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Evaluation of new instruments for screening and diagnosis of tics and tic disorders in a well-characterized sample of youth with tics and recruited controls

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Abstract

Tics and tic disorders can significantly impact children, but limited screening tools and diagnostic challenges may delay access to care. The current study attempted to address these gaps by evaluating sensitivity and specificity of the Motor or Vocal Inventory of Tics (MOVeIT), a tic symptom screener, and the Description of Tic Symptoms (DoTS), a brief diagnostic assessment for tic disorders. Children (n=100, age 6–17 years old) with tic disorders attending a Tourette specialty clinic and a community-recruited sample without tics completed a gold-standard assessment by a tic expert; these evaluations were compared to child self-report and parent and teacher report versions of the MOVeIT, and child and parent versions of the DoTS. The parent and child MOVeIT met or exceeded pre-specified 85% sensitivity and specificity criteria for detecting the presence of tics when compared to a gold-standard tic expert diagnosis. The Teacher MOVeIT had lower sensitivity (71.4%) but good specificity (95.7%) for identifying any tic symptoms compared to gold standard. For determination of the presence or absence of any tic disorder, sensitivity of both parent and child DoTS was 100%; specificity of the parent DoTS was 92.7% and child DoTS specificity was 75.9%. More work may be needed to refine the teacher MOVeIT, but it is also recognized that tic expression may vary by setting. While the MOVeIT and DoTS parent and child questionnaires demonstrated adequate sensitivity and specificity for determining

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Declaration of Interest Statement

None of the authors have conflicts of interest related to the subject matter of this manuscript.

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the presence of tics and tic disorders in this well-defined sample, additional testing in a general population is warranted.

Keywords

Tourette; tic disorders; screening tools; sensitivity; specificity

Introduction

Tics are abnormal involuntary movements and sounds that are discrete, repetitive, and have a waxing and waning course. Tic frequency, severity, and functional impact can range from mild to severe. Up to 25% of children in the United States may experience tics during typical development, though for most children these do not recur or persist (Kurlan et al., 2001; Snider et al., 2002). By contrast, persistent tic disorders may affect between 0.3% to 4% of children; tic and tic disorder prevalence is less well characterized in adults but the prevalence of Tourette syndrome is estimated to be approximately 0.05% of adults (Knight et al., 2012; Scharf et al., 2012; Tinker et al., 2022). According to the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5), a tic disorder diagnosis requires the presence of motor and/or vocal tics before 18 years of age that are not attributable to another medical reason (American Psychiatric Association, 2013). Specific tic disorder diagnoses are based upon the constellation of tic symptoms that are present and the duration of tics: Tourette syndrome is diagnosed when a child has two or more motor tics and at least one vocal tic, with tics present for at least one year; a persistent motor tic disorder, or a persistent vocal tic disorder is diagnosed when only motor or vocal tics, respectively, are present for at least one year. Provisional tic disorder is defined by the presence of any tics for less than one year.

Children with persistent tic disorders can experience a number of adverse impacts to school and peer function; also, some tics may result in injury or discomfort (Gorman et al., 2010; Kurlan et al., 2002; Mathews et al., 2004; McGuire et al., 2013; Storch et al., 2007; Sukhodolsky et al., 2003; Zinner et al., 2012). Once a persistent tic disorder is diagnosed, there are well-established evidence-based guidelines for effective behavioral and pharmacologic management of tics and any co-occurring conditions (Murphy et al., 2013; Pringsheim, Holler-Managan, et al., 2019; Pringsheim, Okun, et al., 2019). Early diagnosis is therefore desirable to facilitate access to care that may prevent or mitigate associated adverse health and psychosocial impacts. However, Tourette syndrome and other persistent tic disorders are often under-recognized, which can delay access to care and prolong impairment (Debes et al., 2008; Shilon et al., 2008; Wand et al., 1992). The most recently updated Diagnostic Interview Schedule for Children (fifth edition; DISC-5) has demonstrated improved accuracy in identifying tic disorders (R. Bitsko et al., in press) but this and other structured diagnostic assessments are lengthy and impractical for regular use in primary care settings (Augustine et al., 2017; Gaffney et al., 1994; Lewin et al., 2014; Storch et al., 2005). In addition, the screening tools currently available to aid in tic identification may not capture sufficient information about tic onset, duration, or other clinical features relevant to informing diagnosis and treatment planning, and/or differentiate among tics and other movement disorders of childhood (Augustine et al., 2017; Martino

et al., 2017). As noted by Chang and co-authors (Chang et al., 2009), clinician-based assessment of tics may not fully capture the waxing and waning time course of tic manifestation and context-dependent (e.g., anxiety, familiarity, activity type) expression of many tics, therefore self- and caregiver report of tics and tic phenomenology is critical. However, existing self-report or proxy-informant measures may have limitations including a pre-specified list of tics to assess, as with the Parent Tic Questionnaire (Chang et al., 2009), or restriction to limited metrics such as global severity or restricted time frame, such as presence of tics within the past week, i.e., the Tic Symptom Self-Report (Scahill et al., 2003). The Motor tic, Obsession and compulsion, and Vocal tic Evaluation Survey, or MOVES (Gaffney et al., 1994) shows promise for identifying tics, but additional items that do not assess tics (i.e., obsessions, compulsions symptoms) may restrict its efficiency for rapid screening; work is presently underway to further refine its use (Lewin et al., in preparation).

Two other recently developed instruments have been designed to enable rapid screening of tics and identification of tic disorders and can be completed by proxy informants (e.g., caregivers, teachers) and as self-report measures. Development of these measures, the "Motor or Vocal Inventory of Tics" (MOVeIT) and the "Description of Tic Symptoms" (DoTS) is presented in companion papers in this special issue (ref, this issue). The current project was initiated as part of an ongoing process to develop and refine measures that could be used for rapid clinical screening and population surveillance of tics and tic disorders. Specifically, we evaluated the sensitivity and specificity of the MOVeIT and the DoTS for identifying the presence of tics and the presence of a tic disorder, compared to a gold standard evaluation by an expert pediatric movement disorder clinician.

