

Dental Implants Leading To Oro Mandibular Dystonia - A Case Report

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Abstract

A rare focal form of dystonia, Oromandibular dystonia (OMD) is caused by prolonged muscles spasms in the mouth, face, and jaw. OMD can develop after dental treatment, as poorly aligned dentures or multiple tooth extraction, immediate implants which may cause an impairment of proprioception in the oral cavity, leading to the subsequent development of dystonia. While chewing, swallowing, and speaking, these repetitive involuntary jaw movements interfere. We report here a case of OMD after dental implants rehabilitation.

Key words: Tooth extraction, Dystonic disorders, Jaw, Implant

Introduction

Dystonia is a neurological movement disorder wherein sustained muscle contractions cause twisting and repetitive movements or abnormal postures. The disorder may be hereditary or may be caused by other factors such as birth related or other physical trauma, infection, poisoning (e.g., lead poisoning), or reaction to pharmaceutical drugs particularly neuroleptics¹.

A rare form of focal dystonia, Oromandibular dystonia (OMD) involve the masticatory muscles, muscles of facial expression, and those of the tongue and pharynx. It is involuntary, inappropriate, repetitive, or sustained muscle contractions cause varying degrees of jaw opening, closing, deviation, protrusion, or retrusion as well as facial grimacing, abnormal tongue or pharyngeal movement.^{2,3}

OMD is a rare, often misdiagnosed disease that is difficult to manage. Its treatment has been limited to minimizing the symptoms of the disorder^{1,2}. The mechanism of OMD is not well-understood. Some cases of OMD after dental treatment have been reported, although the causal relationship between these procedures and dystonia is still unclear^{4,5}.

This case report describe a case of peripherally induced OMD. Here we sought to discuss some aspects of clinical manifestations, diagnostic criteria, mechanisms, and treatment options for OMD as the onset of dystonia occurred after a dental procedure.

Case Report

A 68 year-old male came with a 6 month history of abnormal jaw movements , visible twitching of oro facial musculature



Figure.1 a,b Profile - Patient

causing difficult in speech at rest and function both.(Figure.1a,b) According to the history given, his problem started 6 months ago when implants were restored in upper arch .

On oral examination, Implant supported prosthesis is seen, limited to anteriors only with partial unilateral occlusion was present on right side and multiple teeth were missing on left side. OPG revealed failing dentition in lower jaw (Figure.2) but non tender. Bilateral TMJ joint were palpable and non tender.

Implants were placed 10 months ago after extraction of failing teeth. After 4 months of time period, prosthesis was delivered following which present state of discomfort started. He was healthy, with no significant medical history or family history of

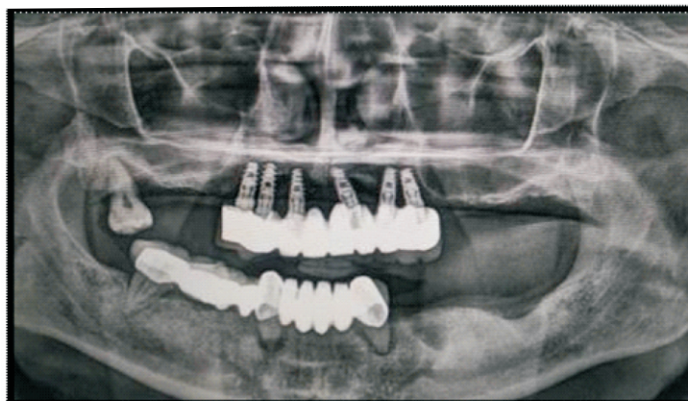


Figure.2 - OPG of The Patient

neurological disorder.

Some examination like CBCT and MRI was conducted but nothing abnormal finding was detected. As far as our clinical examination , it is a real case of muscular dystonia happened with possible association of altered vertical dimension after implant retained prosthesis was delivered, with only occlusion in anterior region.

Patient was advised to remove the prosthesis but in order to reduce the acute discomfort, he was referred to neuro-physician. Upon taking neuro-muscular/ skeletal muscular relaxants , the condition of patient is quite better but medication is still on and even now the exact cause of the situation remains a question mark.

Patient is under follow up and quite apprehensive to remove the cement retained prosthesis.

Discussion

Dystonia is a persistent posture from the co-contraction of agonists and antagonists and is generally considered part of the spectrum of dyskinesia^{2,6}. Dyskinesia consists of adverse effects including diminished voluntary movements and presence of involuntary movements, similar to tics or chorea¹. Dystonia is a neurological movement disorder rarely seen by oral and maxillofacial surgeons⁷ wherein sustained muscle contractions cause twisting, repetitive movements, or abnormal postures^{1,2,5}. The movements may be painful, and some individuals with

dystonia may have tremor or other neurological features. Several different forms of dystonia may affect only one muscle, groups of muscles, or muscles throughout the body^{1,8,9}. The cause of majority of the cases is not known. Hereditary or other factors such as birth related, other physical trauma, infection, poisoning (e.g., lead poisoning), and reaction to pharmaceutical drugs particularly neuroleptics are presumed to be the possible causes⁸⁻¹⁰. There are different ways of dystonia classification - it can be classified by etiology into primary (also referred to as idiopathic, inherited, or familial) and secondary forms (also referred to as acquired). Primary dystonia refers to dystonia that does not have a clear cause. Many instances of dystonia are idiopathic. The secondary form of dystonia develops due to environmental factors, long-term neuroleptic drugs (tardive dystonia), neurological disease, brain injury, Wilson's disease, and peripheral trauma. The most common form is tardive dystonia, which develops as a side effect of long-term treatment with antipsychotic drugs¹¹.

