Neurogenic temporomandibular joint dislocation treated with botulinum toxin: report of 4 cases

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Many patients suffer recurrent episodes of temporomandibular joint (TMJ) dislocation due to an excess of muscle contraction or spasticity in the depressor muscles of the jaw. The manual repositioning using the Nelaton maneuver is the first treatment. Occasionally, it may be necessary to use sedation or general anesthesia to achieve the desired muscle relaxation. In case of recurrence, surgical treatment is indicated. One nonsurgical method of treatment is the local infiltration of botulinum toxin type A. We present 4 cases of recurrent TMJ dislocation in patients suffering from conditions of neurologic origin, with considerable motor deterioration, treated with local infiltration of botulinum toxin type A. In conclusion, the injection of botulinum toxin type A is an effective method in cases of neurogenic TMJ dislocation, with low morbidity and side effects, improving patients’ quality of life. (Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2010;109:e33-e37)

The anterior dislocation of the temporomandibular joint (TMJ) is by far the most frequent form. It occurs when one or both mandibular condyles are displaced in front of the temporal eminence. It may be reducible when it returns spontaneously to the glenoid cavity, or nonreducible when 1 or 2 condyles remain dislocated. In this position, the mouth remains open due to the action of the elevator muscles with or without lateral deviation, depending on whether the dislocation is unilateral or bilateral.

Dislocation of the TMJ is generally of unknown origin, with several theories put forward to explain its onset. Some disorders of collagen metabolism, such as ligamentous hyperlaxity and Ehler-Danlos syndrome might be related. However, there is a group of patients who suffer recurrent episodes of TMJ dislocation due to an excess of muscle contraction-spasticity in the muscles depressing the lower jaw. These patients are frequently associated with severe disorders of the central nervous system, with motor and cognitive deterioration, thus contraindicating surgery under general anesthesia. Daelen defined these cases of TMJ dislocation as neurogenic, to distinguish them from traumatic injuries. Some degenerative neurologic disorders are accompanied by repeated bouts of TMJ dislocation, such as Parkinson disease or multiple sclerosis, as well as posttraumatic cerebral palsy, particularly those occurring with spasticity and orofacial or oromandibular dystonia.

The treatment of acute episodes, in the case of nonreducible TMJ dislocation, consists of the manual repositioning using the Nélaton maneuver. Occasionally, in cases of inveterate TMJ dislocation, it may be necessary to use sedation and even general anesthesia to achieve the desired muscle relaxation.

In cases of recurrence, surgical treatment is indicated using various techniques. These are basically divided into 2 groups:

1) Eminectomy (Myrhaug technique).
2) Interposition techniques (redirecting the tendon of the temporal muscle, alloplastic or bone graft, dislocation fracture of the zygomatic arch).

One nonsurgical method of treatment that has been described is the injection of sclerosing substances in the area adjacent to the joint, including arthroscopic techniques.

In those patients suffering neurogenic TMJ dislocation, the reiteration of the dislocation produces local pain and severe eating difficulties. This, together with the inconvenience of repeated visits to emergency centers, causes anxiety in patients and their relatives. In these cases, TMJ dislocation occurs through a loss of coordination of the muscles in the jaw and hypertonia of the muscles depressing the lower jaw.

The use of local treatment with botulinum toxin type A might be effective in such patients. The toxin produces temporary denervation on the muscles depressing the lower jaw, thus preventing excessive displacement of the
condyle when opening. Botulinum toxin type A is indicated for the treatment of a focal dystonias, and it is effective to treat episodes with excess of muscular activity. We decided to use botulinum toxin type A to treat 4 cases of recurrent TMJ dislocation of neurologic origin after approval by the ethics committee and authorization of each case by the Ministry of Health.

**CASE REPORTS**

We present 4 cases of recurrent TMJ dislocation in patients suffering from conditions of neurologic origin treated with local infiltration of botulinum toxin type A.

**Case 1**

A 26-year-old male who suffered severe head and brain trauma in 2003 as the result of a traffic accident. After the accident, he presented multiple foci of brain contusions, with subarachnoid bleeding in the third and lateral ventricles, the right thalamus, and the right occipital cortex. These injuries led to the presence of chronic subdural haematomas. He also suffered diaphyseal fracture of the right humerus, right shutter ring, right acetabulum, and sacrum.

The clinical consequence of these injuries was the onset of right-side hemiparesia, predominantly brachial, as well as movement dystonia, particularly marked in the orofacial sphere, and widespread spasticity. The most striking manifestation was the presence of recurrent episodes of temporomandibular dislocation when eating, yawning, or speaking, with a strong tendency to relapse and requiring multiple treatments (simple manual reduction under sedation, temporary intermaxillary fixation under general anesthesia) over the course of 2 years.

