

TREATMENT OF REFRACTORY nTOS AND CRPS IN THE SAME UPPER EXTREMITY WITH A SPINAL CORD STIMULATOR

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Background: Neurogenic thoracic outlet syndrome (nTOS) makes up 95% of all thoracic outlet cases. Symptoms consist of pain, numbness, dysesthesia, numbness, and weakness of the affected upper extremity. Current treatment options include medical management, physical therapy, steroid injections, and surgical decompression. Even patients who undergo surgical management do not always have complete resolution. These patients tend to develop chronic pain secondary to refractory nTOS. The treatment for refractory nTOS is the same as for the primary disease. Unfortunately, this exposes the patient to more invasive surgeries that may not resolve their symptoms. This case report is about a patient with refractory nTOS and complex regional pain syndrome (CRPS) in the same extremity, treated with a spinal cord stimulator, allowing him to avoid repeat surgery.

Case Report: A middle-aged man presented to the interventional pain clinic with right upper extremity pain. He had a history of bilateral thoracic outlet syndrome and underwent surgical decompression with good resolution on the left, but not the right side. He continued to display nTOS symptoms, but also developed new symptoms of the right anterior chest consistent with CRPS. He opted for conservative therapy without improvement but did not want to undergo another surgery, even though he had been suffering for many years. A temporary spinal cord stimulator (SCS) was placed for a trial period with complete resolution of pain and weakness. A permanent SCS was implanted. The patient continued to have good results leading to improved mood, activity, and overall quality of life.

Conclusion: Interestingly, this patient had overlapping symptoms of refractory nTOS and CRPS. A SCS was able to treat both pathologies. This revelation is important, not only because the SCS can treat both pathologies, but also because it has potential to become a great minimally invasive alternative treatment for refractory nTOS. This would allow patients to avoid surgery and the distress that comes with it.

Key words: Complex regional pain syndrome, spinal cord stimulator, stellate ganglion block, thoracic outlet syndrome

BACKGROUND

Thoracic outlet syndrome (TOS) was first described in 1956 (1). It is caused by the compression of the neurovascular bundle that lies within the thoracic outlet. The thoracic outlet is bounded by the spinal column, first ribs, and sternum. There are overlying muscles and liga-

ments along with the bony borders. Compression occurs most commonly at 3 distinct points. These include the interscalene triangle, the costoclavicular space, and the retropectoralis minor space. The 3 types of TOS include neurogenic (nTOS), venous (vTOS), and arterial (aTOS). In this article, we will describe the presentation and treat-

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ment for nTOS. Ninety-five percent of thoracic outlet cases are nTOS. This occurs when the brachial plexus and adjacent nerves are affected. Symptoms consist of pain, dysesthesia, numbness, and weakness in the affected extremity. Diagnosis can be done with clinical exam, nerve conduction studies, needle electromyography, computed tomography, magnetic resonance imaging (MRI), brachial plexus block, and scalene muscle injection (2). There are 2 types of nTOS: true and disputed. True nTOS has objective findings such as nerve conduction defects, a defined compression point seen on imaging, and muscle atrophy. Disputed nTOS does not have objective findings, but instead has symptoms suggestive of TOS and provocative tests are normally positive. Conservative management includes medication management, physical therapy, and local anesthetic injections. Surgical decompression is reserved for nTOS patients who have neurologic deficits, disabling pain, or failed conservative therapy. The initial success rate is 90%, but the success rate at 10 years decreases to 64% to 71% (3,4).

Patients who continue to have symptoms after surgical treatment or who develop recurrence are ultimately seen to have incomplete first rib resection, compression of the brachial plexus by an ectopic band, persistent brachial plexus compression by an intact first or second rib, compression by pectoralis minor tendon, scar tissue causing compression, or adherent residual scalene muscle (3). Incomplete resolution of symptoms can be treated with reoperation. The success rate with reoperation is 84% and diminishes to 41% at 10 to 15 years (4). Patients with persistent symptoms may also have sympathetic dysfunction resulting in complex regional pain syndrome (CRPS) due to injury of the sympathetic fibers that run along the vessels and bone within the thoracic outlet. This sympathetic dysfunction can result in atypical chest pain, hyperhidrosis, Raynaud's phenomenon, and vascular insufficiency of the upper extremity (1). The exact incidence of CRPS with nTOS is unknown. CRPS is diagnosed in this subset of patients if they meet Budapest criteria, which include pain that is disproportionate to inciting events. The patient must report symptoms in 3 of the 4 listed categories: sensory, vasomotor, sudomotor, and motor. The patient must display one physical exam finding in at least 2 of the 4 categories. Lastly, there should not be another diagnosis that better explains the symptoms (5).

This case is about a patient with refractory nTOS who also developed CRPS in the same upper extremity. Treatment with a spinal cord stimulator (SCS) was able to provide complete resolution of pain.