Materials and Methods

Participants

Eligible participants were children between the ages of 6–17 years of age with a confirmed diagnosis of Tourette syndrome or a persistent motor or vocal tic disorder (cases) or without a known history of tics or a tic disorder (controls). Full details of the recruitment strategy are described in Bitsko et al. (R. Bitsko et al., in press). Briefly, children with diagnosed tic disorders were recruited through clinical practices where they received care for a primary tic disorder and through regional support and advocacy groups for people with Tourette syndrome; control participants were recruited from community settings and from pediatric and family medicine clinics, specifically member-practices of the Greater Rochester Practice-Based Research Network, a regional consortium of clinical practices that engage with the University of Rochester for patient-centered research activities. All eligible participating children were also required to be enrolled in a traditional public- or private-school setting (not home-schooled) to enable collection of teacher ratings of tics and attention-deficit/hyperactivity disorder (ADHD) symptoms (R. Bitsko et al., in press).

All parents completed a written, informed consent process to provide parental permission for their child's participation in the study and children aged 8 years or older completed either a verbal or written assent process depending on their age (8–12 years old and 13–17 years old, respectively). Because many of the children with tic disorders were recruited through

the investigator's clinics, children and their parents were provided assurance during the consent/assent process that participation decisions would not influence clinical care, and the informed consent discussion was conducted by study personnel who did not have a direct clinical relationship with the child (e.g., study coordinators). Finally, whenever possible, study visits were scheduled so that children in the tic disorder group did not complete study activities with their clinician of record. A modest remuneration was provided to all child-parent dyads and teachers for their participation. The research was approved by the University of Rochester's Research Subjects Review Board (RSRB #00064456).

Approach to assessment of tics and tic disorder diagnosis

Each child and parent independently met with a non-tic-expert clinical assessor to complete self-report and parent proxy versions, respectively, of the Motor or Vocal Inventory of Tics (MOVeIT) and the Description of Tic Symptoms (DoTS) described below. A supplemental file is provided with sample versions of the MOVeIT and DoTS, however these measures are currently under revision and not yet available for general use.

For the present study, both tic measures were administered to all parents and to children ≥ 10 years of age. Clinical assessors were Master's and PhD-level graduate students in Psychology or Family Counseling, supervised by a licensed clinical child psychologist with expertise in tic disorders. The clinical assessor role was to explain the measures, be available to answer participants' questions, and assist children as needed to complete the forms (e.g., help to read questions, understand vocabulary). Clinical assessor training also included providing a basic understanding of tics and diagnostic criteria for tic disorders.

A tic expert also completed a gold-standard diagnostic evaluation of the child to assess for tic disorders, by meeting with the parent and child together to review DSM-5 diagnostic criteria for Tourette syndrome, persistent motor tic disorder, and persistent vocal tic disorder. The tic expert also utilized any observations of the child (i.e., visible tics) noted during the visit. Tic experts were blinded to the child's and parent's DoTS and MOVeIT responses. Tic experts were clinical providers in the URMC Pediatric Movement Disorder clinic and included pediatric neurologists, a pediatric neurology nurse practitioner, and a child psychologist.

Case status of many children with tic disorders was already known to the tic expert prior to the study visit because many of these participants were recruited through the investigator's clinics. However, to ensure standard evaluation of all participants, tic experts performed and documented a structured review of all DSM-5 criteria for a tic disorder. Children and their parents also knew their diagnostic status prior to participation in the study, as this was a clear inclusion criterion for a participant's enrollment in their assigned study group (Tic Disorder, Control). By contrast, the study activities completed with the clinical assessor (MOVeIT, DoTS) were conducted in blinded fashion, and children/parents and tic experts were instructed not to reveal the child's group status (Tic Disorder or Control). In only one instance was the blind accidentally broken when a child's tic disorder diagnosis was inadvertently discussed in front of a clinical assessor. To reduce order effects of assessments and potential bias in administration of index tests, clinical assessors were randomly assigned

to see either parent or child for each study visit, and the sequence of clinical assessor and tic expert assessments was counterbalanced across study visits.

Study visits occurred between January 2016–November 2017. For children who participated during the school year (January–June 2016; September–Nov 2017), classroom teachers were contacted (with written parental consent) to request they complete the MOVeIT. For children in early elementary grades, the main classroom teacher who provided the majority of instruction was approached; for older students receiving instruction from multiple teachers, the English Language Arts or Math teacher was asked to complete the MOVeIT, as they were expected to have extensive contact with the student (in contrast to teachers for a non-core academic or non-academic subject). To encourage a high response rate, teacher received a written request from the study team including the study measures and a pre-paid return envelope, follow-up phone-call reminders, and remuneration for their time and effort.

Measures

Motor or Vocal Inventory of Tics (MOVeIT)—The MOVeIT is a brief screening questionnaire developed to rapidly assess the presence and frequency of various motor or phonic tics (Lewin et al., in preparation). MOVeIT items describe common tics (e.g., "*My child sniffs (or snorts or clears their throat)* ...") and are ranked on a three-level ordinal scale (0 = "*never*", 1 = "*sometimes*", 2 = "*often*"). The MOVeIT can be administered as a 10-item proxy measure to parents or teachers, respectively, or as a self-report measure to children directly, for children who, in the present study, were at least 10 years old. Across the three 10-item versions of the MOVeIT, item content is identical, except that proxy measures are written in the third person, e.g., "*My child* [or, "*The student*"] *makes the same twitches, movements, noises, words or sounds over and over*". The original parent proxy version of the MOVeIT included 4 additional items with similar content, i.e., descriptions of common motor and vocal tics such as sniffing, coughing, or shrugging shoulders (MOVeIT-14). For the current project, the full item bank (14 items) was administered to parents as item reduction work was still in progress at the time that the current project was initiated. Total score on the MOVeIT-10 ranges from 0–20 and on the MOVeIT-14 from 0–28, with higher scores indicating greater endorsement of tics (i.e., more tics, and/or more frequent tics).

Description of Tic Symptoms (DoTS)—The Description of Tic Symptoms (DoTS) (R. H. Bitsko et al., in preparation) is a diagnostic questionnaire to ascertain not only the presence of tics, but also age of onset, persistence, and clinical phenomenology (e.g., tic-related impairment, premonitory urges). All DSM-5 diagnostic criteria for a tic disorder are addressed by the DoTS, except for exclusion in cases where apparent tic symptoms are attributable to medications, drugs, or other medical conditions (American Psychiatric Association, 2013). As with the MOVeIT, the DoTS has parent-proxy and child self-report versions (with identical item content), though a teacher-report version is not available; in the present study, the child version was administered to children at least 10 years of age.

