



Academic, Interpersonal, Recreational, and Family Impairment in Children with Tourette Syndrome and Attention-Deficit/Hyperactivity Disorder

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Abstract

This study describes impairment in academic, interpersonal, recreational, and family financial or occupational domains across children in three mutually exclusive diagnostic groups: ever diagnosed with Tourette syndrome (TS), attention-deficit/hyperactivity disorder (ADHD), and both disorders. In 2014, parents reported on impairment and diagnostic status of children aged 4–17 years ($n = 3014$). Weighted analysis and pairwise *t*-tests showed more children with ADHD (with or without TS) experienced impairment in overall school performance, writing, and mathematics, relative to children with TS but not ADHD. More children with TS and ADHD had problematic handwriting relative to children with ADHD but not TS. More children with TS and ADHD had problematic interpersonal relationships relative to those with ADHD but not TS. Children with TS and ADHD had higher mean impairment across domains than children with either TS or ADHD. Findings suggest assessing disorder-specific contributions to impairment could inform targeted interventions for TS and ADHD.

Keywords Tourette syndrome · ADHD · Children · Academic performance · Social behavior · Interpersonal relations · Cost of illness

Introduction

Tourette syndrome (TS) and attention-deficit/hyperactivity disorder (ADHD) are childhood-onset, neurodevelopmental disorders broadly characterized by disinhibited actions [1]. TS involves involuntary movements and vocalizations (i.e., tics) present for longer than one year, whereas ADHD

is marked by a pattern of inattention and/or hyperactivity-impulsivity lasting at least 6 months [2]. These disorders frequently co-occur and share pathophysiological factors, including frontal neural (e.g., basal ganglia) dysfunction, susceptibility genes, and maternal or pre-perinatal risk factors (e.g., alcohol or smoking during pregnancy, premature birth, low birth weight) [3]. Although these disorders often

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co-occur (i.e., rates of ADHD in children with TS are estimated at 63% and rates of TS in children with ADHD are estimated at 1.2% in population-based samples) [4, 5], they differ with respect to their prevalence within the general population of children (0.3% to 0.9% for TS, 8% to 10% for ADHD) [5–7]. For individuals who have either disorder, functional impairment is common. For instance, children with ADHD often experience impairment in social functioning (e.g., social deficits, low self-esteem), family functioning, and academic functioning in comparison to peers without ADHD [8]. Previous research shows that children with TS experience difficulties in similar functional domains [4, 9, 10].

Identifying the functional impairment associated with each disorder may be clinically beneficial. Interventions for TS include education about tics, behavior therapy (i.e., Habit Reversal Training/Comprehensive Behavioral Intervention for Tics), and medication (e.g. alpha-2-adrenergic agonists, antipsychotics) [11]. Treatments for ADHD include education, parent and classroom behavior training, and stimulant medication [12]. Guidelines on the treatment of TS and co-occurring disorders, such as ADHD suggest prioritizing treatment for the most impairing disorder. In the event that co-occurring conditions are most impairing, guidelines suggest treating them first may indirectly improve the child's management of TS [13, 14]. Better understanding of the specific functional impairment domains, and the disorder (i.e., TS and/or ADHD) the impairment is associated with may guide clinicians in their choice of treatment.

Empirical studies have demonstrated that children with co-occurring TS and ADHD experience greater challenges in academic and social functioning than children with TS alone [10, 15–18]. The clinical picture of impairment increases in complexity when comparing children with co-occurring TS and ADHD to children with ADHD without TS. For instance, one study found that children with co-occurring TS and ADHD experience greater deficits in global functioning relative to children with ADHD alone [19]. In contrast, Lin and colleagues [20] found that children with co-occurring ADHD and TS had fewer social problems than children with ADHD alone. Meanwhile, other reports have found no difference in social or family functioning between children with co-occurring TS and ADHD compared to children with ADHD only [18, 21]. Taken together, these findings suggest that children with co-occurring TS and ADHD have greater impairment than children with TS without ADHD; however, the findings are inconsistent when comparing children with co-occurring TS and ADHD to those with ADHD but not TS. Parsing the respective influences of TS and ADHD on functional impairment may inform the development or prioritization of treatment approaches to alleviate impairment for children with one or both disorders.

Despite the growing amount of research dedicated to understanding the contributions of TS and ADHD on children's functioning, prior research has often been limited to clinic-referred samples of children, which does not necessarily reflect the heterogeneity of symptom severity or impairment within the larger population of children with TS and/or ADHD. Furthermore, little is known about the differential influence of TS and ADHD on impairment with respect to children's recreational functioning and family financial and occupational problems due to TS or ADHD.

In response to these gaps in the literature and the need for more research on impairment among children with TS and their families (both with and without ADHD), the objective of this investigation was to examine functional impairment across academic, interpersonal, recreational, and family financial or occupational domains in a sample of children classified into the diagnostic groups of TS without ADHD (TS-only), ADHD without TS (ADHD-only), and TS and ADHD (TS + ADHD), based on parent-reported data. This investigation utilizes a nationally drawn sample from the 2014 National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome (NS-DATA). Based on the extant literature obtained from clinically referred children, we hypothesized that children in the TS + ADHD group would have greater functional impairment across all four domains compared to either the TS-only or ADHD-only groups.

Methods

Data for this analysis are from NS-DATA, an in-depth survey regarding the diagnostic experiences and treatment utilization of children ever diagnosed with TS and/or ADHD [22]. NS-DATA is a follow-up telephone survey to the 2011–2012 National Survey of Children's Health (NSCH). The NSCH is a population-based survey in which a random digit-dial sample of landline and cell phones was used to select a nationally-representative sample of households with non-institutionalized children aged 0–17 years in the United States (U.S.) [22]. Parents or primary caretakers (herein referred to as parents) were queried about their child's physical health, emotional health, and overall wellbeing [22]. For households with multiple children, one child was randomly selected to be the subject of the NSCH [22].

