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Case Study

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OROMANDIBULAR DYSTONIA: A RARE CASE REPORT

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ABSTRACT

Dystonia can be defined as involuntary, intermittent, repetitive, sustained or spasmodic muscle contraction resulting in abnormal muscle movements and posturing. Oromandibular dystonia (OMD) is mainly concerned with the involuntary hyperkinetic movements of jaw, tongue, muscles of mastication etc. Etiology of this condition is varied like brain injury involving basal ganglia, ischemia of brain, drugs, trauma, metabolic or toxic states, neurodegenerative diseases and predisposing factors include like bruxism, antipsychotic drugs, ill-fitting dentures etc. This condition severely affects the quality of life of patients. With a similar condition, we report a case in our department diagnosed as OMD and treated with monoamine depletors.

KEYWORDS: Hyperkinetic movement, Monoamine depletors, Oromandibular dystonia (OMD).

INTRODUCTION

Oromandibular dystonia (OMD) is focal dystonia involving the mouth, jaw, and tongue causing involuntary mouth closure or opening, deviation of the jaw, facial grimacing, or tongue movements.[1] Deviation of the mandible, dysphagia, dysphonia, subluxation, intraoral soft tissue trauma and bony resorption can be caused by involuntary movement of jaws.[2] As high as 6.9/100,000 cases of OMD were noted and the incidence has been reported up to 3.3 cases per million.[3] Dystonia is thought to be underdiagnosed because of its similarity to other movement disorders, including dyskinesia, hemifacial spasm, muscular effects of temporomandibular disorders, and bruxism.[4]

Dystonia can be anatomically categorized as focal (affecting one or two parts of the body), segmental, generalized and multifocal and based on etiology, it is categorized as Primary dystonia and Secondary dystonia. Primary dystonia is idiopathic or inherited and secondary dystonia is seen following traumatic or surgical incidents, brain diseases, and by taking certain medications.[5] In regard to these cases, the pathology is cleared out using magnetic resonance imaging of the brain and spinal cord.[6]

OMD is seen in frequent association with orobuccolingual dyskinesia's like lip pursing and biting, facial grimacing, tongue protrusion, bruxism and platysma contractions.[7] Mechanism of dystonia is not specifically known but may arise from centrally mediated dysregulation of movement due to defect in the basal ganglia, especially in the sensory-motor regions of the putamen.[8] Management of OMD can be broadly divided into medical management and surgical management. Medical management involve the use of centrally acting medications, chemodenervation using botulinum neurotoxin and surgical management including deep brain stimulation, pallidotomy or myectomy etc.[9] Careful approach to diagnosis and management can totally eliminate the OMD.

CASE REPORT

A 70 year male patient reported to oral and maxillofacial surgery department with a chief complaint of severe pain in upper jaw region since 7days "Fig 1".

Patient was apparently normal seven days back when he gradually developed pain in maxillary alveolar region which was severe, intermittent and radiating towards both temporal regions. Pain was so severe that he

couldn't sleep properly at night. His past medical history was unremarkable but past habit revealed that he was smoker and alcoholic since forty years.

Fig.1
Preoperative front view of patient.

Fig.2
Preoperative lateral view of patient showing hyperkinetic movement of lower jaw.

On clinical examination, multiple ulcers were seen over alveolar mucosa of maxilla. Ulcers were pale pink and surrounded by erythematous halo suggesting healing ulcers. It was observed that remaining lower teeth were impinging over maxillary alveolar mucosa. On clinical basis a diagnosis was made as traumatic ulcers over maxillary alveolar mucosa and extraction of remaining mandibular root pieces were done as treatment.

Seven days later patient reported with similar symptoms. It was observed that his lower jaw was showing hyperkinetic movements which was impinging on healing ulcers Fig 2. A thorough history revealed that patient had history of spontaneous lower jaw movement since 6 months and he was unable to maintain his lower jaw in one position. These movements were episodic in nature. On clinical examination, there was an involuntary movement of mandible in opening and closing of jaw. Irregular movement and inward folding of tongue was also observed. The patient showed signs of dysphagia and dysarthria. Mouth opening was adequate and no abnormality of the temporomandibular joint was noted. No signs of abnormal blinking of eyes were seen and no tenderness observed over the region of the muscles of mastication.