Dystonia can also be classified by age of onset, anatomically by region of distribution such as focal, segmental, multifocal, and generalized, and further into the affected body parts^{2,12,13}. OMD is focal dystonia involving the masticatory muscles, muscles of facial expression, and those of the tongue and pharynx. The condition of muscle get worsened over the year due to involuntary, inappropriate, repetitive, or sustained muscle contractions cause varying degrees of jaw opening, closing, deviation, protrusion, or retrusion as well as facial grimacing, abnormal tongue, or pharyngeal movement.^{2,3}

OMD is classified as jaw opening, jaw closing, jaw deviating, lingual dystonia, or combinations of these^{2,4,8}. Focal OMD is extremely rare. The prevalence of OMD varies, reportedly as high as 6.9 cases per 100,000. Women seem to be affected more frequently than men, with the onset typically between the age of 45 and 70 years¹⁴. The pathophysiology of dystonia is unclear but is thought to originate in the centrally mediated dysregulation of movement due to defect in the basal ganglia particularly in the sensory motor regions of the putamen. The mechanism of peripherally induced dystonia - as illustrated by OMD - is also based on the theory of sensory pathway disruption at the level of the basal ganglia¹⁵⁻¹⁷.

Dental intervention can be regarded as peripheral iatrogenic injury. Several cases of OMD following dental procedures have been reported.^{3,7,18} Thompson et al. reported development of OMD following dental extraction in a woman⁸. Traumatic situations in the mouth - such as poorly aligned dentures, immediate implantation or multiple extractions - may cause an impairment of the proprioception of the oral cavity, leading to the subsequent development of dystonia^{8,19}. The muscle condition worsened over the year. Note, however, that the causal relationship between these procedures and dystonia is still unclear⁴. OMD is difficult to manage, and its treatment has been limited to minimizing the symptoms of the disorder. Note, however, that there are several treatment options that can relieve some of the symptoms of dystonia, so physicians can select a therapeutic approach based on each individual's symptoms^{12,13}. There are various approaches for treating and managing OMD, which include medication, BTX, local anesthetic blocks, dental appliances, behavioral modification and psychological support, and surgical procedures^{12,13}. Oral medication is the usual first line of treatment, but there is no

medication to prevent dystonia or slow its progression. In case of large studies Tetrabenazine, clonazepam, or other oral drugs have been assessed in a systematic way. The results of oral medication for OMD have been largely disappointing. Some authors find the oral medication of baclofen to be useful for OMD^{10,13,18}. BTX injection into the affected muscle with or without EMG guidance is a second-line therapy^{7,13}. Botox has been proven to be superior to medical treatment particularly in focal dystonias²⁰. Injections of small amounts of this chemical into the affected muscles prevent muscle contractions, and they can provide temporary improvement in abnormal postures and movements characterizing dystonia⁷. The toxin decreases muscle spasms by blocking the release of the neurotransmitter acetylcholine, which normally causes muscles to contract. The effect is typically seen a few days after the injections, and it can last for several months before the injections need to be repeated^{2,7}.

Physical therapy, use of splints, stress management, and biofeedback may also help individuals with certain forms of dystonia^{12,13}. Surgical therapies include peripheral and central surgery. Peripheral denervation or myectomy is seldom needed since OMD usually responds well to BTX and must be delayed while other treatment options are effective^{12,13}.

This peripherally induced OMD case followed dental procedure, with no family history of movement disorders, organic brain lesion, or exposure to neuroleptic drugs. Hence in such case the only predisposing factor was the recent dental extraction and prosthetic treatment. The patient experienced a short latency period between the extraction and onset of dystonia, and the symptoms worsened over time. The close association of time, location of the procedure, and onset of symptoms suggests that the onset of dystonia may have been caused by the dental intervention. Because of its rare occurrence, patients with OMD are probably often misdiagnosed, or their diagnosis may be delayed. Consequently, these patients may also receive incorrect dental treatment, and the symptoms may worsen over the years.

OMD may be caused or aggravated by some of the other dental and prosthetic problems. The possible dystonic movements and forces and subtype of OMD should be considered while planning the dental treatment and procedure.

Conclusion

Because of its rare occurrence, patients with OMDs are probably often misdiagnosed, or their diagnosis may be delayed. Consequently, these patients may also receive incorrect treatment, and the symptoms may worsen over the years. This case gives us the lesson that OMD should be born in mind while evaluating a patient for dental procedure.

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