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A Case of Painful Legs and Moving Toes Syndrome Successfully Treated with Spinal Cord Stimulation Therapy

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Background: Painful legs and moving toes syndrome (PLMT) lacks clear diagnostic criteria. Some reports have described possible causes of PLMT. However, in most cases, the etiology is not clear and the therapeutic response is typically poor.

Case Report: A 49-year-old woman underwent osteosynthesis surgery for a bimalleolar fracture of the left ankle joint. After the surgery, she experienced persistent pain in the left ankle joint along with the appearance of involuntary movements of the left lower extremity. She was diagnosed as a case of PLMT. Since the pain and involuntary movements were refractory to medication, she was administered a trial of spinal cord stimulation (SCS). She experienced immediate complete resolution of symptoms after the trial stimulation. Subsequently, we implanted a permanent SCS device. As of the 8-month follow-up, there has been no flare-up of symptoms and she is able to walk without support.

Conclusion: We describe a patient in which SCS was effective in treating PLMT syndrome.

Key words: Involuntary movements, painful legs and moving toes syndrome, spinal cord stimulation therapy

BACKGROUND

Painful legs and moving toes syndrome (PLMT) is characterized by unilateral or bilateral lower extremity pain and complex involuntary movements of the toes and ankle joints. There is a lack of clear diagnostic criteria for PLMT, and the diagnosis is based on clinical manifestations after excluding other causes of involuntary movements. Some reports have described peripheral neuropathy, trauma, and radiculopathy as causes of PLMT (1).

However, in most cases, the etiology is not clear. Symptomatic treatment using drugs and nerve blocks are administered; however, the therapeutic response is typically poor. In this report, we describe our experience with a case of PLMT in which the symptoms did not improve with medication. However, a trial of spinal cord stimulation (SCS) resulted in complete symptom relief. Therefore, we implanted a permanent SCS device. The patient provided written consent for publication of this case report, and there are no conflicts of interest to be disclosed.

CASE

A 49-year-old woman with no remarkable medical history underwent osteosynthesis surgery for a bimalleolar fracture of the left ankle joint in 2020. After the surgery, she had persistent pain in the left ankle joint and involuntary movements in the left lower extremity. In May 2021, she was referred to the Department of Anesthesiology and the Pain Clinic at our hospital because of intractable pain and difficulty in walking due to involuntary move-

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ments even after plate removal surgery.

On physical examination, there was no edema or swelling of the affected limb and no abnormalities in the range of motion of the joints. There were no signs of muscle weakness or sensory disturbance, and her tendon reflexes were normal. The pain was persistent and was described as burning and tingling, and sudden electric shock-like pain. In addition, there were intermittent repetitive involuntary movements in the affected limb once or twice per second, and the ankle joint and toes showed complex movements such as forward and backward flexion, internal and external rotation, and wiggling. The involuntary movements were aggravated when the pain was more severe. Thermal imaging showed no temperature difference between the left and right lower extremities, and nerve conduction tests showed no abnormalities in the tibial and peroneal nerves. Magnetic resonance imaging (MRI) of the brain and lumber spine showed no obvious abnormalities. Based on the above findings, after excluding complex regional pain syndrome and other diseases presenting with involuntary movements, a clinical diagnosis of PLMT was made. She was prescribed analgesics such as oral opioids (tramadol 200 mg/day), Ca2+ channel $\alpha 2\delta$ ligand (mirogabalin 30 mg/day), and serotonin and noradrenaline reuptake inhibitor (duloxetine 20 mg/day); however, there was no improvement in pain. For involuntary movements, anti-Parkinson's disease drugs (allotinol 5 mg/day, trihexyphenidyl 4 mg/day) and an antiepileptic agent (clonazepam 1 mg/day) were prescribed by the neurologist at our hospital, but their effect was poor. We performed a sciatic nerve block in the popliteal fossa, which led to temporary relief in pain and involuntary movements. Since she was refractory to medication, a trial of SCS was performed in June 2021. Two leads with 8 electrodes were used. These were placed in the midline of the dorsal epidural space at the level of the 10th to 12th thoracic vertebrae. Tonic stimulation at a frequency of 50 Hz and a pulse width of 200 μs was administered with intensity adequate to produce paresthesia, and in a manner that the stimulation sensation overlapped the painful area of the left lower extremity.

RESULTS

Immediately after the test stimulation, there was complete resolution of pain and involuntary movements. After 2 weeks of test stimulation, the leads were removed, and the pain and involuntary movements flared up again 2 days later. In August 2021, we decided to implant a permanent SCS device. The 2 leads with 8 electrodes were placed in the same position as the test stimulation (Figs. 1 and 2), and the permanent device was implanted in the left hip.

