Botulinum Toxin A for Spastic Trismus Due to Brain Stem Encephalitis in a Pediatric Intensive Care Setting: A Unique Case Report

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Abstract

Keywords

- interincisal distance
- suction
- oral care
- masseter
- temporalis

Injection of botulinum toxin A to masseter and temporalis muscles improved mouth opening and facilitated suction and oral care in a child with spastic trismus. This is the first such report in a 3.5-year-old child on mechanical ventilator in pediatric intensive care unit setting.

Introduction

Spastic trismus refers to a sustained spasm of the masticatory muscles, in particular the masseter and temporalis, which results in forced jaw closure. Spastic trismus can be caused by brain stem pathology due to involvement of trigeminal nerve.¹ Complications of trismus in a patient in an intensive care unit setting include difficulties in suction, oral care, and oral hygiene. This can lead to aspiration of secretions resulting in life-threatening sepsis. Botulinum toxin A has been found to be effective in the management of spastic trismus.² In this report, we discuss the benefits of botulinum toxin A to improve mouth opening, to facilitate oral suction, and to improve oral care in a child with trismus due to brain stem encephalitis admitted to the pediatric intensive care unit. This is the first report on the use of botulinum toxin A for trismus in a 3.5-year-old child in an intensive care setting.

Case Report

A 3.5-year-old female child was admitted to the pediatric intensive care unit with fever, respiratory distress, respiratory acidosis, and cardiogenic shock and was rapidly intubated and connected to mechanical ventilator. Echo was suggestive of viral myocarditis with ejection fraction of

28%. Magnetic resonance imaging brain and cerebrospinal fluid studies confirmed viral meningoencephalitis.

Patient continued to be under intensive care with ventilator support and tracheostomy and her Glasgow Coma Scale remained 4/15 (eye opening: 1, motor: 2, verbal: 1). Feeding was through nasogastric tube. She developed mild hypertonia in all four limbs and she developed trismus that became severe by 2 months from the date of admission. Oral baclofen was started to treat trismus, but there was no response. Nursing staff reported tremendous difficulties in performing oral suction and they were unable to perform oral hygiene as there was no mouth opening at all (► **Figs. 1A** and **2A**). The child had to be treated for recurrent episodes of aspiration pneumonia.

A decision to inject botulinum toxin A was taken after weighing the possible benefits against the probability of complications of untreated severe trismus including recurrent respiratory infections and sepsis. The same was discussed with the father of the child and consent for injection was obtained. Subsequently, 10 weeks post admission, Botox injection was given in bilateral temporalis and masseter muscles as follows: 100 units of Botox was diluted with 1 mL of normal saline. Temporalis muscle was palpated and two points where the muscle was most prominent were chosen and marked. Twenty-five units of botulinum toxin were divided and injected in these two points. The masseter muscle could be easily palpated as the teeth were already clenched due to

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Fig. 1 (A) Before Botox injection: Interincisal distance = 0; severe trismus with overlapping of upper and lower rows of teeth. (B) Two months post Botox: Interincisal distance = 10 mm. Effect was maintained for a period of 10 months.

trismus. The borders of the muscle were defined. A line was drawn from tragus of ear to angle of mouth and two points were chosen along the ramus of the mandible below this line to avoid the pterygoid muscles and 25 units of botulinum toxin were injected in divided doses in these two points.

Results

The following were the outcome measures used:

1. Increase in mouth opening was assessed by measuring the interincisal distance with calipers.
2. Goal attainment scale³ (GAS) calculation was done using the GAS calculation Excel sheet.⁴

Post botulinum toxin injection, the interincisal distance improved from 0 mm at baseline to 3 mm 2 weeks later, 5 mm after 1 month, and 10 mm after 2 months (► **Table 1**). As a result, it was possible to perform suction and oral care more easily and effectively (► **Figs. 1B** and **2B**). The effect was maintained for a period of 10 months.

GAS scoring was done for the following parameters: (1) mouth opening, (2) oral suction, and (3) oral care. GAS T score improved from 26.2 at baseline to 68.7 after botulinum toxin A injection with a change of 42.5, indicating a favorable outcome.

