

Case Report

High-Level Cervical Spinal Cord Stimulation Used to Treat Intractable Pain Arising from Transverse Myelitis Caused by Schistosomiasis

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The efficacy of spinal cord stimulation (SCS) for treatment of various chronic painful conditions is well established. Very few reports have documented the use of SCS for treatment of chronic pain after spinal cord injury. We present a case showing a good outcome after such treatment, and suggest that high cervical stimulation may be efficacious. A 53-year-old male underwent SCS on the C1-3 level for treatment of intractable neuropathic pain below the T3 level, and in the upper extremities, arising from spinal cord injury resulting from transverse myelitis caused by schistosomiasis. High cervical SCS significantly improved the pain in the upper extremities and at the T3-T10 dermatome level. The patient continues to report excellent pain relief 9 months later. The present case suggests that high cervical stimulation may improve chronic pain in the upper extremities and the T3-T10 dermatome level arising from spinal cord injury.

KEY WORDS : Spinal cord stimulation · Schistosomiasis · Spinal cord injury · Transverse myelitis.

INTRODUCTION

Spinal cord stimulation (SCS) has been regarded as an effective treatment modality for chronic pain patients. The first use of an electrode was reported in 1967 by Shealy⁴. Since then, many physicians have extended the indications for SCS, which now include various pain syndromes, failed back surgery syndrome (FBSS), phantom limb pain, complex regional pain syndrome (CRPS), brachial plexus injury, and angina pectoris. The target level for placement of the SCS electrode differs according to pain location. For example, the lower thoracic level has been commonly used in patients with FBSS^{4,6,14}, stimulation at the thoracolumbar junction has been useful in treating foot pain patients⁶, and the cervico-thoracic junction was appropriate to control angina pectoris pain^{4,6,14}. In addition, the cervical level has been employed for treatment of facial,

neck, and upper extremity lesions arising from cord injury, brachial plexus damage, and CRPS²¹.

Central nervous system schistosomiasis has been described in soldiers and workers in endemic areas. Myelopathy arising from schistosomiasis has been reported in several studies and may cause intractable neuropathic pain^{1,17}. Such pain is inadequately controlled by medication such as NSAIDs, opioids, or anticonvulsants.

We experienced a good outcome after application of cervical cord stimulation to a patient with intractable pain arising from spinal cord injury resulting from transverse myelitis caused by schistosomiasis. Here, we report the outcome and technical details of the treatment of this patient with a pertinent literature.

CASE REPORT

A 53-year-old male patient presented to our hospital with intractable neuropathic pain below the T3 level as a result of spinal cord injury suffered after transverse myelitis caused by schistosomiasis. He had worked in Middle Eastern countries for several years and had developed hypesthesia on legs during that time. After he returned to our country, he experienced motor weakness, voiding

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difficulty, dizziness, and hypesthesia. He underwent resection of schistosomiasis-related granulomas on the cerebella, and the T3-4 level of the spinal cord, at another hospital. Subsequently, he had suffered from neuropathic pain below the T3 level, and motor weakness of grade IV in the lower extremities, for 15 years. His magnetic resonance imaging (MRI) showed a diffuse atrophic change ranging from the lower cervical to lumbar spine (Fig. 1). Pain control could not be achieved medically; all of NSAIDs, opioids, and anticonvulsants had been unsuccessful. A nerve block was performed several times with no significant result. The pain was described as pricking, throbbing, shooting, pressing, and either intermittent or continuous. The pain level was 9-10 on the VAS score. On the Korean McGill Pain Questionnaire, the pain rating index (PRI) was 57/75 and the present pain intensity score (PPI) was 5/5. Functional disability was 62/70 on the Korean Brief Pain Inventory. Medications included 1,800 mg neurontin and 10 mg amitriptyline daily.

The patient was placed in the prone position under general anesthesia. A midline incision was made from theinion to C2 or C3 and exposure was performed from previous incision line. A partial laminectomy of the dorsal ring of C1 and the inner portion of the C2 lamina, without destruc-

tion of the outer shape, was performed. The atlanto-occipital ligamentum flavum was minimally removed. After creating enough space for insertion, a lead (Lamitrode 44; Advanced Neuromodulation Systems, Plano, TX, USA) was placed at the midline of the dura at C1 and C3 (Fig. 2). The lead was fixed to the dura with a 4-0 silk suture applied to two lateral points of the lead. The lead was tunneled to a subcutaneous pocket located lateral to the occipital area. The electrode was connected to a screening cable. After a screening stimulation, the wound was closed. The pain score was reduced by more than 50% during the stimulation trial period. Over the next several days of stimulation the patient felt no change in pain level. However, 4 days after stimulation commenced, pain in the upper extremities was reduced. Paresthetic overlap of the pain segments was achieved. Such segments grew daily to attain the T10 dermatome. Stimulation frequencies for SCS ranged between 2 and 6 Hz because the patient complained of intolerable numbness on whole body when stimulation frequency was high. Amplitudes varied from 0.3 to 0.5 mA. Pulse width could be adjusted from 280 to 410 msec. Maximum effect of pain reduction appeared stimulation on left side electrode at C2-3 level. The patient felt pain reduction with comfortable warm sensation on thoracic area. We therefore implanted an implantable pulse generator. Nine months later, the PRI on the Korean McGill Pain Questionnaire was 21/75 and the PPI 3/5. Functional disability was 43/70 according to the Korean Brief Pain Inventory. The patient's medication was not changed because the patient felt that the medication was much more effective than in the preoperative period, and he felt comfortable continuing with the drugs.

