CASE REPORT

http://dx.doi.org/10.5125/jkaoms.2012.38.6.379
pISSN 2234-7550 · eISSN 2234-5930

Oromandibular dystonia after dental treatments: a report of two cases

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Abstract (J Korean Assoc Oral Maxillofac Surg 2012;38:379-83)

Oromandibular dystonia (OMD) is a rare focal form of dystonia 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 may cause an impairment of proprioception in the oral cavity, leading to the subsequent development of dystonia. These repetitive involuntary jaw movements may interfere with chewing, swallowing, and speaking. We report here two cases of OMD after dental procedures.

Key words: Tooth extraction, Dystonic disorders, Jaw, Tongue, Focal

[paper submitted 2012. 10. 31 / revised 2012. 11. 27 / accepted 2012. 11. 27]

I. 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 neuroleptics[1].

Oromandibular dystonia (OMD) is focal dystonia involving the masticatory muscles, muscles of facial expression, and those of the tongue and pharynx. 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, or any combination of these[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 paper describes 2 cases of peripherally induced OMD. Since the onset of dystonia occurred after a dental procedure, we sought to discuss some aspects of clinical manifestations, diagnostic criteria, mechanisms, and treatment options for OMD.

II. Cases Report

1. Case 1

A 59-year-old female with a 12-month history of abnormal jaw protrusive movement and opening was referred by a local practitioner. According to her, her problem started during the dental extraction of her lower right molar teeth and immediate implantation (Fig. 1), and the symptoms worsened over time. Although the local practitioner tried to adjust the occlusion, the symptom was not relieved. She was healthy, with no significant medical history or family history of neurological disorder.

The intra-oral examination revealed severe dental attrition of her residual teeth and unstable occlusion.(Fig. 2) The extraoral examination confirmed the involuntary protrusive

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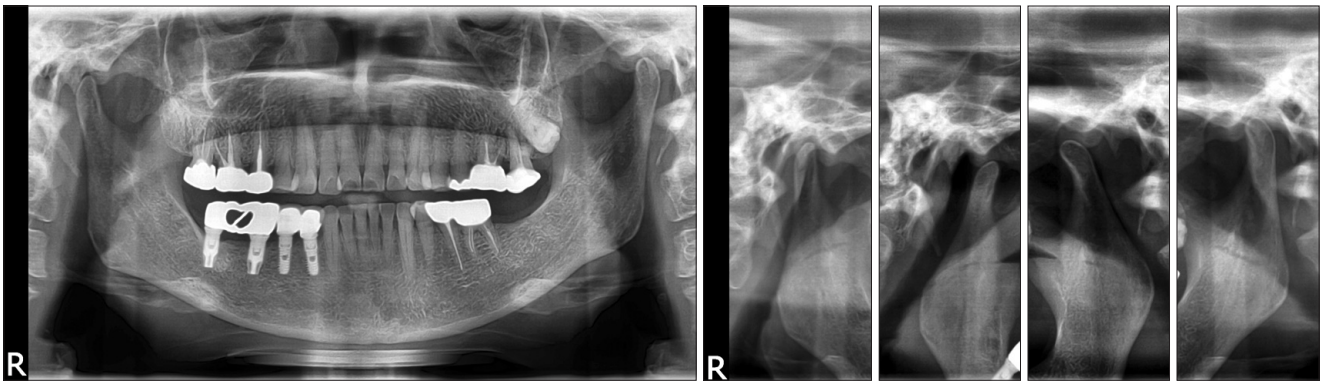

