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Sensory trick splint as a multimodal therapy for oromandibular dystonia

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ABSTRACT

Purpose: Many patients with oromandibular dystonia, which is characterized by involuntary masticatory, lower facial, and/or tongue muscle contractions, experience relief of symptoms through sensory tricks such as eating chewing gum or candy. The aim of this study was to identify the factors influencing the effects of splints in patients with oromandibular dystonia.

Methods: Occlusal splints were inserted in 128 patients (89 women, 39 men) with oromandibular dystonia (102 with jaw closing dystonia, 20 with lingual dystonia, 5 with jaw deviation dystonia, 4 with jaw opening dystonia, 3 with lip dystonia, and 2 with jaw protrusion dystonia). Patients who showed improvement with the use of splints and continued to wear them for at least 3 months were defined as responders. In contrast, patients who showed little or no effect and/or were unable to insert splints were defined as non-responders. Differences in demographic and clinical data were statistically compared between responders and non-responders.

Results: Ninety-eight patients (76.6%) were responders (subjective improvement: 30.5%). Thirty patients (23.4%) were non-responders (subjective improvement: 7.2%). The responders were significantly older than the non-responders (53.8 years vs 47.0 years; $p < 0.05$). Patients with jaw closing dystonia showed the most favorable results. The proportion of patients with sensory tricks was significantly higher in responders than in non-responders (66.3% vs 26.7%; $p < 0.05$).

Conclusions: The sensory trick splint is especially helpful for patients with jaw closing dystonia. It is useful, although partially effective, as an alternative therapy in patients for whom other therapies have been unsatisfactory.

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1. Introduction

Dystonia is a movement disorder characterized by sustained or intermittent muscle contractions that cause abnormal, and often repetitive, movements, postures, or both [1]. Oromandibular dystonia is a focal type of dystonia, involving the masticatory, lower facial, lingual, and/or labial muscles [2–6]. It clinically presents as jaw closing dystonia, jaw opening dystonia, jaw deviation dystonia, jaw protrusion dystonia, lingual dystonia, or a combination of these abnormal movements [2–8].

A variety of involuntary mandibular, lingual, labial, and lower facial movements and dystonic spasms can result in trismus, bruxism, uncontrolled mouth opening, deviation, protrusion,

chewing-like movement, tongue protrusion or dyskinesia, facial grimacing, and nasal or platysma contraction. These symptoms interfere with chewing, swallowing, and speaking, resulting in social embarrassment and cosmetic disfigurement. Dystonic contracture is absent during sleep and is generally aggravated by stress. Oromandibular dystonia is predominantly found in women [4–8] and typically occurs between the ages of 45 and 70 years [4]. The overall prevalence of primary dystonia was calculated as 164.3 per million in a meta-analysis [9] and that of oromandibular dystonia was estimated at 68.9 per million [10]. In most cases, the etiology is elusive. A study by Tan and Jankovic [4] reported that the etiology may be idiopathic (63%), drug-induced (22.8%), peripheral-induced (9.3%), postanoxia (2.5%), neurodegenerative disorder (1.8%), and head injury (0.8%).

There is no gold standard for determining the validity of diagnosis, such as a diagnostic test or biomarker. Oromandibular dystonia is diagnosed on the basis of characteristic clinical features of focal dystonia, such as stereotypy, task-specificity, sensory

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tricks, morning benefit, overflow phenomenon, and co-contraction [3]. In other words, the pattern of muscle contraction and the orientation of abnormal movement are constant in each patient with dystonia. Dystonia often starts with a specific task in its early phase. Sensory tricks are maneuvers that can temporarily ameliorate symptoms. The symptoms tend to be milder in the morning (morning benefit). Overflow phenomenon involves the activation of muscles that are unnecessary for a task. Co-contraction reflects a loss of reciprocal inhibition in muscles. Unfortunately, oromandibular dystonia is often misdiagnosed as temporomandibular joint disorder, bruxism, or psychogenic disease [3,5]. Only 12.5% of the patients had been diagnosed with or were suspected to have dystonia before they visited the author's clinic.

Oromandibular dystonia has been previously treated with various antispasmodic or anticholinergic agents; however, the responses were unsatisfactory [3]. Injection of botulinum toxin is the standard therapy for focal dystonias, such as blepharospasm, cervical dystonia, and oromandibular dystonia [2,4,7–12]. Botulinum toxin is a microbial protein that exists in seven serotypes, designated A through G. Botulinum toxin can block acetylcholine release at neuromuscular junctions, which accounts for its therapeutic action in relieving numerous movement disorders associated with increased muscle tone or muscle overactivity. Muscle afferent block therapy has also been demonstrated to be effective against oromandibular dystonia and can be used as a predictor of the response to botulinum toxin therapy [3,5]. Muscle afferent block therapy by local injection of diluted lidocaine and ethanol was aimed at reducing the effectiveness of the muscle spindle afferents, without causing unfavorable weakness [3,5]. A surgical intervention, namely, coronoidotomy by bilateral resection of the coronoid process, has proven to be very effective against severe jaw closing dystonia combined with trismus, as a last resort for patients refractory to other therapies [13,14]. Several authors have reported that dental appliances were helpful for some patients [15–20]. Various terms have been used for removable intraoral devices in the literature. In this study, the author has used the term “splint” because it is commonly used in dental practice.