Children whose parents indicated on the 2011–2012 NSCH as having ever been diagnosed with TS or ADHD by a healthcare provider were eligible to be included in the NS-DATA sample; data collection occurred January 2014–June 2014 [23]. Additional eligibility criteria for NS-DATA included the following: (1) at the time of NS-DATA, the responding parent confirmed that the target child had ever been diagnosed with TS or ADHD (2) child was

less than 18 years of age at the time of NS-DATA; (3) the child resided in the same household as the responding parent (although the responding parent for NS-DATA did not need to be the same responding parent who participated in 2011–2012 NSCH) [23]. Parents were read a consent statement over the phone indicating that the research was voluntary, data would remain confidential and be used only for research purposes; they received monetary reimbursement for their participation.

For these analyses we created three diagnostic groups to compare disorder-specific impairment: TS-only, ADHD-only, and TS + ADHD. Each child's diagnostic group was determined by their parents answer to the questions: *Has a doctor or health care provider ever told you that your child had [(TS) or (ADHD or ADD¹)]? Children in the TS-only group had ever been diagnosed with TS and never diagnosed with ADHD; children in the ADHD-only group had ever been diagnosed with ADHD and never diagnosed with TS; Children in the TS + ADHD group had ever been diagnosed with TS and ADHD. As a follow-up, parents were also asked, Does [your child] currently have [(TS) or (ADHD)]? Children did not need to have current TS or ADHD at the time of NS-DATA to be eligible and the current disorder status did not contribute to the determination of the diagnostic groups.*

Survey Items

NS-DATA included separate modules for children ever diagnosed with TS and ever diagnosed with ADHD. If a parent endorsed that their child had ever been diagnosed with TS and ADHD, the parent was asked to complete both NS-DATA modules [23]. For the majority of questions that were asked in both modules, the question was only asked once; exceptions are detailed below. The final sample consisted of 30 children in the TS-only group, 2899 children in the ADHD-only group, and 85 children in the TS + ADHD group (total $n=3014$). All data included in these analyses were collected at the time of NS-DATA with the exception of four demographic variables that were carried forward from 2011 to 2012 NSCH (i.e. child sex, minority status, highest level of parent education, and federal poverty level).

Demographics

The demographic and socioeconomic information considered in this study included the children's age, sex, minority status (if identified as Black, Hispanic, or other race/ethnicity [i.e., not non-Hispanic White], the children was included in the minority group), parent education, and

federal poverty level. Parent education was the highest level of education achieved by either parent. For federal poverty level, the Centers for Disease Control and Prevention's National Center for Health Statistics (NCHS) used reported income level and household size at the time of 2011–2012 NSCH to convert household income to percentage of federal poverty level. To account for missing data on income and/or household size, NCHS calculated imputed values using hot-deck imputation to estimate federal poverty level [22]. Approximately 5% or less of each NS-DATA diagnostic group had imputed values for federal poverty level (from 2011 to 2012 NSCH) and analysis of this indicator followed guidance from NCHS [22].

Clinical Characteristics

Parents who reported their child currently had TS or ADHD were also asked, *Would you describe [your child]'s [(TS) or (ADHD)] as mild, moderate, or severe?*; each variable was dichotomized to "mild" and "moderate/severe." For the TS + ADHD group, children were classified as having a current disorder if either TS or ADHD was current at the time of NS-DATA (i.e., this included children with: current ADHD and lifetime TS; current TS and lifetime ADHD; current ADHD and current TS). Current severity was based on the most severe current parent rating of either TS or ADHD. Current TS and ADHD medication status was assessed with the following item: *Is [your child] currently taking medication for [(TS) or (ADHD)]?* and was only asked to parents of children who currently had the disorder and who endorsed that their child had ever taken medication for their disorder. For the TS + ADHD group, children whose parent reported they were taking medication for either disorder were considered to have "medication for current disorder."

Ever diagnosed and current occurrence of other co-occurring psychiatric disorders were measured by parent report to questions on whether a healthcare provider had ever told them that their child had a specified disorder, and if yes, whether the child currently had the specified disorder. The following eleven psychiatric co-occurring disorders were included in our analyses: (1) anxiety disorders, such as generalized anxiety, panic disorder, or a phobia; (2) mood disorders, such as depression or major depressive disorder; (3) bipolar disorder; (4) post-traumatic stress disorder; (5) obsessive–compulsive disorder; (6) oppositional defiant disorder; (7) conduct disorder; (8) autism spectrum disorder or pervasive developmental disorder; (9) intellectual disability; (10) language disorder; and (11) learning disorder. We created two dichotomous variables (present or absent) for ever diagnosed and current other co-occurring psychiatric disorders.

¹ ADD: Attention Deficit Disorder.

