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Surgical Neurology International

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SNI: Pain

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Case Report

Case report: Favorable outcomes of spinal cord stimulation in complex regional pain syndrome Type II consistent with thermography findings

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Received : 22 September 2021
Accepted : 04 November 2021
Published : 08 December 2021

DOI
10.25259/SNI_959_2021

Quick Response Code:

ABSTRACT

Background: Complex regional pain syndrome (CRPS) is a chronic pain disorder that develops as a consequence of trauma to one or more limbs. Despite the availability of multiple modalities to diagnose CRPS, a gold standard technique for definitive diagnosis is lacking. Moreover, there are limited reports describing the use of spinal cord stimulation (SCS) to treat CRPS Type II, given the low prevalence of this condition. Herein, we present the case of a patient with CRPS Type II with novel thermography findings who underwent SCS for pain management after an Achilles tendon repair surgery.

Case Description: A 38-year-old woman was referred to our institute because of chronic left leg pain after Achilles tendon rupture repair surgery. Her case was diagnosed as CRPS Type II based on the International Association for the Study of Pain diagnostic criteria. After an epidural block, thermography showed a significant increase in the body surface temperature of the foot on the observed side. She was subsequently treated with SCS, following which her pain ameliorated. She reported no pain flare-ups or new neurological deficits over 2 years of postoperative follow-up assessments.

Conclusion: SCS could be a useful surgical treatment for medication refractory CRPS Type II as supported by our thermography findings. We may refine surgical indication for permanent implantation of SCS with the presented method.

Keywords: Complex regional pain syndrome Type II, Neurosurgery, Spinal cord stimulation, Thermography

INTRODUCTION

Complex regional pain syndrome (CRPS) is a chronic pain disorder that develops as a consequence of trauma to one or more limbs.[2,21] The clinical manifestations of CRPS include sensory, vasomotor, and autonomic impairments and can be divided into two subtypes: Type I does not involve nerve injury, whereas type II involves significant proven nerve injury and is much less reported.[8,18] For the diagnosis, several tools such as 3-phase bone scans, X-ray imaging, magnetic resonance imaging (MRI), functional MRI, and thermographic devices have been widely accepted for objective diagnosis.[1] In particular, temperature differences are widely regarded as predictors in the diagnosis of CRPS, and thermography has been applied as a diagnostic tool in CRPS.[3,6]

Spinal cord stimulation (SCS) is a useful treatment modality for reducing chronic pain. SCS has been generally reported to be a highly effective method for the treatment of CRPS Type I, but its use in CRPS type II has been reported in only a few cases due to the low prevalence.[14] Here, we present a case with novel thermography findings in a CRPS Type II patient who underwent SCS following Achilles tendon repair.

CASE PRESENTATION

A 38-year-old woman was referred to our institute because of chronic left leg pain after Achilles tendon rupture repair surgery. She ruptured her left Achilles tendon and experienced persistent pain after undergoing open surgery for a ruptured left Achilles tendon at a previous hospital. She received oral treatment and epidural blocks at the previous hospital's pain clinic, but these treatments were not effective. Six months after the surgery, she visited our pain clinic and was diagnosed with left lower extremity CRPS Type II based on the International Association for the Study of Pain diagnostic criteria. Thermography showed that the body surface temperature of the affected foot was 2°C lower than that of the unaffected foot on the healthy side. After an epidural block, thermography showed a significant increase in the body surface temperature of the foot with the pain relief on the observed side. Subsequently, she underwent peroneal nerve dissection during orthopedic surgery, but the pain persisted with decreased temperature of the affected side on thermography [Figure 1 a and b]. She was referred to our department by the pain clinic for the SCS treatment.

The patient was placed in a prone position. A straight skin incision (length, approximately 5 cm) was made in the midline to perform the puncture at the L1/L2 and L2/3 levels. The lead implantation was performed under local anesthesia.

