CASE REPORT

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Spinal Cord Stimulation for the Management of Upper Extremity Pain Associated with Brachial Plexus Injury in Thoracic Outlet Syndrome: Two Case Reports and Review of The Literature

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ABSTRACT Thoracic outlet syndrome (TOS) occurs when the neurovascular structures in the upper extremity are compressed in certain areas of the body such as the interscalene triangle, costoclavicular, and retropectoralis minor space. Neurogenic TOS (nTOS) is the most common type and can be classified as true or disputed nTOS. Pain management for patients with nTOS involves a comprehensive and multidisciplinary approach that includes lifestyle modifications, physical therapy, non-steroidal anti-inflammatory drugs, opioids, peripheral nerve blocks, sympathetic nerve blocks, neurostimulation, intrathecal drug administration, and surgery. Spinal cord stimulation (SCS) that delivers therapeutic doses of electrical current to the spinal cord for the treatment of neuropathic pain has been suggested as a treatment for this syndrome, but not much has been reported in the literature for the efficacy of treatment in nTOS patients. The purpose of these case reports was to evaluate and present the satisfaction of patients with severe chronic pain secondary to nTOS after cervical SCS.

Keywords: Spinal cord stimulation; thoracic outlet syndrome; neck pain; upper extremity; neuralgia

Neurogenic thoracic outlet syndrome (nTOS) which is the most common type of TOS results from compression of the brachial plexus trunks and represents approximately 90-95% of all cases.[1,2] Neck, supraclavicular, and upper extremity pain, paresthesias, weakness, and arm heaviness are the common symptoms of nTOS which are often reproducible with relevant physical activity.

It has been suggested that spinal cord stimulation (SCS) may be a viable treatment option for patients with nTOS who are experiencing unsatisfactory improvement or recurrence of symptoms. However, the efficacy of SCS in nTOS patients is not well established.

We have presented two cases of patients with chronic upper extremity pain and neurological symptoms due to nTOS who underwent cervical SCS. The results suggest that SCS may be an alternative treatment option for patients who have not responded to conservative treatments.

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CASE 1

A 25-year-old patient with nTOS complained of neuropathic pain, weakness, muscle atrophy, temperature, and color changes in right arm. Visual analogue scale (VAS) was 8-9/10. Electrophysiological stud-

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ies showed decreased right median motor and ulnar sensory nerve amplitude, and chronic axonal degeneration involving the right C8 and T1 spinal nerve roots. The patient had a history of transaxillary first rib resection and scalenotomy, release of the brachial plexus and subclavian artery release. The patient had no pain relief despite all the medication, stellate ganglion blocks and pulsed radiofrequency ablation. As the patient's pain recurred bilaterally over time and required repeated intervention, a percutaneous SCS electrode was fixed in the epidural space at the level of the C7-8 vertebra slightly to the right of the midline, as shown in Figure 1. The patient underwent traditional, paresthesia-inducing, tonic SCS and felt paresthesia on the painful area with 2.6 V, 420 µs, 60 Hz. After experiencing 80% pain relief during a successful trial, permanent SCS implantation was performed a week later. The patient's pain score and quality of life were assessed at 1, 6, and 9 months following the procedure. The patient reported a VAS score of 1-2/10 after implantation, and the McGill Pain Questionnaire (MPQ) score decreased from 71 to 25 during follow-up. The patient also reported an improvement in social life quality.

CASE 2

A 39-year-old male patient with nTOS admitted due to severe and persistent pain, atrophy, dry skin, and color changes in the left shoulder, arm, and hand for 8 months. Electromyography revealed conduction defects in the sensory and motor amplitude of the left median and ulnar nerve. The patient had a history of pectoralis major/minor dissection, subclavian vein release, and no benefit from analgesic treatments. The left stellate ganglion block was performed 3 times and resulted in <50% pain relief. Considering the unsatisfactory results with the stellate ganglion block, it was decided to implant cervical SCS. An electrode was fixed at the C2-C4 vertebral level as shown in Figure 2. During the trial period, the patient was satisfied with the procedure with 2.7 V, 300 µs, 90 Hz. After the procedure, the patient reported a VAS score of 3 and improvement in sleep quality and general quality of life. At follow-up, the MPQ score decreased from 82 to 40. Over 3 years, an increase in pain score was observed. However, there was no evidence of lead migration in the patient's cervical radiographs, and the paresthesia overlapped the

