Crocodile tears syndrome: botulinum toxin treatment under EMG guidance

Sibel Kizkin
Selim Doganay
Handan Isin Ozisik
Cemal Ozcan

Faculty of Medicine, Inonu University and Turgut Ozal Medical Center, Malatya, Turkey

Reprint requests to: Dr Sibel Kizkin
Inonu Universitesi Tip Fakultesi
Noroloji Anabilim Dalı
TR-44160 Malatya, Turkey
E-mail: skizkin@inonu.edu.tr

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Summary

Crocodile tears syndrome is one of the rare complications of facial paralysis. There have been several reports of cases in which botulinum toxin was found to be useful in the treatment of crocodile tears syndrome. The adverse effects, due to the paralytic action of botulinum toxin, have been reported to involve the palpebral muscle, lateral rectus and superior rectus. Therefore, we considered that it might be more appropriate to carry out the injection procedure under electromyographic (EMG) guidance, in order to inject botulinum toxin selectively into the lacrimal gland and to protect the above-mentioned muscles.

In conclusion, we recommend EMG guidance in the treatment of crocodile tears syndrome with botulinum toxin.

KEY WORDS: botulinum toxin, crocodile tears syndrome, electromyography.

Introduction

Crocodile tears syndrome, also called gustolacrimal reflex, paroxysmal lacrimation and Bogorad's syndrome, is one of the rare complications of Bell's palsy or traumatic facial paralysis. It was first described by a Russian neuropathologist in 1913, and is characterized by inappropriate and sometimes excessive lacrimation provoked by eating (1,2).

Recently, there have been several reports of cases in which botulinum toxin was found to be useful in the treatment of crocodile tears syndrome (1-6). The adverse effects, due to the paralytic action of botulinum toxin, have been reported to involve the palpebral muscle, lateral rectus and superior rectus (1,3-6). Therefore, we considered that it might be more appropriate to carry out the injection procedure under electromyographic (EMG) guidance, in order to inject botulinum toxin selectively into the lacrimal gland and to protect the above-mentioned muscles.

In this report, we present the findings of the injection of botulinum toxin, under EMG guidance, via a conjunctival approach, in a patient presenting crocodile tears syndrome following idiopathic peripheral facial paralysis (PFP).

Case report

A 19-year-old male patient presented with a 6-month history of left PFP without concomitant medication. Two months after the onset of the PFP, his left eye had begun to present tearing during meals. With the exception of a slightly decreased left nasolabial fold, there were no pathological findings on neurological examination. PFP was categorized as Grade II according to the House Brackmann Facial Nerve Grading System (at rest: normal tone and symmetry; motion: complete closure of the eyelid with minimum effort, minimal oral asymmetry). Blink reflexes were investigated by EMG and a positive response was found after stimulation of the ipsilateral supraorbital nerves, from both the m. orbicularis oculi and m. orbicularis oris, which suggested aberrant reinnervation (Fig. 1). Ophthalmological examination showed normal visual acuity in both eyes. Fundus examination was bilaterally normal. Cornea epithelium was healthy and disclosed no epithelial surface pathology.

The patient was examined clinically, and tearing was assessed by Schirmer’s test before and 1, 4, 12 weeks after the injection. According to this test procedure, one end of a strip of thin filter paper (5 mm wide and 30 mm
long) was inserted into the lower conjunctival sac, while the other end was allowed to hang over the margin of the lower lid. The tears wet the strip of filter paper, producing a moisture measurement. Basal tear secretion and tear secretion after gustatory stimulation (the patient ate a sweet) were measured.

Basal tear secretion (without gustatory stimulation) was found to be 15 mm in the right eye and 17 mm in the left eye. Repetition of the Schirmer’s test during salivary stimulation gave values of 16 mm and over 30 mm, respectively (Tables I and II).

**Table I - Schirmer’s test results without gustatory stimulation.**

<table>
<thead>
<tr>
<th></th>
<th>Right eye</th>
<th>Left eye</th>
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</thead>
<tbody>
<tr>
<td>Before treatment</td>
<td>15 mm</td>
<td>17 mm</td>
</tr>
<tr>
<td>1 week after treatment</td>
<td>16 mm</td>
<td>16 mm</td>
</tr>
<tr>
<td>4 weeks after treatment</td>
<td>15 mm</td>
<td>17 mm</td>
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<tr>
<td>12 weeks after treatment</td>
<td>16 mm</td>
<td>16 mm</td>
</tr>
</tbody>
</table>