Analysis Plan

The co-primary aims of the study were to (a) determine a threshold for achieving a pre-specified 85% sensitivity and 85% specificity of the MOVeIT for the binary decision of

presence/absence of tics (symptom) and (b) assess sensitivity and specificity of the DoTS for the presence of any tic disorder (diagnosis), inclusive of Tourette syndrome, persistent motor tic disorder, and persistent vocal tic disorder, compared to the tic expert assessment. These aims provided the basis for our sample size calculation and power estimate. Calculating sample size for studies of test validity in individuals with a known binary test outcome (i.e., presence/absence of: tics; tic disorder), our sample size estimates were pre-established for 85% sensitivity and 85% specificity, with an estimated precision of +/− 7%, yielding n=101 per group (Hajian-Tilaki, 2014). Based on this sample calculation, we initially planned to enroll approximately n=125 per group to account for potential drop-out, withdrawals, or screen-failures. Analyses for descriptive statistics and group comparisons were performed using Statistica version 13.3 (TIBCO Software Inc., 2017); MedCalc version 20.106 (MedCalc Software Ltd., 2022) was used for calculation of sensitivity, specificity, positive predictive value (PPV), negative predictive value (NPV), accuracy, and for generation of ROC curves and distribution plots by group to determine optimal thresholds for cutoffs on the MOVeIT. Once a MOVeIT total score threshold was determined, we calculated PPV, NPV and accuracy, using a prevalence estimate for tics = 25% based on Snider et al. (Snider et al., 2002). For the DoTS, we applied a population prevalence estimate of 0.8% for Tourette syndrome and 2.8% for any tic disorder (Knight et al., 2012; Scharf et al., 2012). This approach was deemed to be more conservative than use of a prevalence estimate based on the current sample which was intentionally enriched for participants with tics and tic disorders. 95% confidence intervals were calculated for each of the validity measures using the score method with continuity correction (Newcombe, 1998; Tobi et al., 2005). Where appropriate, due to non-normal distribution of the data, non-parametric tests were performed. For analyses comparing Tourette syndrome based on the DoTS to tic expert assessment, children with other tic disorders were excluded.

Results

Sample Characteristics

A total of 100 children enrolled in the study and completed primary outcome measures (tic expert assessment, MOVeIT, DoTS). Consequently, the final sample size for analysis comprised 100 participants in total: n=57 children with a tic disorder (Tourette syndrome, n=52; persistent motor tic disorder, n=4; persistent vocal tic disorder, n=1) and n=43 without a tic disorder diagnosis. Two children who initially enrolled in the Control group based on no known history of tics according to parent report and by medical history were identified by the tic expert as meeting DSM-5 diagnostic criteria for Tourette syndrome and therefore were included in the Tic Disorder group for all analyses. A third child was identified by tic expert assessment as having a history of tics but did not meet diagnostic criteria for a current tic disorder and did not currently have tics, and therefore was retained in the Control group. Table 1 presents the demographic characteristics for the full sample, and by group (cases, controls).

Children < 10 years old did not complete self-report measures (with one exception, a child 9.6 years old completed self-report measures due to erroneous rounding-up when calculating their age), and teacher questionnaires were not completed for participants enrolled during

the summer months; therefore, there were fewer completed MOVeIT and DoTS self-report and teacher questionnaires than parent or teacher questionnaires. Two children who should have completed these questionnaires due to their age > 10 years old had missing data; one did not complete the MOVe-IT and one did not complete the DoTS. The number of participants for which each individual assessment was completed was: tic expert assessment and parent MOVeIT, n=100; parent DoTS, n=98; teacher MOVeIT, n=72; child MOVeIT and child DoTS, n=68. A total 52 children had data on all three MOVeIT forms (parent, teacher, self-report), and both DoTS forms (parent, self-report), along with the tic expert assessment. The response rate for completion of the teacher MOVeIT was 99% (n=72 returned of 73 sent).

MOVeIT screener compared to tic expert assessment

Table 2 presents summary data (mean, standard deviation, median, range) for the MOVeIT scores by informant (parent, child, and teacher) by group (Tic Disorder vs. Controls). For analyses of the parent MOVeIT we evaluated both the 10- and 14-item versions.

Figures 1a–1d show the distribution of MOVeIT total scores by informant by group and illustrate that across all three informant versions (parent [10 and 14 item], self-report, teacher) the control group MOVeIT scores were consistently lower than those of the Tic Disorder group. This distinction is most clearly seen for both of the parent questionnaires, followed by the self-report questionnaire, with less separation between groups for the teacher MOVeIT. There was also a notable difference between parent-proxy and self-report. As shown in Table 2, the mean MOVeIT total score for the Tic Disorder group was significantly higher than that of the Control group for each informant version.

Using the tic expert determination for the presence or absence of tics we evaluated the parent, self-report, and teacher MOVeIT across the range of scores to determine an optimal cut point for sensitivity and specificity (at least 85% for each). Table 3 presents sensitivity, specificity, PPV, NPV, and accuracy for each of the MOVeIT informant versions and Figures 2a–2d presents results of the ROC analysis for each of the MOVeIT versions. For both versions of the parent MOVeIT, a total score >0 (i.e., any tic symptom endorsed) was sufficient to meet the a priori criterion of 85% sensitivity and specificity, but the optimal criterion cutoff score was >4 (see Table 3).

Because the proposed item reduction for the parent MOVeIT (from 14- to 10 items) may have influenced how many children were identified as having any tics (i.e., if the particular items selected for removal would otherwise have been endorsed), we examined whether classification by the parent MOVeIT would be substantially altered by using the 10-item version. Among all 100 participants in the study, 60 were identified with the parent MOVeIT-14 (full item set) as having any tics, and n=58 were identified as having tics using the parent MOVeIT-10. The two participants not identified by the parent MOVeIT-10 as having tics each had a MOVeIT-14 total score=1, i.e., only one item was endorsed as "*sometimes*", and both participants were in the Control group. Regarding the parent MOVeIT-14 and MOVeIT-10 item versions, the AUC values for these two ROC curves were excellent (both AUCs = 1.00, p<.0001). At a total parent MOVeIT score>4 (for either 10- or 14-item measure), sensitivity, specificity, PPV, and NPV each were =100%.