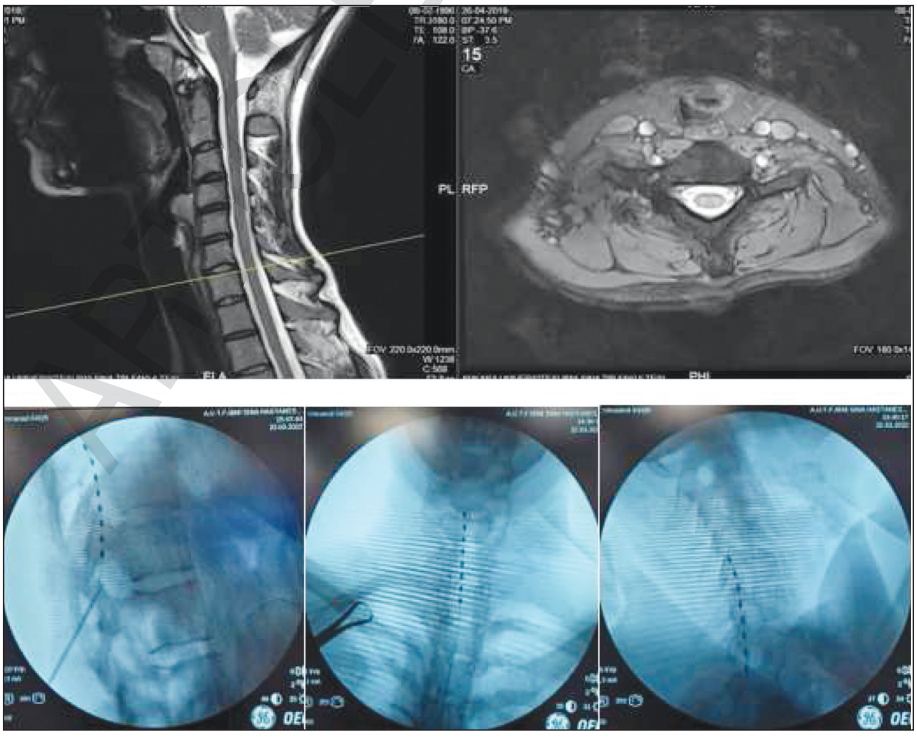

FIGURE 1: Radiographic images of case 1.

