

Case report on Tourette syndrome treated successfully with aripiprazole

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Summary: Tourette syndrome is a childhood-onset neuropsychiatric disorder characterized by multiple motor and vocal tics of at least one year in duration. This case report describes the history of a 16-year-old boy with a 6-year history of Tourette syndrome who was seriously disabled by his symptoms. Treatment with aripiprazole 10mg/d completely resolved his symptoms in about a month allowing him to return to the life that he had been missing because of his illness. In such cases the potential long-term negative effects of using antipsychotic medications need to be weighed against the disruptive effects persistent Tourette symptoms can have on patient's lives.

Keywords: Tourette syndrome, aripiprazole, case report, India

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1. Case history

A 16-year-old male presented to the psychiatric outpatient department with a 6-year history of repetitive involuntary blinking, shoulder shrugging, sniffing, vocalization, and whistling. The intensity and persistence of the symptoms waxed and waned spontaneously but he would never be completely symptom-free. Symptoms tended to be less severe in the summer and when engaged in physical activity but they would exacerbate when he was fatigued, stressed, or sitting idle. He could only voluntarily suppress the tics for a few minutes at a time, but this was associated with severe stress and restlessness. Over the past year new symptoms had emerged (grimacing and foot tapping), he felt increasingly embarrassed in public, his school grades had been deteriorating, and he was irritable throughout the day. Due to his loss of self-esteem he had not been attending school for two months.

He had a normal delivery with no antenatal or postnatal complications, achieved the normal developmental milestones, and was an average student in school. He was in 10th grade and had never failed a grade. His mother reported that prior to the development of tics he had a pleasant personality. There

was no history of psychotic symptoms, hyperactivity, obsessiveness, compulsiveness, or substance abuse. He has an older sister currently being treated for obsessive compulsive disorder but no other family history of mental disorders.

The patient's family had sought treatment 4 years previously (two years after the onset of symptoms). At that time he was treated with 10 mg/d haloperidol in divided doses. This treatment resolved the symptoms and he remained in remission for one year. However, he stopped the medication because of side effects (restlessness, tremors, slurred speech, motor retardation) and the symptoms re-emerged three months later.

Examination at the time of his outpatient visit revealed an irritable, anxious teenager with intact thought processes and higher mental functioning. Nervous system examination revealed excessive eye blinking, neck jerks, nasal sniffing, grunting, throat clearing, shoulder shrugging, grimacing, feet tapping, production of abnormal sounds, and whistling. Physical examination revealed no other abnormalities. Laboratory tests, including complete blood count, sedimentation rate, liver function tests, kidney function

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tests, thyroid function tests, urine analysis, and test for syphilis were all normal. A contrast-enhanced computed tomography scan of the brain was also normal.

He was started on aripiprazole 5mg/d at night but there was minimal improvement in 2 weeks so this was increased to 10mg/d at night. After 2 weeks at this dose there was a significant reduction in his symptoms, his vocal tic had disappeared but some motor tics and occasional grimacing remained. After an additional 2 weeks at the 10mg/d dosage his symptoms had completely disappeared. Subsequently his self-confidence improved and he was able to return to school two months after starting treatment. The family reported no side-effects and there was no evidence of the emergence of obsessive or compulsive symptoms. The treatment plan is to continue taking this dosage for six months (when he will complete his final examinations) and then attempt to gradually reduce (or stop) the medication.

2. Discussion

Tourette syndrome (TS) is characterized by sudden, involuntary, repetitive, non-rhythmic movements (i.e., tics) such as blinking, grimacing, head jerking, or shoulder shrugs. Complex motor tics consist of several simple motor acts occurring in an orchestrated sequence or semi-purposeful movements, such as touching or tapping. Simple phonic tics consist of simple, unarticulated sounds such as throat clearing, sniffing, grunting, and coughing. Tic episodes occur in bouts, which can be exacerbated by stress, fatigue, extremes of temperature, and external stimuli. Intentional movements attenuate tic occurrence over the affected area and concentration in activities tends to diminish tic symptoms.

The patient described in this report had a history of multiple motor and vocal tics that waxed and waned for 6 years, starting at the age of 10. This pattern of symptoms clearly meets the diagnostic criteria for TS,^[1-4] a condition that is more prevalent in males^[5,6] and usually affects the face (with blinking and grimacing) more than other parts of the body.^[7] Other reports suggest^[8] that OCD is more common in individuals with a history of TS. This patient did not manifest obsessive or compulsive symptoms but, given the family history of OCD, it will be necessary to continue monitoring him for the emergence of such symptoms.

Antipsychotic medications are the most effective treatment for tics but the common occurrence of weight gain and the increased risk of metabolic syndrome with chronic use has relegated them to a second-line medication, particularly in children.^[9] Alpha-2 agonists are currently considered first-line treatments; several randomized controlled trials over the last decade have demonstrated the effectiveness of pergolide, tetrabenazine and topiramate.^[10] But these medications are rarely used in India to treat TS, especially in children and adolescents.

We chose to use aripiprazole in this case because of its proven effectiveness in refractory cases,^[11] its relatively benign side effect profile, and the fact that the patient had previously responded to haloperidol (another D2 agonist). Some hypotheses about the etiology of TS focus on abnormalities in dopaminergic and noradrenergic neurotransmission in the frontoparietal network.^[12,13] So aripiprazole's unique mechanism of action as a partial agonist on the D2, 5HT2C, and 5HT1A receptors and as an antagonist of the 5 HT2A receptors may help explain its effectiveness in TS.

In this case the use of aripiprazole was life-changing for the patient; it returned him to society. Given ongoing concerns about the chronic use of antipsychotic medication, even those with limited side effects, long-term follow-up studies are needed to assess the relative cost benefits of different strategies – including the use of low-dose antipsychotic medications like aripiprazole – for treating this disabling condition.

Conflict of interest

The authors declare that they have no conflict of interest related to this manuscript

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Informed consent

The patient and his guardian signed an informed consent form and agreed to the publication of this case report.

阿立哌唑成功治疗抽动秽语综合征 1 例报告

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概述：抽动 - 秽语综合征 (Tourette syndrome, TS) 是一种儿童期发病的神经精神障碍，特征是多发性运动抽动和发声抽动，病程至少 1 年。本病例报告描述了一位 16 岁男孩患有抽动 - 秽语综合征 6 年，症状已使其功能严重受损。阿立哌唑 10 mg/d 治疗一个月左右后，该患者的症状完全消失，恢复了病前的生活。在这种情况下，需要权衡长期使用抗精神病药物潜在的不良

反应与持续的抽动秽语症状对患者生活造成的破坏性影响。

关键词：抽动秽语综合征，阿立哌唑，病例报告，印度

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