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Treatment Use Among Children with Tourette Syndrome Living in The United States, 2014

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Abstract

Treatment of Tourette syndrome (TS) can be complicated by changes over time in tic expression, severity, and co-occurring disorders. Using the 2014 National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome, this study provides descriptive estimates of the use of behavioral interventions and medication among children living with TS. Parent-reported data on 115 children aged 5–17 years ever diagnosed with TS were analyzed to provide descriptive, unweighted results. Overall, 77.4% of children had current or past use of any TS treatment; 59.1% ever used behavioral interventions and 56.1% had ever taken TS medication. Children with "moderate" or "severe" versus "mild" TS, ≥1 co-occurring disorders, and tics that interfered with functioning were significantly more likely to have used one or more TS treatments. Side effects were reported for 84.4% of children who took TS medication. Most parents of children with current TS (87.2%) were satisfied with the management of their child's TS. However, parents of children with "moderate" or "severe" current TS were significantly more dissatisfied compared to parents of children with "mild" TS. Findings from this study could be used to inform efforts to support children living with TS and their families.

Keywords

Tic Disorders, Neurodevelopmental disorders, Movement disorders, Adolescents, Public health, Medication, Behavior therapy

1. Introduction

Tourette syndrome (TS) is a tic disorder characterized by at least one vocal and multiple motor tics that begin during childhood and persist for more than one year (American Psychiatric Association, 2013). Tics are usually first noticed when the child is between the ages of 4 to 6 (Hirschtritt et al., 2015; Khalifa and Von Knorring, 2005; Wolicki et al., 2019), and TS is typically diagnosed around 8 years (Bitsko et al., 2014). The frequency and intensity of tics often waxes and wanes (American Psychiatric Association, 2013), but severity usually increases until the child is between the ages of 10 to 12 years (Bloch and Leckman, 2009; Wolicki et al., 2019), and declines in adolescence (Groth et al., 2017). Co-occurring mental, behavioral, and developmental disorders are common for children with TS, with 80% to 90% of children with TS having one or more co-occurring disorders (Bitsko et al., 2014; Hirschtritt et al., 2015; Wolicki et al., 2019). The most common ever-diagnosed co-occurring disorders are attention-deficit/hyperactivity disorder (ADHD, ranging from 32.6%–62.8%), anxiety disorders (36.0%–44.9%), and 42.3%–66.1% with obsessive-compulsive disorder (Hirschtritt et al., 2015; O'Hare et al., 2016; Wolicki et al., 2019). These characteristics of TS (i.e., changes in tic expression, severity, and impairment from co-occurring disorders over time) pose challenges for the treatment and

management of TS (Murphy et al., 2013; Pringsheim et al., 2019b). Better understanding of the use of TS treatments may inform efforts to support children living with TS and their families.

The American Academy of Child and Adolescent Psychiatry (Murphy et al., 2013) and American Academy of Neurology (Pringsheim et al., 2019b) along with international professional societies from Canada (Pringsheim et al., 2012; Steeves et al., 2012) and Europe (Roessner et al., 2011; Verdellen et al., 2011) have issued clinical practice guidelines for the treatment of TS and other persistent tic disorders. Evidence-based behavioral interventions for TS include psychoeducation on tics such as providing individuals, parents, teachers, and peers with factual information about TS (Murphy et al., 2013; Nussey et al., 2013; Verdellen et al., 2011), school accommodations (Murphy et al., 2013), habit reversal therapy (Murphy et al., 2013; Steeves et al., 2012; Verdellen et al., 2011), and Comprehensive Behavioral Intervention for Tics (CBIT, Pringsheim et al., 2019a), which is structured behavioral therapy for tics. The United States (U.S.) Food and Drug Administration (FDA) has approved three medications for the treatment of TS (haloperidol, pimozide, and aripiprazole).[1] Off-label use of other medications not specifically approved by the FDA is common for the treatment of TS (Pringsheim et al., 2019a; Pringsheim et al., 2019b; Quezada and Coffman, 2018).

Most research regarding children with TS includes samples recruited from specialty clinics for TS and related disorders. Often, these samples are limited to a single geographic area or region and usually include children with more severe symptoms (Bitsko et al., 2014; Knight et al., 2012). The National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome (NS-DATA) was conducted to learn more about the experiences of children and families living with ADHD and/or TS in the U.S. (Zablotsky et al., 2019). Although NS-DATA was used for this analysis, the results presented in this study are not representative nor generalizable to all children with TS within the U.S. The purpose of this study is to provide a descriptive analysis of parent-reported treatment for TS from a geographically diverse sample of children ever diagnosed with TS, with the goal of better understanding characteristics of the child and family that are related to TS treatment received.