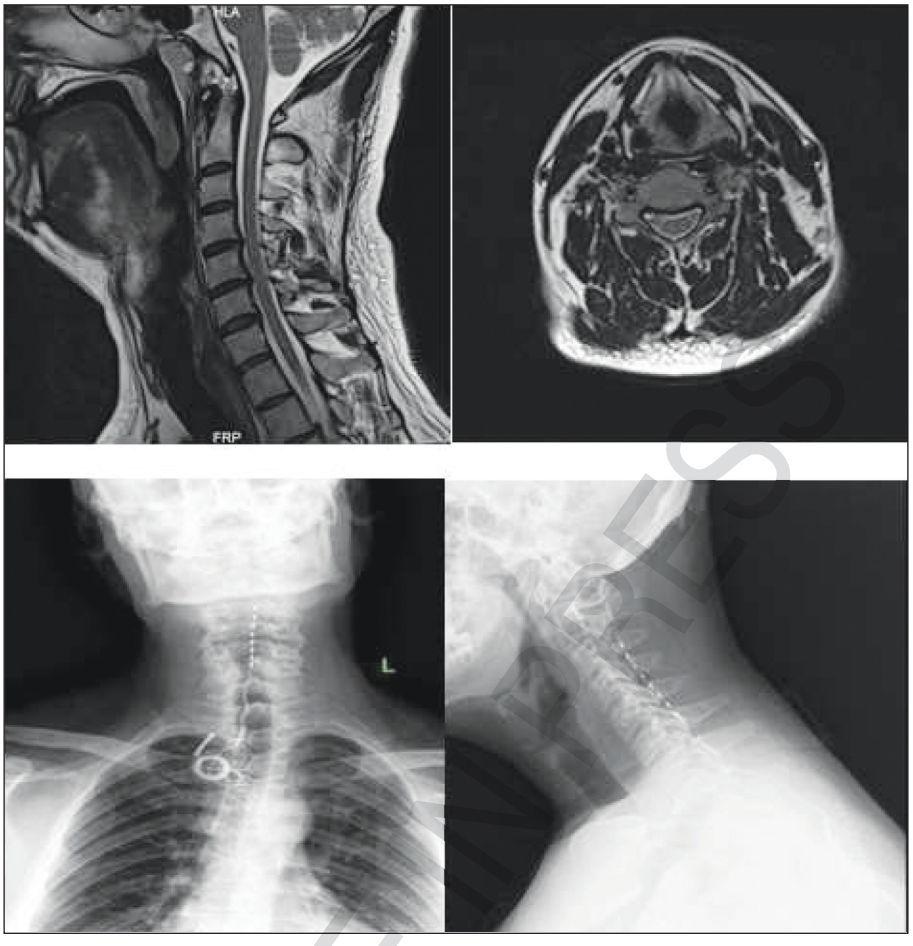

FIGURE 2: Radiographic images of case 2.

patient's painful areas. It is believed that the patient developed tolerance to stimulation over time, and as a result, the SCS generator and lead were removed due to dissatisfaction. After the removal, the patient was discharged with medical treatment and a recommendation for physical therapy.

Informed consent to publication was obtained from the patients.

DISCUSSION

SCS could significantly alter the perception of pain in patients with chronic pain by producing a neuromodulatory effect with low-intensity electrical impulses.[3] It has been stated that SCS can be used in nTOS patients, but there are not many case reports or randomized controlled studies for this syndrome.[1] High frequency-SCS may act on a wider network including both the lateral and medial pain pathways, whereas tonic SCS may principally modulate the lateral pain system.[4] We preferred to use tonic stimulation for pain relief in both patients. After three years with SCS, tolerance to stimulation developed in one of the patients. Perhaps the patient could benefit from burst or high-frequency stimulation, but we did not have a chance to try due to patient-related problems.

There are very few published reports documenting the use of SCS in the treatment of upper limb pain.[5] Due to the narrower epidural space and hypermobility of the cervical spine, lead placement may encounter some resistance, the risk of spinal compression may be higher and paresthesia changes are more likely to occur.[6] In addition, different anatomy of the dorsal columns may cause unwanted paresthesia of the trunk and the lower extremity. A case series

by Vallejo et al. concluded that the use of SCS to treat persistent upper extremity pain after unsuccessful cervical spine fusion surgery resulted in a significant 70-90% reduction in axial neck and upper extremity pain in all patients. SCS has also several advantages such as reducing pain, decreasing the need for repetitive invasive procedures and healthcare consumption, minimizing the use of pain medication, improving patient satisfaction, and enhancing neurological function.[7]

In a recent case study, Hale and Cheng demonstrated the remarkable success of SCS in treating a patient with disputed nTOS and possible pectoralis minor syndrome. The patient was treated with SCS, which resulted in a more than 80% reduction in pain and significant improvement in functional status over 3 years. They stated that this case report offers hope for patients who are suffering from refractory and debilitating symptoms of nTOS. For patients who are unresponsive to surgical treatment, neuromodulation may be the only option that could provide satisfactory relief. Our patients have also reported a significant reduction in pain and improvement in daily activities, similar to the case report. Additionally, the presence of temperature, color, and trophic changes accompanying atrophy in both of our patients suggested that CRPS may have developed in the patients.

The present study has shown that cervical SCS can be a successful method for managing upper extremity chronic pain caused by nTOS. The procedure not only relieved pain but also improved the quality of life in patients. The study found that pain characteristics were similar in both patients and no major adverse events were reported. However, further prospective, randomized, controlled studies are needed to evaluate the efficacy and feasibility of cervical SCS in managing pain in patients with nTOS.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Derya Bayram, Güngör Enver Özgencil;* ***Design:*** *Derya Bayram, Güngör Enver Özgencil, İbrahim Aşık;* ***Control/Supervision:*** *Derya Bayram, İbrahim Aşık;* ***Data Collection and/or Processing:*** *Derya Bayram, İbrahim Aşık, Güngör Enver Özgencil;* ***Analysis and/or Interpretation:*** *Derya Bayram, İbrahim Aşık, Güngör Enver Özgencil;* ***Literature Review:*** *Derya Bayram, Güngör Enver Özgencil;* ***Writing the Article:*** *Derya Bayram, Güngör Enver Özgencil, İbrahim Aşık;* ***Critical Review:*** *Derya Bayram, Güngör Enver Özgencil;* ***References and Fundings:*** *Derya Bayram, Güngör Enver Özgencil, İbrahim Aşık;* ***Materials:*** *Derya Bayram, İbrahim Aşık;*

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