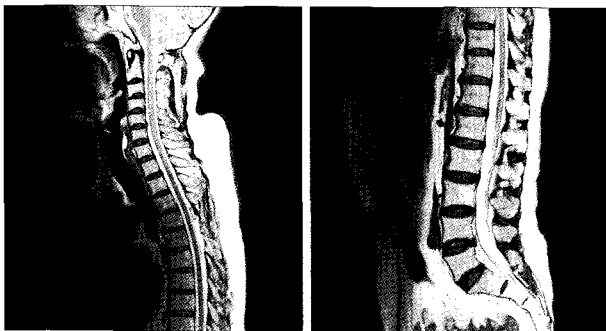


Fig. 1. Sagittal T2-weighted magnetic resonance imaging scan shows a diffuse atrophic change with central T2 high signal intensity ranging from the lower cervical to lumbar spine and epidural calcification and decompression at T4 level.

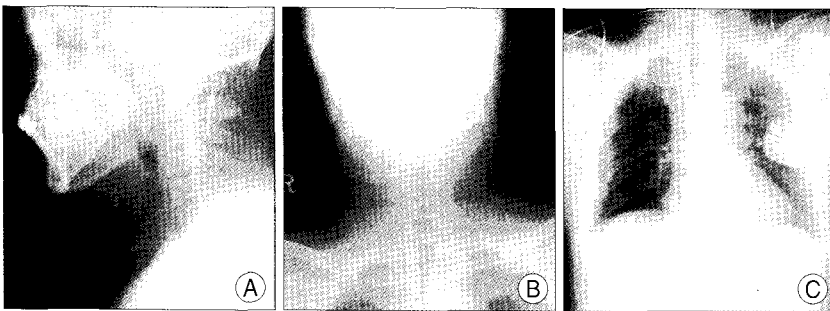


Fig. 2. A and B : Anterior-posterior and lateral view of radiograph showing a electrode (Lamitrode 44; Advanced Neuromodulation Systems, Plano, TX, USA) at C1-3 after partial laminectomy of C1. C : Anterior-posterior view of radiograph showing implantable pulse generator on left subclavicular area.

DISCUSSION

Cervical SCS has been used to treat several distinct conditions, including the neuropathic pain of craniocervical pain syndrome, pain in either lower or upper extremities, chronic neck pain, angina pectoris, phantom pain, and trigeminal neuralgia^{2,4,11,14,23}. Recently, several studies have reported that high cervical SCS improved pain caused by upper extremity ischemia, brachial plexus injury, so-called failed neck surgery, and CRPS^{13,21}. Although the precise mechanism of this effect remains unclear, high-level stimulation might relieve pain by modulation of the dorsal column, the spinothalamic tracts, and the descending inhibi-

tory pain pathway¹³), an increase in peripheral blood flow may also affect the alleviation of pain²⁰.

Cervical spinal cord stimulation has been less studied than thoracic spinal cord stimulation. Cervical epidural stimulation has been explored in an effort to elicit pain reduction in the four extremities and thorax⁹. Some studies have reported that four-extremity pain was reduced when electrodes were placed at the cervical level^{9,11,19,21,23}. Various authors have differed in their designation of the exact target level that yields maximum reduction in four-extremity pain. Vallejo et al.²¹ achieved good results using cervical cord stimulation with electrodes located on the C2-4 epidural space in three patients who had undergone failed neck surgery and who suffered from pain in the upper extremities, neck, back, legs, and feet. Some researchers found that four-extremity pain was reduced only when leads were placed at the high C2 region, over the midline¹¹. Similarly, Hayek et al.⁹ reported that 11 of 12 patients experienced paresthesiae in all four extremities during the test trial period and a significant 38% decrease in pain after SC implantation on C3-C7. The cited authors argued that the cervical electrode could cover the dorsal column fiber tracts and reduce pain in all four limbs, although the treatment target area was slightly different to that of our patient. Simpson et al.¹⁹ studied 41 patients with neuropathic or ischemic pain treated with cervical epidural electrodes and obtained 51% pain reduction in 68% of patients. Whitworth and Feler²³ suggested that a C1-2 sublaminar insertion of paddle leads in a patient with upper extremity pain could offer pain relief; pain control in the lower extremities was not achieved.

