



BOGORAD'S SYNDROME TREATED SUCCESSFULLY WITH ADMINISTRATION OF BOTULINUM TOXIN 'A' INJECTION INTO LACRIMAL GLAND: "A CASE STUDY AND LITERATURE REVIEW."

Neurology

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ABSTRACT

Crocodile sheds tears while eating their victims. Bogorad's syndrome (BS) or crocodile tears is an infrequent complication of facial paralysis characterized by, profuse unilateral lacrimation while eating or drinking on the same side of facial weakness. Here we present a case of 21 year old female who developed BS following Bell's palsy and she was treated with botulinum toxin 'A' injection into lacrimal gland successfully. To the best of our knowledge, this is the first case study from India where botulinum toxin is administered for Bogorad's syndrome accomplishing a desired result.

KEYWORDS

Crocodile tear, botulinum India

INTRODUCTION:

Bogorad's syndrome (BS) commonly called as syndrome of crocodile tears is a rare complication of facial paralysis, characterized by profuse lacrimation on the side of facial palsy during eating. It is also known as 'gusto-lacrimal reflex' or 'paroxysmal lacrimation'. Only 5-10% of patients with facial paralysis develop crocodile tears syndrome. [1], [2] This rare phenomenon usually occurs following a peripheral facial nerve paralysis. To date there are few cases of BS treated worldwide with botulinum toxin. [3],[4],[5],[6] Present case report is the first case from India.

Case presentation: 21-year female presented with profuse lacrimation from left eye while eating food since 6 months. She had a prior history of left Bell's palsy 2 years back, from which she had recovered completely. On examination she had profuse lacrimation while having food, rest of the neurological and ocular examinations were normal. Schirmer's test was used for assessment of Lacrimation. Baseline tear secretion was found to be 14 mm in the right eye and 16mm in left eye [Normal range ≥ 10 mm wetting of paper at 5 minutes], while tear secretion after gustatory stimulation [eating rice] was found to be 15 mm in right eye and beyond 32 mm in left eye. This further confirmed hyper lacrimation in left eye while eating. Based on the above findings diagnosis of gustatory lacrimal reflex, namely BS or crocodile tear syndrome was diagnosed secondary to previous left Bell's palsy. In view of the troublesome nature of BS to our patient, botulinum toxin 'A' administration to lacrimal gland was considered. Accordingly, after obtaining consent, botulinum toxin A (Botox 2.5 U) was injected through sub conjunctival approach into the palpebral lobe of the left lacrimal gland under direct visualization. Prior local conjunctival anesthetic [Proparacaine] drop was instilled into the left eye. We observed immediate disappearance of lacrimation while eating. She remained asymptomatic for 10 months.

DISCUSSION:

Crocodiles shed their tears while consuming their victims, expressing remorse while happily committing the crime. The syndrome of lacrimation that appears during eating was first described by Oppenheim in 1913. [7]. F. A Bogorad a Russian neuropathologist described incidents of "crocodile tears" among human beings and named it as crocodile tears syndrome in 1928. [8] Later Ford termed paroxysmal lacrimation to describe this syndrome. [9] Ford also showed that this syndrome is unassociated with common form of Bell's palsy in which lesion is distal to geniculate ganglion, but occurs when lesion is proximal to geniculate ganglion. [9] Axelsson and Laage-hellman described it as Gusto-lacrimal reflex. [10] Gustatory lacrimal reflex occurs owing to anomalous nervous regeneration occurring after proximal facial nerve injury where autonomic nerve fibers supplying salivary gland misroute to lacrimal glands [Figure 1]. [10]

The syndrome of crocodile tears or BS, develop once the motor paralysis has disappeared. Similar findings were observed in present case. Most often BS occurs in acquired cases such as peripheral facial nerve palsy following trauma, neoplasm, or inflammatory condition of facial nerve or greater superficial petrosal nerve. Rarely syndrome can occur bilateral or congenital. Congenital causes of BS are associated

with abducent palsy or Duane syndrome. [11] The association of the two condition occurs following degeneration or dysgenesis in the surrounding region of abducent nucleus resulting paradoxical innervation of the lateral rectus by fibers from oculomotor nerves and lacrimal gland by fibers sub serving salivation. Thus BS associated with Duane's syndrome is referred as congenital cranial dysinnervation syndrome. BS has also been reported with Ramsay Hunt syndrome [12] and associated with Dandy walker syndrome in Wildervanck syndrome. [13]

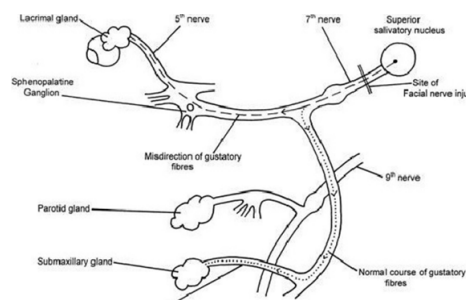


Figure 1: Gustatory lacrimal reflex owing to anomalous abnormal nervous regeneration occurring between autonomic nerve fibers supplying salivary gland and lacrimal glands following proximal facial nerve injury

BS should be differentiated from lacrimation associated with facial paralysis where tearing is because of ectropion, that allows tears to flow out of conjunctival sac. Further facial paralysis with ectropion is unassociated with hypersecretion of the lacrimal gland and is uninfluenced by eating, whereas BS has hypersecretion during eating. Literature review showed several treatments for patients with crocodile tears. These include anticholinergic drugs, intraorbital alcohol or cocaine injection, subtotal resection of the palpebral lobe of the lacrimal gland, and resection of the tympanic nerve proximal to the lesser superficial petrosal nerve. [6] All these approaches were sub optimal for the reason of limited efficacy and morbidity. Botulinum toxin administration into the lacrimal gland is the advanced therapeutic option available for treating crocodile tears syndrome.

Borojerdi et al reported the first successful treatment of crocodile tears by injection of botulinum toxin A directly into the lacrimal gland in 1998. [14] Subsequent studies, [3], [4], [5] reports showed complete or near complete resolution of the crocodile tears within a week with only minor, and reversible complications with botulinum toxin. One study showed long term efficacy and safety of botulinum toxin for 3 years. [6] Transient ptosis (11%) and diplopia (2%) are the complication observed in fewer patients following botulinum toxin injection into lacrimal gland.

Botulinum toxin injection into the lacrimal gland blocks the pre-synaptic release of acetylcholine required for tear production, thus

overcomes the pathologic excess production and relieve tearing symptoms. There are two approaches for botulinum injection to lacrimal gland, transcutaneous and trans- conjunctival injection. Latter approach had fewer complications hence we preferred trans-conjunctival injection [Figure 2]. Our patient had no complications.



Figure 2: Trans-conjunctival approach of giving botulinum toxin to lacrimal gland

CONCLUSION:

Botulinum toxin injection into lacrimal gland is the first choice of treatment approved for Bogorad's syndrome. Procedure is simple, safe and effective. Only small dose of botulinum is needed for immediate and tremendous response.

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