Thus patient was advised MRI scan of brain which revealed that multiple ischemic changes in basal ganglia and mild ischemic changes in bilateral frontoparietal and periventricular white matter "Fig. 3". On clinical as well as radio graphical basis, a diagnosis was made as oromandibular dystonia. Differential diagnoses were myokymia, facial myokymia, meig syndrome and Tourette syndrome. Neurosurgeon's opinion was taken and medicinal approach was started. He was given Tab tetrabenazine 25mg (Monoamine depletors to reduce involuntary movement) twice a day, Tab lorazepam 0.25mg (Benzodiazepines to reduce the anxiety) once a day, Tab aspirin 75mg (antiplatelet aggregating agent to treat the ischemic condition) once a day.

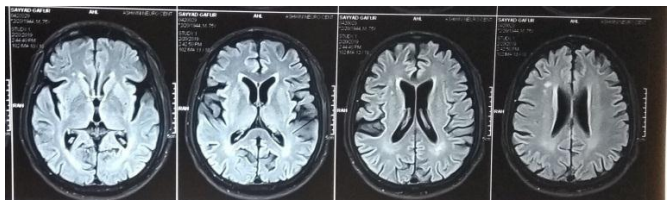

Fig 3
MRI scan of brain showing multiple ischemic changes in basal ganglia.

Follow up of patient was done every week till two months and after that every month till six months. Till the first fifteen days, there was a gradual reduction in involuntary movement of the lower jaw and after two months of therapy involuntary movements of the jaw were totally reduced "Fig 4".

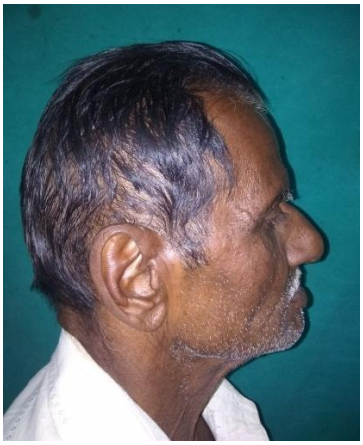

Fig. 4
Postoperative lateral view of patients after 1 month showing reduced hyperkinetic movement.

DISCUSSION

The causes of oromandibular dystonia are mostly unknown but sankhla et al in her studies reported 27 patients with anatomically and temporally related to a prior trauma to the face or mouth. They also describe the predisposing factors like family history of movement disorders, prior exposure to neuroleptic drugs, associated dystonia affecting other regions or essential tremor.[1] Balasubramaniam in his literature said that OMDs are caused due to centrally mediated pathologic changes involving basal ganglia, cerebral cortex, subthalamus, substantia nigra and their communication with other area of brain. He also said that alchol intake may cause OMDs.[9] Various therapeutic modalities which are promising in successfully controlling the symptoms are the therapeutic medications such as tetrabenazine, diazepam, and carbamazepine. Anticholinergic drugs reduce muscle spasm by centrally inhibiting the parasympathetic system.[10]

Gandhi Y reported a similar case treated with medicinal and surgical approach as Tetrabenazine 25 mg and Haloperidol 0.75 mg twice daily, along with a nocturnal bite guard. Simultaneously they treated with botulinm toxin A into masseter and temporalis muscles, followed by coroniodectomy and medial debulking of the masseter on the right side.[2] Papapetropoulos S and Singer C reported a 5 similar case in which they said that combination of medicinal (Tetrabenazine) and Inj. botilinum toxin A are effective.[7]

Leek in his literature said that treatment approaches used to manage OMD include medication, botulinum toxin, local anesthetic blocks, dental appliances, behavioral modification and psychological support, and denervation procedures. Oral medication is the usual first line of treatment, with BTX injection to affected muscle with or without EMG guidance a second-line therapy. They also said that surgical procedures do not have a proven role in treating OMD but being associated with worsening outcome in most cases.[14] Suma Gn reported a similar case and treated with carbamazepine as a medicinal approach.[15] Similarly in our case only medicinal approach was sufficient to resolve the condition. Most of the cases with OMD are successfully treated with Inj. botulinum toxin after medicinal approach.[11-13]

CONCLUSION

Early detection of OMD and understanding of anatomy which is responsible for characteristics clinical sign and symptoms plays a significant role in treatment of this condition. 1st line treatment for this condition is medicinal approach, most of the cases resolved with medicine. If the condition persist then further management will require like Inj. botulinum, surgical procedures as myectomy, pallidotomy etc.

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