how tics/other problems interfere. Another possible explanation is that parents and other family members may be more familiar with and adept at managing tics than individuals at school or in social situations. Therefore, relative to other behaviors, tics may be perceived as less impairing at home.

^{*}p < .05.

^{**}Bonferroni correction: p < .01.

4.2. Differences in tic-related and non-tic-related impairment at the activity level

Subsequent analyses were conducted to examine differences in impairment at the CTIM-P activity level. It was found that impairment in giving oral reports/reading out loud was significantly more attributed to tics than other concerns. This could be expected, as speaking in front of others can be stressful and, consequently, worsen tics [33]. Likewise, tics may also elicit social reactions from peers or result in escape from or avoidance of a task, thereby increasing tics and tic-related impairment [34]. Vocal tics may be especially impactful because they may interrupt or distract affected children as they try to talk to peers. Tic impairment in specific school-related tasks, such as writing, were also more attributed to tics compared to non-tic symptoms. One possible explanation for this is tics involving the hands or arms (e.g., tensing up hands, wrist flick/jerk, and throwing pens) likely interfere with the ability to write [10]. Regarding social activities, being teased by peers and impairment in activities such as going to the movies were more attributed to tics than other concerns. This is not particularly surprising as previous research has shown that many children with PTDs reported being teased because of their tics and affected children that are victims of bullying typically report more frequent, complex, and severe tics [9].

Impairment in several other activities such as getting to school on time, being prepared for class, bathing and grooming, doing household chores, spending the night at a friend's house, and getting along with parents were more attributed to non-tic concerns than tics. These findings are consistent with prior research showing that OCD and ADHD, which are commonly comorbid with PTD, are associated with impairment in these domains. For example, Stewart et al. [35] demonstrated that OCD symptoms often interfere with morning and bedtime routines and lead to frustration and anger between children and parents. Further, Deault [36] showed that families with children who have ADHD tend to have above average levels of conflicted parent-child interactions. In addition, individuals with TD tend to be hypersensitive to a broad range of sensory stimuli and have difficulties with visual motor integration, so these non-tic-related factors could influence bathing and grooming and doing household chores [22].

4.3. Correlations between tic-related impairment on the CTIM-P and tic severity

The hypothesis that YGTSS tic severity scores for motor, vocal, and total tic severity would positively correlate with total tic-related impairment was supported. Motor, vocal, and total tic severity scores on the YGTSS were significantly positively correlated with total tic-related impairment on the CTIM-P. Further, when associations between severity and CTIM-P domains were examined, significant positive correlations were found between total and motor tic severity and tic-related impairment in home, school, and social activities. Vocal tic severity was only correlated with tic-related impairment in social activities. These findings are partially consistent with previous research [12,27]. It is not surprising that more impairment would be attributed to tics when tics are more severe (i.e., higher in number, frequency, intensity, complexity, and interference). It also makes sense that vocal tic severity was related to increased impairment in social activities because it has been shown that peers view children with frequent vocal tics as less "normal" than other children, which can lead to social withdrawal [9]. In addition, vocal tics exhibited at home may be less of an issue because family members have more experience and familiarity with the tics and parents are less likely to respond negatively to tics compared to peers. However, the finding that vocal tics are related to less interference at school is more difficult to explain given that vocal tics may elicit social reactions from peers at school [9,34]. One possible explanation for these discrepant findings lies in the types of questions contained in the school domain on the CTIM-P. In particular, the items in the school domain focus on activities such as getting to school on time, being prepared for class, doing fun things during recess, doing homework, etc. Many of these activities are not related to interactions with peers; thus, it is possible that tic-related impairment is not associated with this domain.