Sensory tricks, one of the characteristic features of focal dystonia, are physical movements or positions that can temporarily interrupt dystonia. Patients might be aware of a particular sensory trick that provides some relief from their symptoms. It is also known as “geste antagoniste.” Moreover, the term “alleviating maneuver” has been introduced recently [21]. The symptoms of oromandibular dystonia can sometimes be temporarily ameliorated via the use of sensory tricks. For instance, gently touching the lips, chin, teeth, or jaws with a handkerchief or mask; chewing gum; talking; biting on a toothpick or tobacco; or placing a finger underneath the chin might cause the symptoms to subside temporarily. Sensory tricks are almost exclusive to dystonia, and they aid in forming a diagnosis. However, if a clinician has no knowledge of the phenomenon, the patient could be easily misdiagnosed as having a psychiatric disorder. The peculiar features can be misinterpreted as indicative of a psychogenic etiology [22].

Splints are effective for some patients with oromandibular dystonia. The response might be related to the sensory tricks of the patient. However, due to the low prevalence of oromandibular dystonia, the effect of dental devices on oromandibular dystonia has been previously reported mostly in single case reports or case series [15–20]. Unfortunately, important information or data for clinicians is lacking. In this report, the author evaluated the factors influencing the effectiveness of a splint for the treatment of oromandibular dystonia.

2. Material and methods

2.1. Patients

This is a retrospective study conducted in a single institution and is based on the findings of an independent physician. The author fabricated and inserted splints in 160 patients with oromandibular dystonia (112 women and 48 men, mean age \pm SD: 53.9 ± 16.4 years) from 2007 to 2016. Oromandibular dystonia was diagnosed on the basis of characteristic clinical features of focal dystonia and electromyographic findings [3,14]. Clinical features included stereotypy, task-specificity, morning benefit, and co-contraction. Patients with oromandibular dystonia exhibit stereotypical jaw muscle contractions according to the subtype (jaw closing, jaw opening, jaw deviation, jaw protrusion, or tongue protrusion dystonia) of oromandibular dystonia. Symptoms of oromandibular dystonia are often task-specific; they mostly appear at the time of speaking, chewing, or swallowing. Sensory tricks are various voluntary maneuvers that ameliorate dystonic postures or movements. Morning benefit refers to the tendency of patients with dystonia to show milder symptoms in the morning. Co-contraction reflects a loss of the reciprocal inhibition of muscles, causing involuntary simultaneous contractions of agonist and antagonist muscles. Patients' chief complaints were very variable according to the subtypes of oromandibular dystonia; they included masticatory disturbance, dysarthria, muscle pain, dysphasia, discomfort, and cosmetic problems.

All patients involved in this study provided written, informed consent after receiving a full explanation of the planned treatment. This study was performed in accordance with the Declaration of Helsinki and was approved by the institutional review board and ethics committee of Kyoto Medical Center.

2.2. Treatment

The treatment scheme of oromandibular dystonia is shown in Fig. 1. After careful examination, for mild to moderate cases, the author prescribed anticholinergic or antispasmodic agents or other relevant drugs. Patients with tardive dystonia who were referred from psychiatrists or neurologists tended to deny additional oral medicine. If the patient denied oral medication, or if response to the oral medication was unsatisfactory, the author blocked involuntary muscle contraction by an intramuscular injection. At first, muscle afferent block therapy [3,5], by injection of a local anesthetic (0.5% of lidocaine), was tried on hyperactive muscles (Fig. 1). In cases of favorable results with the muscle afferent block, the author continued the therapy. Further, if the muscle afferent

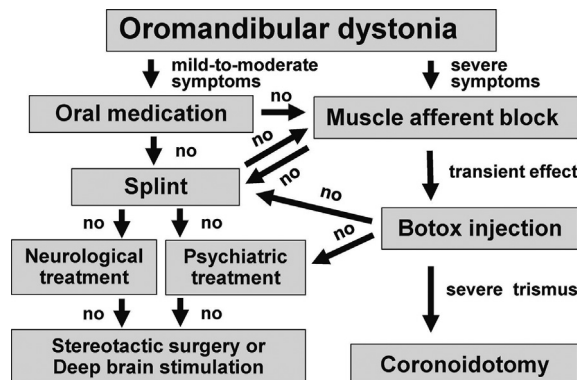


Fig. 1. Flow-chart of the treatment of oromandibular dystonia.

Oromandibular dystonia should be treated with multimodal therapies based on the patients' symptoms by a multidisciplinary team.

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