



Tourette's From a Neuropsychological Perspective

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Abstract

Purpose of Review Pediatric neuropsychologists have interest in the relationship between neurodevelopmental disorders and chronic health conditions, and the developing brain. With growing survivorship of children with such conditions, pediatric neuropsychologists are increasingly looking to the literature for guidance regarding evaluation and treatment approaches, including those for Tourette's syndrome. The purpose of this article is to raise awareness of the complexity in the clinical presentation of Tourette's so that patient care is optimal, and generation of literature for future study of the elusive aspects of the disorder is encouraged.

Recent Findings The published research regarding Tourette syndrome encompasses many aspects of the disorder. Those that are of relevance to pediatric neuropsychologists will be discussed.

Summary Pediatric neuropsychologists must seek out relevant information about Tourette's syndrome for the purposes of patient care. In this article, a brief review of current literature highlights the most salient aspects of this complex disorder to be considered for evaluation and treatment purposes.

Keywords Tourette's syndrome · Pediatric neuropsychology · Comorbidity · Neuropsychological evaluation · Treatment and intervention

Introduction

Neuropsychology is a specialty area of psychology that is concerned with learning and behavior in relationship to the systems and structures of the brain. Because humans are not born with all brain systems established, pediatric neuropsychology is specifically concerned with how learning and behaviors are affected by factors that may compromise the integrity of cognitive functioning associated with particular systems and structures of the developing brain. Pediatric neuropsychologists therefore frequently assist children who are experiencing challenges in learning, attention, behavior, socialization, or emotional control; a disease or inborn developmental disorder that affects the brain in some way; or a brain injury from an accident, birth trauma, or other physical stress [1].

The study of children with neurodevelopmental disorders and chronic health conditions in relation to neuropsychological functioning is growing [2•]. The need for pediatric neuropsychologists and evidenced-based evaluations and interventions for these populations has coincided with advances in medicine that allow children with once terminal medical conditions to now survive further into childhood, adolescence, and even adulthood. The survival of a child who was once given minimal to no chance of life beyond a few days or several months following birth is to be celebrated. However, the survival of countless children with a various neurodevelopmental disorders and chronic health conditions has often come with the cost of life-long compromise of many central nervous system functions, either due to the condition itself or medical interventions necessary to treat or manage the condition.

At the same time, practicing pediatric neuropsychologists have increasingly become more cognizant of the psychosocial and emotional effects that neurodevelopmental disorders and chronic health conditions have on children, and how both psychosocial and emotional aspects, and the medical aspects of the conditions have on a child's quality of life [3]. Understanding the effects of these factors on the child's cognitive, behavioral, and emotional development can guide

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health care providers toward the most effective interventions, and ultimately maximize the child's quality of life [4••]. Therefore, while many think of psychometric testing and evaluation as being the sole consideration of a pediatric neuropsychologist, many aspects of a neurodevelopmental disorder or chronic health condition must be considered by the pediatric neuropsychologist in order to evaluate and treat the child optimally.

One neurodevelopmental disorder that has been amply studied, and of interest to pediatric neuropsychologists for a number of years, is Tourette's syndrome. Tourette's syndrome is a chronic tic disorder that has an onset before 18 years of age. Tics are involuntary, fast, repetitive motor movements or vocalizations. Those with the disorder experience multiple motor tics and at least one vocal tic [5].

Whether conducting diagnostic evaluations or collaborating with a medical professional regarding treatment of a mutual patient with a condition about which they may not be familiar, pediatric neuropsychologists should look to the literature for guidance to better understand its neurological and other medical aspects. Further, when reviewing the literature about the disorder for the purpose of patient care, it is the task of the pediatric neuropsychologist to also understand its psychosocial and emotional characteristics. Tourette's syndrome is a very complex condition with medical, psychiatric, neuropsychological, and psychosocial factors that must be considered in the interest of patient care. Awareness of the comprehensive aspects about Tourette's syndrome will help pediatric neuropsychologists with evaluation and treatment planning, while also spawning interest in generating future research that increases understanding of this complex disorder.

Consider the following vignette, which is a typical case from this author's experience:

When Teddy was a 6-year-old boy, he presented with an ongoing history of exhibiting a difficult temperament, and becoming easily "overstimulated" in unstructured or chaotic environments. Throughout elementary school, his teachers had expressed concern to his parents regarding Teddy's difficulty with attention and concentration during independent seat work, and inability to sit still during larger group instruction. His peers often became annoyed with Teddy's impulsive invasion of personal space. Whereas Teddy was able to acquire academic readiness skills easily during primary elementary grades, Teddy's academic performance was falling behind his peers' in 4th and 5th grades. Due to what appeared to be involuntary grunting, throat clearing, eye blinking, and facial grimacing, symptoms of ADHD, and what seemed to be compulsive rituals (e.g., walking back and forth in a doorway exactly five times before walking all the way through), Teddy was diagnosed with Tourette's syndrome in 3rd grade.

