CASE REPORT

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Oromandibular Dystonia Treatment With Aripiprazole in an Adolescent Patient 2-Year Follow-Up: A Case Report

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ABSTRACT

Oromandibular dystonia (OMD) is a movement disorder that can cause considerable functional and psychosocial disability such as dysphagia, dysarthria, breathing difficulty, weight loss, social withdrawal, and depression. Oromandibular dystonia has no known cure. Many different treatment approaches can be used for the treatment of OMD. In this case report, we present a 16-year-old female adolescent patient with OMD, who showed a significant improvement with the use of 5 mg/day aripiprazole during the 2-year follow-up.

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INTRODUCTION

Dystonia is a movement disorder characterized by involuntary sustained or intermittent muscle contractions causing abnormal, often repetitive movements, postures, or both.[1] Oromandibular dystonia (OMD) is a focal dystonia involving the masticatory muscles, muscles of facial expression, tongue, and pharynx. The clinical presentation of OMD is varying degrees of jaw-opening, closing, malposition, as well as facial grimacing, abnormal tongue or pharyngeal movement, or a combination of these.[2-4] Functions of speech, facial expression, chewing, and swallowing can be affected in OMD and can cause considerable functional and psychosocial disability such as dysphagia, dysarthria, breathing difficulty, weight loss, pain, social withdrawal, and depression.[5,6] Oromandibular dystonia may be classified according to location (jaw-opening, jaw-closing, perioral, lingual, pharyngeal, or a combination) or cause; drug-induced (tardive dystonia), idiopathic (with blepharospasm Meige syndrome), neurodegenerative disorders, secondary basal ganglia/rostral brain stem disorders (head trauma, subcortical infarcts, infections, tumors, metabolic conditions), or peripherally induced (trauma, dental interventions).[2,5]

Oromandibular dystonia has no known cure and many different treatment approaches including medication, chemodenervation with botulinum neurotoxins (BoNTs) injections, local anesthetic injections, behavioral modification, and dental and surgical appliances can be used, alone or in combination, according to the cause.[1,2,7] Botulinum neurotoxins injections into the affected muscles have been reported to be statistically superior to medical therapy--particularly in focal dystonias--and are considered the treatment of first choice for oromandibular dystonias.[1,6,8] However, BoNTs injections in OMD treatment show 49.8% favorable response and a 27.1% occurrence of adverse events such as dysphagia, dysarthria, jaw weakness, and loss of smile.[9]

Aripiprazole is an atypical antipsychotic agent that is approved by the FDA for the treatment of schizophrenia, bipolar I disorder, and Tourette's disorder. It has a unique pharmacological profile that provides partial agonistic activity at dopamine D2 receptors (D2R) and serotonin 5-HT1A receptors (5-HT1AR), as well as antagonistic activity on serotonin 5-HT2A and 5-HT2C receptors.[10] We report herein the first case of an adolescent patient in whom OMD improved following treatment with aripiprazole.

CASE PRESENTATION

A 16-year-old girl who was taking no relevant medication and had no previous family history of movement disorders experienced sudden onset of speech impediment, deviation of the mouth toward the right side, limitation of mouth opening, tongue movements, and facial grimacing after the extraction of the right lower molar tooth 6 months previously. She was referred by the dentist to the pediatric neurology clinic. Facial paralysis was considered as the diagnosis in the neurological exam and electromyography (EMG) was ordered. The EMG recordings did not confirm

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the diagnosis of facial paralysis and the cranial magnetic resonance imaging revealed no abnormalities. Then she was referred to child psychiatry for differential diagnosis. She had depressive symptoms involving feelings of sadness, irritability, increased appetite, difficulty in concentrating, social withdrawal, and thoughts of death in the preceding 2 months. She was diagnosed with major depressive disorder (DSM-V) with scores of 34 on the Beck Depression Inventory (Clinical Global Severity of illness rating scale score: 5). Meanwhile, she was prescribed fluoxetine (20 mg/day). After the treatment with fluoxetine for 4 weeks, the depressive symptoms slightly diminished but the neurological symptoms persisted. Therefore, aripiprazole (5 mg/day) was added to the patient's medical treatment. After the aripiprazole treatment, the neurological symptoms gradually disappeared in a week and depressive symptoms were reduced in 3 weeks (Clinical Global Improvement Change rating scale score: 1). The patient was re-evaluated by a neurologist for diagnosis, and the neurological examination confirmed a diagnosis of OMD. Informed consent was obtained from the patient to report her case in this case study. The treatment was planned to continue, but she stopped taking her medication in the 4th month of the treatment, thinking she had been cured. Her symptoms, including speech impediment, deviation of the mouth, limitation of mouth opening, and tongue movements recurred. Since she did not have depressive symptoms, only aripiprazole (5 mg/day) treatment was resumed and her neurological symptoms diminished in a week. In the 16th month of the treatment, she stopped taking her medication for the second time, and the dystonia symptoms recurred. The treatment with aripiprazole (5 mg/day) was resumed once more and her medications have been maintained successfully for more than 2 years. During the follow-up, there was no side effect observed.

DISCUSSION

Many cases of peripherally induced OMD have been reported after dental treatment, but its pathophysiological mechanisms are not well understood.[7,9] The mechanism of dystonia is thought to be a central dysregulation of movement due to a defect that impairs physiological inhibitory control of the basal ganglia over the thalamus and brainstem.[10] The mechanism of peripherally induced dystonia is also thought to be a dysregulation in local neuronal circuitry, leading to alteration of synaptic transmission in the basal ganglia.[10]

Aripiprazole is used effectively in the treatment of Tourette's disorder and other tic disorders, though its mechanism of action is not fully known. Some studies and reports have demonstrated the efficacy of aripiprazole in the treatment of tardive dystonia, Pisa syndrome, and tardive dyskinesia (treatment with amantadine 200 mg/day).[11-14] On the other hand, some studies have also reported that aripiprazole induced parkinsonism, acute dystonia, tardive dystonia, and dyskinesia.[15-17] Both the induction and amelioration of movement disorders by aripiprazole may be related to its 'adaptive' pharmacological activity, meaning that its action as a full, moderate, or partial agonist for neurotransmitter receptors depends on the endogenous neurotransmitter levels and signaling status.[10] In addition, although somnolence, nausea, vomiting, extrapyramidal symptoms (EPS), and metabolic side effects such as weight gain may occur with aripiprazole, similar to atypical antipsychotics, the severity of EPS and metabolic side effects is less due to receptor specificity.[18]

In our case, a significant improvement was observed during the 2-year follow-up with the use of 5 mg/day aripiprazole. During the 2-year low-dose aripiprazole treatment, there were no other movement disorders or possible side effects, and relapse of symptoms during the discontinuation period and the improvement of symptoms with the initiation of the drug support the safety and efficacy of aripiprazole.

We suggest that low-dose aripiprazole may be effective and safe in the treatment of peripherally induced OMD, of which the favorable response rate in the treatment as the first choice is approximately 50%.[9]

Informed Consent: Written informed consent was obtained from the patient to report her case in this case study.

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