For this reason, after obtaining the family’s written consent, he was injected in September 2007 with 25 UI botulinum toxin type A (Botox; Allergan, Irvine, Calif), under electrical control (Botox Injection Amplifier) in each of the external pterygoid muscles. The technique used in this patient was that described by Daelen et al. With the patient in a sitting position, the location of the sigmoid notch and the condyle of the jaw were determined on either side by palpation. Next, the needle connected to the electrical amplifier was inserted 1 cm in front of the condyle, obliquely backwards, down, and in. When the patient was asked to open his mouth, slight movements were made with the needle to capture the signal corresponding to the contraction of the external pterygoid muscle. Then, after aspiration, the contents of the syringe were injected. No side effects were noted during administration or in the subsequent interval. Approximately 10 days after the treatment, the episodes of TMJ dislocation ceased. This allowed the patient to significantly improve in independence to carry out physiologic functions and in his ability to communicate (Fig. 1).

In October 2008, he was treated again, after reappearance of the TMJ dislocation. Forty UI of botulinum toxin type A (Botox) was injected into both external pterygoid muscles and 10 UI into the anterior faces of both digastic muscles. After 1 week had elapsed, moderate dysphagia appeared, accompanied by oropharyngeal candidiasis, probably secondary. He was treated with a soft diet and Nystatin mouthwash, and the condition disappeared in 2 weeks. Once more, the episodes of TMJ dislocation disappeared, and he remained in remission for 10 months to the time of writing.

All of these treatments were administered on an outpatient basis.

**Case 2**

A 72-year-old female patient, diagnosed as having Alzheimer and Parkinson diseases, had severe intellectual impairment and widespread spasticity. Since May 2008, she had presented repeated bouts of inveterate TMJ dislocation associated with eating, yawning, and even sleeping, leading her to be repeatedly taken to our hospital’s Emergency Unit for its reduction, on occasions under general anesthesia, in view of the associated spasticity that hindered simple manual reduction, even under sedation.

In January 2009, after obtaining the family’s consent, the patient was subjected to general anesthesia in the operating theater by means of nasotracheal intubation.

No systemic muscle relaxants were used. With the assistance of the electrical amplifier, per the technique described by Moore and Wood, the mandibular opening reflex was explored by striking the chin with a reflex hammer (Fig. 2). The capture of the amplified electrical signal produced by this reflex allowed the localization of the infiltration point. Twenty-five units of Botox was injected into both external pterygoid muscles (Fig. 3). No immediate or late-onset side effects were observed. The patient was discharged the next day.

Over the next 7 months, the patient did not present any episode of TMJ dislocation. The patient’s quality of life and autonomy notably improved during this period, according to the testimony of her relatives.

**Case 3**

A 88-year-old woman presented to us for recurrent bilateral TMJ dislocation that had started several months before the initial referral. She had diagnosed Alzheimer and Parkinson diseases with widespread spasticity.

In February 2009, after obtaining the family’s written consent, she was subjected to sedation in the operating the-
We injected 25 UI Botox into both external pterygoid muscles.

Over the next 6 months, the patient did not present any episode of TMJ dislocation.

**Case 4**

A 23-year-old male suffering from Steinert disease (myotonic dystrophy) began to develop recurring episodes of TMJ dislocation in October 2007. The dislocation occurred while performing minimal movements of mouth opening, including, e.g., daily dental hygiene. He frequently had to use our emergency department for reduction, making his normal life impossible.

In March 2009, after giving written consent, he was treated by injection of 25 UI Botox into each lateral pterygoid muscle under local anesthesia. The muscle was localized using the Plexygon 7501.31 nerve stimulator (Vygon) connected to an injection needle (Locoplex 17°, 23 gauge, 35 mm; Vygon). Applying a stimulation of 1 mA, frequency 2 Hz, 300 μs, it was assumed that the tip of the needle penetrated the muscle when a rhythmic movement of the mandible occurred in synchronicity with the stimulation (Fig. 4).

No side effects were noted. The patient presented with a TMJ dislocation episode 5 months after treatment, and a new dose of Botox was administered.