In middle school, the gap between Teddy and his peers' academic performance became wider. However, Teddy did not qualify as a student in need of special education services when his school district evaluated him. At the initiation of his neurologist and general pediatrician, consultation was initiated with a pediatric neuropsychologist, who conducted an evaluation to determine Teddy's unique cognitive strengths and weaknesses, and to monitor his academic performance over time. Following consultation with the parents, the pediatric neuropsychologist was also able to consult with his educators to help them understand Tourette's syndrome and to make appropriate accommodations for Teddy with respect to instructional and behavioral intervention accordingly.

Clearly, this vignette describes a case that includes a number of factors that require attention and consideration that should include, but also go beyond, interpretation of the scores obtained from a neuropsychological evaluation. The following highlights the aspects of Teddy's clinical presentation that should be identified by and addressed by the pediatric neuropsychologist. Recent literature representing each aspect is briefly discussed.

Medical Aspects of Tourette's Syndrome

As indicated above, pediatric neuropsychologists have interest in and can benefit from understanding essential information regarding the medical aspects of the disorder they are evaluating or treating. For example, the neuropathology of Tourette's is important to guide, among other things, pediatric neuropsychological evaluation in order to be aware of and better understand any phenotypical functional impairments when evaluating a patient with Tourette's syndrome, or of specific areas of focus when conducting an evaluation.

As suggested by Martino et al. [6••], there is a great deal of literature regarding the medical aspects of Tourette's syndrome that focuses on the management of tics, as well as other symptoms often ascribed to comorbid conditions. The symptoms of Tourette's syndrome are typically managed through the use of psychotropic medications. Typical side effects of medications that a patient may be taking for a known diagnosis of Tourette's upon evaluation, or as treatment progress is monitored, is important information for pediatric neuropsychologists to understand. Therefore, they may seek out literature regarding pharmacological treatment for control of tics and other symptoms typical in Tourette's patients.

While more recent neuroimaging technology (e.g., structural brain MRI, fMRI, SPECT) has improved the understanding of the neuropathology of Tourette's syndrome, many questions remain, as findings continue to implicate an increasing

number of structural and systemic abnormalities [7•]. It has been widely accepted that particular structures and systems of the brain are very likely to be implicated in Tourette's, such as decreased volume in the left side of the structures of the basal ganglia. Also, the literature also supports frontostriatal circuit dysfunction that occurs in a chain reaction of dysfunctional brain systems; decreased striatal metabolism and decreased white matter in the lateral orbitofrontal, the anterior cingulate, and the dorsolateral prefrontal cortices changes the activity and metabolism of these regions of the frontal lobes. To complicate matters further, it has been unclear as to whether impairment and dysfunction in these systems and structures are causally related to Tourette's syndrome or comorbid disorders [8]. Perhaps the lingering questions regarding the neuropathology of Tourette's syndrome have created at least one need for rigorous study pharmacological approaches to treating tics. Traditional (e.g., Haldol) and atypical (e.g., Risperdal) antipsychotics are commonly used pharmacological interventions, but have been found to cause unpleasant and often intolerable side effects (e.g., lethargy, weight gain). It has been suggested that alpha-2 agonists tend to be better tolerated, yet reduce tic severity without eliminating the tics themselves [9]. Other studies have shown some medications to be no more effective than placebo assignment in clinical trials agonists in some patients with Tourette's [10].

Comorbid Conditions Common to Tourette's Syndrome

The presence of comorbid conditions is one of the aspects of Tourette's syndrome that creates inconsistency in severity across clinical presentations, to the extent that it has been suggested that there may be more than one phenotype of Tourette's [8]. Comorbid conditions have further been found to be more difficult for some individuals with Tourette's syndrome to manage than the tics themselves [6••]. Comorbidities are so frequently present in the Tourette's population that it is difficult to generate large samples for research, as children with "pure" Tourette's syndrome are very challenging subjects to recruit [11].

The most common comorbid conditions that occur in children with Tourette's syndrome are Attention Deficit Hyperactivity Disorder (ADHD) and Obsessive/Compulsive Disorder (OCD). Not only are they the most common, but they are likely to create the most functional impairments [12]. Children with Tourette's syndrome represent about 1% of the pediatric population, and it is estimated that about 86% have at least one comorbid psychiatric disorder, and 57.7% have two or more comorbid disorders, beginning early in life and being present throughout the lifetime [13••]. In a relatively large comprehensive study ($n = 1374$), Hirschtritt et al. [13••] found that 54.3% of the sample met the diagnostic criteria for

ADHD, 50% for OCD, and 29.5% for all three of Tourette's syndrome, ADHD, and OCD. Other comorbidities were each accounted for in 30% of the Tourette's sample, including mood, anxiety, and disruptive behavior disorders; however, it was unclear as to whether those disorders were secondary to the ADHD and OCD. In another study [14•], it was suggested that learning disabilities are likely represented in an unspecified percentage of children with Tourette's syndrome.

