DOI: 10.7860/JCDR/2022/52959.15957

Case Report

Physical Medicine and Rehabilitation Section

Botulinum Neurotoxin Injection as a Therapeutic Option in Thoracic Outlet Syndrome Caused by a Supernumerary Scalenus Muscle: A Case Report

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ABSTRACT

Thoracic Outlet Syndrome (TOS) is a neurovascular syndrome and may occur as a result of compression of brachial plexus elements and/or subclavian vessels as they traverse the cervicoaxillary canal. One of the causes of TOS is the existence of a supernumerary scalene muscle, usually denominated as scalenus minimus. Botulinum Toxin Type A (BoNT-A) acts by binding presynaptically to high-affinity recognition sites on the cholinergic nerve terminals and decreases the release of acetylcholine, suppressing muscle overactivity. BoNT-A reduces contraction in injected muscles, causing focal chemodenervation and has been described as a non surgical effective treatment in selected cases of TOS. Here, is the case of a 44-year-old woman diagnosed with refractory TOS, who presented with paresthesia in the fourth and fifth fingers of her right hand, associated with a feeling of lack of coordination in the same hand. On Magnetic Resonance Imaging (MRI), a supernumerary scalene muscle was identified as a probable cause of TOS. She was treated with chemodenervation of the scalenus anterior muscle (30 Units) and scalenus medius muscle (30 Units), using incobotulinumtoxin A. The patient had a very significant clinical improvement after Anterior Scalene (AS) and Middle Scalene (MS) muscle BoNT-A injections. Incobotulinumtoxin A neurotoxin injection is a therapeutic option in TOS.

Keywords: Anatomic variation, Botulinum toxin type A, Brachial plexus, Scalenus minimus

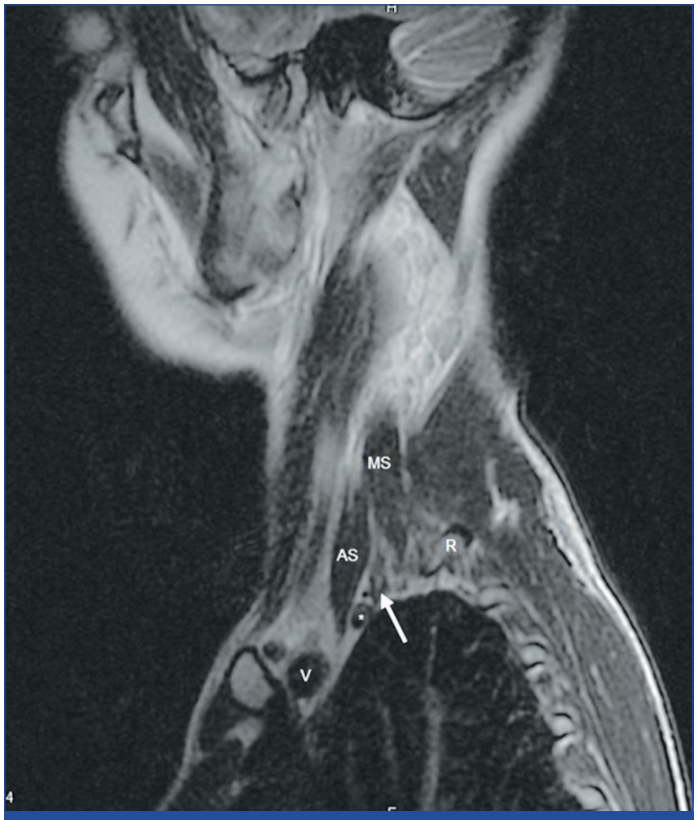
CASE REPORT

A previously healthy 44-year-old woman presented with paresthesia in her right fourth and fifth fingers, oedema, and cyanosis of the hand, which she described as “tingling and discomfort with a bluish discoloration of the skin”. The patient was right-hand sided, and she worked as a bank officer. Her complaints had started insidiously about a year before the patient presented to the clinical neurophysiology department.