2. Methods

NS-DATA is a follow-up survey to the 2011–2012 National Survey of Children's Health (NSCH), a nationally representative parent-report survey conducted within the U.S. that examined the physical and emotional health of children (Bramlett et al., 2017; Zablotsky et al., 2019). NS-DATA was conducted in 2014 by the U.S. Centers for Disease Control and Prevention's National Center for Health Statistics (NCHS), with support from the National Center on Birth Defects and Developmental Disabilities. Children under 18 years of age whose parent had reported that their child was ever diagnosed with ADHD or TS at the time of the 2011–2012 NSCH were eligible for NS-DATA. The responding parent or guardian (hereinafter referred to as “parent”) had to report that the child had ever been diagnosed with TS or ADHD at the time of NS-DATA. The NS-DATA responding parent did not need to be the 2011–2012 NSCH respondent, but had to live in the same household with the child at the time of the survey. NS-DATA had a TS module and an ADHD module; parents of children who had ever received both diagnoses were invited to complete both modules. Our study examined only data from the TS module, which took an average of 32 minutes to complete and included children from 44 of the 50 U.S. states (Zablotsky et al., 2019).[2] In total, interviews were completed by parents of 115 children ever diagnosed with TS, of which 78 had children who had ever received a TS diagnosis at the

time of the 2011–2012 NSCH, according to parent report. The remaining 37 children had a parent who reported only an ADHD diagnosis on the 2011–2012 NSCH but reported their child had been diagnosed with TS when recontacted for NS-DATA (for additional details, see Wolicki et al., 2019).

Current or past use of specified TS treatments was the main outcome of interest for these analyses. We also present TS treatment use and its association with covariates such as current TS status, ever-diagnosed and current co-occurring disorders, as well as worst and current TS severity. We analyzed co-occurring disorders as a group rather than by individual disorder (i.e., presence or absence of any co-occurring disorders and mean number of co-occurring disorders). A list of the 15 co-occurring disorders included in the summary variable can be found in Table 1. For each co-occurring disorder, parents were asked if the child had ever been diagnosed with the specified disorder, and if yes, asked if the child currently had that disorder.

Table 1. Summary variables and NS-DATA verbatim survey questions for analyses related to co-occurring disorders, treatments for Tourette syndrome (TS), and adverse side effects related to medication taken for TS

Content-similar summary variables	**NS-DATA verbatim survey questions included in each group**
Ever-Diagnosed or Current Co-occurring Disorders *n* = 115	Has a doctor or other health care provider ever told you that your child had:*
	Attention-deficit/hyperactivity disorder (ADHD) or ADD?
	Anxiety disorder, such as generalized anxiety disorder, panic disorder, or a phobia?
	Autism Spectrum Disorder or Pervasive Developmental Disorder?
	Bipolar disorder?
	Conduct disorder?
	Eating disorder, such as anorexia or bulimia?
	Intellectual disability?
	Intermittent explosive disorder?
	Language disorder?
	Learning disorder?
	Mood disorder, such as depression, or major depressive disorder?
	Obsessive-compulsive disorder or OCD?
	Oppositional defiant disorder or ODD?
	Post-traumatic stress disorder or PTSD?
	Substance use disorder?
Current or Past Use of Specified Treatments for TS *n* = 115	Has your child ever received:*
	Behavior treatment based outside of school for Tourette syndrome?†
	Comprehensive Behavioral Intervention for Tics (CBIT) or habit reversal therapy for Tourette syndrome?†
	School-based behavioral treatment, support, or accommodation for Tourette syndrome?†
	Any other treatment for Tourette syndrome?†
	Has your child ever taken medication for Tourette syndrome?
Side Effects Experienced Due to TS Medication *n* = 64	Please think about side effects from medications that your child has ever taken for Tourette syndrome, even if your child does not currently take them now. Due to Tourette syndrome medication, has your child ever experienced:

	Being mentally slowed down or sluggish, for example, thinking slowly or being less attentive?
	Being physically slowed down or sluggish, such as moving slowly?
	Body twisting, squirming, or other new body movements, not including tics?
	Sleep problems or insomnia, such as trouble getting to sleep or staying asleep?
	Weight gain?
	Any other side effects?