It is first important for pediatric psychologists to understand the comorbidities for the purpose of differential diagnoses. Specifically, it is difficult to determine if ADHD and OCD symptoms are part of the presentation of the patient's Tourette's syndrome, or if they are independent conditions in and of themselves. It has been suggested that genetic analysis and thorough understanding of familial history of psychopathology can be a helpful way to determine whether ADHD and OCD are independent conditions or related to the child's Tourette's syndrome [13••]. Secondly, the pediatric neuropsychologist must also understand the social/emotional/behavioral functional impairment of the child created by mood, anxiety, and disruptive behavior disorders, and the need to focus part of the evaluation on those aspects of the child's clinical presentation. Finally, the pediatric neuropsychologist may have need to screen for learning disabilities, or as was the situation in Teddy's case, to look for underlying neuropsychological factors that might cause lags in academic achievement that are not evident using IQ/Achievement score discrepancies.

Interest in Patterns of Neuropsychological Impairments Common to Tourette's Syndrome

The aforementioned challenges created by comorbidities can make Tourette's syndrome a very neuropsychologically complex disorder. For each patient with Tourette's for whom a pediatric neuropsychologist provides care, there are also often differences in the extent of the severity of the comorbidities, how or if the comorbidities are being treated pharmacologically, and the relationship between the comorbidities and severity of presentation of symptoms related to Tourette's syndrome.

When evaluating a child with Tourette's syndrome, it is important for a pediatric neuropsychologist to consider the extent to which particular tics themselves can impair the child's performance. In workshops conducted with teachers and other educators to improve understanding of the many facets of Tourette's syndrome that may present challenges in the classroom, this author has illustrated this point with an interactive exercise. The participants are given a long, boring reading passage that is written in rather small font and single spaced. First, the author has asked that the participants read the passage, and when the author claps, to quickly extend their necks back. During the exercise, the author claps in

intermittent time intervals, but a several times within the amount of time the participants are told it should take them to read the passage. At the end of the allotted time, the author collects the paper and hands out a page of questions for the participants to answer about the passage. Invariably, no one in the group has had an opportunity to finish the passage and cannot answer any of the questions. During subsequent discussion, participants typically report having to go back and re-read what they had read just prior to the author's clap, making it cumbersome to get through the passage; frustration in being interrupted; and mild, temporary neck pain. The workshop participants then understand the plight of a student with Tourette's trying to get school work done; their exercise spanned 5–7 min, compared to a 6-h school day of a student with Tourette's. The same may be the case during a neuropsychological evaluation lasting several hours. Therefore, when interpreting data regarding constructs such as visual attention, problem-solving, and comprehension, pediatric neuropsychologists need to be mindful of the extent to which observed motor tics may have interfered with task performance.

Needless to say, there is considerable variability in neuropsychological impairments reported in numerous studies. Some studies have reported, such as inhibitory control, attention, visual-motor impairment, manual dexterity, strategic memory, working memory, verbal fluency, and shifting and cognitive flexibility as being commonly reported [8]. Others have additionally pointed to abstract reasoning and some aspects of complex problem-solving [15]. However, the studies have been riddled with methodological limitations (e.g., difficulty recruiting "pure" TS subjects with no comorbidities, small sample sizes), and the challenge of understanding whether the neuropsychological deficits discovered can be ascribed to TS or comorbid conditions, particularly ADHD [8]. When reviewing the literature regarding neuropsychological impairments, pediatric neuropsychologists should also be mindful that the majority of the studies at least prior to 1999 have been performed on adults with Tourette's syndrome [11].

Behavioral Treatments for Tourette's Syndrome

Pediatric neuropsychologists may look to the literature regarding Tourette's to make recommendations upon diagnosis following neuropsychological evaluation, or to treat a patient with Tourette's syndrome following referral from an evaluating psychologist or physician. Given the literature's ambiguity about effective pharmacological interventions and the undesirable side effects of those most frequently prescribed, methods of intervention outside of the realm of medication have also been widely studied. For example, a meta-analysis of various behavior therapies used with individuals within the Tourette's population, found behavior therapy to be effective

in reducing the severity of tics, and to offer treatment effects that were comparable to medication in many subjects [16]. In a critical review of research regarding behavioral therapy for chronic tic disorders, Edwards and Specht [17•] highlighted the effectiveness of comprehensive behavioral intervention for tics, a relatively new and therefore scarcely researched treatment approach that integrates the widely used and researched treatment of habit reversal training into its many components. According to Edwards and Specht, the components of comprehensive behavioral intervention for tics include psychoeducation, habit reversal training, relaxation, contingency management, and generalization training/relapse prevention. Both articles discussed above called for more research on behavioral therapy to better understand why these therapies are effective, and with which patients the therapies are most likely to be most effective.

