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Case Report

Percutaneous spinal cord stimulator for persistent neuropathic pain following cervical intramedullary ependymoma surgery

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ABSTRACT

This report presented a case of cervical spinal cord stimulation (SCS) application in a patient who experienced progressive arm and neck pain following cervical intramedullary ependymoma resection. A 40-year-old female patient with a history of cervical ependymoma surgery presented to our outpatient clinic with complaints of neck, back, and upper limb pain, as well as tingling and numbness in both hands. Conservative treatments were ineffective, and preimplantation pain was rated 10/10 on the numeric rating scale. Following cervical SCS implantation, pain decreased to 4/10. A successful outcome was achieved through SCS implantation, with electrodes placed percutaneously proximal to the lesion level, for the first time in the cervical region.

Keywords: Central neuropathic pain, chronic pain, intramedullary ependymoma, neck pain, spinal cord stimulator.

Approximately 60% of patients undergoing intramedullary tumor surgery experience moderate to severe central neuropathic pain, irrespective of tumor type.[1] Factors such as syringomyelia, tumor-induced damage to the dorsal horn and corticospinal pathways, central sensitization, and surgical manipulation contribute to the development of central neuropathic pain, which imposes substantial limitations on patients' quality of life.[1-3]

Spinal cord stimulation (SCS) is a neuromodulation method that stimulates neural structures through electrical impulses to inhibit nociceptive transmission and reduce pain.[4] This invasive technique is employed in treating post-laminectomy syndrome, complex regional pain syndrome, phantom pain, postherpetic neuralgia, ischemic pain, angina, and many chronic refractory neuropathic pain conditions.[5] For this purpose, electrodes connected to an internal pulse generator are placed in the epidural space.[4]

Although thoracic and lumbar SCS are commonly used for managing low back and lower extremity pain, there is relatively limited data on the use of cervical SCS for chronic neuropathic pain affecting the neck and upper extremities, with most cases reported following failed neck surgeries.[6-8] Literature on SCS applications for central neuropathic pain caused by intramedullary tumors is also limited.[9-11] Moreover, when reviewing SCS applications for central neuropathic pain caused by tumors with cervical origin, thoracic SCS applications implanted distal to the lesion level are evidently present.[9,10] The current report presented a patient who experienced exacerbated arm and neck pain following cervical intramedullary ependymoma surgery and underwent SCS implantation with the leads placed, for the first time, proximal to the lesion level in the cervical region.

CASE REPORT

A 40-year-old female presented with bilateral arm numbness, tingling, and pain. Six months prior, the patient had presented to an external center with a five-month history of burning, stinging, and pain in

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both hands and arms. Cervical magnetic resonance imaging (MRI) revealed an intramedullary mass at the T1 level (Figures 1a, b), leading to C7-T1 laminoplasty, duraplasty, and tumor excision, with pathology confirming a Grade 2 ependymoma. One year postoperatively, the patient was referred to our clinic due to worsening symptoms. The patient reported pain that was predominantly localized to the medial aspects of both upper limbs, radiating to the fourth and fifth digits. She described persistent burning, stabbing, and electric shock-like sensations in the aforementioned regions. On physical examination, allodynia was detected in the same areas. These findings were consistent with neuropathic pain within the C8-T1 dermatomal distribution. However, detailed neurological examination revealed hypoesthesia across the entire C4-T1 dermatomes bilaterally. The patient also reported diffuse paresthesia involving the entire circumference of both arms. Motor examination revealed no deficits, the Spurling test was bilaterally negative, and deep tendon reflexes were normoactive. Cervical MRI demonstrated no residual mass or root compression (Figures 2a, b).