She had no known history of musculoskeletal injuries in the affected limb and no known changes of radiculopathy, disc herniation or spondylarthrosis. The symptoms were not continuous as paresthesia was more severe during the night as she slept with her right hand on the back of the head with an abducted shoulder. All the symptoms were triggered and worsen by the execution of physical activities, such as walking and playing tennis, and with daily life overhead movements. About six months before being referred to the clinical neurophysiology department, the patient noticed a decrease in her right-hand fine motor skills, which she noticed in tasks such as brushing teeth or writing, due to loss of sensation, numbness, and decreased coordination. These complaints would last for 10-15 minutes, around four times a week.

Right upper limb Electromyography (EMG) and nerve conduction studies did not reveal any abnormalities. Brachial plexus and cervical spine MRI showed the existence of a right accessory scalene muscle between the AS and MS muscles, with a significant reduction in the interscalene space [Table/Fig-1]. Computed tomography angiography revealed compression of the right subclavian vein at the costoclavicular junction with a shoulder abduction maneuver.

The patient had previously been diagnosed with TOS. About six months prior to the first appointment at the clinical neurophysiology department, she started first-line treatment with physiotherapy and anti-inflammatory drugs (naproxen 500 mg every 12 hours, for periods always shorter than 10 days), that proved insufficient,



[Table/Fig-1]: Magnetic Resonance Imaging (MRI) of the brachial plexus and costoclavicular interval, sagittal plane, T2 Fast Spin Echo. A scalene minimus muscle (white arrow) is present, causing narrowing of the scalene triangle.
Note the Anterior Scalene muscle (AS), Middle Scalene muscle (MS), subclavian artery (*) and vein (V), and first rib (R). A partial pass-through brachial plexus is admitted, with roots of C5 and C6 passing through the AS muscle

after three months of treatment. Physiotherapy sessions included relaxation techniques and deep friction massage. In order to open the thoracic outlet by raising the shoulder girdle and opening the

costoclavicular space, the patient was prescribed strengthening exercises of the levator scapulae, sternocleidomastoid and upper trapezius, serratus anterior and lower trapezius. The treatment also included stretches and postural correction exercises. Symptoms persisted, although less continuous, with longer intervals between the onset of oedema and paresthesia (about two episodes per month). According to the patient's description, functionality remained compromised. She had difficulty lifting weights above her head, opening lids and reported decreased sensitivity and coordination during repetitive tasks. At that point, the patient was referred to our hospital.

On physical examination, she presented with normal muscle strength and sensitivity, and deep tendon reflexes were symmetrical. All passive and active articular ranges of motion were preserved. The patient had a negative Adson maneuver [1] as the radial pulse was not markedly diminished nor absent during neck extension and cervical rotation towards the symptomatic side. Ulnar nerve compression and stretching tests did not reproduce symptoms. Ulnar Nerve Tinel test [2] was negative. Regarding functionality, the patient scored 46/80 on the Upper Extremity Function Scale (UEFS) [3]. The UEFS consists of eight questions that are scored out of a possible 10 points. The scale is calculated by summing the points from each individual question, with a range of 0 (no disability) to 80 (maximum disability).

Injection of incobotulinumtoxin-A (BoNT-A) was then proposed by the medical team and was accepted by the patient. EMG-guided chemodenervation of the scalenus anterior muscle (20 units-U of BoNT-A) and scalenus medius muscle (20 U of BoNT-A) was performed using a two-channel EMG machine (Medelec® Synergy; Oxford Instruments, UK). Follow-up was carried out through reassessment appointments. The intensity of paresthesia was measured using a Visual Analogue Scale (VAS). Since, this condition affected functionality, UEFS was applied. Follow-up appointments were scheduled one month after the first injection, at three months, at four and at six months.

At the first re-evaluation, she presented a very significant clinical improvement, with total oedema regression and very rare paresthesia complaints. UEFS score dropped significantly from 46/80 to 8/80, revealing a very satisfactory functionality gain. At three months assessment, she was treated again with chemodenervation of the scalenus anterior muscle (30 U) and scalenus medius muscle (30 U), using BoNT-A. Because chemical denervation is reversible, BoNT-A has temporary effects, with the muscle being progressively reinnervated by nerve sprouting. Usually, intramuscular BoNT-A treatment requires 3-5 months intervals between injections, depending on the anatomical region and symptoms. At the six months assessment, the patient reported significant clinical improvement and improved quality of life. More specifically, the patient reported a decrease in the intensity of paresthesia from 8/10 to 1/10 during physical exercise and from 4/10 to 0/10 at rest, using the VAS. She had a UEFS score of 4/80. The patient will return for follow-up appointments and eventually repeat incobotulinumtoxin toxin injections, if needed. The patient was asked to carefully read and sign an informed consent, regarding publishing her clinical data and MRI.