Changes in pain and clinical symptom scores assessed as pain-related parameters before and after SCS implantation are shown in Table 1. The numeric rating scale score showed a dramatic improvement from 9 to 0. In addition, various pain-related parameters and life-disability rating scales also showed improvement; these included the pain disability assessment scale, which assesses the degree of disability due to chronic pain, the Pain Self-Efficacy Questionnaire, Hospital Anxiety and Depression Scale, Pain Catastrophizing Scale, EuroQol-5 dimension questionnaire, Locomo 25 for the assessment of activities of daily living, and Athens Insomnia Scale. After SCS implantation, she was able to walk independently without assistive devices. As of the 8-month follow-up after SCS implantation, there has been no recurrence of pain or involuntary movements.

DISCUSSION

PLMT is a rare syndrome characterized by unilateral or bilateral lower extremity pain and involuntary movements. The pain is typically severe and uncomfortable, and is associated with a variety of symptoms, including numbness and tightness. The speed and frequency of the involuntary movements suggest that it is a type of hyperkinetic movement disorder, but it does not fit into any of the involuntary movement types. Our patient was previously diagnosed as having tremor, myoclonus, athetosis, and dystonia, but none of these conditions were applicable. She presented with complex movements that seemed to be a mixture of tremor and dystonia. The correlation of involuntary movements with pain intensity (2) and reports of similarities and differences between PLMT and other diseases that present with involuntary movements suggest a relationship between pain and involuntary movements (3). In our patient, pain and involuntary movements occurred in parallel.

The etiopathogenesis of PLMT is not clear. Some reports have described upper limb causalgia, disc herniation, lumbar nerve injury, herpes zoster, and peripheral nerve disease as potential causes (4). Therefore, several mechanisms may be involved in its causation, such as sympathetic nervous system dysfunction and blood flow disturbances. In addition, damage to peripheral nerves induces plastic changes in the central nervous system,

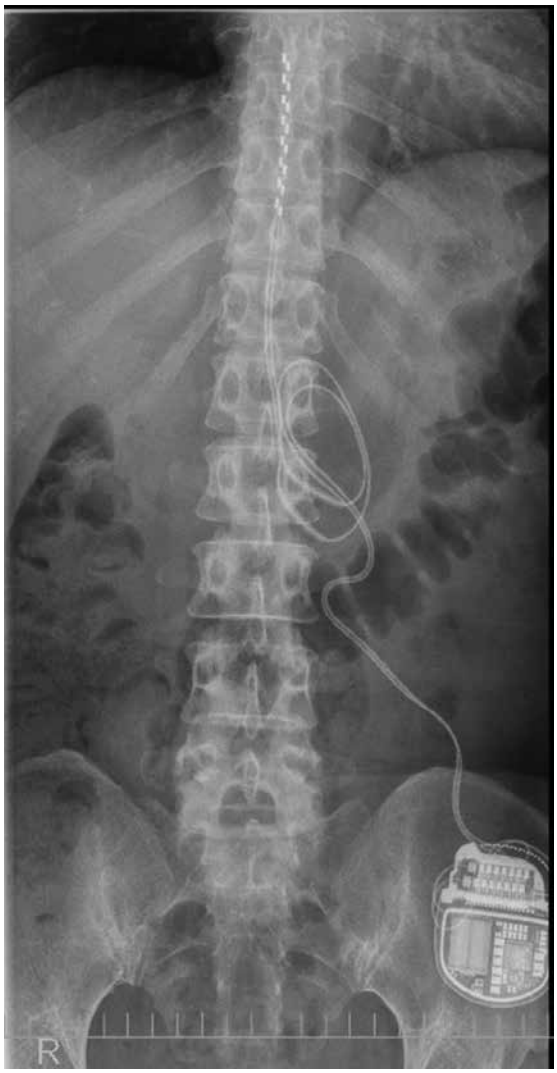

Fig. 1. Lumbar-thoracic spine x-ray after SCS implantation (frontal view). Two leads with 8-electrodes are placed in the midline of the dorsal epidural space at the level of the 10th to 12th thoracic vertebrae. The generator is implanted in the left hip.