The patient had a documented history of a somatosensory nervous system lesion (intramedullary tumor), which represents a

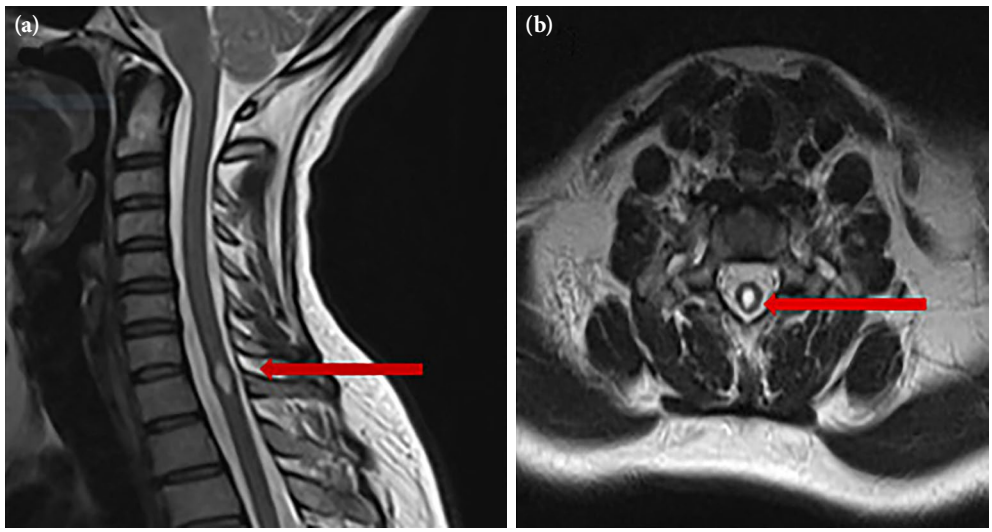

Figure 1. **(a)** Sagittal and **(b)** axial sections of the intramedullary ependymoma identified on preoperative cervical magnetic resonance imaging indicated by arrows.

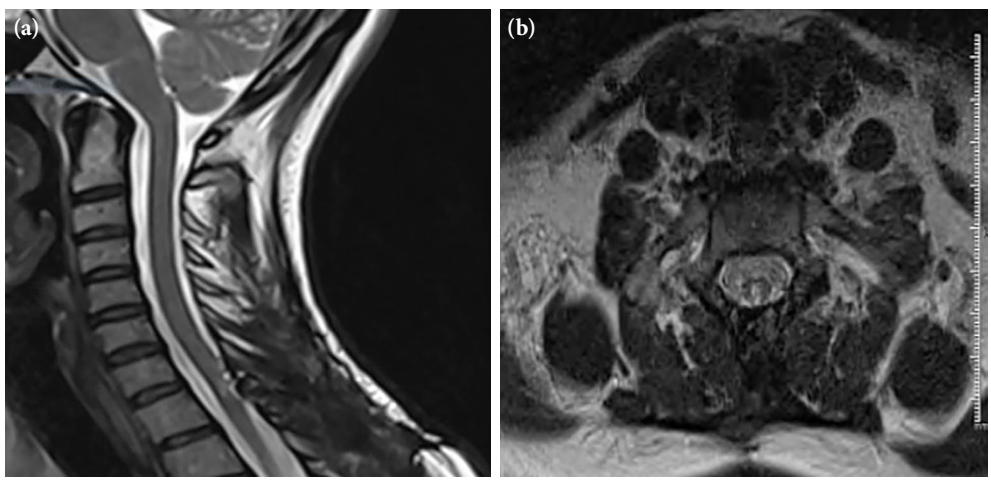

Figure 2. **(a)** Sagittal and **(b)** axial sections of postoperative cervical MRI of the patient following ependymoma resection.