**DISCUSSION**

Botulinum toxin type A is a double-chain protein, 150 kD in weight, produced by *Clostridium botulinum*, an anaerobic bacterium responsible for botulism. It prevents the release of acetylcholine into the synaptic cleft, temporarily blocking neuromuscular transmission.\(^{14}\) It acts selectively on the peripheral cholinergic nerve endings\(^{15}\) to produce muscle relaxation, diminished compression of the muscle vessels, and occasionally a reduction in the concentration of excitatory neuropeptides.\(^{16}\)

Botulinum toxin type A was first indicated for the treatment of strabismus in patients \(\geq 12\) years old, blepharospasm, hemifacial spasm, and associated focal dystonias. Later, new indications were approved, such as cervical dystonia (for the reduction of signs and symptoms of spasmodic torticollis), dynamic equine foot deformity caused by spasticity in children \(\geq 2\) years old with cerebral palsy, arm spasticity in patients who have suffered a cerebrovascular accident,\(^{11}\) and, recently, axillary hyperhidrosis.

In the literature, there is an increasing number of reports on the applications of botulinum toxin for the treatment of disorders of the TMJ. Bakke et al.\(^{17}\) described its use in severe clicking of the TMJ associated with anterior disc displacement, with good results.

The treatment with botulinum toxin type A is contraindicated in the case of hypersensitivity to any of the
components in its formulation and when there are widespread disorders of muscle activity; e.g., it should not be used in patients with myasthenia gravis or Eaton Lambert syndrome, when aminoglycoside antibiotics or spectinomycin are or will be administered, or when there is inflammation or infection at the injection site.\textsuperscript{18}

In 1997, Moore and Wood\textsuperscript{13} and subsequently Daelen\textsuperscript{12} described the treatment of TMJ dislocation with botulinum toxin in patients affected by recurrent TMJ dislocation and neurologic disorders. The technique used by the authors is based on the work of Brin et al.,\textsuperscript{19} who first described the injection of botulinum toxin type A into the external pterygoid muscles under electromyographic control for the treatment of oromandibular dystonia.

In the present cases, it is reasonable to assume that during injection there is a certain diffusion of botulinum toxin into the pterygoid space, affecting not only the lateral but also the medial pterygoid muscle. The denervation of the medial pterygoid muscle is compensated by other masticatory muscles, such as the masseter or the temporal muscles. The denervation of the lateral pterygoid muscle is not compensated, thus preventing movements of the condyle that lead to dislocation.

In 1998, Daelen described 5 cases of neurogenic TMJ dislocation treated with botulinum toxin, with scant undesirable effects.\textsuperscript{20} Only 1 of our cases presented some side effects: the appearance of transitory dysphagia, probably due to the toxin spreading from the anterior fasses of the digastric muscles. From the perspective of our current experience, we do not consider it to be necessary to administer the drug in this location, because there is no need for it and because of the possible onset of dysphagia. Other injection techniques include intraoral route with electromyographic guide, as described by Martínez-Pérez and García Ruiz-Espiga.\textsuperscript{21}

Case 2 presented particularly difficult characteristics, in view of the impossibility of injecting the toxin into the patient while conscious, owing to a lack of cooperation resulting from the underlying disorder. For this reason, we used the technique described by Moore and Wood in 1997,\textsuperscript{13} which allows the medication to be administered in a controlled fashion under general anesthesia. However, the outcome in this case was the most satisfactory, as indicated by the improvement in the patient’s quality of life. The greatest difficulty lies in not using systemic muscle relaxant drugs and in observing the mouth opening reflex, which can easily be confused with the mouth closure reflex (masseter reflex). Probably, at present, the most effective technique is the use of the nerve stimulator, as is described in case 4.

The doses used (between 25 and 40 UI) depend on the type of drug (Botox in this series), because the other preparation available on the market, Dysport, is \textsuperscript{22} \sim 4 times less active than Botox,\textsuperscript{22} although there is no fixed dose relationship. In view of the scarcity of literature on the subject, there are no references about the dose per individual or the titration of botulinum toxin type A. Therefore, the doses used in this series can be considered to be valid, although, as mentioned above, they depend on the preparation used and different formulations are not interchangeable.

The irrelevance of side effects, together with the dramatic results on these patients’ quality of life, makes TMJ dislocation treatment with botulinum toxin type A an extraordinarily useful resource as an alternative to well tried surgical techniques that are difficult to apply in patients with severe neurologic disorders. Moreover, the etiology of TMJ dislocation in these cases is confirmed by the efficacy of botulinum toxin type A. There is no limit on the number of doses if a minimum interval of 3 months is respected between applications. Respecting this time interval minimizes the formation of neutralizing anti–botulinum toxin type A antibodies, thus maintaining its efficacy without limit of time.

In conclusion, the injection of botulinum toxin type A in the external pterygoid muscles under instrument control is an effective method in cases of neurogenic TMJ dislocation, with scant morbidity and side effects, improving patients’ quality of life and their interaction with their surroundings.

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