

A Case of Posttraumatic Cervical Dystonia Treated with OnabotulinumtoxinA

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Abstract

Cervical dystonia (CD) is a condition that typically presents with cervical muscle spasm, producing head tilt and cervical rotation. CD is most often idiopathic, however, in a small number of patients, CD occurs within one day to one year after mild to severe trauma. This type of CD is further classified as posttraumatic CD. OnabotulinumtoxinA (Botox) injections are considered to be a controversial treatment for posttraumatic CD and have produced variable result. This report describes the case of a 32-year-old female presenting with a two year history of posttraumatic CD and associated head, neck, and shoulder pain after obtaining a severe head injury during a motorcycle accident. OnabotulinumtoxinA was used to successfully treat her posttraumatic CD muscle spasms and associated chronic pain. Three months after her first and second ONA treatments, the patient reported at least 50% improvement in her overall pain symptoms and a noticeable reduction in cervical paraspinal muscle spasms.

Keywords

Posttraumatic Cervical Dystonia, Traumatic Dystonia, Botox Injections, OnabotulinumtoxinA, Chronic Pain

1. Introduction

Cervical dystonia (CD) is the most common type of adult-type focal dystonia and is typically marked by palpable muscle spasm of the head, neck, and shoulder musculature that typically result in unilateral head tilt and contralateral cervical rotation [1] [2]. Associated symptoms include neck and shoulder pain, persistent head tremor, and nystagmus [1]. A review of over 260 patients with CD

revealed that only 62% of patients with CD presented with typical CD, which is marked with cervical tilt and rotation, which is the typical presentation of CD [2]. The other 38% of patients who do not show typical symptoms of CD fall under the category of atypical CD and can be difficult to diagnose [2]. If there is a delay in diagnosis or CD remains untreated, chronic pain can develop leading to debilitating effects for patients [3]. In two separate studies, 90% of patients with CD reported having pain that they attributed to CD [4] [5].

Several causes of CD have been identified including idiopathic [2] [6], medication-induced [7], and posttraumatic [3] [8]. Posttraumatic CD presents after mild to severe traumatic injuries ranging from whiplash to blunt force trauma, and the onset can occur between 1 day and 1 year after the traumatic incident [8]. Posttraumatic CD caused by peripheral trauma is controversial and must meet certain criteria. These criteria include that the trauma must be severe enough to produce at least two weeks of local symptoms, the dystonia must be related to the region that was injured, and the onset must be within days to 1 year after the trauma [8]. The pathophysiology of CD is thought to involve the activation of agonist and antagonist muscle spasms of cervical musculature [3] [9]. One current theory, which has been supported by an MRI study, indicates that CD is a result of pathologic sensorimotor activation throughout the central and peripheral nervous system [10].

Long-term treatments that have been used for CD include pharmaceuticals including anticholinergic agents, OnabotulinumtoxinA (ONA) injections [11], surgery, and non-pharmaceutical therapies such as physical therapy, osteopathic manipulative treatment, acupuncture, and massage therapy. Current research and preliminary clinical trials support the use of ONA as first line treatment for CD [11] [12]. ONA, brand name Botox, is designed to replicate the effects of Botulinum Toxin A by causing flaccid paralysis through inhibition of acetylcholine release at the neuromuscular junction. The mechanism of ONA pain relief is still under investigation, however, leading theories indicate that it involves the inhibition of peripheral and central nerve sensitization leading to a decrease in secondary nociceptive neuron activation [13]. Once injected into a spasmodic muscle, ONA can provide immediate muscle spasm and neuropathic pain relief for a period of up to three months after the ONA injection [14]. Although pain may persist, repeat injections of ONA have been associated with significant reduction of CD and several types of neuropathic pain [12]. Some adverse side effects of local ONA injections include temporary local muscle paralysis and injection site injuries such as bruising and bleeding. Therapeutic dosage of ONA injections for cervical dystonia treatments is less than 50 units per injection site [15]. Systemic toxicity is not commonly experienced, and it has been estimated that a dose of 2800 units of ONA would be necessary to cause systemic effects in a 70 kg patient [15].

This case study demonstrates a successful ONA treatment of posttraumatic CD, which is defined as a greater than 30% reduction in pain duration and in-

tensity [16]. A few case studies have been produced detailing the use of ONA to treat post-traumatic CD with mixed results, and we hope to add to this limited number of case reports by detailing a successful treatment of posttraumatic CD with ONA.

2. Case Presentation

A 32-year-old female presented for initial evaluation with a history of chronic head, neck, and shoulder pain associated with daily headaches. The pain began after a motorcycle accident two years prior to her initial evaluation where the patient was not wearing a helmet. Since the accident, the patient has noticed a right-sided head tilt and states that in order to turn her head she has to turn her entire body. She describes her pain as aching, burning, throbbing, and shooting. She also reports bilateral hand weakness with intermittent numbness and tingling of all of her digits. On the Visual Analogue Scale (VAS) for pain, the patient reports her pain is a 7 - 8/10 in severity. The patient's headaches occurred more than 15 days per month with each incident lasting between 4 - 6 hours. During these incidents, the patient reports light and sound sensitivity with nausea and vomiting. Her pain and headache symptoms are exacerbated by lifting, bending, weather, standing, and sitting and relieved by rest, lying down, and sitting. She underwent unsuccessful treatment with Tylenol, Gabapentin, Triptans, Beta Blockers, Ibuprofen, Excedrin Migraine, Bayer, Cyclobenzaprine, Tramadol, Osteopathic Manipulative Therapy, and Physical Therapy.

