Botulinum toxin type A combined with cervical spine manual therapy for masseteric hypertrophy in a patient with Alzheimer-type dementia: a case report

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Received 3 July 2012; received in revised form 8 October 2012; accepted 12 October 2012

Key indexing terms:

Botulinum toxin
type A;
Stroke;
Physiotherapy;
Muscle spasticity;
Masseter muscle,
cervical spine;
Alzheimer disease

Abstract

Objective: The purpose of this case study is to present the findings of combining botulinum toxin type A (BoNT-A) and cervical spine manual therapy to address masseter muscle spasticity in a patient with Alzheimer-type dementia.

Case Report: A 78-year-old woman with bilateral spasticity of the masseteric regions for 2 years was referred for physiotherapy. She had trismus and bruxism, and could neither close nor open her mouth normally; thus, she was unable to be fed orally in a normal manner.

Intervention and Outcome: The patient underwent combined treatment with BoNT-A and cervical spine manual therapy. A medical physician (neurologist) performed the BoNT-A injections into 2 points at the center of the lower third of the masseter muscle. A physical therapist performed manual therapy interventions targeted at the cervical spine. Manual therapy started the day after the BoNT-A injection and continued for 5 sessions per week for a total period of 2 weeks. Clinical outcomes were measured including spasticity (Modified Ashworth Scale), functionality (Barthel Index), and jaw opening. Outcomes were conducted at baseline, 2 weeks after treatment, and at 2-month follow-up session after finishing the treatment. The patient improved in all of the outcomes at the end of treatment, and these

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results were maintained during the follow-up. After treatment, the patient was able to feed with minimal caregiver dependency because oral feeding was possible.

Conclusion: The patient in this study responded positively to a combination of BoNT-A and manual therapy, resulting in decreased masseter muscles spasticity and improved trismus and bruxism.

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Introduction

Bruxism is often noted in patients with altered states of consciousness, but its occurrence after brain injury is unknown.¹ Resolution of bruxism may be associated with improvement in the level of consciousness.² It seems that there is an intimate functional relationship between the temporomandibular joint and the cervical spine, as suggested by their anatomical and biomechanical interrelationships, although current evidence is conflicting.³ Eriksson et al⁴ reported that mouth opening is accompanied by head-neck extension and mouth closing is accompanied by head-neck flexion, with a precise coordination between jaw-neck movements dependent on the speed of the movement.⁵

Botulinum toxin type A (BoNT-A) is widely used for treating spasticity and other forms of muscle overactivity.⁶ Botulinum toxin type A is derived from *Clostridium botulinum*; and when injected into a spastic muscle, it inhibits acetylcholine release, causing a blockade of the neuromuscular patches without affecting the antagonist muscles.⁷ Botulinum toxin type A has been used in the past with mild improvements in conjunction with physiotherapeutic approaches.^{8,9} However, to our knowledge, publication of this combination with manual therapy has not been reported. There are only a few randomized controlled studies and some systematic reviews^{10,11} reporting a favorable effect of BoNT-A on craniofacial and neck pain as well as bruxism. Nevertheless, previously published studies have not investigated the effect of BoNT-A on neck pain caused by bruxism because they only assessed jaw pain due to bruxism or neck pain due to cervical dystonia.¹²

The purpose of this case study is to present the findings from combining BoNT-A and cervical spine manual therapy for a patient with masseter muscle spasticity and Alzheimer dementia disorder.

Case report

A 78-year-old woman (weight, 52 kg; height, 154 cm) with Alzheimer-type dementia since the age of

65 years and a bilateral spasticity of the masseteric regions that persisted for 2 years presented for care. The patient was referred to the Department of Physical Therapy, Residenza Sanitaria Assistenziale "A. Maritano," Sangano, Italy, for trismus and bruxism. She could neither close nor open her mouth normally. Clinical examination revealed a mouth opening of 0 mm. The patient received oral tube feeding at home because normal oral feeding was impossible. On pretreatment, the Barthel score was 60 and the Modified Ashworth Scale (MAS) was 5. Extraoral examination revealed bilateral hypertrophy of the masseter muscles. Magnetic resonance imaging and ultrasonographic imaging showed no underlying pathology.

