Effect of botulinum toxin type A on the cervical sympathetic trunk in chronic relapsing inflammatory optic neuropathy

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To the Editor: Optic neuritis (ON) usually ends with one episode, but recurs in 3% to 5% of patients. Among recurrent patients, those that emerge negative from multiple sclerosis or neuromyelitis optica (NMO) workup are diagnosed with chronic relapsing inflammatory optic neuropathy (CRION). This disease group, first named by Kidd et al.,[1] refers to a clinical situation in which inflammatory optic nerve inflammation on both sides occurs repeatedly and worsens when steroids or immunosuppressants are discontinued. Early diagnosis is important in this group because the risk of blindness is significant, and as such, treatment is focused on preserving vision. However, because a significant number of CRION patients suffer from ocular pain, alleviation of pain should not be overlooked.

The cervical sympathetic trunk (CST) is an anatomical structure through which all of the sympathetic nerves of the face and neck pass.[2] A cervical sympathetic block (CSB) is commonly used as an alternative to control pain. When only a local anesthetic is used, the analgesic effect may be temporary, although it can be prolonged via radiofrequency treatments. Botulinum toxin (BTX) administered to the lumbar sympathetic trunk has been reported to lengthen the analgesic period, and an injection of BTX during CSB may confer similar effects, although this method has received little attention. The case presented here is the first in which BTX was successfully used concurrently with CSB for the treatment of CRION-induced ocular pain. This case was approved by the institutional review board of our institution (IRB No. KC20ZISI0004).

A 34-year-old woman diagnosed with CRION 3 years earlier visited the clinic presenting with biocular pain due to ON. Although the ON was previously treated at another hospital, the patient experienced persistent biocular pain over the past 2 years. Her numeric rating scale (NRS) pain score was 8–10/10; the pain was exacerbated by eye movement and wind exposure. After a neurological examination, it was concluded that she was completely blind in the right eye, and the left eye was gradually losing sight. NMO-immunoglobulin G was seronegative. Sella magnetic resonance images showed contrast enhancement of the bilateral inflamed optic nerves. Medical records from the previous hospital proved that she had responded to immunosuppressive treatment and experienced relapse upon dose reduction of immunosuppressive treatment. Although she continued immunosuppression treatment under consultation with the neurology department, proper pain treatment had never been applied. Initially, CSB using a local anesthetic was performed several times under ultrasound guidance, but the analgesic effect did not last >1 day. Pulsed radiofrequency of the CST (42°C, 120 s, four cycles) was performed on both sides at regular intervals but did not affect.

Next, CSB with BTX was performed under ultrasound guidance. First, after placing a 13 to 6 MHz linear probe in the neck, the location of the CST was confirmed at the surface of the longus capitis at the level of the fourth cervical vertebra.[3] The needle was placed on the longus capitis, and a mixture of BTX and bupivacaine was injected into the site [Figure 1]. Because her right eye had no vision, the patient asked for the left side to be treated first. Fifty units of BTX (Botox®, Allergan Inc., Irvine, CA, USA) were mixed with 5 mL of 0.5% bupivacaine and injected into the left CST. Approximately 3 days after the injection, the NRS pain score decreased from 10 to 5, with the effect lasting for 4 weeks. She could open her left eye, which was previously not possible due to severe pain. One week after the injection, however, she complained of dysphagia and neck weakness, which lasted for 4 weeks and resolved without targeted treatment. In the second trial, 25 units of BTX mixed with 5 mL of 0.2% bupivacaine were injected around the left CST. Similar to the first trial, the patient complained of dysphagia and neck weakness. In the third trial, 7.5 units of BTX mixed...
with 0.3 mL of 0.2% bupivacaine were injected into the left CST. Neither analgesia nor side effects were observed. In the fourth trial, 10 units of BTX mixed with 1 mL of 0.2% bupivacaine were injected into the left CST. The analgesia lasted approximately 4 weeks without any side effects. Finally, the patient was treated successfully on both sides.

This case report demonstrates that BTX injected around the CST is effective for managing ocular pain arising from CRION-induced ON. CSB is commonly applied to alleviate head and neck neuropathic pain.[2] Because the effect of a single sympathetic nerve block can be short, an alternative to prolong the duration of analgesia is pulsed radiofrequency of the CST. This method did not affect the patient in this study, presumably because of the difficulty in evoking sensory perception via sensory stimulation while performing pulsed radiofrequency in the CST. Our next step was to inject BTX into around the CST, although no report has been published regarding the utility of this procedure for alleviating ocular pain. In a study using rabbits, CSB with BTX caused miosis (duration of 1 month) without pathological changes.[3]

Because few studies have been assessed injections of BTX during CSB, the optimal dose and volume of BTX are unknown. Also, side effects have been scarcely reported. In studies pertaining to cervical dystonia, BTX injection into the cervical muscle is preferred.[4] When cervical dystonia is treated with onabotulinumtoxinA, the incidence of dysphagia is 7.1%. When BTX is administered into the sternocleidomastoid muscle, dysphagia may result from pharyngeal muscle weakness due to the regional spread of BTX.[4] Chang et al[5] hypothesized that the occurrence of dysphagia following BTX injection into the sternocleidomastoid results from hyperactivation of the suprahyoid and infrahyoid muscles to compensate for a weakened sternocleidomastoid. In reports on cervical dystonia, the total dose of onabotulinumtoxinA has varied, ranging from 60 units to 374 units. In this case report, the doses were smaller than those used for cervical dystonia. The dysphagia that followed CSB, even at the lower dose of 50 units, may have been because the extent of dysphagia was greater when administered over the fascia between the muscles than when administered intramuscularly. To confirm the safest and most effective BTX dose and volume, a clinical study with a larger number of patients is necessary.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s)/patient’s guardians has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the article. The patients/patient’s guardians understand that their names and initials will not be published and due efforts will be made to conceal the identity of the patient, although anonymity cannot be guaranteed.

**Conflicts of interest**

None.

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