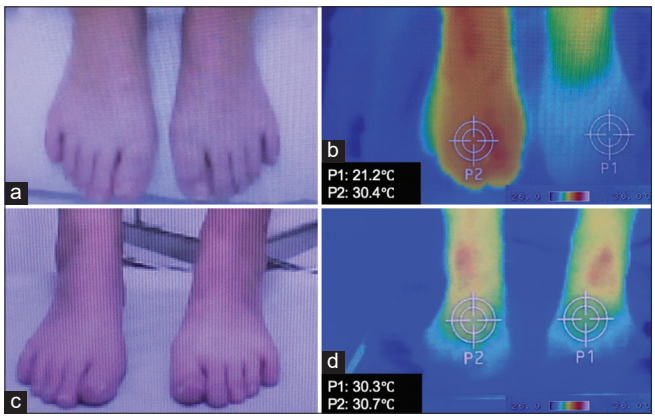

Figure 1: (a and b) Despite peroneal nerve dissection, thermography showed a decreased temperature of the affected side. (c and d) Postoperative findings included an improvement in the temperature decreasing on the affected side that had been observed preoperative thermography.

The first stimulating electrode was placed from the L2/L3 intervertebral space on the left side of the midline so that the tip spanned the level of the upper end of Th9. Stimulus-evoked sensations were confirmed to coincide with the pain site. The second electrode was then implanted from the L1/L2 intervertebral space to the right (approximately midline) of the first electrode [Figure 2]. The test stimulation elicited a paresthesia in the painful area opposite to the first electrode. As with the first electrode, the fascial puncture site was sutured and fixed, and the C-arm was used to confirm that both electrodes were located in the dorsal part of the spinal cord. Both electrodes were connected to an extension cable, and a part of the extension cables was externalized through the left side of the abdomen. In this case, Medtronic SCS percutaneous leads (1 × 8 Compact SureScan MRI - model 977A275, Medtronic, Minneapolis) were used. Following a week of the successful test stimulation period, the patient elected for the permanent implantation of the pulse generator (Intellis, model 97715, Medtronic, Minneapolis).

The patient showed a favorable postoperative course without complications. The numerical rating scale score was 8 points preoperatively, but it improved to 2 points postoperatively. Postoperative findings included an improvement in the temperature decrease on the affected side on preoperative thermography [Figure 1c and d]. The patient has shown no pain flare-ups or new neurological deficits over 2 years of postoperative follow-up assessments. The SCS parameters were followings: amplitude 3.9–9.0 mA, pulse width 60 μs, frequency 1000 Hz, cathode 4 and 7, anode 6 [Figure 2].

DISCUSSION

CRPS is a chronic neurological pain disorder involving the limbs that are characterized by severe pain along with sensory, autonomic, motor, and trophic abnormalities.[2,21] CRPS is thought to be induced by surgery, trauma, or minor injury and has a varying course, ranging from mild and self-limiting

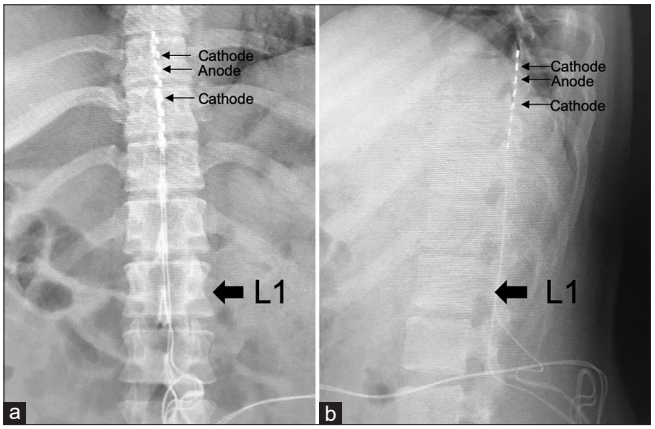

Figure 2: Plain X-ray ((a) anteroposterior view; (b) lateral view) of postoperative lumbar X-ray.

to severe chronic pain, which impairs activities of daily living. CRPS can be classified into two subtypes: Type I and II, which are characterized by the absence or presence of identifiable specific nerve injury. CRPS Type I usually develops after an initiating harmful event is not limited to damage to a single peripheral nerve and is disproportionate to any inciting event. In contrast, CRPS Type II can be defined as burning pain, allodynia, and hyperpathia occurring in a region of the limb after specific partial injury of a nerve or related branches.[7]

In a population-based study, the estimated overall incidence rate of all CRPS was 26.2/100,000 person years (95% CI: 23.0–29.7).[4] Female patients were affected approximately 3 times more often than males; the highest incidence occurred in female patients aged 61–70 years.[4] Another population-based study showed that the incidence rate of CRPS type I was 5.46/100,000 person-years at risk, whereas that of CRPS Type II is 0.82/100,000 person-years. A retrospective study by Rewhorn *et al*. evaluating the occurrence of CRPS following elective ankle and foot surgery in 390 patients showed that the overall incidence was 4.4%: 3.6% for CRPS Type I, and 1.8% for CRPS Type II.[17] These studies suggest that the number of cases of CRPS type II is much lower than that of CRPS Type I, which implies that our case is rare.