DISCUSSION

The TOS is defined as an association of clinical signs and symptoms caused by congenital or acquired compression of the brachial plexus or subclavian vessels as they pass through the superior thoracic aperture [4]. TOS can be classified according to aetiology, symptoms, clinical presentation, or anatomy [5]. The most widely used classification is based on the anatomical structures being compressed, dividing it in neurogenic, venous or arterial. Neurogenic TOS (NTOS) is caused by compression of the brachial plexus roots at the interscalene triangle and/or retropectoral space [6]. Symptoms may include pain, upper limb oedema, decreased manual dexterity, paresthesia or numbness in the neck, shoulder, arm, or hand [5,7]. A differentiating feature is that these symptoms worsen with shoulder elevation, overhead movements, or outstretched positions of the arm [5,7,8].Compression in TOS can be caused by a variety of structures, such as cervical ribs, congenital fibrous and muscular band anomalies, supernumerary scalene muscles, or first rib abnormalities [9]. A supernumerary scalene muscle, denominated as scalenus minimus, may be present in the lower part of the interscalene triangle [10]. The incidence of scalenus minimus muscle ranges between 7.8-71.7% [11,12].

The TOS first-line treatments include physiotherapy, anti-inflammatory drugs and analgesic agent injections [12]. Scalenectomy, first rib resection or pectoralis minor tenotomy are reserved for severe cases. Torriani M et al., showed that botulinum toxin injection of AS and pectoralis minor muscles provided temporary relief of symptoms and functional recovery in over two thirds of 41 subjects with suspected NTOS [7]. Foley JM et al., reviewed several case reports and studies using BoNT-A injection to reduce scalene muscle hypertrophy and brachial plexus compression [13]. BoNT-A acts by binding to high-affinity recognition sites on muscle cholinergic nerve terminals, decreasing the release of acetylcholine, hence reducing muscle contraction. Also, analgesic effects of BoNT-A have been described [14]. Due to its analgesic features and its ability to treat muscle overactivity, BoNT-A has been tested in TOS cases refractory to conservative treatment.

In this case report, diagnosis was relatively simple as the patient presented the classic symptoms of oedema, paresthesia, decreased manual dexterity, all aggravated by overhead movements. The identification of an accessory scalene muscle on MRI allowed the identification of a cause, with high sensitivity and specificity. Differential diagnosis would be a cervical disc injury, cervical discogenic pain syndrome, cervical radiculopathy or elbow and forearm overuse injury. Treatment with botulinum toxin was effective. Repeating the injection three months after the first, is a sign of therapeutic success, as the patient showed a significant clinical improvement with incobotulinumtoxin. Botulinum toxin always has a temporary effect, because chemodenervation is reversible. To our knowledge, this is the first case report to describe in detail the complaints, functionality, physical examination and therapeutic approach with incobotulinumtoxin chemodenervation in a case of TOS caused by the existence of a supernumerary scalene muscle, including a six-month follow-up.

CONCLUSION(S)

This case highlights the therapeutic efficacy of incobotulinumtoxin injection in the treatment of TOS caused by an accessory scalene muscle, with compression of the brachial plexus. The patient presented a significant reduction of oedema and paresthesia and showed important gain in functionality.

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PLAGIARISM CHECKING METHODS: [Jain H et al.]
- Plagiarism X-checker: Oct 21, 2021
- Manual Googling: Jan 13, 2022
- iThenticate Software: Jan 18, 2022 (12%)

ETYMOLOGY: Author Origin

AUTHOR DECLARATION:
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Oct 20, 2021**
Date of Peer Review: **Dec 16, 2021**
Date of Acceptance: **Jan 14, 2022**
Date of Publishing: **Feb 01, 2022**