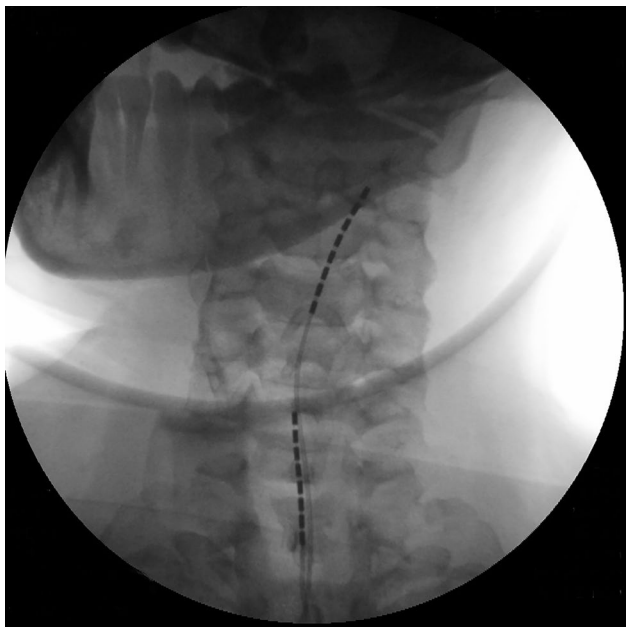

Figure 3. Anteroposterior imaging showing the final position of the percutaneous leads of the cervical spinal cord stimulation.

necessary condition under the current definition for establishing a diagnosis of neuropathic pain.[3] In addition to hypoesthesia, allodynia was identified on physical examination, and the reported pain was anatomically consistent with the neuroanatomical distribution of the sensory findings. Furthermore, the patient's DN4 (Douleurs Neuropathiques en 4 Questions) score was recorded as 6 prior to SCS. In light of these findings, a diagnosis of postoperative neuropathic pain was established.[12]

Pharmacologic management included pregabalin (150 mg twice daily) and tramadol (100 mg three times daily), with pain rated 10/10 on the numeric rating scale. Although attempts were made to increase the dosages, the patient developed lower extremity edema and somnolence, which necessitated a return to the previously tolerated doses. Accordingly, the medications were administered at the highest dosages the patient could tolerate. Pregabalin, which had been initiated at an external center prior to tumor surgery, was continued for approximately one year until the time of SCS implantation. Tramadol hydrochloride was used for an estimated duration of eight months. A T2-T3 interlaminar epidural injection was also ineffective, leading to the recommendation of cervical SCS implantation.

The patient underwent bilateral SCS electrode implantation in the cervical epidural space for a trial period. Under monitored sedoanalgesia, and fluoroscopic guidance, access to the epidural space was obtained via the T1-T2 interlaminar space. The first electrode was placed at the lower C2 vertebral body, and the second at the left paramedian side of C6 (Figure 3). Patient feedback confirmed that the paresthesia produced by electrical stimulation corresponded to the painful regions without upper extremity muscle contractions. The electrodes were anchored and sutured to the fascia. The extensions connected to the electrodes were attached to a temporary battery, and the surgical procedure was completed.

The SCS System (Precision; Boston Scientific Corporation, Valencia, CA, USA) was programmed to FAST (Fast-Acting Sub-perception Therapy) mode. The patient's pain intensity, assessed as 10/10 according to the numeric rating scale prior to the SCS application, decreased to 4/10 by the end of the first week. Subsequently, the patient underwent reoperation for permanent implantation at the end of the first week. The extension cables were removed, and the electrodes were connected to the internal pulse generator, implanted subcutaneously on the left iliac crest. Pain intensity remained at 4/10 during both the first- and third-month follow-ups, increasing to 5/10 at the six-month follow-up. The DN4 score, initially 6 prior to SCS implantation, was 0 at the first month, 2 at the third month, and 3 at the sixth month. The patient's oral medication regimen remained unchanged throughout the follow-up period. A written informed consent for publication was obtained from the patient.