During the encounter, the patient reported nausea, joint pain and arthritis, and numbness without dizziness. Her past medical history is significant for a heart murmur and thyroid disorder. Along with her motorcycle accident, the patient was involved in a motor vehicle accident 5 years prior where she experienced whiplash and fractured her clavicle, which required surgical repair. She drinks socially, smokes 1 pack of cigarettes per day, and uses marijuana occasionally. For medications, the patient uses Ibuprofen, Levothyroxine sodium, and Tylenol. Her allergies include Penicillin, Amoxicillin, and paper tape.

During her exam, the patient was alert and oriented to person, place, and time. She did not have any neurological deficits, and she had 5/5 strength throughout bilateral upper and lower extremity myotomes and full range of motion in all major joints except her neck in all planes and right shoulder. Paraspinal muscle spasm was palpated throughout her cervical, thoracic and lumbar regions with tenderness bilaterally in her trapezius, posterior neck, and occiput. We noted a right cervical head tilt and left cervical rotation with persistent involuntary spasm of the cervical paraspinal groups associated with pain upon palpation leading to a "110%" replication of pain location and type, including her right shoulder pain. Based upon patient history and physical exam findings, the patient was diagnosed with posttraumatic CD with associated post-concussive migraine headaches and spasm. The patient elected to undergo ONA injections

as treatment.

3. Management and Outcome

One hour prior to the procedure, the patient's vitals were stable, and she was sedated with oral Alprazolam. After pre-procedure checklists and monitoring were performed, trigger points were identified, marked, and scrubbed with Chlorhexidine. Then, ONA injections were performed in procerus, bilateral corrugator, frontalis, temporalis, occipitalis, cervical paraspinal and trapezius muscle groups using a 30 gauge 0.5 inch needle, delivering a total of 155 units of ONA divided equally between points. Prior to each injection, aspiration was negative for blood, CSF, or air. Once the procedure was finished, the patient's skin was cleaned and sterile bandages were placed. No complications were noted during the procedure and the patient's vitals remained stable. The patient reported a pre- and post-procedure VAS pain score of 7/10 and 9/10, respectively.

On the same day as her initial ONA treatment, the patient was traumatically assaulted, and her head was slammed onto the pavement multiple times. She did not lose consciousness, however, she experienced gradually worsening head and neck pain that was affecting her quality of life. At an urgent care center, she reported "the worst headache of her life." An x-ray was performed revealing no acute changes of her head and neck.

Upon follow up after 4 weeks, the patient noted that her headaches lessened in both frequency and intensity, but her neck pain had increased. She had not experienced any side effects or complications due to the procedure. Her VAS pain score was 8/10 at the visit. Aside from the increased pain, the patient noted a significant increase in her neck range of motion and states that her headaches may have been so intense before that she did not notice her neck pain. The patient states that she "feels like a new person" and that for the first time in 3 years she can vacuum her house, do daily activities, and take her kids to school. The patient's significant other stated that he noticed "a total difference." Physical exam revealed improvement in neck and shoulder range of motion, and her CD, although still present, was improved without persistent involuntary spasm of cervical paraspinal muscle groups. Four weeks after the first ONA injections, the patient attributed at least 50% improvement in overall pain symptoms to her ONA treatment.

The patient was followed up with again two months after her initial ONA treatment. At this appointment, she continued to have at least 50% reduction in overall pain symptoms and cervical paraspinal muscle spasms. Based on this positive procedure outcome, she was scheduled to receive a second round of ONA injections 12 weeks after her first injections. When she arrived for her second round of ONA treatment, she stated "I am a normal person again, I can do laundry, clean my house, and do things with my kids." Three months after her second round of ONA injections, she has continued to show greater than 50% reduction in her pain, chronic migraines, and cervical paraspinal muscle spasms with no short or long-term side effects.

4. Discussion

In this paper we discussed a rare presentation of posttraumatic CD with associated debilitating migraine headaches for two years after a motorcycle accident. Our patient's chronic cervical muscle spasms likely contributed to her chronic migraine headaches, and the use of ONA was successful in treating her post-traumatic CD spasms and associated migraine headaches. Although she indicated a higher post-procedural VAS pain score, she attributed this increase to a higher self-awareness that she could have only achieved with the relief of her chronic migraines. Pain management treatments can be considered to be therapeutic when an improvement of greater than 30% is achieved [16] [17]. Her first treatment provided 50% or more relief overall pain and spasm symptoms for three months, and a second treatment was able to continue her improved of more than 50% for another three months. These six months of improvement were well above the 30% cutoff and indicate that ONA treatment is a successful therapeutic treatment for her posttraumatic cervical dystonia and associated pain.

Posttraumatic CD has been shown to induce chronic migraine headaches that are often difficult to diagnose and treat [18]. A study produced in 1993 documents two patients who were treated with ONA for posttraumatic CD with only a "mild" improvement in symptoms [3]. This report documents a successful treatment of posttraumatic CD with ONA injections, and it provides support for further research into whether or not ONA should be a standard treatment for posttraumatic CD. The next step in researching this issue would be a study that compares the percentage and duration of headache and pain relief that patients with posttraumatic CD experience after ONA injections.

While posttraumatic CD can often be difficult to diagnose and manage, physicians should consider the use of ONA for pain management and spasm relief. In this report, we describe a patient with posttraumatic CD that was treated successfully with two rounds of ONA injections, which have provided a total of six months of at least 50% improvement in overall pain and cervical paraspinal spasm symptoms.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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