Figure 2c and 2d respectively show the ROC curves for the child self-report and teacher proxy report MOVeIT. The AUC values for these two ROC curves were excellent (child MOVeIT AUC = 0.96, p<.001) and good, respectively (teacher MOVeIT AUC = 0.85, p < .001). For the child self-report MOVeIT, the a priori threshold for 85% sensitivity and specificity was met with a total score >2 (sensitivity = 92.5%; specificity = 89.3%). For the teacher MOVeIT, the requirement for 85% sensitivity and specificity was not met at any MOVeIT score, but a total score >2 provided sensitivity=71.4% and specificity=95.7%.

DoTS compared to tic expert

The parent DoTS (completed by N=98 parents) correctly identified 60 children with a tic disorder based on expert assessment. The child DoTS identified n=46 children with a tic disorder. Of note, two participants were identified by the child DoTS as having a single motor and vocal tic each and meeting all other DSM-5 criteria for a tic disorder that were assessed by the DoTS. Hence, these two participants might be considered on the basis of the child DoTS to have a persistent motor tic disorder *and* a persistent vocal tic disorder, but not Tourette syndrome because only one motor tic symptom was endorsed. Each of these two children were determined by the tic expert to meet DSM-5 criteria for Tourette syndrome (i.e., having at least *two* motor tics as well as at least one vocal tic). Table 4 shows the breakdown of tic disorder diagnoses rendered by the parent and child DoTS, respectively, cross-referenced to the gold-standard diagnosis established by the tic expert.

Because the preponderance of children with a tic disorder diagnosis based on the reference standard assessment were deemed to have Tourette syndrome (n=52), we first evaluated sensitivity and specificity of the DoTS for identifying the presence of Tourette syndrome and then separately, evaluated the DoTS for identifying the presence of any tic disorder, inclusive of both Tourette syndrome and either a persistent motor or persistent vocal tic disorder. Full results (sensitivity, specificity, PPV, NPV, and accuracy) are shown in Table 5. For identifying Tourette syndrome, the parent DoTS demonstrated excellent sensitivity (96.2%) and specificity (97.6%) when compared to the tic expert determination. The child DoTS demonstrated good sensitivity (72.2%) and excellent specificity (93.1%) for identification of Tourette syndrome. The parent DoTS also demonstrated excellent sensitivity (100%) and specificity (92.7%) for identification of *any* tic disorder when compared to a tic expert determination of the same. The sensitivity of the child DoTS for determination of any tic disorder compared to the tic expert was also excellent (100%) though specificity was lower (76.5%) and did not meet the a priori threshold of 85%. The estimated PPV was low, and estimated NPV high, for both the parent and child DoTS, for determination of the presence/absence of Tourette syndrome and for any tic disorder. This was not unexpected considering the relatively low prevalence for both Tourette syndrome (<1%) and for tic disorders in general (<3%), which increases the probability of false positives (lowering PPV) and decreases the probability of true negatives (increasing NPV) in the sample.

As noted above, two children enrolled in the control group with no known history of tics were re-classified as having tic disorders. One child was <10 years old, so did not complete the self-report MOVeIT or DoTS. Both children had non-zero total Parent MOVeIT scores (parent MOVeIT-14 total score =12 and 13, respectively; parent MOVeIT-10 total score =9

for each child), and for each child both motor and vocal tics were endorsed by their parents. The child who was old enough to complete both measures (age 15 years) had a self-report MOVeIT score =12 and endorsed both motor and vocal tics. There were no tics endorsed on the teacher MOVeIT for this teenager, but the younger child's teacher did endorse presence of both motor and vocal tics (teacher MOVeIT total score = 8). According to the parent DoTS for each of these participants, and for the teenager who completed the self-report DoTS, diagnostic criteria for Tourette syndrome were met: childhood onset of tics, at least one vocal and at least two motor tics, and duration at least one year. This determination aligned with the tic expert's assessment of these two participants, who were both deemed by the expert clinician to meet all DSM-5 diagnostic criteria for Tourette syndrome. Thus, for these two subjects, the DoTS and MOVeIT were largely successful at identifying the presence of tics, and likely presence of a tic disorder.

Discussion

In the present study, a tic symptom-screening tool (MOVeIT) and tic disorder diagnostic tool (DoTS) each demonstrated good to excellent (depending on informant and measure) sensitivity and specificity for identification of tics and tic disorders respectively, in a well-defined sample of children with and without tic disorders. Compared to an expert clinical assessment, the accuracy of parent report of tics was marginally higher than that for child or teacher report. Based on the pattern of children's responses on the DoTS, it is clear that although all children in the Tic Disorder (Cases) group were aware that they had tics, a subset did not recognize the joint presence of motor and vocal tics and/or the presence of multiple motor tics which would match to their clinical diagnosis of Tourette syndrome. While it is possible that some children due to young age or developmental immaturity, may have lacked awareness of their tics, the children in this subset ranged from 10–15 years old. Of note, the MOVeIT does not specify any timeline for tic presence. In addition, while the DoTS includes items that ask respondents to report on their age of onset of tics, tic persistence and duration, and the presence of current versus lifetime tics, other DoTS items lack specificity with regard to timeline. Thus, another possibility is that the inconsistency across DoTS items may have introduced uncertainty for children in considering their complete tic history. This inconsistency could account for the fact that some children with a diagnosis of Tourette syndrome instead reported the presence of only motor or only vocal tics, respectively, which may have been more predominant at the time of the study visit.

Classroom teachers and other school personnel may be unaware of a child's tics or tic diagnosis unless informed by the child or their caregiver. Also, if tics are mild, infrequent, of minimal intensity or complexity (e.g., not readily distinguished from everyday voluntary movements or sounds), and do not interfere with a child's functioning, they may go unnoticed in a busy classroom setting. These factors may help to explain why the teacher MOVeIT had lower sensitivity for identifying tics compared to the parent and child versions of this symptom screening measure. The MOVeIT may be useful for school-based surveillance studies, but further work could refine its ability to detect tics in the classroom setting. For clinical screening, the teacher MOVeIT would not need to be deployed; indeed, the present guidelines for evaluation of tics and tic disorders does not require input

from teachers or other outside informants beyond the child's parent, unlike a common comorbidity, ADHD, where symptom presence in multiple settings is a requirement for the diagnosis (American Psychiatric Association, 2013).