All survey questions are verbatim from the 2014 National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome (NS-DATA).

∗ If the responding parent answered "yes," a follow-up question asking if their child currently had specified co-occurring disorder or was currently receiving specified treatment was also asked. Both the current co-occurring disorders (presence or absence) and current use of treatment for TS (presence or absence of use) were also analyzed as a group.

† These four survey questions about use of behavioral interventions for TS were also grouped together to create a dichotomous (presence or absence of use) for current and past use of any specified behavioral interventions as treatment for TS.

Table 1 also presents the specified TS treatments reported on in NS-DATA, four of which were behavioral interventions, one of which was medication use. We created dichotomous summary variables (presence or absence) for current or past use of TS treatment including any of the four behavioral interventions and/or medication for TS and use of any behavioral interventions for TS (Table 1). To determine severity of TS, parents were asked to describe their child's current TS, as well as when the TS symptoms were at their worst, as “mild,” “moderate,” or “severe.”

Some questions in NS-DATA were only asked for subgroups of children, which are detailed below. Specific questions about medication use for TS (e.g., child's age when first started taking TS medication, a list of current types of medication taken for TS, and side effects ever experienced due to TS medication, were side effects troublesome enough to stop taking the medication[3]) were only asked of parents who had reported “yes” to their child ever taking medication for TS (*n* = 64). Parents of children who ever took TS medication were asked to report if their child had ever experienced six specified adverse side effects (see Table 1 for verbatim survey questions) due to TS medication in general (not specific to a type or dosage of medication for TS). We present analyses of the six side effects individually as well as the mean number reported per child. Parents of children with current TS at the time of NS-DATA (*n* = 82) were asked about the accessibility of TS treatment and their overall level of satisfaction with the treatment and management of their child's TS. Parents who reported that their child had past TS (i.e., responded “no” to the survey question “does your child currently have TS?”, *n* = 29[4]) were asked if they believed treatment helped the tics go away or if the tics seemed to go away on their own. The responding parent was also asked to report the child's age and health insurance status at the time of NS-DATA. Other demographic information about the child and the household was imported from the 2011–2012 NSCH. Multiple imputation was used to estimate the federal poverty level for participants with missing data used to construct that indicator (less than 10% of the sample) from the 2011–2012 NSCH; imputations are calculated by NCHS and provided in the dataset (Bramlett et al., 2017).

Due to small sample size and confidentiality concerns, we collapsed most response options into fewer categories for our analysis. Although the sample for NS-DATA was drawn from across the U.S., it cannot be assumed to be nationally representative of all children with TS. Therefore, we analyzed this small probability sample as a convenience sample. We present an unweighted, descriptive analysis of this cross-sectional, structured interview survey including percentages, means, and standard deviations (SD), when appropriate. We used Fisher's exact test and t-tests for all statistical comparisons. Outcomes with small sample sizes (i.e., fewer than 5) were either collapsed into other categories or are not presented to protect confidentiality of the respondent families. Any responses of “don't know” or refusal to answer the question were categorized as missing throughout our analysis. All analyses were conducted using SAS version 9.4 (RTI International; Cary, NC).

3. Results

3.1. Sample characteristics, TS severity, and co-occurring disorders

As previously reported (Wolicki et al., 2019), most children included in the TS module of NS-DATA were male (82.6%), non-Hispanic white (70.4%), and had at least one parent who attended some college or

more (74.8%). The mean child age at the time of NS-DATA was 13.2 years (SD = 2.9). More than half of children (59.7%) were living in households with income greater than 200% of the federal poverty level.

According to parent report, most children (92.9%) had continuous insurance coverage over the past year. Among children with continuous insurance coverage (*n* = 105), 52.9% had private insurance and 47.1% had public insurance. When "[TS] symptoms were at their worst," TS was most often rated by parents as "moderate" or "severe" (80.9%) as opposed to "mild" (19.1%). Most children (89.6%) had one or more ever-diagnosed co-occurring disorders and 85.2% of children currently had one or more co-occurring disorders at the time of NS-DATA.