Table 1 Verbatim questions and response options from the 2014 National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome (NS-DATA) to assess for impairment in academic, interpersonal, recreational, and family financial or occupational domains

Impairment domains	Response options
<i>Academic</i>	
How would you describe [your child]’s overall school performance? ^a	
How would you describe [your child]’s performance in reading? ^a	
How would you describe [your child]’s performance in writing? ^a	
How would you describe [your child]’s performance in mathematics? ^a	Problematic
How would you describe [your child]’s handwriting, that is, [his/her] ability to form letters and numbers that are clear and can be recognized?	Somewhat problematic
<i>Interpersonal</i>	
How would you describe [your child]’s relationship with [his/her parents]? ^a	Average
How would you describe [your child]’s relationship with [his/her siblings]? ^a	Above average
How would you describe [your child]’s relationship with [his/her peers]? ^a	Excellent
<i>Recreational</i>	
How would you describe [your child]’s participation in organized activities such as teams? ^a	
<i>Family financial or occupational</i>	
Has [your child]’s [(TS) or (ADHD)] caused financial problems for your family?	
Have you or other family members stopped working because of [your child]’s [(TS) or (ADHD)]?	Yes
Have you or other family members cut down on the hours you work because of [your child]’s [(TS) or (ADHD)]?	No
Have you or other family members avoided changing jobs because of concerns about maintaining health insurance for [your child]’s [(TS) or (ADHD)]?	

^aThese questions are from the Vanderbilt ADHD Parent Rating Scale [24]

Domains of Impairment

Impairment due to TS and/or ADHD was measured with indicators related to the following four domains: (1) Academic, (2) Interpersonal, (3) Recreational, and (4) Family Financial or Occupational. Most of the questions in the academic, interpersonal, and recreational domains came from the Vanderbilt ADHD Parent Rating Scale. The specific questions [24] included in each impairment domain are listed in Table 1. For the academic, interpersonal, and recreational domains, parents were instructed to think about a time in the past six months when their child was not taking any medication for TS, ADHD, or any other medication for other difficulties with their emotions, concentration, or behavior. Help text was provided for interviewers to explain that “school includes preschool or daycare” if parents asked in response to the questions related to academics. Unless otherwise specified, for our analysis, answer options of *problematic*, and *somewhat problematic*, were categorized as “problematic” while *average*, *above average*, and *excellent* were categorized as “not problematic.”

Academic Impairment

Questions about children’s overall school performance and their specific performance in reading, writing, mathematics, and handwriting were asked of parents and rated on the same problematic scale (i.e., “problematic” vs. “not

problematic”); see Table 1. Handwriting was included with academic impairment due to associations between TS [25] and ADHD [26] and difficulties with handwriting. Grades were queried with *Overall, would you consider [your child] an A student, B student, C student, D student or F student?* and were collapsed into three categories, “A/B student,” “C student,” and “D/F student.” Due to small sample sizes in the TS-only group, the analysis of grades is presented by collapsing TS-only and TS + ADHD groups, and comparing that to the grades for the ADHD-only group.

Interpersonal Impairment

Parents were queried about the nature of their child’s relationships with parents (i.e., themselves), siblings, and peers; see Table 1. The three interpersonal relationship questions were collapsed into a single derived variable, and children were rated as having “interpersonal impairment” if a parent endorsed that their child had any problematic relationships.

Parents of children ever diagnosed with TS (TS-only and TS + ADHD groups) were asked *Has your child ever been treated differently because of [his/her] tics, for example, being bullied or discriminated against, or treated rudely?* If yes, parents were then asked *Who has discriminated against or bullied the child? Were they siblings, other children, business owners or employees, teachers, family members or*

some other adults? and told to report all that apply. This series of questions was not included in the ADHD module of NS-DATA and therefore results are only presented for TS-only and TS + ADHD diagnostic groups.

Recreational Impairment

Parents reported on their child's participation in recreational activities (i.e., organized activities such as teams) on the same problematic scale (i.e., "problematic" vs. "not problematic"); see Table 1. If a parent indicated their *child does not participate in organized activities* this was coded as missing for our analysis (TS-only and TS + ADHD, $n \leq 5$ and ADHD-only, $n = 117$).

Family Financial or Occupational Impairment

Family financial or occupational impairment was assessed using a composite variable based on four questions about family financial problems and work considerations due to their child's TS or ADHD; verbatim questions are listed in Table 1. A response of *yes* to any of the four questions reflected "family financial or occupational impairment due to their child's disorder." The family financial or occupational impairment questions were asked specific to each disorder; for children in the TS + ADHD group, a response of *yes* to any of the financial or occupational problems for either disorder was considered "family financial or occupational impairment" in our analysis.

Overall Impairment by Diagnostic Groups

Overall impairment was measured using the sum of the domains in which children had any parent-reported impairment (possible range 0 to 4); mean (M) impairment scores were calculated and compared across diagnostic groups (TS-only, ADHD-only, TS + ADHD).

Analyses

Due to confidentiality concerns related to the smaller sample size of the NS-DATA TS module, data from the TS module could only be accessed through the NCHS Research Data Center.² Therefore, the datasets for the diagnostic groups were analyzed separately. Weighted prevalence estimates and 95% confidence intervals (CI) were calculated for each of the three diagnostic groups. For the TS-only and

TS + ADHD groups, we used a weight variable specific to children with TS and for the ADHD-only group, we used the weight variable in the ADHD public-use dataset; all weight variables were calculated and provided by the NCHS. While analyses incorporating sample weights are traditionally intended to produce estimates reflective of a population of children, sampling challenges, small sample size, and confidentiality concerns among those children with TS limited the generalizability of our sample. While we performed a weighted analysis, results are not representative of the U.S. population of children with TS. With that said, we followed all analytical guidance in the NS-DATA methods report and performed weighted analyses using SAS-callable SUDAAN VERSION (RTI International; Cary, NC) to account for the complex survey design [27]. Differences between the three diagnostic groups (e.g., TS-only vs. ADHD-only; ADHD-only vs. TS + ADHD; TS-only vs. TS + ADHD) were compared using pairwise t-tests. T-tests were manually calculated in Excel using weighted estimates and standard errors that were calculated with the consideration for the complex survey design of the NS-DATA. Throughout the analysis, p-values of less than 0.05 were considered to be significant. If a parent refused to answer or responded "don't know" to a question the response was considered missing. For all collapsed derived impairment indicators children were only classified as missing if they were missing responses to all questions used to create the derived indicator.