Prompt, accurate diagnosis of tics and tic disorders enables affected individuals and their families to seek early and appropriate access to clinical care and to be monitored for risk of emerging comorbidities that may develop over time (Hirschtritt et al., 2015). Except for a provisional tic disorder, a tic disorder diagnosis requires that tics persist for at least one year, meaning this duration is expected between the initial determination that tics are present and confirmation of diagnosis. However, prior investigations have reported an average delay to diagnosis (beyond this one-year observational period) ranging from almost one year up to a decade, depending on the sample under study (Baümer et al., 2016; Debes et al., 2008; Hollis et al., 2016; Malaty et al., 2019; Wolicki et al., 2019). Factors that may be associated with prompter diagnosis include: earlier onset of tics, presence of motor (vs. vocal) tics, more noticeable tics and greater tic severity, male sex, or presence of co-occurring conditions - particularly obsessive-compulsive disorder (Park & Kim, 2021; Shilon et al., 2008). A potential contributor to delays in diagnosis may be lack of access to a provider who is knowledgeable about tics and tic disorders. Although over 75% of children may be initially seen by their primary care provider (PCP) when parents have concern for tics, the majority will ultimately be diagnosed by a specialist such as a pediatric neurologist, psychiatrist, or developmental and behavioral pediatrician (Hollis et al., 2016; Wolicki et al., 2019). Unfortunately, there is a critical shortage of these and other pediatric subspecialty providers in the United States with few specialists available and excessive wait times – in up to a third of practices, wait times for appointments may exceed 4 months, further delaying diagnosis and care (Keller et al., 2020; Majersik et al., 2021). As well, a recent study found that approximately half of children with a tic disorder were seen by at least 3 different providers prior to diagnosis (Majersik et al., 2021; Rimsza et al., 2018; Wolicki et al., 2019). PCPs are uniquely positioned to screen for tics and tic disorders, as they are the initial point of contact for most children receiving medical care. The MOVeIT and the DoTS have the potential to provide PCPs with accurate and sensitive tools to screen for tics and tic disorders, reducing the number of children who experience multiple referrals and excessive wait times prior to obtaining their tic disorder diagnosis.

There are several limitations of the present study. First, the overall final sample of N=100 fell short of the planned target. In addition, though the choice of case-control study design was intentional, recruitment of children with tic disorders (Cases) was directed at those receiving care from a tic disorder clinic or from a primary care provider with special expertise in tic disorders, which likely resulted in a sampling bias towards enrolling Cases with high clinical needs, i.e., more severe or functionally impactful tics. This recruitment strategy may have contributed to the stark difference between the Cases and Controls in the mean MOVeIT scores and may also have contributed to high sensitivity and high specificity for the parent and child MOVeIT and DoTS. In a general population setting where tic symptom and tic disorder status is not known to children, parents, or teachers, we anticipate lower accuracy of these measures. Also, because diagnosis status was known to the children in the Cases group and their parents, accurate endorsement of tics on the MOVeIT and endorsement of diagnostic criteria for a tic disorder (and other tic phenomenology) on the

DoTS was more likely to occur, contributing to high agreement between the ratings by participants in this group and the tic expert assessment. The children in the Cases group, drawn from a Tourette specialty clinic, represented a broad range of clinical need, including some who maintained regular contact with our clinic for management of troublesome symptoms, and some who were seen only for an initial diagnosis and/or only saw our providers occasionally for a 'check-in' visit but did not generally experience bothersome tics.

We acknowledge that because the prevalence of a disorder will influence measurement of PPV and NPV, the DoTS and MOVeIT may not perform as well for these metrics in the general population, where the tic disorder prevalence is low (Yi et al., 2004); however, we attempted to address this by using the expected population prevalence rather than sample prevalence in calculating PPV and NPV. Finally, our sample was predominantly White and non-Hispanic. A study of comparative estimates of Tourette syndrome among different ethnic groups reported a higher prevalence of diagnosed Tourette syndrome (0.39%) in non-Hispanic White children compared to Hispanic (0.16%) and non-Hispanic Black children (0.15%), based on parent's retrospective report (Bitsko et al., 2022; Bitsko et al., 2014; Charania et al., 2022) that disparities in health care access and cultural distinctions in symptom interpretation or significance may influence who is counted as having a tic disorder (Eapen et al., 1997; Flores et al., 2008; Mathews et al., 2001; Robertson, 2008). Future investigations could redress these gaps with enrollment that enriches the sample for underrepresented minority groups.

Despite these limitations, the MOVeIT and DoTS appear to demonstrate reasonable sensitivity and specificity when screening for tics and most features of tic disorder diagnoses, respectively. Other work is examining the diagnostic utility of the MOVeIT tic screener in other pediatric clinical populations at high risk for tics and other common pediatric movement disorders (i.e., stereotypy) and in a general pediatric setting. Yet to be established is whether the MOVeIT and DoTS will function best in tandem; a recently initiated project by our group will administer the DoTS as a focused follow-up diagnostic tool to children whose MOVeIT scores cross a pre-specified threshold for the likely presence of tics. Ultimately however, use of these measures jointly or independently in real-world settings will likely depend upon the specific intended concepts of interest (i.e., tics vs. tic disorders) and context of use (e.g., clinical screening and/or diagnosis; population-level surveillance). In addition, comparison to other established gold-standard clinical assessments, such as the Yale-Global Tic Severity Scale (Storch et al., 2005) could be considered in future research. Finally, item reduction work is ongoing to determine the most utile item set (for MOVeIT and DoTS) to further improve streamlining and accuracy of the measures. Once finalized, these two questionnaires could be useful aids to the primary care physician to quickly identify children at risk for tics and tic disorders. Both instruments are easy to administer, and the DoTS provides most information needed to establish a clinical diagnosis of a tic disorder, facilitating use by clinical providers in the primary care setting. Since, as noted above, the majority of children with new-onset tics present first to their PCP, the MOVeIT and DoTS may aid in early identification of tics and tic disorders and in doing so, likewise facilitate early treatment to mitigate the associated adverse health and psychosocial impacts to, and improve outcomes for children with persistent tic disorders.