Table 1 Time line showing improvement in interincisal distance, post Botox injection

Period after Botox injection	Interincisal distance
Baseline	0 mm
2 weeks	3 mm
1 month	5 mm
2 months	10 mm
Till 10 months	10 mm

Discussion

In trismus, spasticity of muscles of mouth closing, namely masseter, temporalis, and medial pterygoid muscles, results in mouth closure and inability to open mouth. In brain stem lesion, trismus occurs due to abnormalities in the programming of the nucleus of trigeminal nerve.⁵ Trismus in brain stem lesions can happen even before the onset of generalized spasticity. It can also occur in the absence of generalized spasticity.⁶ In our patient, the trismus became severe by 2 months after admission.

Fig. 2 (A) Pooling of secretions, before Botox injection, due to inability to perform effective suction. (B) Suction after Botox.

Review of literature using PubMed showed only one report⁷ on the use of baclofen for trismus in vegetative patients but we could not find any report on injection of botulinum toxin A for trismus in such patients. However, it is not uncommon in rehabilitation practice to inject botulinum toxin A for spasticity management in vegetative patients⁸ to facilitate nursing care. For example, botulinum toxin A injection is used to correct hip adductors spasticity to facilitate perineal hygiene, catheterization, and bed positioning.

In our patient, the spasticity in the muscles of mastication was more prominent than the generalized spasticity. Further oral baclofen treatment did not help to relieve the trismus. Since our goal was to facilitate mouth opening, it was appropriate to inject the masseter and temporalis muscles with botulinum toxin A.

Mor et al have discussed electromyographic localization for injecting botulinum toxin in masseter and temporalis muscle to treat temporomandibular myofascial pain.⁹ Quezada-Gaon et al concluded that using ultrasonogram for guiding injection¹⁰ in masseter muscle improves outcome in patients with masseteric hypertrophy compared with clinical marking. In our young patient on ventilator, these techniques were not applicable. Hence, the muscles were injected by palpation and clinical marking technique.¹¹ While injecting the masseter muscle to avoid injecting the facial muscles, the ramus of the mandible could be palpated with the needle and then the needle withdrawn 1 or 2 mm before injecting, as described by Ahn et al in their study on botulinum toxin injection for masseter reduction.¹²

Seo et al used a dose of 50 units to masseter and 20 units to temporalis for trismus in a 62-year-old female patient with brain stem stroke.¹³ In our patient, we decided on the dosage of 25 units each to temporalis and masseter muscles based on severity of the spasticity in the temporalis and masseter muscles and weight of the patient.

We used the interincisal distance and the GAS score to measure outcome in our patient.

Interincisal distance is the distance between the lower border of upper incisors and the upper border of lower incisors. Mouth opening as measured by interincisal distance improved progressively from 3 mm at 2 weeks to 5 mm at 1 month and 10 mm at 2 months after injection. Subsequently, suction and oral hygiene could be performed with ease by the nursing staff. Spillane et al¹⁴ reported an increase in interincisal distance to 2.5 cm after three sets of injections of Botox that were maintained up to 1 year in a 9.5-year-old boy with stroke-induced trismus. In our patient, the results were maintained up to 10 months with a single set of injection.

Goal Attainment Scale is a method of scoring the extent to which patient's individual goals are achieved in the course of intervention. Three to four goals are set and they are scored based on a mathematical formula giving weightage to the importance of the goal and difficulty in achieving it and T score is calculated (GAS T). In our case, we chose the following as our goals: (1) mouth opening, (2) oral suction, and (3) oral hygiene. In our patient, GAS T improved from 26.2 at baseline to 68.7 at outcome. The change in GAS obtained by subtracting the outcome GAS from baseline GAS was high at

42.5, indicating a favorable outcome in obtaining our goals for the injection.

Conclusion

In spastic trismus, botulinum toxin A injection to masseter and temporalis muscle relieves spasticity and increases mouth opening. In intensive care unit, it can be a valuable tool to treat trismus early and effectively to facilitate suction and oral hygiene and hence prevent life-threatening complications. Ours is the first report on such a treatment in a very young child of age 3.5 years in a pediatric intensive care setting.

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