Results

Demographics

Demographic information is presented by diagnostic group (TS-only, ADHD-only, and TS + ADHD) in Table 2. Children ranged in age from 4 to 17 years and most were adolescents (12–17 years of age; range 68.5–88.2% across diagnostic groups) and male (range 69.9–86.2%). Children in the TS-only group were more likely to be in the older age group ($p < 0.01$) and male ($p = 0.04$) compared to children in the ADHD-only group. While only 10.4% of children in the TS-only group were of a racial/ethnic minority group (i.e., Black, Hispanic, or other race/ethnicity), significantly more children were of a minority race/ethnicity group in the ADHD-only group (37.6%, $p < 0.01$) and the TS + ADHD group (32.4%, $p = 0.02$). Indicators of parental education and federal poverty level did not significantly differ by diagnostic group.

Clinical Characteristics

Diagnostic groups did not significantly differ with respect to the proportion of children with current TS and/or ADHD at the time of NS-DATA (range 70.8–86.2%; see Table 2).

² The dataset for NS-DATA ADHD module is publicly available; the TS module requires a data agreement with the National Center for Health Statistics' Research Data Center. More information about accessing the NS-DATA datasets can be found at www.cdc.gov/nchs/slraits/ns_data.htm and www.cdc.gov/rdc.

Table 2 Demographic and clinical characteristics among youth ever diagnosed with Tourette syndrome (TS) and attention-deficit/hyperactivity disorder (ADHD)

	TS-only (n = 30) % (CI)	ADHD-only (n = 2899) % (CI)	TS + ADHD (n = 85) % (CI)	TS-only vs. ADHD-only <i>p</i> -value	ADHD- only vs. TS + ADHD <i>p</i> -value	TS-only vs. TS + ADHD <i>p</i> -value
Demographics						
Child age (% 12–17 years)	88.2% (67.8%–96.3%)	68.5% (65.5%–71.4%)	69.9% (53.3%–82.6%)	< 0.01	0.85	0.07
Sex ^a (% male)	86.2% (63.1%–95.8%)	69.9% (66.9%–72.7%)	79.8% (63.2%–90.2%)	0.04	0.15	0.53
Minority status ^a (% minority race/ethnicity) ^b	10.4% (2.9%–31.0%)	37.6% (34.4%–40.9%)	32.4% (20.7%–46.7%)	< 0.01	0.44	0.02
Parental education ^a (% either parent attended at least some college or more)	77.9% (49.1%–92.8%)	66.0% (62.7%–69.1%)	68.1% (51.9%–80.9%)	0.29	0.78	0.46
Federal poverty level (FPL) ^{a,c} (% living in households with income ≤ 200% FPL)	28.2% (10.1%–57.8%)	50.7% (47.5%–53.9%)	51.0% (36.8%–65.1%)	0.08	0.97	0.12
Clinical characteristics						
Current TS and/or ADHD ^d	70.8% (41.2%–89.3%)	86.2% (83.9%–88.2%)	84.4% (67.6%–93.3%)	0.23	0.78	0.34
Moderate/severe current TS and/or ADHD ^e	35.5% (12.1%–68.8%)	69.5% (66.2%–72.5%)	69.5% (53.7%–81.8%)	0.03	> 0.99	0.05
Medication for current TS and/or ADHD ^f	27.3% (6.5%–67.1%)	67.2% (63.7%–70.5%)	82.4% (70.3%–90.2%)	0.02	< 0.01	< 0.01
Other co-occurring psychiatric disorders^g						
Ever co-occurring	62.9% (35.6%–83.9%)	66.3% (63.3%–69.3%)	95.8% (90.5%–98.2%)	0.80	< 0.01	0.01
Current co-occurring	48.3% (23.6%–73.7%)	55.9% (52.7%–59.0%)	90.4% (82.7%–94.9%)	0.58	< 0.01	< 0.01

Data from the 2014 National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome (NS-DATA) and 2011–2012 National Survey of Children's Health (NSCH). All data are parent-reported

CI 95% confidence interval; Pairwise t-tests were used to calculate differences between the three diagnostic groups

Proportions reflected weighted values for each diagnostic group

^aData for these indicators are from 2011 to 2012 National Survey of Children's Health (NSCH), all other values are from NS-DATA

^bYouth were categorized as minority status if identified as Black, Hispanic, or other (i.e., not non-Hispanic White)

^cFederal poverty level reflects the proportion of household incomes that were less than or equal to 200% federal poverty level, using multiply imputed data for approximately 5% or less for each diagnostic group

^dReflects proportion of youth in each diagnostic group with a current disorder (i.e., TS or ADHD) at the time of NS-DATA. For the TS + ADHD group, proportion reflects youth with a current disorder of TS and/or ADHD

^eReflects proportion of youth in each group with moderate/severe current disorder (TS and/or ADHD). For TS + ADHD group, the proportion reflected the worse severity of either current disorders

^fReflects proportion of youth currently taking medication(s) for their current disorders (TS and/or ADHD)

^gOther co-occurring psychiatric disorders include anxiety disorders; mood disorders; bipolar disorder; post-traumatic stress disorder; obsessive-compulsive disorder; oppositional defiant disorder; conduct disorder; autism spectrum disorder or pervasive developmental disorder; intellectual disability; language disorder; learning disorder

Of children with a current disorder, a higher proportion in the ADHD-only (69.5%, $p = 0.03$) and TS + ADHD (69.5%, $p = 0.05$) groups had a moderate/severe disorder, compared to 35.5% in the TS-only group. A significantly higher proportion of children in the TS + ADHD group

(82.4%) were currently taking medication for their disorder, relative to children with ADHD-only (67.2%, $p < 0.01$) and TS-only (27.3%, $p < 0.01$).

