Case Report

Tourette Syndrome and Bipolar Disorder: Unique Problems with Pediatric Comorbidity

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ABSTRACT

Tourette syndrome and bipolar disorder are frequent comorbidities in pediatric age group. They provide a clinician with certain unique challenges. While on one hand the tics mask manifestation of affective symptomatology, the latter makes it difficult to elicit tics with certainty. Data suggest that they might share genetic and neurobiological basis and this is currently an area of extensive research. These clinical and biological overlaps provide grey areas in our understanding, which not only complicates the diagnosis, but also poses problems with management.

Key words: *Bipolar disorder, pediatric comorbidity, tourette syndrome*

INTRODUCTION

Tourette syndrome is a neurodevelopmental disorder that is characterized by both multiple motor and one or more vocal tics present some time during the illness with onset before 18 years.[1] A large majority of the patients with Gilles de la Tourette syndrome are known to have psychiatric comorbidities with attention-deficit hyperactivity disorder and obsessive compulsive disorder being the most common ones, followed by mood and personality disorders.[2,3] Though mood symptoms are frequent in this group, the diagnosis of bipolar disorder is complicated by their inherent unreliability in children. However, authors have reported as high as 20-30% comorbidity between the two.[4,5] While mood disorders might aggravate tics, the latter also has reciprocal effect on the former; leading to spiralling deterioration.[6] We report an interesting case of Tourette syndrome with bipolar affective disorder, which highlights these interactions and the clinical challenges encountered.

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CASE REPORT

Master BG, presently a 15-year-old adolescent male from lower socioeconomic class of suburban India, with unremarkable past, family, birth or developmental history, and an easy going premorbid temperament, presented to us with symptoms with insidious onset and fluctuating course of 6 years' duration characterized by prolapsed mass per anum, abnormal repetitive movements, and fluctuations in mood; with inappropriate sexual behavior and abusive behavior for last 4-5 months.

The symptoms started with complaints of repeated squatting and straining for passing stools at the age of 9 years. Over the next few months, the frequency gradually rose to more than 20 per day (with no change in consistency of stool or any other gastrointestinal symptoms). There was a simultaneous deterioration

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in his academic performance, interaction with friends and family members, and a reduction in his interest toward leisure activities. The boy would appear sad and his appetite reduced. He was found to spend more and more time in the washroom, and after his parents noticed these changes they also found a soft, single, red, fleshy painless mass protruding out of the his anal opening. He was treated by faith-healers and quack practitioners for next few months, but in vain.

As the straining behavior improved spontaneously, first glimpses of abnormal movements were noted in the boy. He was found to repeatedly curl his toes and oppose his fingers. He also started taking a step forward and two steps backward whenever he walked, walked in tandem, stomped his feet alternately, and repeated syllables of words that he spoke. He started repeatedly blinking, spitting, and stroking his hair with his hand in a combing like fashion for no apparent reason. This persisted for about one to 2 years, during which the mood had improved and he remained euthymic.

After around 2 years of onset, the boy developed repeated self-grooming and hand washing behavior. He started counting with fingers for no purpose. These symptoms persisted for around 2 years, when the other behaviors had shown improvement. However, with this, his mood became elevated. He grew irritable, became over familiar, and developed abusive behavior. He started masturbating and disrobing in public. The symptoms of elevated mood gradually deteriorated to present admission when he had prominent features of increased appetite, increased sexuality, reduced sleep, inflated self-esteem, and an elated affect. During this time, the boy never reached premorbid levels in terms of his mood. The repetitive behaviors persisted with a fluctuating course, with a few predominantly present at any given time. He had developed repeated barking sounds around a year ago and started making pouting and sniffing gestures with his mouth, which persisted to current admission.