3.2. Current or past use of any specified behavioral interventions and medication as treatments for TS

Of the 115 children in the sample, 77.4% had ever received treatment for TS; 37.4% of children had current or past use of both behavioral interventions and medication, 21.7% had current or past use of only behavioral interventions, and 18.3% had current or past use of only medication (Table 2). More than half of the children (59.1%) had ever received one or more behavioral interventions as treatment for TS and 35.7% had ever received two or more behavioral interventions for TS (Table 2). More than four out of ten children (42.1%) had ever received "school-based behavioral treatment, support, or accommodation"; 35.1% had received "behavioral treatment based outside of school"; 17.1% received "Comprehensive Behavioral Intervention for Tics (CBIT) or habit reversal therapy." In response to an open-ended question, parents reported that about one in five children (18.6%) had ever received another non-pharmacological treatment for TS including psychotherapy, counseling, holistic treatments, and dietary changes. Over half, 62.6%, of children had a "formal educational plan" (i.e., Individualized Education Program or IEP or a 504 accommodation plan); however, this survey question did not specify whether the educational plan was related to TS. A little over half (56.1%) of children had ever taken medication for TS (Table 2). The mean age when children started taking medication for TS was 8.3 years (SD = 2.9). No significant difference in age at the time of diagnosis was found between children who had ever taken medication for TS (mean = 7.6 years, SD = 2.6) and those who had not (mean = 7.8 years, SD = 2.9, $p = 0.62$).

Table 2. Use of Specified Treatments for Tourette syndrome (TS), *n* = 115

Current or Past Use of Any TS Treatment*	**%†**	***n***
Never Received Treatment for TS	22.6	26
Received Both Specified Behavioral Interventions and Medication for TS	37.4	43
Received Only Specified Behavioral Interventions for TS	21.7	25
Received Only Medication for TS	18.3	21
Current or Past Use of Specified Behavioral Interventions		
1 or more Behavioral Treatments for TS§	59.1	68
2 or more Behavioral Treatments for TS	35.7	41
Current or Past Use of Behavioral Interventions by Type§		
School-based treatment, support, accommodation¶	42.1	48
Behavior treatment based outside of school¶	35.1	40
Comprehensive Behavioral Intervention for Tics (CBIT) or habit reversal therapy¶	17.1	19
Any other non-pharmacological TS treatment¶**	18.6	21
Current Use of School Support (IEP or 504 plan)¶¶	62.6	72
Current or Past Use of Medication for TS	56.1	64

Data from the 2014 National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome (NS-DATA).

∗ Participants were only classified as missing if they were missing answers to all questions used to create this derived variable.

† All percentages are unweighted and not nationally representative.

§ The category of "1 or more Behavioral Treatments for TS" includes children in the "2 or more Behavioral Treatments for TS."

¶ These values are not mutually exclusive.

∗∗ Any other non-pharmacological TS treatment included psychotherapy, counseling, holistic treatments, and dietary changes.

†† Survey question was not specific to TS: "Does the child currently have a formal educational plan, such as an Individualized Education Program, also called an IEP or a 504 plan?".

3.3. Side effects due to medication for TS

Parent-reported side effects due to TS medication were common. Of the children who ever took medication as treatment for TS (n = 64), parents reported that 84.4% of children had one or more side effects; 50.0% had 3 or more side effects due to their TS medication. Specific side effects included "being mentally slowed down or sluggish" (58.1%), "being physically slowed down or sluggish" (48.4%), "experiencing sleep problems" (42.0%), "experiencing weight gain" (40.6%), and "experiencing body twisting, squirming, or other new body movements not including tics" (27.0%). Approximately four in ten children (42.2%) had other side effects, such as headaches, mood and behavioral changes, weight loss, seizures, and loss of appetite. The mean number of side effects per child was 2.5 (SD = 1.6). No difference was found in the mean number of side effects by worst TS severity ("mild," mean = 2.6, SD = 1.7 vs. "moderate" or "severe," mean = 2.5, SD = 1.6, p = 0.95).

Of the children who ever took medication as treatment for TS (n = 64), half (50.0%) had side effects that were "troublesome enough to stop taking the [TS] medication," according to parent report. Among children with current TS, who had ever stopped a TS medication due to side effects (n = 26), 42.3% were currently taking medication for TS at the time of NS-DATA. Children who stopped taking a medication due to side effects had significantly more side effects reported by parents, compared to children who did not stop taking medication due to side effects (mean = 3.3, SD = 1.3 vs. mean = 1.8, SD = 1.6, $p < 0.01$).