Fig. 1. Radiological examination at the initial visit.
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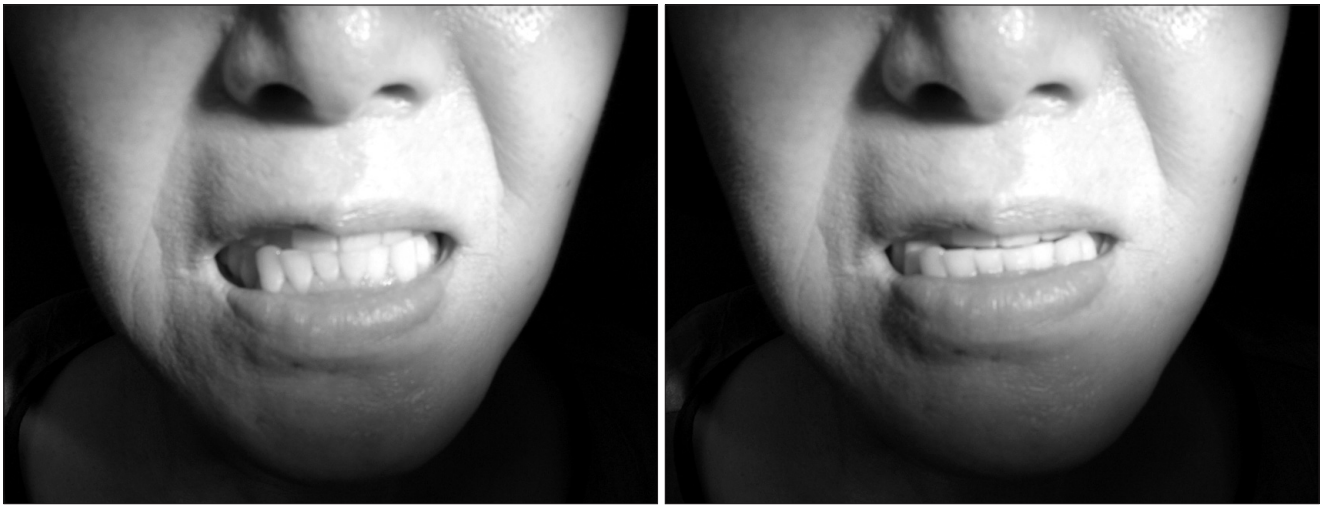

Fig. 2. Clinical photo revealing unstable occlusion.
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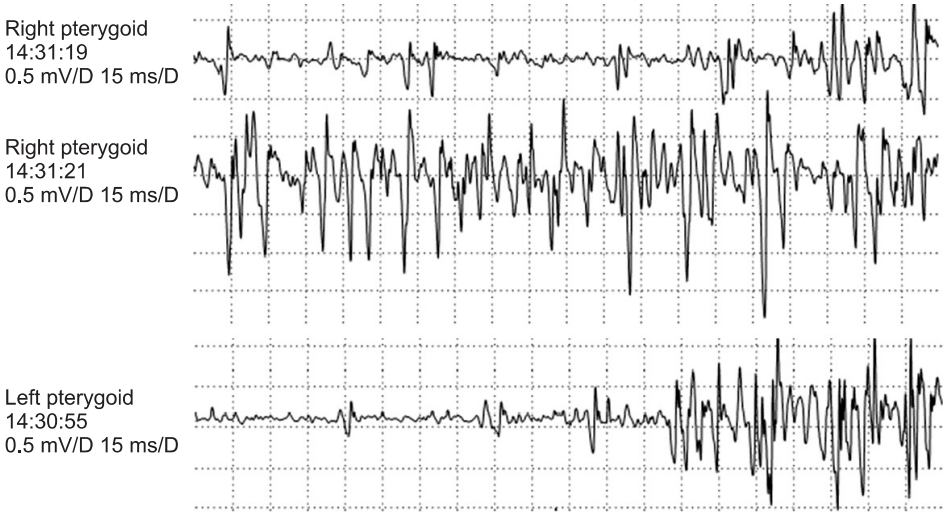

Fig. 3. Electromyography activity was typically reflected as significant high-frequency and high-voltage activity of motor unit potentials with either sustained or short-duration bursts of discharge patterns at rest when normally electrically inactive.
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and opening movement with slight deviation toward the right side.

She was prescribed baclofen (Baclan; Yooyoung Pharmaceutical Co., Ltd., Seoul, Korea) 10 mg three times a day and analgesics for the relief of chronic muscle spasm. Despite the treatment with a muscle relaxant for three weeks, the symptoms persisted.

She was referred to a neurologist for electromyography (EMG).(Fig. 3) The EMG recordings revealed that her problem was due to a spasm of the lateral pterygoid muscle. Botulinum toxin (BTX) A (Meditoxin; Pacific Pharma, Seoul, Korea) was injected at a dose of 30 units per muscle via an extraoral approach.(Fig. 4) A week later, she showed a definite reduction of the dystonic movement, becoming symptom-free. No side effect was observed save for 26 mm of active range of motion. A diagnosis of jaw opening and protrusive OMD was made.