Additionally, if used in population-based surveillance studies, these measures may help improve precision of population prevalence estimates of pediatric tics and tic disorders which can serve as the basis for directing resources to better identify and mitigate the impacts of tic disorders upon children and families.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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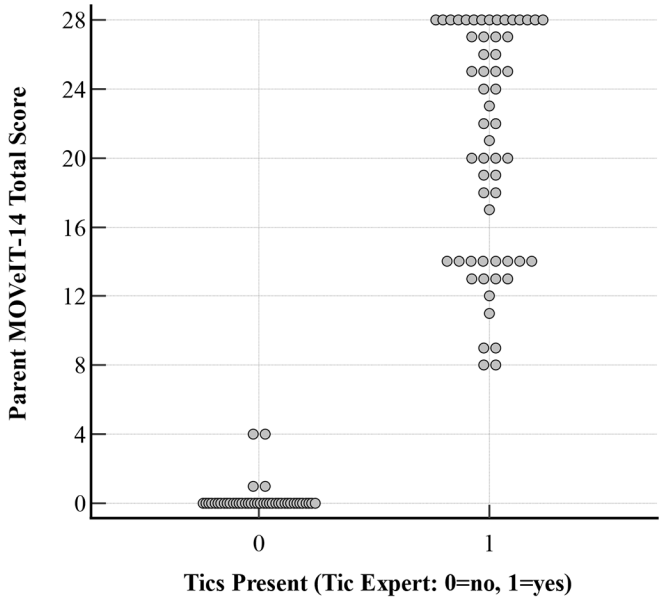

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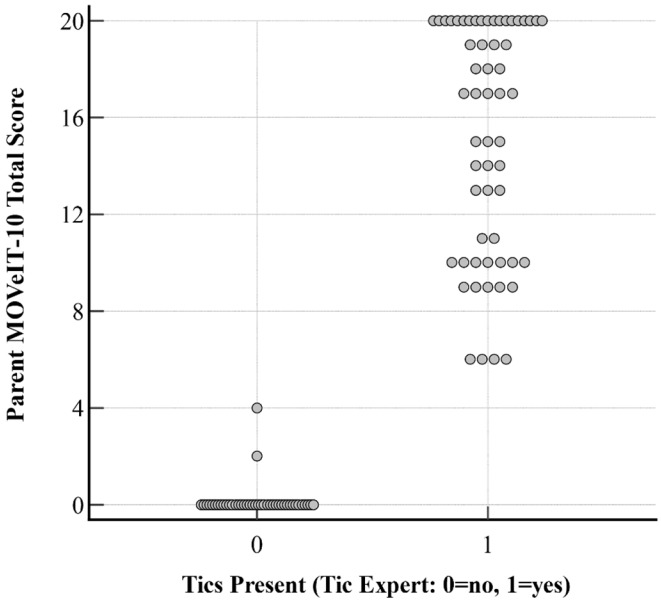

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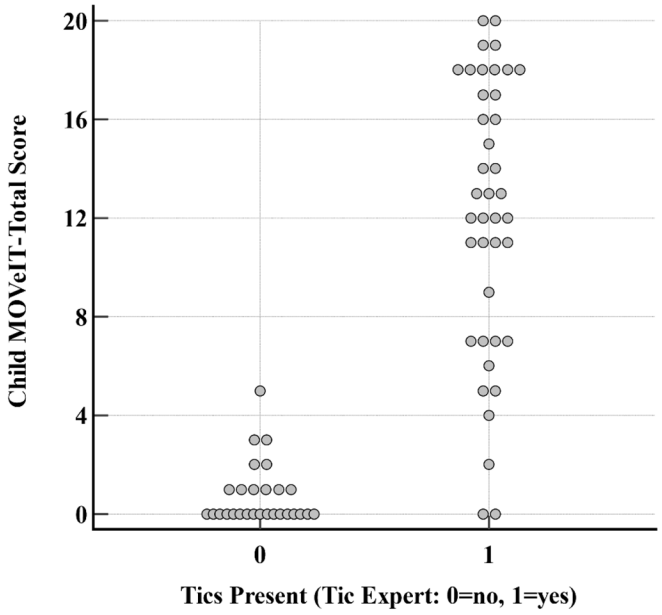

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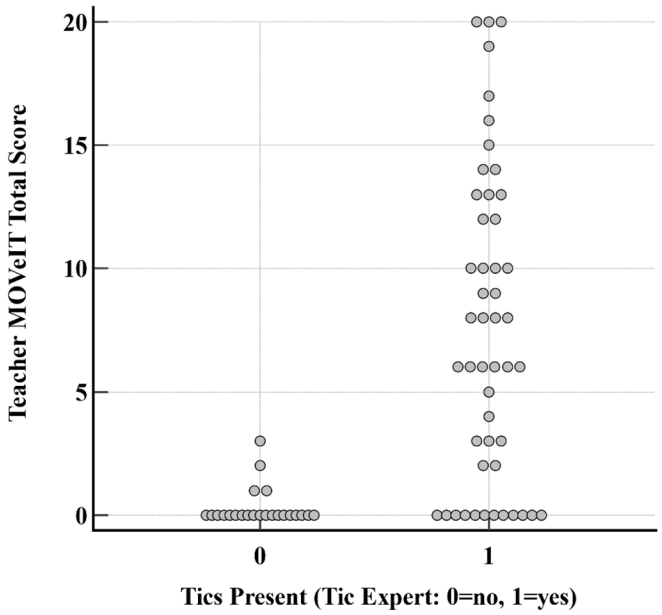


Figures 1a-1d.

