

Tourette syndrome and comorbid early-onset schizophrenia

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Abstract

Objective: A study of the shared phenomenology between Tourette syndrome (TS) and schizophrenia. **Method:** An illustrative case report is presented. We used a chart review of 399 clinically ascertained patients with TS to identify 10 cases meeting criteria for schizophrenia. From our 10 patients, salient clinical characteristics were then tabulated. We then extracted similar clinical characteristics from a previously published series of patients with comorbid TS and schizophrenia in order to combine cases and allow for a comparison between childhood-onset schizophrenia (COS), adolescent-onset schizophrenia (AdoLOS), and adult-onset schizophrenia (AduOS) cases in these groups. **Results:** We found 10 cases of schizophrenia (all were males) in the 399 TS patients for a prevalence rate of 2.5% (95% CI 0.96–4.04). Mean age of tic onset for TS diagnostic criteria ranged from 2–14 years with a mean of 8.2 years. The mean age of diagnosis for schizophrenia was 14.2 (range 9–23 years). We found six cases of schizophrenia with onset of positive psychotic symptoms by 13 years of age, two cases with onset after 13 years of age and before

18 years of age, and two cases with onset after 18 years of age. Attention deficit hyperactivity disorder was present at a higher rate (70%) than one would expect in a clinically ascertained group of patients with TS. Comparison between COS, AdoLOS and AduOS in our pooled cases noted a sex bias skewed toward males. Catatonic symptoms may be more likely in child or adolescent onset cases and negative symptoms more likely in AduOS cases. **Conclusions:** The 2.5% prevalence of schizophrenia in our TS sample exceeds the 1% expected rate of schizophrenia in the general population (chi-square=9.14; $P=.0025$). The six cases of COS (before 13 years of age) exceeds the expected rate of 1–2 per 100,000 (chi-square=4499; $P=.0001$). The 752-fold increase in observed rates of comorbid TS and COS over expected rates suggests a role for unknown common underlying etiologic factors. Based on clinical features, patients with TS and comorbid COS, AdoLOS, or AduOS do not have different conditions. We conclude with suggestions for further research.

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Introduction

Tourette syndrome (TS) is a neuropsychiatric developmental disorder, the hallmarks of which are multiple motor and vocal tics of at least a year's duration [1]. TS is thought to represent a genetically mediated condition with a broad range of severity, much of which may be attributable to the neuropsychiatric comorbidities with which it commonly occurs [2]. Comorbid disorders including attention deficit-

hyperactivity disorder (ADHD) and obsessive-compulsive disorder (OCD) are common [3]. We have previously noted additional patterns of comorbidity between TS and autistic disorder, Asperger syndrome, and bipolar disorder [4–8]. We have also have examined the comorbidity between TS and schizophreniform symptomatology, and TS and childhood-onset schizophrenia (COS) [8,9].

As conceptualized in the *Diagnostic and Statistical Manual of Mental Disorders, 4th Edition, Text Revision (DSM-IV-TR)*, specific symptom criteria for schizophrenia encompass positive symptoms such as delusions, hallucinations, disordered thinking, and disorganized behavior. The negative symptoms include flat affect, poverty of speech, and lack of ability and persistence in goal directed activity [1].

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Although commonly cited in the literature, age of onset criteria for COS vary widely. Prevalence rates of schizophrenia suggest that a childhood onset (based on age 14 or 13 years and younger) is very rare [10,11]. Studies in Sweden reported a rate of COS of 1.6 per 100,000 [12,13]. Another study in North Dakota found a prevalence rate of 1.9 per 100,000 [4,14]. In New Zealand, only two cases with an onset under age 12 years were identified in a population of 130,000 [15]. Thus, current prevalence estimates suggest a rate of 1 to 2 cases per 100,000 for COS (onset by 13 years of age).

In our clinical work, we have been struck by the phenomenologic overlap in symptoms of our TS patients and our schizophrenia patients. Our patients with schizophrenia, particularly of the catatonic and disorganized (hebephrenic) subtypes often exhibit motor or vocal tics, including complex echophenomena, and palilalia. Similarly, some of our patients with TS have engaged in complex posturing reminiscent of catatonia, or in mirror gazing reminiscent of disorganized schizophrenia. Many of our TS patients report brief and fragmentary auditory hallucinations. The vivid, eidetic imaging of echolalia and palilalia that some patients with TS describe has considerable overlap with the auditorization of thought, or “echo des pensees” of Schneider’s so-called first rank symptoms, which often present with schizophrenia, but not exclusively so [16]. One of our TS patients interpreted his echokinesis in response to the movement of others as those people having control over his bodily movements, reminiscent of a delusion of somatic passivity, another so called first rank symptom of schizophrenia. These clinical similarities have caused us and others to question just what the relationship between TS and schizophrenia might be [9,17–24].

