



Eileen Watt

Indiya Sangani, Fiona Crawford and Toby Gillgrass

The Role of a Dentist in Managing Patients with Dystonia

Abstract: Oromandibular dystonia is a neurological condition where the muscles of the mouth and jaw contract in involuntary spasms. Dystonia currently has no cure, but there are treatment options to manage symptoms. This case report describes the treatment of a 41-year-old male who presented at University of Glasgow Dental Hospital and School with established oromandibular dystonia. The use of removable appliances as a treatment option for this condition will be discussed.

Clinical Relevance: This case highlights the potential role of the dental practitioner in providing oromandibular dystonia sufferers with an effective and relatively easy treatment modality in the form of a removable appliance.

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Dental treatment of oromandibular dystonia has not been widely reported in dental literature to date. This report will outline a case of oromandibular dystonia that was treated within Glasgow Dental Hospital and School. We hope to describe this debilitating condition, its signs and the beneficial effect of removable appliances in this case.

Background

Dystonia is a neurological movement disorder, affecting both children and adults, and its prevalence is reported to be 1 in 900. Dystonia can affect different parts of the body and is named accordingly,

that affecting the oral region being described as oromandibular dystonia.

Dystonias may be classified according to the age of onset, or whether they are task specific. Focal (affecting one or two parts of the body) task specific dystonia affects one part of the body and only occurs during that performance or task.¹ Typically, it affects the hands and may be related to a sporting activity or playing a musical instrument with the hands, or it can affect the oral musculature in wind or brass players. Adult-onset dystonia is usually focal, while early-onset dystonia often generalizes to affect multiple parts of the body.²

Oromandibular dystonia describes the condition that affects the muscles that move the face, tongue and/or jaw, resulting in involuntary spasms or forceful contractions of these muscles. Women are more affected than men and it commonly occurs between the ages of 40 and 70. It is a physically and psychologically disabling condition that tends to present in those who use the muscles of the mouth more than the average person, classically involving brass and woodwind musicians.³ Dystonia that affects the facial muscles and lips of musicians who play wind instruments is called embouchure dystonia.

Embouchure refers to the adjustment of the mouth to fit the mouthpiece of a wind instrument. Embouchure dystonia may be further subdivided into the anatomy affected: lip, tongue, jaw, etc.⁴ The aetiology of the condition is poorly understood, although current theory suggests it involves the over activity of the central cortex within the brain.⁵ This may be caused by the repetitive nature of making sounds in music, that acts to strengthen the normal sensory pathways to the point where they become overactive. Peripheral injury, from trauma or stings, has been reported as a trigger, as has dental intervention, such as extensive crown and bridgework.⁶ Classically, embouchure dystonia occurs in the fourth decade and insidiously leads to professional disability within three years.⁴

Muscles affected in each case of oromandibular dystonia may vary. In this case, the lips producing lip-lock, where the lips are clamped shut; the tongue, where it is extended outwards and sideways and latterly; and the jaw, where there was involuntary lateral movement, were all noted (Figure 1).

This condition currently has no cure. Common treatment modalities include:

Eileen Watt, BDS, MFDS RCPS(Glasg), Senior House Officer, **Indiya Sangani**, BDS, MFDS RCS(Edin), Senior House Officer, **Fiona Crawford**, BDS, DDS, LFHom(Dent), Consultant in Oral Medicine and **Toby Gillgrass**, BDS, FDS RCS(Edin), MSc(Glas), FDS(Orth) RCS(Edin), Consultant in Orthodontics, Glasgow Dental Hospital, 378 Sauchiehall Street, Glasgow, G2 3JZ, UK.

- Botulinum toxin injections of the affected area to reduce muscle activity;
- Medication (clonazepam, baclofen);
- Deep brain stimulation surgery;
- Sensory and motor retraining; and
- Cognitive behavioural therapy.

All these treatments are designed to reduce the hyperactivity of the muscles.³ ‘Sensory tricks’, such as pinching, or touching the skin associated with the area, have been found to be beneficial in some patients, although the mechanism of action is unknown. Similar effects have been found by using dental splints, probably due to similar proprioceptive stimulation.^{7,8}

This paper looks to increase awareness of the condition so that the signs can be recognized, and summarizes how, in this case, dental appliances have been used to reduce symptoms.

Case report

Our report describes a 41-year-old professional tuba player, who had been referred to the Oral Medicine Department of the Glasgow Dental Hospital in October 2008 by his General Medical Practitioner, as he was having difficulty in adjusting his mouth to play one or two specific notes. After consultation, a provisional diagnosis of embouchure dystonia was made and the patient was referred for an MRI scan (Figure 1).

The patient never returned for follow-up. However, he was re-referred by his General Medical Practitioner in August 2010 with markedly progressive symptoms. It was established that the patient had felt unable to have an MRI as a result of him feeling claustrophobic during the procedure.

A more detailed history described how he had been a tuba player for 20 years before the dystonia had caused him to give up his profession. He reported how he had first noticed a slight difference in his tuba playing in 2002. This difference was minimal and only whilst playing one particular note. Between 2002 and 2008, the patient practised the affected notes in an attempt to try and improve on what he presumed to be a deficiency in his technical ability. It was in July 2008 that the symptoms started

to interfere with his career, as he began to have involuntary oromandibular contractions whilst playing and was therefore forced to give up playing altogether. A diagnosis of focal task specific dystonia was confirmed by his neurologist in December 2008; however, by October 2009 the contractions were now affecting him during speech.

On presentation at the Orthodontic Department, the patient had discovered that his symptoms were alleviated by occluding on a pen inbetween his premolar teeth (Figure 2).

In an attempt to replicate this effect, a lower pressure formed retainer with an acrylic block was constructed (Figure 3).⁶ It was noted by the patient that the effectiveness of this retainer was of limited time duration and lost with continued use. Therefore, splints were alternated with blocks on the left and the right. Because the patient essentially clenches on the appliance to help

prevent the spasms, the longevity of the blocks is limited and they need replacing frequently. Other appliances have been attempted with varying effectiveness for the patient. Figures 4 and 5 show a lower removable appliance with acrylic pellets adapted to induce muscle relaxation and prevent spasms. The most effective appliance to date is the one with acrylic pellets in the lower posterior buccal sulcus (Figure 5). The advantage of this particular type was the longevity of the pellets, as they were not occluded upon and they had limited effects on speech.

Follow-up

The patient continues to be followed-up within our hospital setting and new appliances continue to be developed in order to provide the most effective treatment modality and limit the fatigue effects of each successive appliance.



Figure 1. Affected muscles in continuous spasm.



Figure 2. Pen between teeth showing muscles now relaxed.



Figure 3. (a) Patient biting on bite block of lower splint; (b) lower splint in mouth; (c) lower splint on model.



Figure 4. Lower removable appliance with labial pellets.

Comment

Our report presents a classical picture of embouchure dystonia, presenting in the fourth decade and forcing the patient to give up professionally within three years. The patient presented with a ‘sensory trick’ to reduce his symptoms and we constructed dental appliances to mimic this, as well as searching for other sensory trick areas. This case highlights the potential role of the dental practitioner in providing oromandibular dystonia sufferers with a relatively simple treatment modality in an attempt to reduce their symptoms. Similar results have been reported using



Figure 5. Lower removable appliance with buccal pellets.

splints, but of different design from those described here.^{7,8} We wish to bring this condition to the wider dental population and would welcome constructive correspondence with others who have found similar success with other sufferers.

Conclusions

This condition, although rare, is one where the dental team may be able to make an early diagnosis and appropriate referral. The treatment modality outlined using removable dental appliances may provide beneficial effects of symptoms.

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