The patient and her family agreed to undergo the combined treatment. Before the procedure, the nature and the established use of BoNT-A as well as its potential adverse effects were explained; and signed informed consent was obtained from the patient. Consent for publication in this study was also given.

Physical examination included the MAS for grading spasticity,^{13,14} the Barthel Index,¹⁵ and the active pain-free mouth opening¹⁶ to assess functionality. The MAS has been criticized, and both its reliability and validity have been challenged because it is unable to distinguish between the neurological and mechanical contributions to the stiffness levels observed during passive movement.¹⁷ It is, therefore, an indirect measure of spasticity because it cannot establish the presence of (or rule out) abnormal muscle activation.¹⁸ Physical disability was measured with the Barthel Index. The range of this instrument is between 0 and 60, with lower scores indicating more severe disability.¹⁹ For mouth opening, the patient in a supine position was asked to "open the mouth as wide as possible without causing pain."²⁰ At the end position of mouth opening, the distance between upper and lower central incisors was measured in millimeters.²⁰ The mean of 3 trials was calculated and used for the analysis.²⁰ The measurements were performed before the BoNT-A inoculation, at the end of manual therapy treatment (2 weeks after the

Fig 1. (A) Upper cervical flexion mobilization; (B) C2-C5 central posterior-anterior mobilization. (Color version of figure is available online.)

injection), and at the 2-month follow-up period after the end of treatment.

The medical physician (neurologist) carried out a treatment consisting of BoNT-A injection (Xeomin, Merz Pharmaceuticals, Milan, Italy) in both masseter muscles (200 IU). One hundred international units of BoNT-A was injected equally into 2 points at the center of the lower third of the masseter muscle that were located 1 cm from each other. The muscle was localized by the standard anatomical landmarks used in needle electromyography.²¹ Standard dilutions of BoNT-A in isotonic sodium chloride solution were used.

The physical therapist carried out the manual therapy treatment protocol including interventions targeted at the cervical spine. Manual therapy started the day after the BoNT-A injection and was continued for 5 sessions per week for a total period of 2 weeks. Throughout the course of treatment, the neurologist and physical therapist were in constant contact.

During the 10 treatment sessions, the patient received the following manual therapy techniques:

Upper cervical flexion mobilization²²

The patient was supine with the cervical spine in a neutral position. The therapist contacted the occipital bone with the first finger and the medial aspect of one hand and over the frontal region of the patient's head with the other hand. The mobilizing force was delivered by flexing the upper cervical region using a combination of cephalic traction with the occipital hand and caudal pressure with the frontal hand (Fig 1A).

C2-C5 central posterior-anterior mobilization²⁰

The patient was supine with the cervical spine in a neutral position. The therapist placed the tips of his indexes on the posterior surface of C2 to C5 spinous processes, while the thumbs were gently placed around the patient's neck. A grade III (large-amplitude movement that moved into the resistance limiting the range of movement) posterior-anterior technique was applied centrally to the C2, C3, C4, and C5 spinous processes (Fig 1B).

The mobilizations were applied at a rate of 2 oscillations per second. The mobilizations were performed for a total of 9 minutes, divided into 3-minute intervals with a 1-minute rest in between.²⁰

The outcomes are summarized in Table 1. The clinical presentation of the patient improved following the treatment if compared with her clearly deteriorated feed function after Alzheimer-type dementia (Fig 2A). The patient's compliance with the treatment was 100%, showing full tolerance. Spasticity also decreased after treatment. The MAS score at the masseteric region was 2 after treatment. At follow-up, these results were maintained: the MAS of the masseter muscles decreased to 1, indicating an increase of functionality (Table 1). Therefore, the spasticity was reduced by 60%

Table 1 Changes in outcome measures of the patient

	Pretreatment	Posttreatment	Follow-up
MAS			
Masseter right	5	2	1
Masseter left	5	2	1
Mouth opening	0 mm	15 mm	20 mm
Barthel Index	60	58	58

Fig 2. (A) Extraoral view of the patient before treatment; (B) extraoral view of the patient 2 weeks after treatment. (Color version of figure is available online.)

at the end of the treatment and by 80% at 2-month follow-up.