3.4. Clinical characteristics of children with current or past use of TS treatment

Shown in Fig. 1, the percent of children with any current or past use of TS treatment (specified behavioral interventions and/or use of TS medication) was significantly higher among children with "moderate" or "severe" worst TS when compared to children with "mild" worst TS (84.9% vs. 45.5%, $p < 0.01$). Fig. 1 shows the percent of children with current or past use of TS treatment was significantly higher among those ever diagnosed with one or more co-occurring disorders compared to children without a specified co-occurring disorder (80.6% vs. 50.0%, p = 0.03). Among children with tics that interfered with functioning (i.e., their "ability to do things that other children could do"), the percent of children with current or past use of TS treatment was significantly higher than children with tics that did not interfere with their functioning (87.3% vs. 66.0%, p = 0.01). No significant difference was documented in current or past use of TS treatment by children with current or past TS (p = 0.60) or "tic noticeability to strangers" ($p > 0.99$).

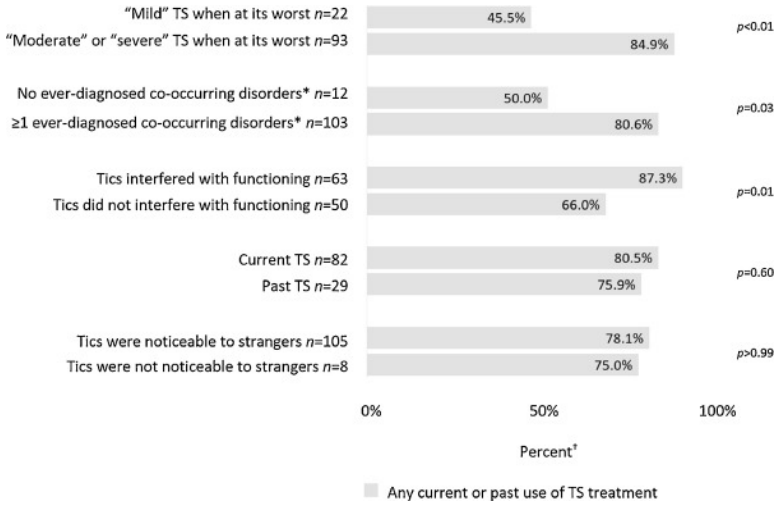

Fig. 1. Percent of Children with Tourette Syndrome (TS) Who Had Any Current or Past Use of Behavioral Interventions or Medication as Treatment for TS by Clinical Characteristics, by Parent-report, *n* = 115
Data from the 2014 National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome (NS-DATA).
† All percentages are unweighted and not nationally representative.
* Co-occurring disorders included: Attention-deficit/hyperactivity disorder, anxiety disorder, autism spectrum disorder or pervasive developmental disorder, bipolar disorder, conduct disorder, eating disorder, intellectual disability, intermittent explosive disorder, language disorder, learning disorder, mood disorder, obsessive-compulsive disorder, oppositional defiant disorder, post-traumatic stress disorder, substance use disorder.

Examination of the same factors by current or past use of specified TS behavioral interventions or TS medication revealed similar patterns of significant differences as those for current or past use of TS treatment. Specifically, compared to children with "mild" worst TS, children with "moderate" or "severe" worst TS were more likely to have current or past use of behavioral interventions (64.5% vs. 36.4%, $p = 0.03$) and current or past use of TS medication (62.0% vs. 31.8%, $p = 0.01$). Children with tics that interfered with functioning (according to parent report) were more likely to have current or past use of behavioral interventions for TS ($p = 0.06$) and medication use ($p = 0.02$) compared to children whose tics did not interfere with functioning. Also, children with current or past use of medication for TS had significantly more ever-diagnosed co-occurring disorders than children who never took TS medication (mean 4.6 disorders, SD = 3.0, vs. 3.3 disorders, SD = 3.2, $p = 0.03$). No significant differences in the current or past use of behavioral interventions for TS or use of TS medication were found by child's sex (male vs. female), age (5–12 years vs. 13–17 years), race/ethnicity (non-Hispanic white vs. other), or insurance type (private vs. public insurance).