Achilles tendon rupture repair surgery has been reported to cause nerve-related complications. The overall incidence of nerve-related complications in open and percutaneous Achilles tendon rupture repair surgery ranges from 3% to 18%.[9,16] Generally, the nerve-injury rate in the percutaneous group is significantly higher than that in the open surgery group.[22] In the present case, the patient underwent open surgery, but nerve injury during the surgery resulted in CRPS. Thus, nerve injuries resulting in CRPS are still a complication of open Achilles tendon rupture repair.

A diagnosis of CRPS is supported by the relatively higher temperature in the pain-affected area, which can be primarily attributed to increased sympathetic activity. Krumova *et al*. reported that a skin temperature of two degrees Celsius or greater in pain-affected areas demonstrated a diagnostic sensitivity of 73% and specificity of 94% in the diagnosis of CRPS.[11] Although their findings require further evaluation due to the lack of cases and clinical implications, our case also showed an increase of two degrees Celsius or greater in the skin temperature at the pain-affected area on thermography, suggesting that temperature measurements can be used as a specific diagnostic method. In addition, the temperature difference in the lower extremities improved after an epidural block, suggesting that it may be a favorable outcome in the treatment of CRPS. However, it should be noted that the evaluation using thermography is susceptible to the influence of outside temperature. Further evaluation is needed to support the clinical implications.

Neuromodulation has been shown to play an important role in treating CRPS, especially in patients who are unresponsive to multiple medical management modalities and sympathetic blockade. Santon reported that if patients do not respond to conventional treatment within 12–16 weeks, SCS should be considered for surgical management.[19] Kemler *et al*. reported that SCS and physiotherapy were significantly more effective for pain relief than physiotherapy only over the 6-month and 2-year follow-up periods.[10] Another study reported that in the majority of patients, SCS contributes to sustained improvements in functional capability, quality of life, and pain management.[5,13] Although fewer reports have described SCS for CRPS Type II, a retrospective report evaluating 32 patients with CRPS, including 6 patients with CRPS Type II, found that after 8 years of follow-up, 23 patients (71.8%) continued to show successful long-term pain relief.[12] This case showed successful SCS treatment for CRPS Type II, which is relatively rare and will contribute to the establishment of surgical treatment for CRPS Type II that is unresponsive to multiple pain management modalities.

Various mechanisms of action for SCS have been suggested. The effects of SCS on the vascular symptoms of CRPS are thought to occur via two main mechanisms: antidromic activation of spinal afferent neurons and inhibition of the sympathetic nervous system.[15] Peripheral vasodilation after SCS, which involves antidromic release of calcitonin gene-related peptide, and possibly nitric oxide, from small-diameter sensory neurons expressing the transient receptor potential V1 receptor, has been extensively studied in animal models.[20] Our case showed an improvement in the temperature in the affected limb after SCS. This could be related to the mechanism of peripheral vasodilation after SCS by antidromic activation of spinal afferent neurons and inhibition of the sympathetic nervous system.

CONCLUSION

SCS could be a useful surgical treatment for medication refractory CRPS type II as supported by our thermography findings. We may refine surgical indication for permanent implantation of SCS with the presented method.

Ethical approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

This study was partially supported by Japan Society for the Promotion of Science (JSPS) Grant-in-Aid for Scientific Research (C) (Grant number: 18K08956), and the Central Research Institute of Fukuoka University (Grant number: 201045).

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Oda K, Morishita T, Shibata S, Tanaka H, Hirai N, Inoue T. Case report: Favorable outcomes of spinal cord stimulation in complex regional pain syndrome Type II consistent with thermography findings. Surg Neurol Int 2021;12:598.