The boy has had multiple medical consultations in the past for his symptoms. He was initially treated with risperidone at 0.5 mg/day in 2009, which he discontinued within a few days due to poor perceived effect. Subsequently, he was given levodopa 100 mg+ carbidopa 25 mg per day, which he took for around 10 months. This was discontinued for a deterioration in symptoms, and he began taking olanzapine 5 mg/day. This resulted in some improvement in repetitive behavior but was discontinued after 3 months due to excessive sedation. He was then seen at our institute and given oxcarbazepine 450 mg and risperidone 0.5 mg per day which he discontinued within 1 month due to poor results. Following this, he started taking sodium valproate 300 mg/day, and risperidone 2.5 mg/day, to which he responded well and continued for next 1 year. However, all the symptoms persisted and when he stopped these medicines, they aggravated to present admission.

The boy was investigated with complete blood count, renal function test, liver function test, serum electrolytes and lipid levels, thyroid function test, urine for abnormal metabolites, electrocardiography, quantitative electroencephalography, computed tomography brain, magnetic resonance imaging brain, and ultrasonography whole abdomen, and also received a slit-lamp examination to rule out K-F rings. All of these were within normal range.

On admission, C-YBOCS "Obsession" score was 9, "Compulsion" score was 14 and a total score was 23. Score on YMRS was 39. Though the boy had 3 motor and 2 vocal tics at this time, his severity rating on YGTSS revealed mild impairment (score of 20). Children Global Assessment Scale (CGAS) score was 44 indicating a moderate degree of interference in functioning. His IQ, assessed on Malin's Intelligence Scale for children (MISIC) revealed a VQ of 70, a PQ of 69 and Full Scale IQ of 70.

The boy did not give any explanation for these behaviors, but reported that if he did not perform them, it made him feel tensed. He does not recognize them as being senseless and persisting without a cause, though he agrees to them being his own thoughts and not being imposed upon him by some external agency. However, there is no account of active resistance to these thoughts neither are they being perceived as unpleasantly repetitive. He does not describe any fear of harm being bestowed upon his near and dear ones if he does not perform them, any particular distress with asymmetry, or any account of repetitive images, impulses or ideas coming to his mind. There is no history of significant head injury, seizure episodes or of substance abuse.

The boy was started on haloperidol 5 mg, and carbamazepine 400 mg per day. After a period of 2 weeks, he started showing improvement in repetitive spitting, snorting, sniffing, and pouting. His mood symptoms also improved, but over next 1 week, deteriorated again. The treatment was reevaluated, carbamazepine was increased to 800 mg per day and was subsequently augmented with lithium carbonate. The mood symptoms showed improvement after 2 weeks on lithium, with the YMRS score reducing to 17 on serum lithium level of 0.8 mEq/L. Simultaneous psychoeducation to the parents regarding patient's illness and coping skills training, activity scheduling

for the boy to channelize the increased physical energy, and a schedule for differential reinforcement of other behaviors (DRO) and alternate behavior (DRA) helped rehabilitate the boy, and he was discharged after 6 weeks with a satisfactory improvement.

DISCUSSION

Although there is adequate evidence of the comorbid occurrence of Gilles de la Tourette syndrome and bipolar disorder from studies,[7] the etiology of the same remains unexplained. The two disorders may share several features such as abnormalities in neurotransmission involving noradrenaline, dopamine, and serotonin. Kerbeshian *et al*.,[8] reported that the candidate region for primary genetic factors resulting in the phenotype of the two disorders could be 16q22-23. The association has also been explained by a modulation defect,[9] in certain areas of the brain such as, the cortical-limbic system, ventral striatum, and sensory motor cortical regions. This construct is indirectly supported by literature reporting frequent precipitation of mania or bipolar disorder following stimulant use in patients with tics.[10]

Etiology aside, our case demonstrates certain obvious hurdles that a clinician might face with this comorbidity in pediatric age group. While on one hand an account of the characteristic features of tics might not be elicited; features of repetitive self-grooming, stomping, spitting, or other manifestations of tics might mimic motor hyperactivity of mania. It also poses problems with management, for if the boy is not able to recognize the premonitory urges, it also precludes the use of behavioral modification techniques. We suggest that under such circumstances, symptoms of increased appetite, hypersexuality, decreased need for sleep, and affective display might have better discriminative power.

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