3.5. Current use of TS treatment among children with current TS

Nearly three-quarters (73.9%) of children had current TS at the time of NS-DATA. The mean age of children with current TS (13.3 years, SD = 2.8) was similar to those with past TS (13.4 years, SD = 3.0). Of children with current TS (*n* = 82), 63.4% currently had "mild" TS and 36.6% currently had "moderate" or "severe" TS. Fig. 2 compares treatment types, co-occurring disorders, and treatment access and satisfaction for children with "mild" current TS to "moderate" or "severe" current TS. A higher percent of children with "moderate" or "severe" current TS were currently receiving one or more behavioral interventions for TS ($p < 0.01$) and were currently taking medication for TS ($p = 0.10$)

than children with “mild” current TS. Regardless of the level of current TS severity, a similar percent of children currently had one or more co-occurring disorders (p = 0.74).

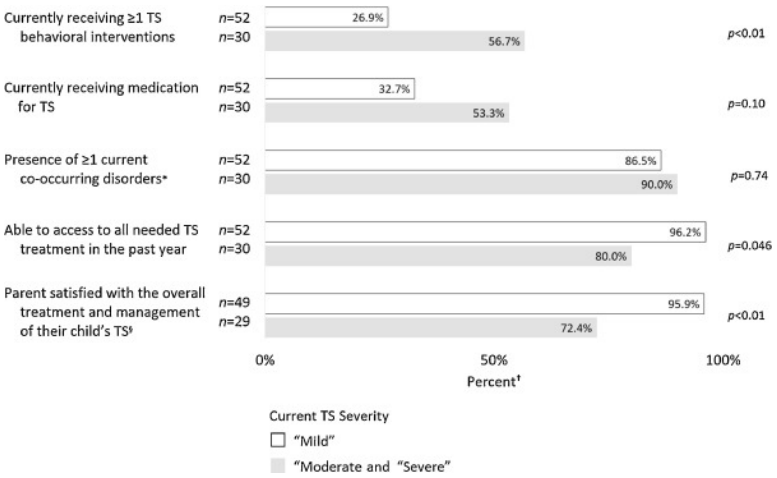

Fig. 2. Clinical Characteristics by Current Severity of Tourette Syndrome (TS) Among Children with “Mild” or “Moderate” and ”Severe” Current TS, by Parent-report, n = 82
Data from the 2014 National Survey of the Diagnosis and Treatment of ADHD and Tourette Syndrome (NS-DATA).
† All percentages are unweighted and are not nationally representative.
* Co-occurring disorders included: Attention-deficit/hyperactivity disorder, anxiety disorder, autism spectrum disorder or pervasive developmental disorder, bipolar disorder, conduct disorder, eating disorder, intellectual disability, intermittent explosive disorder, language disorder, learning disorder, mood disorder, obsessive-compulsive disorder, oppositional defiant disorder, post-traumatic stress disorder, substance use disorder.
§ Parent selected “very satisfied” or “somewhat satisfied” regarding the overall treatment and management of their child's TS.

Among children with current TS (n = 82), 37.8% were currently receiving one or more behavioral interventions for TS; 40.2% were currently taking medication for TS. Among children currently taking medication for TS, 12 children were currently taking an antipsychotic medication, including the three medications approved by the FDA for the treatment of TS (aripiprazole, haloperidol, or pimozide), as well as two medications not approved by the FDA for TS (quetiapine and risperidone). Fewer than half of children taking an antipsychotic medication were taking one of the FDA-approved medications for TS. Other medications children were currently taking as treatment for their TS at the time of NS-DATA include alpha adrenergic receptor agonist agents (clonidine or guanfacine, n = 13); anticonvulsants/mood stabilizers/antidepressants[5] (n = 9); and central nervous system stimulants (n = 7). The medications listed are not mutually exclusive; each child could have been taking multiple medications for TS from one or more classes. For the 33 children currently taking medication for TS, a parent was responsible for making sure their child took their TS medication for 91.0% of children. Parents reported that 87.9% of children (n = 29) currently taking a TS medication did not resist taking their TS medication in the past year.

3.6. Parent level of TS treatment satisfaction among children with current TS

Overall, most parents of children with current TS (90.2%) reported that in the past 12 months they were able to get the TS treatment their child needed. Similarly, approximately seven out of eight

parents (87.2%) reported being “very satisfied” or “somewhat satisfied” with the overall treatment and management of their child's TS. No significant differences were found in satisfaction based on the current or past use of TS treatments by type (TS treatment, any specified behavioral interventions for TS, or any TS medication, $p > 0.99$ for all comparisons) or the child's age at the time of NS-DATA (5–12 years of age vs. 13–17 years, $p = 0.49$). When compared by current severity, parents of children with “mild” current TS were more likely to report being able to access needed TS treatments ($p = 0.046$) and being satisfied (i.e., “very satisfied” or “somewhat satisfied”) with the treatment and management of their child's TS ($p < 0.01$) compared to parents of children with “moderate” or “severe” current TS (also in Fig. 2).