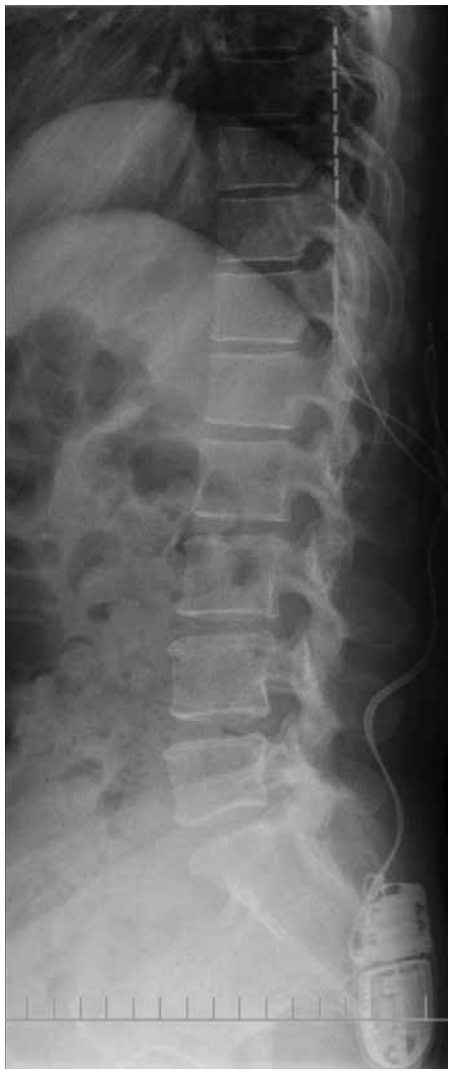

Fig. 2. Lumbar-thoracic spine x-ray after SCS implantation (lateral view). The right side shows the dorsal side and the left side shows the ventral side. The location of the lead in the dorsal epidural space can be seen.

leading to abnormal central nervous system function, which causes pain and involuntary movements (5). It has been speculated that impulses from peripheral nerve lesions produce involuntary movements by excitation of anterior horn cells via spinal interneurons (2). However, these mechanisms are only speculative and not well-characterized. A report has also described an association of PLMT with immunological diseases (6). Therefore, multiple factors and mechanisms may be involved in the causation of this syndrome. In the present case, the PMLT was triggered by trauma and surgery. Therefore, complex regional pain syndrome (CRPS) was one of the differential diagnoses. However, there was no edema, swelling, or limitation of joint range of motion, and there was no temperature drop in the affected limb on thermography, which excluded CRPS. In 2012, Hassan et al (1) reported a series of PLMT cases (n = 76) in which drug therapy (antiepileptic drugs, selective serotonin

Table 1. Changes in clinical endpoints before and after SCS implantation.

	Pre-SCS	Post-SCS
NRS	9	0
PDAS	38	17
PSEQ	12	53
HADS A/D	24 (15/9)	10 (4/6)
PCS	52	38
EQ-5D	0.418	0.649
Locomo 25	58	21
AIS	16	2

Abbreviations: AIS, Athens Insomnia Scale; EQ-5D, Euro QOL-5 dimension; HADS A/D, Hospital Anxiety and Depression Scale; NRS, Numeric Rating Scale; PCS, Pain Catastrophizing Scale; PDAS, Pain Disability Assessment Scale; PSEQ, Pain Self-Efficacy Questionnaire; SCS, spinal cord stimulation

reuptake inhibitors (SSRI), and opioids) and nerve blocks (epidural block and lumbar sympathetic ganglion disruption) were ineffective (1).

SCS is a pain-treatment method used for neuropathic pain such as postoperative spinal pain syndrome and peripheral vascular disorders, but there are few reports of its use for PLMT. A previous case report described SCS implantation in a 59-year-old woman with bilateral lower extremity PLMT caused by a lumbar disc herniation (7). A 4-electrode lead was implanted at the 10th to 11th thoracic vertebrae level and tonic stimulation at a frequency of 100 Hz and a pulse width of 192 μs was administered. There was improvement in involuntary movements and pain over a 13-month period. Another report described successful use of SCS implantation in a 51-year-old man with bilateral lower limb PLMT who developed depression during treatment (8). A 4-electrode lead was implanted at the level of the 10th to 11th thoracic vertebrae and tonic stimulation at a frequency of 50 Hz and a pulse width of 300 μs was administered. However, SCS was ineffective in a patient with PLMT associated with herpes zoster myelitis (9).

Although the mechanism by which SCS suppresses pain is still unclear, it has been reported that SCS suppresses ascending pain transmission at the level of the dorsal horn of the spinal cord via interneurons from dorsal cord stimulation (10) and centrally activates the descending pain inhibitory system (11). In this case, since sciatic nerve block in the popliteal fossa prior to the trial of SCS led to the disappearance of pain, neuropathy of the peripheral nerve was considered to be the main cause of pain. Therefore, we speculate that the suppression of the ascending transmission of pain by the tonic stimulation of SCS led to pain resolution. According to a previous report, SCS affects the spinal cord anterior horn cells via the spinal reflex arch (12). Therefore, we hypothesized that the relief of involuntary movements was due to suppression of the excitation of anterior horn cells by SCS via interneurons from the spinal cord dorsal horn by pain stimulation. Although SCS may not be indicated for all cases of PLMT due to the variety of background diseases, we believe that it should be considered in cases where nerve block at the level of peripheral and spinal nerves is effective. Further accumulation of experience of PLMT treatment will provide more definitive evidence.

CONCLUSION

We describe a patient in which SCS was effective in treating PLMT syndrome. As of the 8-month follow-up after SCS implantation, there has been no flare-up of pain or involuntary movements.

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