Table 3 Impairment across academic, interpersonal, recreational, and family financial or occupational domains among youth with Tourette syndrome (TS) and attention-deficit/hyperactivity disorder (ADHD)

	TS-only (n = 30) % (CI)	ADHD-only (n = 2899) % (CI)	TS + ADHD (n = 85) % (CI)	TS-only vs. ADHD-only <i>p</i> -value	ADHD- only vs. TS + ADHD <i>p</i> -value	TS-only vs. TS + ADHD <i>p</i> -value
Academic						
Problematic overall school performance ^{b,c}	3.7% (1.1%–12.4%)	45.7% (42.4%–48.9%)	54.0% (38.2%–69.0%)	< 0.01	0.30	< 0.01
Problematic reading performance ^{b,c}	– ^d	39.8% (36.7%–43.0%)	40.3% (26.1%–56.3%)	– ^d	0.95	– ^d
Problematic writing performance ^{b,c}	16.9% (4.4%–47.5%)	43.7% (40.5%–46.9%)	67.0% (51.5%–79.6%)	0.01	< 0.01	< 0.01
Problematic mathematics performance ^{b,c}	12.2% (4.0%–32.0%)	49.9% (46.7%–53.1%)	57.5% (40.9%–72.5%)	< 0.01	0.36	< 0.01
Problematic handwriting ^b	45.0% (21.1%–71.5%)	40.7% (37.6%–43.9%)	58.6% (44.0%–71.8%)	0.76	0.01	0.39
Interpersonal						
Problematic relationships with parents, siblings, and/or peers ^{b,c}	30.0% (9.9%–62.7%)	45.0% (41.8%–48.3%)	62.7% (46.3%–76.7%)	0.30	0.03	0.05
Youth treated differently because of tics	40.8% (17.5%–69.1%)	– ^e	57.9% (40.6%–73.4%)	– ^e	– ^e	0.30
Recreational						
Problematic participation in organized activities ^{b,c}	20.4% (6.9%–47.0%)	29.1% (26.2%–32.1%)	43.8% (29.0%–59.7%)	0.39	0.07	0.07
Family financial or occupational						
Family financial or occupational problems due to their child's disorder	34.9% (14.5%–62.8%)	41.6% (38.5%–44.9%)	56.1% (39.5%–71.4%)	0.61	0.08	0.17

Data from the 2014 National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome (NS-DATA). All data are parent-reported CI 95% confidence interval; Pairwise t-tests were used to calculate differences between the three diagnostic groups

^aProportions reflected weighted values for each diagnostic group

^b“Problematic” performance was derived from answer options of *problematic* and *somewhat problematic*

^cThese questions are from the Vanderbilt ADHD Parent Rating Scale

^dFive or fewer youth had parent reported problematic reading performance and this estimate was suppressed for confidentiality

^eThis query was only included in the TS module of NS-DATA, not the ADHD module

Other Co-occurring Psychiatric Disorders

Regarding the presence of other co-occurring psychiatric disorders, significant group differences emerged (see Table 2). By parent report, a higher proportion of children in the TS + ADHD group had ever been diagnosed with and currently had one or more other co-occurring psychiatric disorders compared to children in other diagnostic groups (TS + ADHD, 95.8% ever-diagnosed, 90.4% current co-occurring disorders; ADHD-only, 66.3% ever-diagnosed, $p < 0.01$; 55.9% current co-occurring disorders, $p < 0.01$; TS-only, 62.9% ever-diagnosed, $p = 0.01$; 48.3% current co-occurring disorders, $p < 0.01$). TS-only and ADHD-only groups did not statistically differ in the proportion of children with ever-diagnosed or current co-occurring psychiatric disorders.

Impairment Domains by Diagnostic Group

Academic Impairment

As displayed in Table 3, significantly more children in the TS + ADHD and ADHD-only groups had problematic performance in multiple academic domains. When compared to the TS-only group, significantly more children in the TS + ADHD and ADHD-only groups had problematic performance in overall school performance, and specifically in writing and mathematics (all p -values ≤ 0.01 , see Table 3). The two ADHD groups did not significantly differ from one another regarding problematic academic performance except for writing; significantly more children in the TS + ADHD group (67.0%) experienced impairment in writing compared to the ADHD-only group (43.7%, $p < 0.01$). A similar proportion of children in the

Table 4 Overall mean impairment across academic, interpersonal, recreational and family financial or occupational domains among youth with Tourette syndrome (TS) and attention-deficit/hyperactivity disorder (ADHD)

	TS-only (n = 30) M (CI)	ADHD-only (n = 2899) M (CI)	TS + ADHD (n = 85) M (CI)	TS-only vs. ADHD-only <i>p</i> -value	ADHD-only vs. TS + ADHD <i>p</i> -value	TS-only vs. TS + ADHD <i>p</i> -value
Ever-diagnosed with TS and/or ADHD	1.4 (0.7–2.1)	1.9 (1.8–2.0)	2.5 (2.0–2.9)	0.21	<0.01	0.02
Current TS and/or ADHD ^a	1.7 (0.9–2.5)	2.0 (1.9–2.1)	2.6 (2.1–3.1)	0.45	<0.01	0.05
Mild current TS and/or ADHD ^b	1.8 (0.7–2.9)	1.6 (1.5–1.8)	2.1 (1.1–3.0)	0.74	0.33	0.70
Moderate/severe current TS and/or ADHD ^c	1.5 (0.8–2.2)	2.2 (2.1–2.3)	2.9 (2.5–3.3)	0.09	<0.01	<0.01

