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Oromandibular dystonia seen during pramipexole treatment: A rare case

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Abstract

Dystonia is an abnormal, often repetitive, bending/twisting behavioral disorder characterized by continuous or intermittent muscle contraction. Oromandibular dystonia (OMD) is a type of dystonia involving chewing, mouth circumference, tongue, and platysma muscles. OMD is divided into different clinical types, including jaw opening OMD, jaw closing OMD, and mixed type OMD. OMD may either be primary or secondary to other diseases. The average patient age is between 50 and 60 years, and several studies have shown that it is more common among women. Dystonia may occur either as idiopathic (primary) or resulting from neurodegenerative diseases and other secondary dystonia. OMD can cause difficulty in speaking, chewing, and swallowing and produce pain during these movements. Therefore, OMD can lead to deterioration in an individual's daily life and social relationships. Although dopaminergic drugs can be used in the treatment of dystonia, the aim of the study was to report that these drugs may also be a factor in further development of dystonia and to attract the attention of clinicians to this anomaly.

Keywords: Oromandibular dystonia, Pramipexole, Dopamine agonist, Restless leg syndrome

Introduction

Dystonia is a continuous or intermittent muscular contraction-induced movement disorder that can often involve repetitive motions and involuntary bending/twisting. Dystonic movements are characterized by bending or sometimes tremors. Dystonic movements almost always become intensified during voluntary acts. Expansion of dystonic posture to close and even remote muscles during a volitional act is defined as 'overflow' [1]. Dystonia is divided into two categories depending on its etiology and clinical features, such as age of onset, body distribution, time-related features, and co-occurrence with other movement disorders or neurological or systemic symptoms [2]. A diagnosis of dystonia is based on its clinical features.

Oromandibular dystonia (OMD) is a type of dystonia involving masticatory, circumoral, lingual, and platysma muscles. OMD has different clinical sub-types, including jaw-opening OMD, jaw-closing OMD, and mixed type OMD. OMD can be a primary disorder or secondary to another disease. Patients are classified as having focal, segmental, multifocal, and generalized dystonia depending on the bodily distribution of dystonia. While OMD may appear focally, it is mostly a part of segmental and generalized dystonia [3]. The average age of incidence occurs between 50 and 60 years of age. Various studies have reported that OMD occurs more in females [3, 4]. OMD may lead to difficulty in speaking, chewing, and/or swallowing and cause pain during these activities that lead to impairment of daily life and social relationships [5].

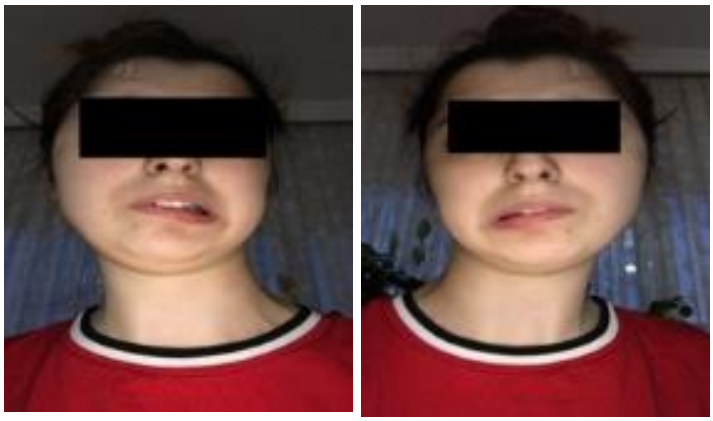
The aim of this article was to report that while dopaminergic drugs have a role in dystonia treatment, oromandibular dystonia may appear as an adverse acute reaction during pramipexole treatment and to bring this anomaly to the attention of clinicians.

Case presentation

A 25-year-old single female high-school graduate presented to the Neurology Polyclinic with complaints of an abnormal and displeasing sensation that had been occurring for a couple of years on her feet, especially during rest periods and at night. These complaints were forcing her to move her legs uncontrollably as her complaints were relieved when she stood up and wandered around. The patient had no other characteristics in her medical history. The patient received pramipexole 0.250 mg 1x1 tb treatment. The patient presented to our hospital's emergency department two days after the start of treatment as she had deviation to the right and left in perioral muscles and lips (Figures 1A and 1B). The patient was administered one ampoule (5 mg) of intramuscular biperidene treatment in the emergency department. The patient's complaints were relieved after administration of the biperiden treatment. The patient's neurological examination performed in the emergency department was completely normal except for the finding of dystonia in the perioral muscles. No pathology was found based on patient's routine blood tests, brain magnetic resonance imaging, electroencephalography, and electromyography (MRI, EEG and EMG, respectively) examinations performed in the neurology clinic for determining the etiology of her dystonia. The patient was discharged with recommendations.