4.4. Correlations between tic-related impairment and the dimensions of motor and vocal severity

Additional exploratory analyses examined whether tic-related impairment was correlated with the various dimensions of motor and vocal tic severity measured by the YGTSS (i.e. number, frequency, intensity, complexity, and interference). Motor tic intensity, complexity, and interference significantly correlated with tic-related impairment in school activities. Such results are consistent with work by Espil et al. [5] and Wadman et al. [10] who demonstrated that these dimensions predicted impairment in school productivity by influencing students' attention and writing skills. Likewise, motor tics that interrupt the flow of behavior can be difficult to manage in a school and/or social environment. For instance, hand tics (e.g., tensing up hands, wrist flick/jerk, and throwing pens) can make taking tests and doing homework extremely challenging [10]. Results also showed that motor tic complexity correlated with tic-related impairment in home and social activities. Given that complex tics can be very noticeable, and that noticeability of tics is related to negative peer perception ratings [6], such a relationship between motor complexity and social impairment is not surprising.

Further, vocal tic number, intensity, and interference were significantly positively correlated with tic-related impairment in social activities. It is not surprising that problems with peers are associated with a greater number of vocal tics because having multiple different tics gives peers multiple ways in which an affected child could be impacted socially. Also, in social situations, if other people react to a child's numerous vocal tics by talking to the child about his/her tics, the vocal tics will continue to increase and become more impairing [37]. Presumably, this is even more likely to occur when the tic is intense and exaggerated in character because the tic will be more noticeable to others. Likewise, tics that interrupt speech and disrupt intended action or communication (i.e., interfering) can be problematic when a child is trying to interact with his/her peers and could lead to bullying and difficulty in making friends.

4.5. Limitations

The current study had several limitations. First, the sample size (*n* = 58), though similar to previous studies examining the CTIM-P (e.g., [12]) was still relatively small and mostly comprised of white, non-Hispanic males. Second, this study did not collect children's perspectives on their own tic-related and non-tic-related impairment. This is potentially problematic because the parents who participated in this study may have been unable to directly observe their children's behavior in school or some social situations. Thus, it is unclear whether parents have an accurate understanding of their children's behaviors and impairment in those situations. Future research would benefit from also examining children's perspective on their tic-related versus non-tic-related impairment. This may be done by utilizing self-report measures like the CTIM-C. Moreover, the study did not control for comorbidities or medication, though one-third of the sample (32.8%) were taking one or more medications during the course of their participation in the study. Further, there was not a measure quantifying the severity of non-tic-related problems, preventing us from assessing the correlation between severity of these concerns and impairment due to these concerns. Lastly, it should be noted that the correlations between tic severity and tic-related impairment were exploratory. As a result, such findings should be considered more as hypothesis generating than hypothesis testing. It is our hope that these results will lead to future research questions examining how the different dimensions of tics influence impairment.

4.6. Implications

This study showed that parents of children with PTDs attribute impairment in home activities more to other concerns than to tics; however, parents reported no difference between tic-related and non-tic-related impairment in school or social activities. As a result, if children with tics are most impaired at home, the results

would suggest that non-tic-related concerns should likely be the focus of treatment. However, since impairment attributed to tics and to other non-tic problems seems to be high in both school and social activities, intervention strategies should focus on alleviating both tic-related and non-tic-related issues, as such an approach may improve functioning more than treating one symptom cluster or the other. In addition, although tics did not appear to yield greater impairment than non-tic-related problems across the three domains, for particular items, impairment was more attributed to tics than to non-tic-related problems. This may be beneficial clinically as it could help clinicians become aware of the activities that tend to be most impaired by tics.

The exploratory analyses done in this study lead to additional questions about whether the dimensions of motor and vocal tic severity differentially impact impairment in children with tics. Further research needs to be done to pinpoint the aspects of tics that are most impairing to children, so those can be the focus of treatment. Also, it would be interesting if future research could examine impairment attributed to tic-related and non-tic-related problems in children of different ages, as children of different ages face unique challenges due to changing social environments (e.g., in adolescence, vast changes occur in the social environment, contributing to challenging school and peer relationships for some children).

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Conflicts of interest

Dr. Woods receives royalties from the Oxford University Press, Springer Press, and Guilford Press. Also, Drs. Woods and Himle give paid talks for the Tourette Association of America. Finally, Dr. Mouton-Odum is the President of PsycTech, Ltd., a private practice that receives revenue from <u>TicHelper.com</u>.

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