Dot diagrams for the distribution of MOVeIT Parent-14, Parent-10, Child, and Teacher total scores, by group (Tic Disorder vs. Control)

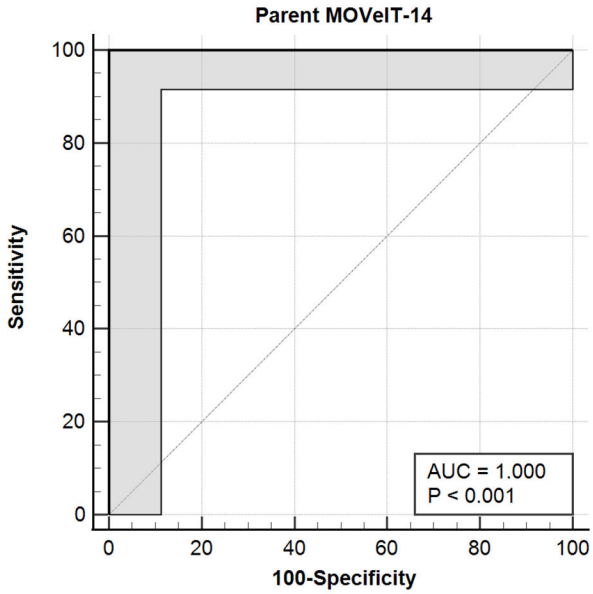
1a. Parent MOVeIT-14 total score by presence or absence of tics, according to tic expert

1b. Parent MOVeIT-10 total score by presence or absence of tics, according to tic expert

1c. Child MOVeIT total score by presence or absence of tics, according to tic expert

1d. Teacher MOVeIT total score by presence or absence of tics, according to tic expert

a.



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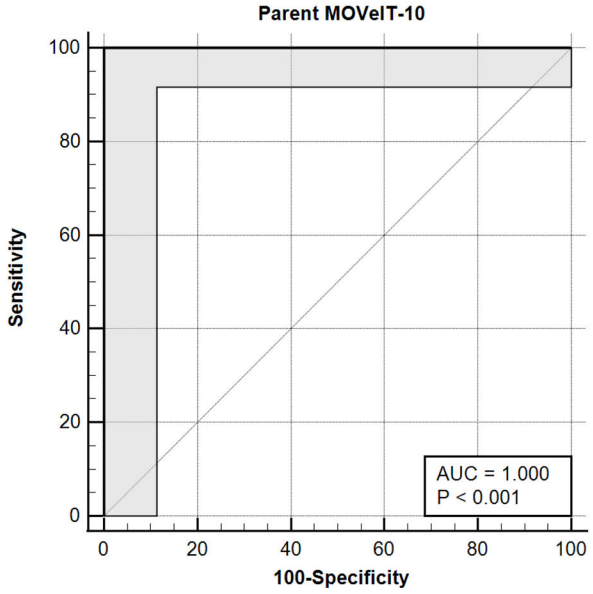

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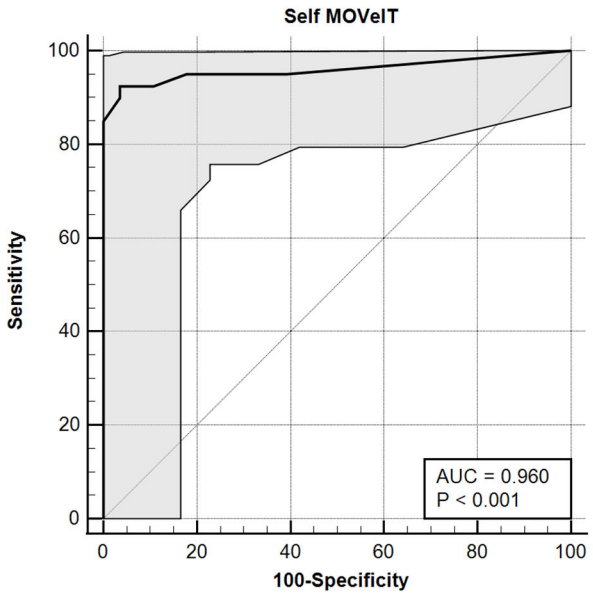

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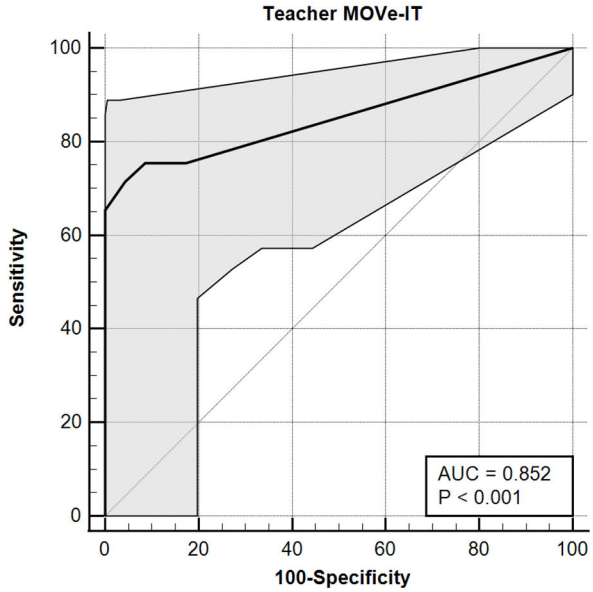

Figures 2a-2d.

Receiver Operating Characteristic Curve analyses with 95% Confidence Intervals of MOVeIT total scores by informant (parent, child, teacher) for distinguishing between participants with and without tics based on tic expert assessment (Reference Standard)

2a. ROC Curve for Parent MOVeIT-14

2b. ROC Curve for Parent MOVeIT-10

2c. ROC Curve for Child MOVeIT

2d. ROC Curve for Teacher MOVeIT

Table 1.

Demographic characteristic of all participants and by tic disorder status.

		All Children N = 100	Children with a Tic Disorder (Cases) (n=57)	Children without a Tic Disorder (Controls) n = 43	Children with vs. without a Tic Disorder
Age (years)	Mean (SD) Range	12.0 (3.3) 6.2 – 17.9	12.0 (3.2) 6.6 – 17.9	12.1 (3.3) 6.2 – 17.5	$t^1(df, 98) = 0.17$, 95% CI [−1.2, 1.4], *ns*[2]
Sex (n, %)	Females	37	13 (22.8%)	24 (55.8%)	Cases < Controls, $z^3 = -3.38$, p< .001
	Males	63	44 (77.2%)	19 (44.2%)	Cases > Controls, $z^3 = 3.38$, p<.001
Race (n, %)	American Indian	1	0	1 (2.3%)	point probability[4], p<.01
	Asian	3	0	3 (7.0%)	
	African American	5	1 (1.8%)	4 (9.3%)	
	White	91	56 (98.2%)	35 (81.4%)	
Ethnicity (n, %)	Hispanic	7	2 (3.5%)	5 (11.6%)	*ns* [2]

[1] Student's t test for independent samples; *df* = degrees of freedom

[2] *ns* = not significant

[3] Two-sample Z-test for proportions

[4] Extended version of Fisher's exact test (Uitenbroek, 1997)

Table 2.