Figure 1A: Oromandibular dystonia (OMD) in the lips and perioral muscles in the form of left shift

Figure 1B: OMD in the lips and perioral muscles in the form of right shift



Discussion

Dystonia is a movement disorder characterized by repetitive, bending, twitching, painful, and prolonged muscle contractions in the affected body part. OMD is a focal form of dystonia where the facial, lingual and jaw muscles are affected. While dystonia may be idiopathic (primary), it may also be induced by neurodegenerative diseases and other secondary issues associated with dystonia.

In the literature, no case reports concerning the occurrence of pramipexole-induced oromandibular dystonia can be found. Only a few case reports on pramipexole-induced limb dystonia and antecollis in Parkinson's disease are available. In one case, extremity dystonia with sub-acute onset following pramipexole treatment was reported in a Parkinson's disease patient receiving levodopa/carpidopa treatment. In a previous case report on pramipexole-induced extremity dystonia in Parkinson's disease, the authors presumed that although pramipexole showed a rather low affinity for serotonergic receptors, dystonia in the extremities occurred resulting from a potentially reactive fibrosis appearing at the serotonergic 5 HT2A and 5HT2B receptors [6]. However, the further increase in dystonia severity following cessation of pramipexole treatment suggests that the source of dystonia cannot be explained by a simple fibrotic complication. Moreover, abnormal activation of D1 receptors is a mechanism that could play a critical role in the occurrence of dystonia [7]. Activation of D3 receptors in the striatum has synergistic effects on D1 receptor-mediated transmission. Pramipexole shows a more evident affinity primarily for D3 receptors [8]. Antecollis is a form of dystonia that was first been shown in multi-system atrophy (MSA) patients, and it is a helpful indicator used in the diagnosis of MSA disease [9]. Antecollis is relatively rare in Parkinson's disease, and its pathophysiology is uncertain. However, several central and peripheral mechanisms have been suggested to be responsible for causing this condition [10]. In literature, antecollis cases associated with pramipexole treatment in Parkinson's disease have been reported [11]. In another case report, a female patient receiving trihexiphenidyl, pramipexole, and resagiline triple treatment was reported to have jaw dystonia characterized by lateral deviation to the right and left that was seen after the addition of levodopa/carpidopa to her current treatment. This jaw dystonia was reported to have occurred at the time when levodopa/carbidopa treatment reached its peak level in the blood. It was observed that while jaw dystonia disappeared with the gradual reduction in levodopa/carbidopa treatment, the on–off phenomenon appeared in the patient. Controlled release forms of Levodopa/carbidopa were found to be ineffective in the treatment of dystonia [12]. In the literature, two case reports concerning oromandibular dystonia associated with levodopa/carbidopa treatment are available. One of these patients was treated for Parkinson's disease, and the other was treated for progressive supra-nuclear palsy [13, 14]. Levodopa-induced dyskinesia is mainly and typically characterized in the choreiform and is noticed seen when Levodopa reaches its peak dose level. Dyskinesia may appear in the form of dyskinesia less frequently and is presumed to be in the form of dystonia in biphasic dyskinesia with a wearing off pattern. Dystonia, which Levodopa can induce as a peak dose phenomenon, is quite rare [15].

Conclusion

Clinicians should consider dopamine agonists as a factor leading to the development of dystonia because this condition can be reversed when recognized early. In our case, it was not possible to explain how pramipexole caused acute dystonia. Yet, it is considered that OMD could appear due to abnormal stimulation in D1 receptors resulting from pramipexole stimulation of D3 receptors. Moreover, the genetic makeup of the patient is assumed to be a factor that is influential in the development of pramipexole-induced acute dystonia. Further studies could yield a better understanding of the pathophysiology of the development of pramipexole-induced OMD.

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