3.7. Parent perceptions for children with past TS

In exploratory analyses, parents of children with past TS at the time of NS-DATA ($n = 29$) were almost equally divided on their perception of what seemed to help their child's “tics go away.” Fifteen parents (53.6%) reported that “treatment helped the tics go away,” 13 parents (46.4%) reported “the tics seemed to go away on their own,” and 1 parent answered “don't know.”

4. Discussion

This study yielded several findings. First, over three-fourths of children (77.4%) had used at least one TS treatment (i.e., specified behavioral interventions and/or medication). Although nearly 1 in 4 children had not received treatment for TS, providers might simply be following the American Academy of Neurology's practice guideline recommendations of “watchful waiting” as an acceptable form of treatment for children who are not exhibiting impairment due to tics (Pringsheim et al., 2019b). Over 60% of children with TS had current formal education plans (e.g., IEP or 504 plan). While NS-DATA did not ask parents about whether the educational plan was specifically due to TS, an IEP or 504 plan may be another way to address TS impairment (Murphy et al., 2013). Educating teachers, school administrators, and peers and classmates about TS has been shown to encourage positive attitudes toward people with TS, and may in turn, improve functioning among people with TS (Nussey et al., 2013; Pringsheim et al., 2019b; Tourette Association of America, 2020).

Second, as is common for children with TS (Bitsko et al., 2014; Hirschtritt et al., 2015; Wolicki et al., 2019), a majority of children in the sample had one or more co-occurring disorders and the mean number of co-occurring disorders was significantly higher among children who had ever received TS treatment compared to children who had never received treatment. A recent survey of parents conducted by the Tourette Association of America reported that for 42% of children with TS, dealing with co-occurring disorders was the greatest challenge for TS management (Malaty et al., 2019). Other research has shown that prioritizing treatment for an impairing co-occurring disorder may indirectly improve the child's management of TS as well (Eddy et al., 2011; Roessner et al., 2011). Practice guidelines recommend an assessment for common co-occurring disorders when evaluating children for TS (Murphy et al., 2013; Pringsheim et al., 2019b) due to the high prevalence of co-occurring disorders, which has implications for TS treatment.

Third, similar to previous studies (Leucht et al., 2013; Pringsheim et al., 2011), we found that side effects due to medication for TS were common. Parents in our study reported that among children who had ever taken medication for TS, almost 85% had one or more side effects and half had stopped

taking a TS medication due to troublesome side effects. Discontinuing or changing medication over the course of TS is common; reasons for switching medication may be due to loss of benefit, side effects, or impairment from co-occurring disorders (Carulla-Roig et al., 2018; Farag et al., 2015). The American Academy of Neurology and American Academy of Child and Adolescent Psychiatry practice guidelines for tic disorders recommend that providers share information to patients about potential side effects, and periodically re-evaluate treatment decisions based on current treatment effectiveness, tic severity, and presence of side effects (Murphy et al., 2013; Pringsheim et al., 2019b).

Fourth, parents of children with current TS reported high levels of access to needed treatment and satisfaction with the overall treatment and management of their child's TS. These findings align with recent research from the same survey (NS-DATA), and a separate parent survey conducted in 2018 that reflects the time from onset to TS diagnosis may be decreasing (Malaty et al., 2019; Wolicki et al., 2019). Decreases in the time to diagnosis have been associated with improved access to needed treatment, support, and services (Council on Children with Disabilities et al., 2006). Furthermore, the demographic characteristics of the NS-DATA TS module sample suggest a higher socioeconomic status and in general, people with higher socioeconomic status have better health and health outcomes (National Research Council and Institute of Medicine, 2009). Parents of children with more severe TS ("moderate" or "severe") were significantly less likely than parents of children with "mild" TS to report having access to needed treatments and were less satisfied with the treatment and overall management of their child's TS. The reasons for reduced access and lower satisfaction among those with more severe TS cannot be determined from these data. Other research suggests the most available TS treatments were unsuccessful in treating children with more severe TS and parents were unable to access more specialized TS treatment options (Hollis et al., 2016). Currently available treatments may not be satisfactory in all cases of severe TS. Overall, a high percentage of parents of children with TS reported accessible treatment and satisfaction with care, but access to care may remain a problem for some (Hollis et al., 2016).