DISCUSSION

While SCS is considered to be effective for managing peripheral neuropathic pain, its efficacy in treating central neuropathic pain remains a subject of debate.[13] Some cases have reported beneficial outcomes from SCS in patients with spinal cord injuries resulting in central neuropathic pain;[14] however, the literature on SCS applications for central neuropathic pain induced by intramedullary tumors is limited.[9-11] For instance, Noordhof et al.[15] described a patient experiencing persistent back pain and numbness in both legs following T7-level meningioma surgery, for whom analgesia was achieved through leads placed at the T5 level. Lee et al.[11] implanted leads at the T1 and T2 levels in a patient suffering from persistent pain after the excision of a meningioma at the T5 level, resulting

in a positive response to SCS treatment. Benedetti[9] implanted SCS leads at the T8 level to treat lower extremity pain associated with myelomalacia that developed after C6 ependymoma surgery, resulting in a reduction of the patient's lower extremity pain. Additionally, Eisenberg and Brecker[10] documented a case involving persistent pain in the right upper and lower extremities despite improvements in motor deficits following meningioma surgery at the foramen magnum. In this instance, a unilateral lead was placed at the T12 level to address severe pain in the right lower extremity, resulting in nearly complete pain relief.

The narrower epidural space in the cervical region, in comparison to the thoracic and lumbar regions, coupled with the development of epidural fibrosis following surgery, complicates lead placement in the cervical area.[15] In the present case, C7-T1 laminoplasty, duraplasty, and mass excision were performed upon detection of an intramedullary ependymoma at the T1 level. It was anticipated that epidural fibrosis and spinal adhesions, which may have developed as a consequence of the surgical removal of the tumor, could complicate the direction of the lead over the surgical level via a percutaneous approach (Figure 2). The neurosurgeons were consulted regarding the possibility of placing a surgical paddle lead for SCS. However, when this was deemed too risky by the surgeons, a comprehensive review of the literature was conducted.

In the case reported by Benedetti,[9] although there was pain after excision of the ependymoma detected at the C6 level, the pain felt in the lower extremities was attributed to myelomalacia, and leads were implanted at the T8 level, which is significantly lower than the tumor surgery site. Therefore, fibrous tissue did not present an obstacle to lead placement in that case. Similarly, in the case described by Eisenberg and Brecker,[10] the lead was placed in a different region from the site of tumor surgery. The only documented case in the literature involving the placement of a percutaneous lead proximal to the surgically excised tumor was reported by Lee et al.[11] and was performed at the thoracic level.

Our review of the literature reinforced our belief that, in this case, paddle lead placement via a surgical approach may be more technically feasible. In such instances, we recognize that the risks of hematoma, dural tears, and neurological injury are increased with percutaneous implantation. However, the refusal of neurosurgeons to perform the procedure, combined with the patient's ongoing severe pain despite conservative treatment, ultimately led us to pursue percutaneous SCS implantation. The first electrode was cautiously advanced to a right paramedian position at the lower endplate of the C2 vertebra. Although the second electrode was intended for placement on the left, resistance was encountered. Attempts to advance the electrode from the right to the left epidural space were unsuccessful due to hardened tissues. Ultimately, the upper end of the left electrode was positioned at the C6 vertebral level. However, paresthesia from both electrodes covered all painful areas.

Central neuropathic pain associated with intramedullary tumors is likely similar to the mechanisms underlying spinal cord injury. Similar to spinal cord injury, it is reasonable to place electrodes in intact regions above the level of the lesion when intramedullary tumors are present. This approach is thought to modulate the nociceptive transmission from the pain generator at the lesion level to the brain. Therefore, in the current case, we performed cervical SCS implantation proximal to the level of the lesion. Additionally, several mechanisms of action for SCS have been identified beyond the original gate-control theory. These mechanisms include the suppression of wide dynamic range neurons through antidromic activation of the dorsal column, inhibition of excitatory neurotransmitter release, and stimulation of GABA (gamma-aminobutyric acid) release.

In conclusion, this case is significant as it marks the first instance of SCS implantation, in which electrodes were placed in the cervical region to manage persistent neuropathic pain following intramedullary tumor surgery. Furthermore, the current case is valuable as it represents the second instance of SCS, where leads were placed proximally to the lesion level using a percutaneous approach, following a previous procedure at the thoracic level. It is essential to consider that cervical SCS implantation may serve as an effective treatment option for chronic neuropathic pain resulting from intramedullary tumors or the surgical interventions associated with them.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

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