Data from the 2014 National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome (NS-DATA). All data are parent-reported *M* mean, *CI* 95% confidence interval; Pairwise t-tests were used to calculate differences between the three diagnostic groups

^aMeans were calculated using the sum of the domains in which youth had any parent-reported impairment (possible range 0 to 4) and reflected weighted values for each diagnostic group

^bReflects mean impairment score for youth in each diagnostic group with current TS or ADHD at the time of NS-DATA. For the TS + ADHD group, mean impairment score reflects youth with a current TS and/or ADHD. Sample size of youth in each diagnostic group: TS-only n = 22; ADHD-only n = 2436; TS + ADHD n = 75

^cReflects mean impairment score of youth in each diagnostic group with current TS or ADHD categorized as mild. For TS + ADHD group, the mean impairment score reflects the worse severity rating of either current disorder. Sample size of youth in each diagnostic group: TS-only n = 15; ADHD-only n = 799; TS + ADHD n = 22

^dReflects mean impairment score of youth in each diagnostic group with current TS or ADHD categorized as moderate/severe. For TS + ADHD group, the mean impairment score reflects the worse severity rating of either current disorder (TS and/or ADHD). Sample size of youth in each diagnostic group: TS-only n = 7; ADHD-only n = 1620; TS + ADHD n = 53

ADHD-only (39.8%) and TS + ADHD groups (40.3%) experienced problematic performance in reading, see Table 3. The proportion of children who experienced problematic handwriting ranged from 40.7 to 58.6% among the three diagnostic groups, with significantly more children in the TS + ADHD group with problematic handwriting compared to children with ADHD-only ($p = 0.01$).

When comparing grades among children ever diagnosed with TS (TS-only and TS + ADHD groups) and children in the ADHD-only group, patterns were similar across the diagnostic groups. Over half of the children were reported to be A/B students (children with TS, 56.3%, CI 41.9–69.7% vs. ADHD-only 55.7%, CI 52.5–58.8%, $p = 0.93$; data not shown in table).

Interpersonal Impairment

With respect to interpersonal functioning, more children (62.7%) in the TS + ADHD group experienced interpersonal impairment (i.e., problematic relationships with parents, siblings, and/or peers) than children in the ADHD-only group (45.0%, $p = 0.03$) and children in the TS-only group (30.0%, $p = 0.05$), see Table 3. In only the NS-DATA TS module, an additional question asked if children had ever been treated differently because of

tics (e.g., being bullied, discriminated against, or treated rudely). Of all children ever diagnosed with TS (TS-only and TS + ADHD groups), 53.5%, CI 38.4–67.9%, had ever been treated differently because of tics, according to parent report (not shown in table). When parents reported on who treated the children differently because of tics ($n = 71$), they most often reported being treated differently by other children (e.g., their peers), 92.4%, CI 71.2–98.4%; teachers, 40.1%, CI 23.7–60.8%; other adults (i.e., not family), 21.9%, CI 9.9–41.7%; siblings, 6.7%, CI 1.4–25.7%; less than 5% of children with TS had been treated differently because of their tics by business owners or employees or other family members.

Recreational Impairment

No statistically significant differences were identified between diagnostic groups for children who experienced recreational impairment due to their disorder (TS-only, 20.4%; ADHD-only, 29.1%; TS + ADHD, 43.8%, p -values range from 0.07 to 0.39, see Table 3).

Family Financial or Occupational Impairment

No statistically significant differences were identified between diagnostic groups for family financial or occupational problems related to their child's TS and/or ADHD (TS-only, 34.9%; ADHD-only, 41.6%; TS + ADHD, 56.1%, p -values range from 0.08 to 0.61, see Table 3).

Overall Impairment by Diagnostic Groups

Overall, when combining the four impairment domains (academic, interpersonal, recreational, and family financial or occupational) to generate a mean impairment score due to TS and/or ADHD, the three diagnostic groups differed significantly (see Table 4). The TS + ADHD group (children ever diagnosed with both TS and ADHD) had a significantly higher mean impairment score ($M=2.5$) than either the TS-only group ($M=1.4$, $p=0.02$) or the ADHD-only group ($M=1.9$, $p\leq 0.01$), while the TS-only and ADHD-only groups were not significantly different from each other ($p=0.21$). For children with current TS and/or ADHD, the mean impairment score was greatest for children in the TS + ADHD group ($M=2.6$), followed by the ADHD-only group ($M=2.0$), and then the TS-only group ($M=1.7$). For children with a mild current disorder, there were no significant group differences in the mean impairment score (range of the means 1.6–2.1, see Table 4). However, for children with a moderate/severe current disorder (TS and/or ADHD), the TS + ADHD group ($M=2.9$) had a higher mean impairment score compared to both TS-only ($M=1.5$, $p<0.01$) and ADHD-only ($M=2.2$, $p<0.01$) groups.