There are many reports regarding the efficacy and use of cervical cord stimulation for pain^{9,11,19,21,23}. However, the exact correlation between the spinal levels of the lead position and the stimulating area have not yet been determined, although there have been many variations in that correlation. Barolat et al.³ pointed out the mapping data of the sensory response to epidural stimulation in 106 patients. Although, in the results the authors did not suggest in detail the stimulating area according to each spinal level, the result graphs reveal a stimulation coverage percentage of less than 20% in the four extremities, the abdomen, lower back and buttock by cervical spinal cord stimulation³. Also theoretically, pain reduction in the four limbs and in the body could be explained by the anatomical arrangement of sensory fibers within the dorsal columns²¹. Barolat et al.³ reported that stimulation of the chest wall can achieve pain reduction primarily through laterally placed lead at the thoracic level. However, according to these graphs, there was a possibility of obtaining stimulation of the chest wall.

In our case, we achieved pain relief in the thoracic area. We initially planned to achieve pain control in the four extremities and below the T3 level via cervical spinal stimulation of C1-3 level, however, our patient obtained pain relief on the upper extremities and at the bodily T3-T10 level. As our patient complained of numbness without pain relief in the four extremities and in the body using high frequency stimulation, we used low frequency stimulation. This finding was also reported in another study⁷. These authors achieved this type of coverage using high frequency stimulation with the lead position at the C2 level.

Hayek et al.⁹ suggested the reasonable hypothesis of paresthesiae coverage of the four extremities : first, the lead can be placed closer to the spinal cord due to the relatively lesser thickness of dorsal cerebrospinal fluid at the cervical level; second, fasciculus gracilis and cuneatus fibers have a favorable orientation for epidural electrical stimulation; third, large fibers assume a superficial position which is suitable to neurostimulation according to the rearrangement of nerve fibers with continuous changes in the dorsal column fiber positioning.

Recently, some clinicians prefer to use of percutaneous lead for spinal cord stimulation. In our patient, we performed the surgical electrode insertion via laminectomy rather than by percutaneous electrode insertion. As the patient had already undergone suboccipital craniectomy, we were concerned about possible adhesion at the C1 level and in the extended area. In addition, percutaneous procedures at the cervical level were not familiar for neurosurgeons. As electrode placed via laminectomy can have significant long-term effectiveness for epidural stimulation²². Moreover, complications such as lead migration and positional stimulation occurred more frequently with percutaneous lead insertion than with surgical type lead insertion²².

Few reports on the effect of spinal cord stimulation for treatment of chronic pain after spinal cord injury have appeared. Our patient achieved a good outcome after high cervical SCS treatment of chronic neuropathic pain caused by spinal cord injury arising from transverse myelitis subsequent to schistosomiasis.

Over the past 40 years, the prevalence of pain after spinal cord injury has ranged from 18-90% in various reports^{15,16}. Several theories have been put forward to explain the mechanisms of neuropathic pain. Siddall¹⁸ reviewed the literature and suggested that the mechanisms of neuropathic pain may arise from 'downstream' changes in damaged nerve roots as well as 'upstream' changes in the brain. Hulsebosch et al.¹⁰, in another review, suggested that the mechanisms of remote microglial activation and pain signaling in 'below-level' central pain are dorsal horn neuron

hyperexcitability, central sensitization, microglial activation, and locally upregulated chemokine synthesis. Complete injury was significantly more likely to result in chronic pain than incomplete injury, and was associated with increased pain severity¹⁵).

The management of neuropathic pain following SCI includes surgical approaches, pharmacological options, neurostimulation, and psychological and environmental management. Surgical techniques such as dorsal root entry zone, cordotomy, and cordomyelotomy can reduce pain but these approaches are now rarely used because they are associated with serious complications. Adequate pain control is often difficult to achieve pharmacologically. Neurostimulation treatments include SCS, transcutaneous electrical nerve stimulation (TENS), and motor cortex stimulation. Siddall¹⁸ reported on the efficacy of SCS and TENS used to treat SCI pain. Ravenscroft et al.¹⁶ reported one case where dorsal column stimulation relieved SCI pain but other reports had described less favorable outcomes⁵). Cioni et al.⁵ reported good short-term relief of paraplegic pain. In our patient, excellent pain reduction continued for at least 9 months. To control chronic pain in an SCI patient, a multidisciplinary approach is necessary.

The efficacy of SCS in the treatment of transverse myelitis has been described in several reports. Hamid et al.⁸ reported pain reduction after SCS in a patient with radiation-induced transverse myelitis. Laffey et al.¹² observed a good outcome after SCS treatment of neuropathic pain caused by idiopathic acute transverse myelitis.

CONCLUSION

In conclusion, we suggest that high-level cervical stimulation can provide effective pain relief in patients with persistent neuropathic pain caused by spinal cord injury resulting from schistosomiasis. More studies on the long-term efficacy of such treatment are warranted.

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