Parent, teacher, and self-report scores on the Motor or Vocal Inventory of Tics (MOVeIT) screener for tics by tic disorder status.

	Tic Disorder Mean (SD) Median, Range [informant n]	Controls Mean (SD) Median, Range [informant n]	Mann-Whitney U Test for Means (2-tailed) (Tic Disorder vs. Controls)
Parent MOVeIT-14	21.0 (6.5) 22.0, 8 – 28 [n=57]	0.4 (1.6) 0.0, 0 – 9 [n=43]	U=2.5, z = 8.51 *
Parent MOVeIT-10	15.4 (4.7) 17.0, 6 – 20 [n=57]	0.3 (1.1) 0.0, 0 – 6 [n=43]	U=49.0, z = 6.40 *
Self (child) MOVeIT-10	12.3 (5.6) 13.0, 0 – 20 [n=39]	0.8 (1.4) 0.0, 0 – 5 [n=29]	U=158.5, z = 4.98 *
Teacher MOVeIT-10	7.6 (6.3) 7.0, 0 – 20 [n=48]	0.3 (0.8) 0.0, 0 – 3 [n=24]	U=1.5, z = 8.52 *

* p<.00001

Table 3.

Comparison of the Motor or Vocal Inventory of Tics (MOVeIT) screener 14-item parent-report and 10-item parent proxy report, child self-report, and teacher-proxy report to expert clinician assessment, for presence of any tics (motor or vocal).

MOVeIT version (criterion score)	n	Sensitivity 95% CI	Specificity 95% CI	PPV 95% CI	NPV 95% CI	Accuracy 95% CI
Parent MOVeIT-14 (sum>4)	100	100.0% 93.8, 100%	100.0% 91.6, 100.0%	100.0% _[c]	100.0% _[c]	100.0% 96.4,100.0%
Parent MOVeIT-10 (sum>4)	100	100.0% 93.8, 100%	100.0% 91.6, 100.0%	100.0% _[c]	100.0% _[c]	100.0% 96.4,100.0%
Child MOVeIT-10 (sum>2)	68[a]	92.5% 79.6, 98.4%	89.3% 71.8, 97.7%	92.5% 80.8, 97.3%	89.3% 73.6, 96.1%	91.2% 81.8, 96.7%
Teacher MOVeIT-10 (sum>2)	72[b]	71.4% 56.7, 83.4%	95.7% 78.1, 99.9%	84.6% 44.4, 97.4%	90.9% 86.5, 94.0%	89.6% 80.1, 95.6%

PPV=Positive Predictive Value; NPV = Negative Predictive Value

[a] Child MOVeIT-10 only administered to children ≥ 10 years old, except for one participant age 9.6 years (due to rounding when calculation of chronological age)

[b] Teacher MOVeIT-10 only administered to classroom teachers of participants who enrolled in the study during the academic year

[c] 95% Confidence Intervals not provided by MedCalc for this NPV value because CI range is negligible

Table 4.

Tic Disorder diagnoses based on parent and child report on the Description of Tic Symptoms (DoTS), as compared to tic expert diagnosis.

Tic Disorder Diagnosis, n		
Tic Expert (N=100)	**Parent DoTS (N=98)**	**Child DoTS (N=68)**
Tourette syndrome, n=52	Tourette syndrome, n=50 persistent motor tic disorder, n=2	Tourette syndrome, n=26 persistent motor tic disorder, n=5 persistent vocal tic disorder, n=3 persistent vocal + motor tic disorder, n = 2 not included[1], n=16
persistent motor tic disorder, n=4	persistent motor tic disorder, n=4	persistent motor tic disorder, n=2 none, n=2
persistent vocal tic disorder, n=1	persistent vocal tic disorder, n=1	persistent vocal tic disorder, n=1
none, n=43	Tourette syndrome, n=1 persistent motor tic disorder, n=1 persistent vocal tic disorder, n=1 none, n=38 not included[1], n=2	Tourette syndrome, n=2 persistent motor tic disorder, n=1 provisional tic disorder, n=4 none, n=22 not included[1], n=14

[1]Children who were assessed by the tic expert, but for whom a parent or self-report DoTS form was not completed

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Table 5.

Comparison of the description of tic symptoms (DoTS) parent-proxy and child self-report and expert clinician assessment, for presence of (a) Tourette syndrome (TS) and (b) any tic disorder.

Criterion	n	Sensitivity 95% CI	Specificity 95% CI	PPV 95% CI	NPV 95% CI	Accuracy 95% CI
(a) Tourette syndrome[a]						
Parent DoTS	93	96.2%	97.6%	24.1	100.0%	97.6%
		86.8, 99.5%	87.1, 99.9%	4.4, 68.8%	_[c]	92.0, 99.6
Child DoTS	65	72.2%	93.1%	7.8%	99.8%	92.9%
		54.8, 85.8%	77.2, 99.2%	2.1, 24.6%	99.6, 99.9%	83.8, 97.8%
(b) Any tic disorder[b]						
Parent DoTS	98	100.0%	92.7%	28.2%	100.0%	92.9%
		93.7, 100%	80.1, 98.5%	11.7, 53.9	_[c]	85.9, 97.1%
Child DoTS	68	100.0%	75.9%	10.7%	100.0%	76.5%
		91.0, 100%	56.5, 89.7%	5.9, 18.5%	_[c]	64.7, 86.0%

CI=confidence interval

[a] TS population prevalence estimated at .8% (Knight et al., 2012; Scharf et al., 2012)

[b] Any tic disorder population prevalence estimated at 2.8% (Knight et al., 2012)

[c] 95% Confidence Intervals not provided by MedCalc for this NPV valu