Discussion

We examined functional impairment across four domains (academic, interpersonal, recreational, and family financial or occupational) among children from the U.S. who had ever been diagnosed with TS, ADHD, or both. We observed some demographic and clinical differences in the present study. Children with TS without ADHD were older and more likely to be male relative to children with ADHD without TS. A higher proportion of children with ADHD (i.e., the TS + ADHD and ADHD-only groups) were minorities relative to children with TS alone. Previous studies have highlighted the issue of undiagnosed or underreported cases of TS among children in racial or ethnic minorities [4, 28, 29]. While this was not an aim of our analysis, the higher proportion of children with ADHD in racial or ethnic minorities may provide an opportunity to identify children with TS indirectly via a commonly co-occurring disorder, ADHD. A higher proportion of children with current ADHD (with or without TS) used medication for TS or ADHD and had

moderate/severe symptoms relative to children with TS alone. Co-occurring psychiatric disorders were also more common among children in the ADHD groups compared to children in the TS-only group. This pattern of clinical characteristics is similar to previous findings for medication status [18] and co-occurring psychiatric disorders or symptoms [18, 19, 30], which provide context for interpretation of results related to impairment.

This analysis generally revealed that, based on parent report, the proportion of children with functional impairment was greatest among the TS + ADHD group, followed by the ADHD-only and TS-only groups. This pattern was observed for most academic measures (i.e., overall school performance, writing, mathematics, and handwriting). Impairment experienced by children with ADHD, regardless of the presence or absence of co-occurring TS, aligns with prior research indicating greater academic difficulties [15] and disruptive behavior [18] in children with ADHD, with or without TS, relative to TS alone. Writing and handwriting performance findings showed increased impairment in children with co-occurring TS and ADHD relative to ADHD alone. Difficulties with written expression and handwriting are common in both children with TS [25] and children with ADHD [26]. However, TS may directly disrupt the flow of hand and arm movements depending on tic expression [31], increasing the risk of handwriting difficulties among children with TS compared to children without TS. From an assessment perspective, the American Academy of Pediatrics ADHD practice guidelines suggest that clinicians screen for impairment in school performance among children with ADHD [12]. In addition, these data support TS treatment guidelines stating that children with co-occurring TS and ADHD who experience academic impairment may benefit from interventions directed toward both disorders [11, 32]. For example, ADHD symptoms may be addressed with educational strategies, medication management (particularly $\alpha 2$ adrenergic agonists) and parent behavior training, while TS symptoms may be addressed with interventions focused on tics (i.e., habit reversal training/comprehensive behavioral intervention for tics) [11, 32].

With regard to problematic interpersonal relationships with parents, siblings and/or peers, more children with TS and co-occurring ADHD experienced interpersonal problems relative to children in the ADHD-only and TS-only groups, and just over half of the children ever diagnosed with TS (TS-only and TS + ADHD groups) were treated differently because of their tics. Prior findings examining differences in social impairment have been mixed when comparing interpersonal or social problems among children with TS and/or ADHD. Some studies have shown increased social problems in individuals with TS and co-occurring ADHD relative to TS alone [19, 28]. Consistent with our findings, one study showed increased social impairment in

children with TS and co-occurring ADHD relative to those with ADHD alone [19]. Yet other studies have shown no significant differences in social problems between children with ADHD and children with TS and co-occurring ADHD [18, 20, 33] or even reduced social impairment in children with TS and co-occurring ADHD relative to those with ADHD alone [20]. The differences in results across studies may be in part due to variation in demographic and clinical characteristics (e.g., age, disorder severity, medication status, comorbidity) and definitions of diagnostic groups across samples. In our study, interpersonal problems may have also been influenced by co-occurring disorders beyond ADHD and TS (e.g., autism spectrum disorder, conduct disorder). In general, recommendations by the American Academy of Child and Adolescent Psychiatry Committee for care of children with tic disorders include assessment and monitoring of social relationships to determine children's need for interventions to improve social skills [32]. Also, considering how each disorder might impact children's functioning in multiple settings and with different types of people such as family members, peers, and teachers may inform treatment decisions [12, 32].

Within recreational and family financial or occupational domains, there were no significant differences in impairment across diagnostic groups. Despite this, findings highlight that a large proportion of children with TS and/or ADHD experience recreational impairment, ranging from 20.4 to 43.8%. Furthermore, an even larger proportion of families experience financial or occupational impairment due to their child's TS and/or ADHD (34.9% to 56.1%). However, point estimates of impairment were higher among children with co-occurring TS and ADHD as compared to children with either ADHD or TS. Recreational activities comprise a considerable portion of children's daily lives (e.g., playing sports, participating in clubs) and contribute to positive and healthy development [34]. Therefore, fostering engagement in recreational activities may be included in the management of TS or ADHD, regardless of whether the disorders co-occur [35]. Similarly, families of children with TS and/or ADHD often experience problems in financial or occupational domains [29, 36–38]. For example, parents may need to miss work to take their child to appointments and the costs of health care services may result in financial strain. In addition, the presence of one or more co-occurring disorders in either of TS or ADHD increases health care needs [39, 40]. Our findings underscore the need to consider the family context in treatment [12, 32]. Future research could examine the degree to which early intervention and increased access to services may also help to address family financial or occupational challenges.

We also calculated and explored differences in the mean impairment score (i.e., the mean number of domains in which children experienced impairment). Our findings

revealed that children ever diagnosed with both TS and ADHD (TS + ADHD group) had a significantly higher mean impairment score relative to children ever diagnosed with TS or ADHD (i.e., TS-only and ADHD-only groups), with no significant differences between the two groups of children with either disorder alone. For children with a current disorder, children with TS and co-occurring ADHD had a higher mean impairment score relative to children with ADHD alone and children with TS alone. Taken together, these results suggest that there may be an exacerbated effect of TS and ADHD, whereby the co-occurrence of these disorders may contribute more to functional impairment than either disorder alone. Further, we found differences in the mean impairment score for children with moderate/severe TS and/or ADHD. Specifically, children in the TS + ADHD group with moderate/severe disorders had a significantly higher mean impairment score relative to children in the TS-only and ADHD-only groups with a moderate/severe disorder. We did not find significant differences among the three groups for children with mild symptoms. These findings emphasize the added value of considering symptom severity in understanding the unique contributions of each of these disorders to functional impairment.