**Table II - Schirmer’s test results with gustatory stimulation.**

<table>
<thead>
<tr>
<th></th>
<th>Right eye</th>
<th>Left eye</th>
</tr>
</thead>
<tbody>
<tr>
<td>Before treatment</td>
<td>16 mm</td>
<td>over 30 mm</td>
</tr>
<tr>
<td>1 week after treatment</td>
<td>15 mm</td>
<td>14 mm</td>
</tr>
<tr>
<td>4 weeks after treatment</td>
<td>15 mm</td>
<td>16 mm</td>
</tr>
<tr>
<td>12 weeks after treatment</td>
<td>16 mm</td>
<td>15 mm</td>
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</table>

Before botulinum toxin injection, the conjunctiva mucosa was anesthetized with proparacaine HCL (Alcaine 0.5%; Alcon, Belgium). The EMG needle was inserted in the palpebral portion of the lacrimal gland following ectropionization of the upper eyelid. Six units of botulinum toxin (Botox; Allergan, Irvine, Calif.) were injected into the palpebral portion of the lacrimal gland (Fig. 2), after it had been established that muscle activity was not detectable by EMG (Fig. 3). The needle only entered the lacrimal gland. The treatment was not painful. Within 4 days of the botulinum toxin injection, the patient noticed a reduction of tear secretion during eating, and a complete disappearance after 7 days. Side effects such as eye burning, ptosis, double vision and visual problems were not observed after botulinum toxin injection.

One week after treatment (Tables I and II), the Schirmer’s test gave values of 14 mm (left eye) and 15 mm (right eye) during gustatory stimulation (eating a sweet). The patient had no discomfort 12 weeks after the procedure.

Discussion

Crocodile tears are seen in 3.3 to 6.5% of patients following Bell’s palsy (7,8). The syndrome may also occur following acoustic neurinoma surgery, in maxillofacial trauma, lepra, Paget’s disease, vascular diseases and cervicoaculoacoustic syndrome (3,9,10).

A misdirection of regenerating parasympathetic fibres is widely accepted as the pathogenetic mechanism underlying crocodile tears syndrome. Gustatory fibres from facial or glossopharyngeal nerves may be misled through n. petrosus superficialis major to the lacrimal gland, causing the eye on that same side to present tearing when the patient eats or drinks (1,3,6). It may rarely be accompanied by loss of taste and by facial spasm.

Many methods have been used to treat crocodile tears including drugs such as antihistaminics, anticholinergics (8), intraorbital alcohol or cocaine injection (1), Vidian neurectomy (11,12), and subtotal resection of the lacrimal gland (13). However, none of these treatment options has been unanimously accepted due to persistent side effects in some of them (e.g., eye burning in Vidian neurectomy), or inadequate results.

Recent reports suggest that botulinum toxin injection into the lacrimal gland of patients with crocodile tears syndrome may effectively abolish the reflex symptom. Botulinum toxin-A affects neurotransmission by inhibiting the release of acetylcholine at the neuromuscular junction and at cholinergic autonomic nerve terminals (1,5, 6,14,15). Botulinum toxin has been applied both tran-
EMG-guided botulinum toxin treatment of crocodile tears

scutaneously and transconjunctivally to the lacrimal gland in patients with crocodile tears syndrome (2,5,6, 14). Riemann et al. (6) injected 2-5 U botulinum toxin into the palpebral portion of the lacrimal gland, obtaining a noticeable reduction in the patient’s complaint in a week; moreover, no side effects were observed. Montaya et al. (1), on the other hand, first applied 10 U transcutaneously. When this proved ineffective, an additional 10 U botulinum toxin was administered transconjunctivally. But it was reported that this application caused side effects such as eye burning and ptosis resulting from involvement of the m. levator palpebra. However, the desired result was achieved with a lower dose using the transconjunctival rather than the transcutaneous technique.

Botulinum toxin injection may have a paralytic effect on m. levator palpebra, m. rectus superior, and m. rectus lateralis due to the proximity of these muscles to the lacrimal gland. To avoid these side effects, we performed transconjunctival botulinum toxin injection under EMG guidance. Symptoms completely improved one week after application and no complications occurred in the related eye and adnexes. Follow-up ophthalmological examination with Schirmer’s test 12 weeks after injection showed persistence of the botulinum toxin effect. In conclusion, the use of EMG guidance could increase the efficiency and safety of botulinum injection in cases with crocodile tears syndrome. We recommend EMG guidance in the treatment of crocodile tears syndrome with botulinum toxin.

References