Mouth opening also improved after the treatment. Two weeks after the BoNT-A injection, the patient had exhibited a great improvement in trismus showing a mouth opening of 15 mm, with no bruxism reported. After the 2-month follow-up period, the patient showed signs of good recovery from trismus; and mouth opening was 20 mm. The patient was able to feed better and with minimal caregiver dependency.

Finally, the Barthel Index also improved. It was decreased up to 58 after 2 weeks of treatment and maintained at the same level at the 2-month follow-up period. Two weeks after the BoNT-A injection, she could feed better with caregiver dependency; spasm was reduced; and mobility and activities of daily living improved (Fig 2B).

Discussion

This case report showed that the combination of BoNT-A injection and manual therapy was able to improve severe masseteric hypertrophy in a patient with dementia. The subject had experienced her symptoms for 2 years before beginning the treatment. Before treatment, her spasticity and mouth opening had failed to respond to various medical therapies and dental procedures, providing further evidence of the severity of her condition. In the current study, 2 weeks after the application of BoNT-A in conjunction with manual therapy to the cervical spine, she demonstrated an improvement in spasticity, trismus, and bruxism. At the 2-month follow-up period, the improvement was maintained. Different treatment modalities have been reported to be useful for severe masseter muscles spasticity and related disorders,²³

but there is no general agreement as to which is the best therapeutic option.¹

Although no central nervous system structures associated with teeth grinding have been identified, it has been speculated that, in some cases, bruxism may be a part of dystonia because both share similar pathophysiology.²⁴ Bruxism involves jaw clenching and grinding of the teeth, commonly causes myofascial pain, and is often resistant to treatment.² A higher prevalence rate of bruxism has been reported in cranial-cervical dystonia compared with healthy control.²⁵ Patients with neurological disorders who are in a comatose state can also have severe bruxism.²⁴ The activation of phasic jaw activity may depend on the interaction between the motor, limbic, and autonomic systems, resulting in either disinhibition or facilitation of a central bruxism generator.¹ Cases have also been reported of total recovery from bruxism.^{1,26} Some authors showed that jaw muscle paralysis induced by BoNT-A may disrupt the feedback loop from the trigeminal motor nucleus and inhibit the central bruxism generator.¹ Santamato et al² described the clinical improvement of a case report with nocturnal bruxism and related neck pain after a bilateral treatment with BoNT-A injections in the masseter and temporal muscles.

Previous studies concerning BoNT-A treatment of bruxism describe successful treatment of bruxism associated with TMD or traumatic brain injury.^{27,28} In fact, bruxism and temporomandibular disorders are usually associated with neck pain and muscle hyperactivity without specific anatomical modification.^{29,30}

To the best of the author's knowledge, the current study is the first case report in which manual therapy to the cervical spine combined with BoNT-A therapy showed promising results by improving masseter muscles spasticity and mouth opening in a patient with masseteric hypertrophy.

Limitations

The current case report demonstrates only a single case; and, therefore, the management protocol cannot be generalized to all cases of severe bruxism. A cause-and-effect relationship cannot be inferred through one case report. Because of this and also the lack of a placebo intervention, further studies are necessary to establish whether this patient would have improved without treatment; however, this is unlikely because of the severity and the duration of her symptoms.

Perhaps the biggest disadvantage of BoNT is that the treatment effect seems to be limited to 6 months, after which the original condition returns.³¹ Therefore, patients have to be informed about the recurrence rate after the procedure. Unlike surgical excision of muscle tissue that reduces the actual number of muscle cells, BoNT-A reduces muscle volume temporarily.³² Tan and Jankovic¹ reported that the total effect of each injection lasted up to 19.1 weeks. However, the BoNT-A treatment combined with manual therapy improved the effects during the follow-up sessions; the effects were maintained up to 6 months.³³ Moreover, as with any case report, the findings may have been due to several factors, including the natural history of the condition, concurrent life events, and other treatment modalities used by the patient in the same period. The results of this case report are promising and justify further clinical controlled studies to confirm the current results.

Conclusion

The current case report showed that combined manual therapy and BoNT-A injection decreased masseter muscles spasticity and improved trismus and bruxism in a patient with Alzheimer dementia, enabling her to be fed normally.

Funding sources and potential conflicts of interest

No funding sources or conflicts of interest were reported for this study.

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