CASE

A pleasant man in his 40s presented to our clinic with right upper extremity pain. The patient had a history of cervical degenerative disc disease status post cervical fusion in 2009. He was doing well until 2013 when he developed weakness, numbness, and paresthesia in the trapezius, shoulder, and bilateral upper extremities. The symptoms worsened with lateral neck rotation and abduction of arms greater than 90 degrees. His nerve conduction study was normal. He was eventually diagnosed with nTOS by vascular surgery and underwent bilateral first rib resection and scalenectomy in 2013. The surgery provided improvement in the left upper extremity, but there was minimal improvement in the right upper extremity. In addition to the persistent pain, numbness, and weakness of the right upper extremity, he developed right anterior chest pain, paresthesia, hypersensitivity, swelling, and allodynia. He continued to have decreased grip strength and dropped objects regularly. Due to the severe hypersensitivity of the chest and upper extremity, he was unable to keep a shirt on comfortably or shake hands with others. His pain became difficult to control with conservative measures. Given concern for cervical radicular symptoms consistent with MRI findings, he underwent revision of C6-C7 in 2018 by neurosurgery. The patient had improvement of symptoms, but continued to have persistent pain, numbness, and right anterior chest pain. Repeat cervical MRI ruled out any additional structural defects. He continued to be dependent on nonsteroidal anti-inflammatory drugs (NSAIDs), acetaminophen, muscle relaxers, narcotic pain medication, and neuropathic pain medications. Further workup was suggestive of refractory nTOS with findings of scarring and granulation tissue surrounding the right brachial plexus. He completed more than 6 weeks of physical therapy for refractory nTOS without significant relief. Local anesthetic injections for brachial plexus blockade provided only partial relief. He did not wish to undergo any more surgeries and was eventually referred to interventional pain medicine. Due to high suspicion for CRPS, diagnostic stellate ganglion block was performed and provided moderate but short-term relief. Given adequate response to local anesthetic brachial plexus and sympathetic blockade, the patient underwent a SCS trial. The SCS trial was one week with the Medtronic (Minneapolis, MN) device. Multiple programs were trialed including traditional tonic, high-frequency paresthesia-free, and burst stimulation. The optimal program for this patient was traditional tonic stimulation with paresthesia. During the trial period, the patient had a 100% reduction in symptoms. The tip of

the SCS lead was placed at the middle of the C3 vertebral body (Fig. 1) with entry point in the lumbar region. He then proceeded with implantation of the SCS with resolution of symptoms. An implantable pulse generator (IPG) was placed in the left lower gluteal region. He was able to tolerate wearing a shirt without hypersensitivity. He could shake hands and hold objects without discomfort. His activity level and mood significantly increased. He was able to stop taking all oral pain medications.

DISCUSSION

Refractory or recurrent nTOS can be caused by many factors including scarring and adhesions around the brachial plexus, as seen on this patient's MRI (1). Brachial plexus blockade can be used as a diagnostic tool for nTOS. A positive response with brachial plexus blockade is predictive of a good outcome with surgical decompression (3). This patient had partial relief with brachial plexus blockade, making a diagnosis of persistent nTOS likely. In addition, he developed new symptoms in the anterior chest and arms including edema, allodynia, and hyperesthesia after surgical decompression. His symptoms met the Budapest criteria for CRPS. A stellate ganglion block was performed with partial relief because there was a component of sympathetic dysfunction associated with his set of symptoms. Sympathetic fibers travel along the subclavian artery as well as the bony prominence surrounding the brachial plexus, which places patients with TOS at higher risk for CRPS (1). Chronic compression from nTOS as well as trauma from surgeries can cause damage to sympathetic fibers. A history of scalenectomy is also associated with higher rates of postoperative CRPS (6). It is important to note that this patient had a positive diagnostic block for nTOS and CRPS. However, both diagnostic blocks did not provide complete resolution of pain. Instead, only partial relief was obtained and interestingly, only symptoms of nTOS and CRPS were relieved with each respective block. This led us to believe the patient had overlying pathologies including both nTOS and CRPS.

CONCLUSION

SCS has been used successfully in the past for failed back surgery syndrome, CRPS I & II, postherpetic neuralgia, and inoperable critical limb ischemia (7). Only recently has SCS been used for nTOS. Hale et al (8) reported the first case of refractory nTOS that was successfully treated with SCS. Our patient presented with refractory nTOS and CRPS. We were able to successfully treat both pathologies with spinal cord stimulation. We believe patients may be an



Fig. 1. Two spinal cord stimulator leads placed at the level of the C3 vertebral body.

ideal candidate for spinal cord stimulation if they have positive responses to diagnostic peripheral nerve blocks for nTOS and sympathetic blocks for CRPS. Given the paucity of literature on this topic, further studies are required to determine which patients would adequately respond to this therapy.

Letter of Authorship Contribution

The authors listed in this case report all meet criteria for authorship. All authors certify that they have participated sufficiently in the work to take public responsibility for the content.

K.T. and K.S. conceived the presented ideas.

K.T. performed literature review.

K.S. carried out the procedure and followed the medical course on follow-up visits.

K.T. performed extensive chart reviewed.

K.T. and K.S. formulated the paper's format.

K.T. drafted up the case report.

K.S. performed critical revisions of the case report and provided the final approval of the version to be published.

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