Broadly, whether a child presents with TS, ADHD, or a combination of both disorders, conducting evidence-based assessments to evaluate symptom severity can help determine the degree to which TS and/or ADHD symptoms are interfering with functioning (see McGuire and colleagues [41] for a review on evidence-based assessments of tics and Pelham et al. [42] for a review of evidence-based assessment of ADHD symptoms). Evidence-based assessment can incorporate scales that evaluate impairment associated with each disorder separately. For instance, the Child Tourette Syndrome Impairment Scale [43–45] could help reveal the degree to which specific difficulties are related to TS or ADHD symptoms. Comprehensive Behavioral Intervention for Tics (CBIT) is a recommended, first-line intervention targeting tic symptoms [32]. Additional TS treatments address impairment and distress associated with tics and any co-occurring disorders. For example, Living with Tics [46, 47] is a modularized intervention developed to treat tic-related psychosocial problems in children through combined habit reversal training and behavioral strategies (i.e., problem solving, cognitive restructuring, parent training). This type of modularized intervention is in accordance with guidelines stating treatment for tic disorders should address impairment and emotional distress related to tics and any co-occurring disorders [12, 32]. Similarly, several evidence-based interventions (e.g., stimulant medication, school-based intervention, summer treatment program, parent training) exist for children with ADHD to manage core symptoms and related functional impairment [12, 48].

Our study design had numerous strengths including initial population-based sampling in NSCH, ethnographic and geographic diversity, and numerous indicators of impairment across multiple domains. We were also able to examine impairment across a wide age range of 4–17 years. However, potential limitations exist. First, we had a relatively small sample size of children in the TS-only group, which may have limited our power to detect small to moderate differences among diagnostic groups in some impairment domains, and did not allow for analyses of impairment based on diagnostic group or demographic characteristics (e.g., age). Data for children with TS were treated as a convenience sample for the analyses. However, the smaller sample size among the TS-only group is consistent with the relatively low prevalence of TS in the general population [6]. Second, because the diagnostic group were determined by the child having ever been diagnosed with the disorder (i.e., TS and/ ADHD), children in the diagnostic groups were mixed with respect to the presence of current TS and/or ADHD. This means that children in the TS + ADHD group included four subpopulations (i.e., ever [but not current] TS, ever [but not current] ADHD; current TS, ever [but not current] ADHD; ever [but not current] TS, current ADHD; and current TS, current ADHD). Therefore, these different subgroups within the diagnostic groups may have different clinical profiles. Third, while our analysis includes sample weights, results are not representative of the U.S. population of children with TS or ADHD. Fourth, the results reported here are based on parent-reported data that have not been validated against medical or educational records and may be subject to recall or social desirability bias. Fifth the NS-DATA sample did not include a control sample of children without TS or ADHD. Sixth, impairment may have been influenced by differences in demographic and clinical characteristics along with presence of co-occurring disorders beyond ADHD and TS. Future research accounting for these differences may be beneficial. Seventh, these analyses were not adjusted for multiple comparisons and therefore may be susceptible to Type I error (i.e., incorrectly identifying an association as significant when there is not a difference between the two groups); future studies evaluating these relationships in different datasets may help further establish the evidence for which associations are consistently observed. Finally, the questions regarding academic (except for handwriting), interpersonal, and recreational domains are part of a validated measure for ADHD impairment [24], and may not be optimal for detecting impairment due to TS. In addition, when responding to impairment questions in the academic, interpersonal, and recreational domains, parents were instructed to think about a time in the past six months when their child was not taking medication for TS, ADHD, or other difficulties with their emotions, concentration, or behavior. Therefore, the survey wording may limit generalizability of the findings for children taking medication, as some children experience reduced impairment while taking medication.

Summary

The present study examined impairment in academic, interpersonal, recreational, and family financial or occupational domains in 3014 children aged 4–17 years classified into three diagnostic groups, including children ever diagnosed with TS without ADHD ($n = 30$), ADHD without TS ($n = 2899$), and TS and ADHD ($n = 85$). Parents completed the 2014 National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome. Analyses included measures of child demographics, TS and ADHD severity, TS and ADHD medication status, co-occurring psychiatric disorders, problematic school performance (i.e. overall, reading, writing, mathematics, handwriting), interpersonal relationships, and recreational participation, in addition to family-related financial or occupational problems. This descriptive analysis expands previous research using a nonclinical, nationally drawn sample of children with TS and/or ADHD to reveal a general pattern around disorder related impairment. More children with co-occurring TS and ADHD may experience impairment in multiple domains, followed by children with ADHD without TS, and finally children with TS without ADHD. Findings support previous studies about the use of evidence-based clinical assessments that may distinguish disorder-specific contributions to impairment across multiple domains [43–45]. Findings draw attention to the potential need for tailored interventions to address specific domains of impairment associated with TS and ADHD.

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Compliance with Ethical Standards

Conflict of interest Emily J. Ricketts, PhD has received research support from the National Institute of Mental Health (NIMH), the Tourette Association of America (TAA), and the BFRB Precision Medicine Initiative granted by the TLC Foundation for Body-Focused Repetitive Behaviors. She has received honoraria from the TAA, and serves on their Diversity Committee. Sara Beth Wolicki, MPH, CPH and Melissa L. Danielson declare that they have no conflict of interest. Michelle Rozenman, PhD has received research support from the NIMH, Uni-

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Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animal participants performed by any authors. Informed consent was obtained from all individual participants included in the study.

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