This study is not without limitations. The survey is comprised of parent-reported indicators that may be subject to recall or social desirability bias. In addition, parent reports reflect their perception of the child's experience. The data collected were not validated by medical records or clinical assessment. A study of parent-reported indicators for children with autism spectrum disorder found that the parent's perception of the child's disorder was associated with the impact of the disorder on the family and not necessarily the direct impact of the disorder felt by the child (Zablotsky et al., 2015). Although that analysis was specific to children with autism spectrum disorder, similar reporting tendencies are a potential limitation in this study. In another study examining children's impairment due to tics, parent-reported impairment was moderately related with the clinical assessment of impairment (Storch et al., 2007). The granularity of our findings was limited because we needed to collapse or combine many indicators throughout our analysis to ensure participants' confidentiality due to the small sample size. In addition, the medications listed are the parent-reported medications the child was currently taking for TS at the time of NS-DATA and might not include medications the child had previously taken for TS or co-occurring disorders. By survey design, the TS module of NS-DATA included children with a parent-reported diagnosis of TS or ADHD at the time of the 2011–2012 NSCH interview. Approximately two-thirds of this study's analytic sample had received a parent reported TS diagnosis at the time of the 2011–2012 NSCH and the remaining third reported receiving a diagnosis of TS at the time of NS-DATA.

Therefore, since most of the children were diagnosed with TS before the 2011–2012 NSCH; these estimates may not reflect current diagnostic and treatment practices for TS. Finally, the NS-DATA sample was nationally drawn, but the results are unweighted and not nationally representative and therefore may not be generalizable (Zablotsky et al., 2019).

Even with these limitations, this study contributes to the understanding of treatment used among children with TS. Given the emerging evidence base and updated practice guidelines around TS treatment options, continued research into the accessibility of TS treatments including Comprehensive Behavioral Intervention for Tics (CBIT), other behavioral interventions and accommodations, and medication may inform where additional training or treatment resources may be needed. Ultimately, this paper may serve as a resource to inform the continued monitoring of TS treatment and management among many children and their families living in the U.S.

Conflicts of interest and sources of funding

The findings and conclusions in this report are those of the authors and do not necessarily represent the official position of the Centers for Disease Control and Prevention.

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CRediT authorship contribution statement

Sara Beth Wolicki: Data curation, Formal analysis, Writing - original draft, Visualization, Project administration. **Rebecca H. Bitsko:** Conceptualization, Methodology, Validation, Writing - original draft,

Writing - review & editing, Supervision, Project administration. **Joseph R Holbrook:** Conceptualization, Methodology, Validation, Writing - original draft, Writing - review & editing. **Melissa L. Danielson:** Conceptualization, Methodology, Validation, Writing - original draft, Writing - review & editing. **Benjamin Zablotsky:** Validation, Writing - review & editing. **Lawrence Scahill:** Conceptualization, Writing - review & editing. **John T. Walkup:** Conceptualization, Writing - review & editing. **Douglas W. Woods:** Conceptualization, Writing - review & editing. **Jonathan W. Mink:** Conceptualization, Writing - review & editing.

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[2]Data from the two modules of NS-DATA are available separately. Data access for the TS module requires an agreement with the National Center for Health Statistics' Research Data Center. More information is available at www.cdc.gov/rdc. The NS-DATA survey and data for the ADHD module are publicly available at www.cdc.gov/nchs/slaits/ns_data.htm.

[3]For this analysis, 10 children had no side effects due to TS medication and due to the survey skip pattern, were not eligible for the question, "Were these side effects troublesome enough to stop taking the medication?" For comparisons of the number of side effects, children in this group (i.e., those who did not have any side effects due to TS medication), originally missing, were considered as "no" to the question "were these side effects troublesome enough to stop taking the medication?," since they did not experience any side effects due to TS medication to be troublesome.

[4]Only the 29 survey participants who indicated "no" to the question, "Does your child currently have Tourette syndrome?" were eligible for the subsequent questions. Four survey participants indicated they "don't know" and therefore were not eligible.

[5]Because of small sample size, these medications were combined into a single group to ensure participants' confidentiality.