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CLINICAL TECHNIQUES AND TECHNOLOGY

VIDEO

Use of an oral sensory feedback device in the management of jaw-opening dystonia

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Sponsorships or competing interests that may be relevant to content are disclosed at the end of this article.

Jaw-opening dystonia (JOD) is a type of oromandibular dystonia in which patients suffer from prolonged lateral pterygoid muscle contraction causing sustained jaw opening. Patients have difficulty eating and speaking, often drool uncontrollably, and suffer from a poor quality of life.[1] Therapeutic options for this disease are limited, making this a challenging problem to address.[2]

We describe the use of a novel sensory feedback device (SFD, Allergan Pharmaceuticals, Irvine, CA) in addition to conventional treatment with oral medication and botulinum toxin A (Botox) injections in the management of JOD. This device provides immediate relief of dystonic symptoms and allows patients to depend less on other methods of treatment.

Figure 1 Oral sensory feedback device.

METHODS

A retrospective chart review was performed at the University of Southern California Department of Otolaryngology after obtaining institutional review board approval. Two males and three females with idiopathic JOD were identified over the last five years. The average age of our patients was 57 years with an age range of 42 to 76 years.

Our treatment approach was three tiered. A standard medical regimen was prescribed by the neurologist. Transoral injections of Botox into the lateral pterygoid muscles, which are persistently contracted in JOD, were performed in the second tier of treatment. Patients actively opened their mouths, and two sites within each lateral pterygoid muscle with the largest amounts of electromyogram activity were injected with equivalent doses of Botox. The physician performing Botox injections was blinded as to whether patients were using an SFD. The total dosage of Botox injected per visit was recorded.

In the third tier of treatment, each patient was fit with an oral SFD (Fig 1). The device was clear and custom molded to fit along the patient's mandibular teeth. A peg of silicone one centimeter in height and spanning three molar teeth provided pressure to the corresponding maxillary molars. Patients were educated to bite down on the device to overcome the activity of the lateral pterygoid muscle. Application of this device allowed for rapid relaxation of the affected dystonic muscle group. Patients wore this device during the day and soaked it in cleaning solution at night.

RESULTS

All five patients with JOD treated with the SFD were able to immediately overcome symptoms with intraoral placement of the device (Video 1, online at www.otojournal.org). These patients tolerated the device well and were compliant with daily wear. After use of this device, we noted that patients required Botox injections less frequently as demonstrated in Figure 2. The patients, who initially required injections with Botox every 2.8 months, required Botox every 5 months, on average, after using the SFD ($P = 0.0098$). We also found that after employing this device patients required less Botox for resolution of their symptoms. Prior to intervention patients were treated, on average, with a total of 78 units of Botox per office visit. After continued usage of the SFD, patients required, on average, 64 units of botox, a statistically significant difference ($P = 0.026$). Patients have been followed between 3 and 5 years after treatment initiation.

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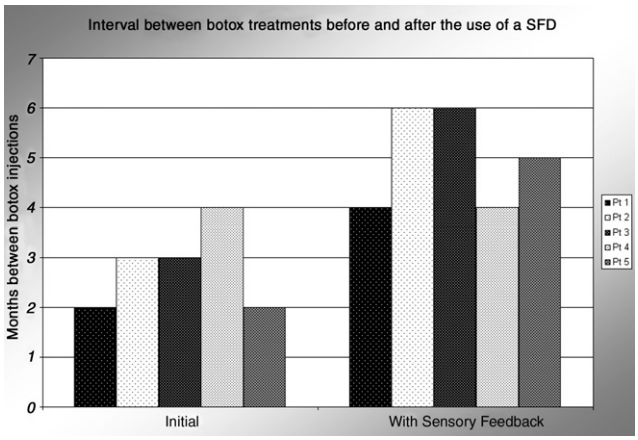

Figure 2 Graph showing the interval, in months, between Botox injections before and after use of the sensory feedback device.

DISCUSSION

JOD is a devastating disease. Those afflicted are unable to close their mouth, leading to drooling and difficulty with masticating and swallowing. Patients have trouble articulating words and are often left unintelligible. Classically, management of JOD has been challenging and one of the most difficult dystonias to treat.[2] Two other series detail the treatment of JOD with Botox injections.[2,3] As many as 40 percent of patients experienced adverse effects from Botox including dysphagia, breathy voice, painful chewing, and velopharyngeal insufficiency. Brin and colleagues[3] described a patient with JOD who developed antibodies to Botox after aggressive treatment, leaving the patient with limited therapeutic options.

The geste antagoniste, or "sensory trick," is a feature common to many oromandibular dystonias and has been described in JOD.[4] Tactile or sensory stimulation in the region of the affected muscle group causes relaxation of dystonic muscles. Sensory tricks are a hallmark of dystonia and are dramatic when present. In our five patients the sensory trick was triggered by applying light pressure on the molar teeth, which caused lateral ptyergoid muscle relaxation, allowing patients to overcome their dystonia and close their mouths. The mechanism of the sensory trick of dystonia has been often studied but is still largely unknown.

Frucht and colleagues[5] described a geste device that fits on a patient's mandibular molar to treat jaw-closing dystonia. Similar to the patients reported here, this patient achieved immediate resolution of symptoms and used the device as an adjunct to Botox treatment.

To our knowledge this is the first reported series of patients with JOD successfully managed with an SFD that reduced the need for other treatments. Our patients have been followed for 3 to 5 years, during which time we have noted less of a demand for Botox and a persistent effect of the SFD.

CONCLUSION

Our approach for management of JOD is multidisciplinary, with patients being treated by both the neurologist and the otolaryngologist. The combination therapy includes medical management, periodic Botox injections and, most importantly, an SFD. The device is easy to use, inexpensive, and noninvasive. It is worn by patients throughout the day, sparing them the functional and social sequela of oromandibular dystonia. With use of the SFD, patients are treated less frequently with Botox, sparing them painful injections and the inconvenience of frequent office visits. Other patients with JOD may benefit from being examined for the presence of a geste antagoniste and being treated with an SFD.

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AUTHOR CONTRIBUTIONS

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DISCLOSURES

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SUPPLEMENTARY DATA

Supplementary data associated with this article can be found, in the online version, at doi:10.1016/j.otohns.2009.04.009.