Psychoeducational Interventions for Tourette's Syndrome

Psychoeducational interventions have not been widely studied with the Tourette's syndrome population specifically but are widely used, and have been found effective in other populations [18••]. In this author's experience as a practicing pediatric neuropsychologist, psychoeducational intervention with the Tourette's population has two important applications. The first is, upon diagnosis, the involved health care provider must educate the family and patient (if age appropriate) about Tourette's syndrome. This may include, but not be limited to, informing patients and families about what aspects of brain function are best known to be affected by Tourette's syndrome, the nature of tics (e.g., changes in kinds of tics, such as no longer eye blinking but now facial grimacing), common behavioral characteristics, and commonly diagnosed comorbid conditions [18••]. Parents often come to pediatric neuropsychologists at the recommendation of their child's primary care doctors after all other stones to find answers that will allow them to help their child have been unturned [1]. In this author's experience, they are often frustrated, confused, uncertain of what their child's future holds, and experiencing mental health disorders themselves secondary to the challenges they have been facing with a child whom they love but do not understand. They want answers and insight. It is this author's impression that psychoeducational intervention is one of the most important services a pediatric neuropsychologist can provide to a family with a child that has just been diagnosed with Tourette's syndrome. Other components of psychoeducational intervention can include offering the books targeted for parents to read regarding Tourette's syndrome (i.e., bibliotherapy), and providing them with contact information for their local chapter of the Tourette Syndrome Association.

Another psychoeducational intervention that has been very effective in this author's experience is psychoeducation within the affected child's classroom. In this approach, the child's classmates and teacher are given a presentation about Tourette's syndrome so that they can better understand the atypical behaviors in which the affected child engages. In one study conducted by Nussey et al. [19], psychoeducational intervention for Tourette's syndrome implemented in four different classrooms improved the knowledge and attitudes of peers, was well received by peers, and appeared to improve the self-confidence of the student with Tourette's syndrome. Furthermore, the students with Tourette's became more accepting of their condition. In this author's experience, the intervention can be conducted through a presentation provided by an adult (e.g., parent, teacher, representative from the local Tourette Syndrome Association), video, or depending on the child's confidence level, by the affected child themselves. In the study described above, the peers in the classroom watched a slideshow presented by the teacher, and three of the four children with Tourette's then answered peer questions and participated in a class discussion following the presentation. This author has been present in one of many psychoeducational interventions recommended to be conducted in a classroom setting, and the experience was very much as the Nussey et al.'s study described. Pediatric neuropsychologists should consider recommending this intervention, particularly if the tics and disruptive behaviors of a patient with Tourette's syndrome are causing them difficulty with peer relations.

While psychoeducational intervention is widely used, in the interest of practitioners using empirically based interventions, additional research that supports psychoeducational intervention is clearly needed.

Conclusion

When providing patient care for children with Tourette's syndrome, pediatric neuropsychologists need to be aware of the complexity of the disorder, and all of the factors that must be considered, including clinical symptoms, comorbid conditions, cautionary interpretation of neuropsychological data, and treatment options. It will be important for collaboration between treating physicians and pediatric neuropsychologists to be ongoing regarding the factors that not only affect the symptoms of Tourette's syndrome but also for ways that impact the child's psychosocial well-being and quality of life.

The literature available regarding Tourette's syndrome may pose more questions as it provides answers. For example, as more evidence becomes available regarding neuropathology, neuropsychological impairment exclusive to Tourette's may be more apparent and guide pediatric neuropsychologists in conducting neuropsychological evaluations. However, it has been suggested that comorbid conditions sharing the same

dysfunction in structures and systems of the brain implicated in Tourette's syndrome make it difficult to understand which disorder is primary in creating any evident impairments.

This author is hopeful that the many aspects necessary for consideration when providing patient care for children with Tourette's syndrome that were highlighted in this article will increase the quality of patient care, encourage use of the evidence-based practices, and motivate scholars in the field to continue with efforts to address confounding factors that make well-controlled research so difficult. With new insights into many questions for which there are currently no clear answers, the elusiveness of Tourette's syndrome will be diminished, and improved quality of life for these patients and their families can be achieved.

Compliance with Ethical Standards

Conflict of Interest The author declares that she has no conflict of interest.

Human and Animal Rights and Informed Consent This article does not contain any studies with human or animal subjects performed by any of the authors.

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