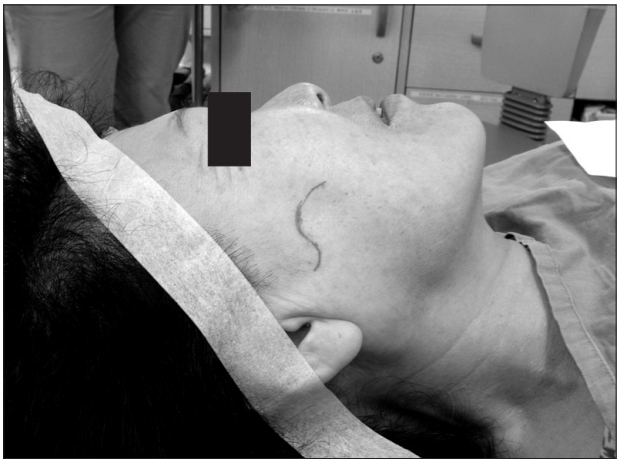

Fig. 4. Botulinum toxin injection was done via the extraoral approach.
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2. Case 2

A 57-year-old female with no previous history of movement disorders and who was taking no relevant medication experienced sudden onset of involuntary jaw tremor, limitation of mouth opening, and protrusive tongue movements following the extraction of both lower molar teeth and immediate partial denture 6 months ago.(Fig. 5) The symptoms worsened over time and interfered with her speaking and eating.

The clinical examination revealed the denture to be generally unstable. She had spontaneous tongue movements even when she was not speaking. The movements worsened when her dentures were removed.

She had no other relevant medical and family history. She was referred to the neurologist for a neurological exam. The cranial magnetic resonance imaging revealed no abnormalities, and the result of the neurological examination was normal save for the presence of abnormal movement. A diagnosis of lingual protrusive OMD was made.

She was prescribed baclofen, analgesics, and occlusal stabilization appliance. After oral medication and oral appliance therapy for 1 month, the symptom improved, and she was referred to a prosthodontist for the repair of her old, ill-fitting denture.

At the follow-up 1 month later, mild tongue tremor with protrusion and jaw tremor were observed. BTX injection into the genioglossus muscle was recommended, but the patient refused because of financial problems and the symptoms did not cause any disability. Only the oral medication of baclofen was continued.

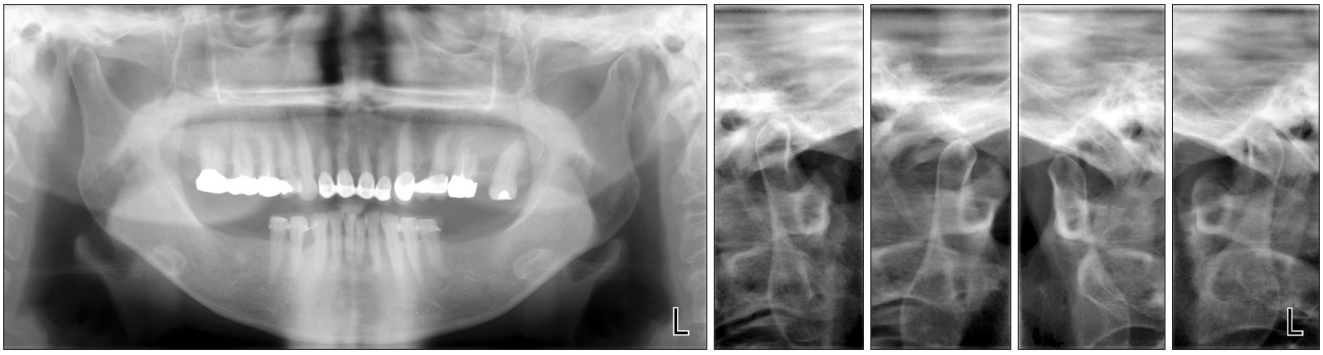

Fig. 5. Radiological examination at the initial visit.
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III. Discussion

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 chorea[1]. Dystonia is a neurological movement disorder rarely seen by oral and maxillofacial surgeons[7] 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 causes[8-10].

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 drugs[11].

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. 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 or any combination of these, with the muscle condition worsening over the year[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 years[14].

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 ganglia[15-17].

Dental intervention can be regarded as peripheral iatrogenic injury. There have been several reports of OMD following dental procedures[3,7,18]. Thompson et al. reported one woman who developed OMD following dental extraction[8]. Traumatic situations in the mouth - such as poorly aligned dentures 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 unclear[4].

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]. Treatment approaches used to manage OMD 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. Tetrabenazine, clonazepam, or other oral drugs have been assessed in a systematic way in large studies. 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 dystonias[20]. 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 dystonia[7]. 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].

These 2 peripherally induced OMD cases followed dental procedure, with no family history of movement disorders, organic brain lesion, or exposure to neuroleptic drugs. The only predisposing factor was the recent dental extraction and prosthetic treatment. These 2 patients 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. Thus, in planning the dental treatment and procedure, the subtype of OMD and the possible dystonic movements and forces should be considered.

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