The onset of *DSM-IV-TR* Tourette’s disorder, or TS, by criterion definition is during the childhood or adolescent years with a peak age of tic onset at 6–7 years of age [1]. Schizophrenia has a broader range of age of onset [1]. COS is thought to be symptomatically similar to and appears to be continuous with adolescent-onset schizophrenia (AdolOS) and adult-onset schizophrenia (AduOS), according to Nicolson and Rapoport [25]. However, their cohort of COS patients had more premorbid developmental abnormalities, more cytogenetic abnormalities, and a greater burden of family history of paranoid and schizotypal personality disorders than later onset cases, pointing to perhaps a greater developmental neurobiologic vulnerability. Their finding that an earlier age of onset of schizophrenia is associated with more severe psychopathology and neurocognitive deficits has been supported by others [26,27]. It has been hypothesized that the typical age of onset of schizophrenia in later adolescence to early adulthood is a manifestation of neurotransmitter ontogeny interacting with neurobiological vulnerability [25]. An earlier age of onset would be consistent with a developmental period of increased vulnerability to neurobiological and genetic risk factors.

In 1946, the venerable Margaret Mahler described the case of G.K., a boy with “the tic syndrome” who later went on to

develop schizophrenia [28]. In more recent decades, there have been multiple case reports and small case series of patients with comorbid TS and schizophrenia [9,17–21,23,24]. From an epidemiologically defined sample in North Dakota, we have published a prevalence rate of schizophrenia in boys aged 2–12 years old with TS of 8.7% [9]. In a recent paper, Cavanna et al. [29] also noted that 15% of patients in their clinical population with TS also met criteria for schizotypal personality. They found increased rates of obsessiveness and anxiety in this group with comorbid TS and schizotypal personality [29]. Muller et al. [20] also have reported on 5 adults with comorbid TS and schizophrenia. All of their patients first showed symptoms of TS, followed by symptoms of schizophrenia. Four of these had an adult onset of their schizophrenia, and one had an onset in later adolescence. The course of the tic disorder was chronic or worsening over time. They speculated that TS and schizophrenia might co-occur at a rate greater than expected by chance. In their review of the clinical and pathophysiological similarities between TS and schizophrenia, they posited that common biological mechanisms likely play a key role in the shared phenomenology between these clinical syndromes.

To date, the majority of case series reports of comorbidity for TS and schizophrenia have focused on patients with primarily later AdolOS/AduOS. In this study, we estimate the prevalence rate for schizophrenia and shared phenomenology in a clinical population of primarily child and adolescent patients with TS.

Case illustration from subjects of study

U.T. is a 14-year-old boy who has been in our care since age 7. He was first seen for treatment of hyperactivity, inattentiveness, and learning difficulties, not responsive to stimulants. His pervasive hyperactivity dated back to his earliest years. His tics preexisted the use of mixed salts of amphetamine, but seemed to worsen following the start of that medication. The patient’s pattern of waxing and waning multiple motor and vocal tics dates back to 5 years of age. They have included the blinking of his eyes and pursing of his lips. He would repeatedly clear his throat, causing the parents often to wonder if he had allergies and was congested. He had difficulties with swearing, usually in anger, as well as episodic sexual exhibitionism. At age 7 years, his learning disability included a pattern of attempting to read from right to left and letter and number reversals. He would have rapid shifts in mood and sleep onset insomnia. U.T. was treated with clonidine 0.05 mg qam and 0.2 mg qhs, with some improvement in attention and tics. Due to periods of brief blanking of consciousness, he had a neurological evaluation and was diagnosed with epilepsy, for which he was successfully treated with carbamazepine. He admitted that since age 5 years, he has had 2 “imaginary friends.” They had names, but he did not hear them or see them. At age 8 years, the patient indicated that he was hearing voices in his head when no one else was about. At

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