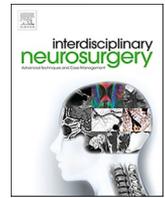




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Effectiveness of spinal cord stimulation for improvement of microcirculation and fixed dystonia in complex regional pain syndrome: A case report

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ABSTRACT

Complex Regional Pain Syndrome (CRPS) is a form of chronic pain affecting one arm or leg. Spinal cord stimulation (SCS) is one of the few effective treatments for CRPS. However, it is difficult to objectively determine the appropriate conditions of CRPS and predict the efficacy of SCS.

In this case report, we report a 21-year-old woman with CRPS type I, who had a history of meniscus injury in the left knee at age 15. Her motor and sensory symptoms in the left lower extremity improved dramatically with SCS. Thermography imaging showed a significant change in the skin temperature of the left lower limb before and after surgery, which can be useful in objectively assessing the therapeutic effect.

1. Introduction

Complex regional pain syndrome (CRPS) is a form of chronic pain affecting one arm or leg. It typically occurs after soft tissue or bone injury, or after surgery or stroke. There are two types of CRPS: Type I in which there is no nerve damage – also known as reflex sympathetic dystrophy – and type II associated with distinct nerve injury. In addition to chronic pain, affected patients manifest various symptoms that do not correspond to a single innervated area. Currently, the most commonly used criteria for diagnosis of CRPS, established by the International Association for the Study of Pain, include chronic pain, allodynia or hyperalgesia, vasomotor symptoms, abnormal sweating or edema, and motor symptoms in the absence of another diagnosis (Supplementary Table). Motor symptoms include local weakness, dystonia, a restricted range of motion, and atrophic changes in the nails and skin, these symptoms being diverse and altering during the course of the disease. Spinal cord stimulation (SCS) is one of the few effective treatments for CRPS. However, due to the unknown pathophysiology of CRPS, it is difficult to determine the most appropriate indications for SCS or predict its efficacy. In the present case report, we describe a 21-year-old woman with CRPS type I in whom motor and sensory symptoms in the left lower extremity improved dramatically after SCS treatment, demonstrating the efficacy of thermography for a non-invasive, prompt and objective

assessment of CRPS, which will lead early surgical intervention and favorable outcome.

Written informed consent was obtained from the patient for this case report.

2. Case presentation

The patient was a 21-year-old female nursing assistant, who had a history of meniscus injury in the left knee diagnosed with MR imaging at age 15, which had improved after conservative treatment. She also had medical history of bronchial asthma, hyperventilation syndrome, and panic disorder, as well as being allergic to multiple drugs, including loxoprofen, pregabalin, eperisone and limaprost alfadex, in addition to buckwheat, latex, and grapefruit. She had no particular family history.

At the age of 20, her left knee pain reappeared without any cause and she consulted various orthopedists, but no abnormal findings were detectable by MR imaging. Her left knee pain worsened with bad weather and strong exercise. Four months after onset, her left lower limb often turned purple and cyanotic after bathing or in an upright position. At the seventh month after onset, she became unable to walk without support and the left ankle joint became constricted to a varus and extended position (Fig. 1). She was unable to perform dorsiflexion or plantar flexion voluntarily in the left ankle joint, and passive movements

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Fig. 1. The patient's lower legs before surgery. The skin of her left leg turned deep purple and the left ankle joint was constricted.

were also restricted due to severe pain.

Various antinuclear antibodies and rheumatoid factors were negative, and the absence of other collagen tissue diseases or autoimmune disorders were confirmed by rheumatologists. The results of neurophysiological examinations including a nerve conduction velocity test

were within the normal ranges.

MRA revealed a decreased blood flow signal from the distal part of the left popliteal artery to the peripheral side (Fig. 2) while vascular ultrasound examination demonstrated no arterial stenosis or venous thrombosis in the lower extremities. Thermography showed a significant decrease in the skin temperature of the left lower leg (Fig. 3A).

After the onset, various medication including acetaminophen, pregabalin and duloxetine were administered by an orthopedist, vascular surgeon, rheumatologist and anesthesiologist, but were not effective while some of them caused allergic response. Finally, with a diagnosis of CRPS and fixed dystonia, the anesthesiologist referred her to the neurosurgery department of our institution and SCS was introduced considering efficacy.

3. Surgical intervention

Two spinal cord stimulation electrodes (Medtronic Co.) were implanted in the epidural space under local anesthesia. According to the sensation evoked in the patient, the tip of the left paramedian lead was placed at the Th12 level and the tip of the other lead in the midline was placed at the Th11 level. Using stimulation with paramedian electrodes the evoked paresthesia covered her painful left ankle joint. Using stimulation with the other medial electrodes, the induced paresthesia covered an area from the buttocks on both sides to the tip of left lower limb. The test stimulation started with a stimulation width of 300 μ s and a frequency of 5 Hz just after electrode surgery. The intensity was set above the sensory threshold but did not create an uncomfortable sensation.

On the day after the operation, the pain, numbness and stiffness in the left lower leg had improved. Three days later, the patient became independent in wheelchair transfer, dressing and standing. During the trial period of 9 days, she got to feel more comfortable by modifying the stimulation frequency from 5 Hz to 50 Hz. After the test stimulation, an implantable pulse generator (Intellis, Medtronic Co.) was placed under general anesthesia subcutaneously just above the right lateral gluteal fascia, on the unaffected side. For both of the leads, the stimulation conditions were set with a width of 300 μ s and a frequency of 50 Hz; the intensities were set from 1.5 to 2.4 mA, depending on the patient's posture and degree of comfort. After rehabilitation for three weeks, she became able to walk independently without any support and had no pain in her lower left leg, and was discharged (see movie). Postoperative thermography showed no remarkable difference in skin temperature



Fig. 2. Magnetic resonance angiography (MRA) images of blood vessels in both lower limbs show severe stenosis of the distal part of the left popliteal artery (arrow).

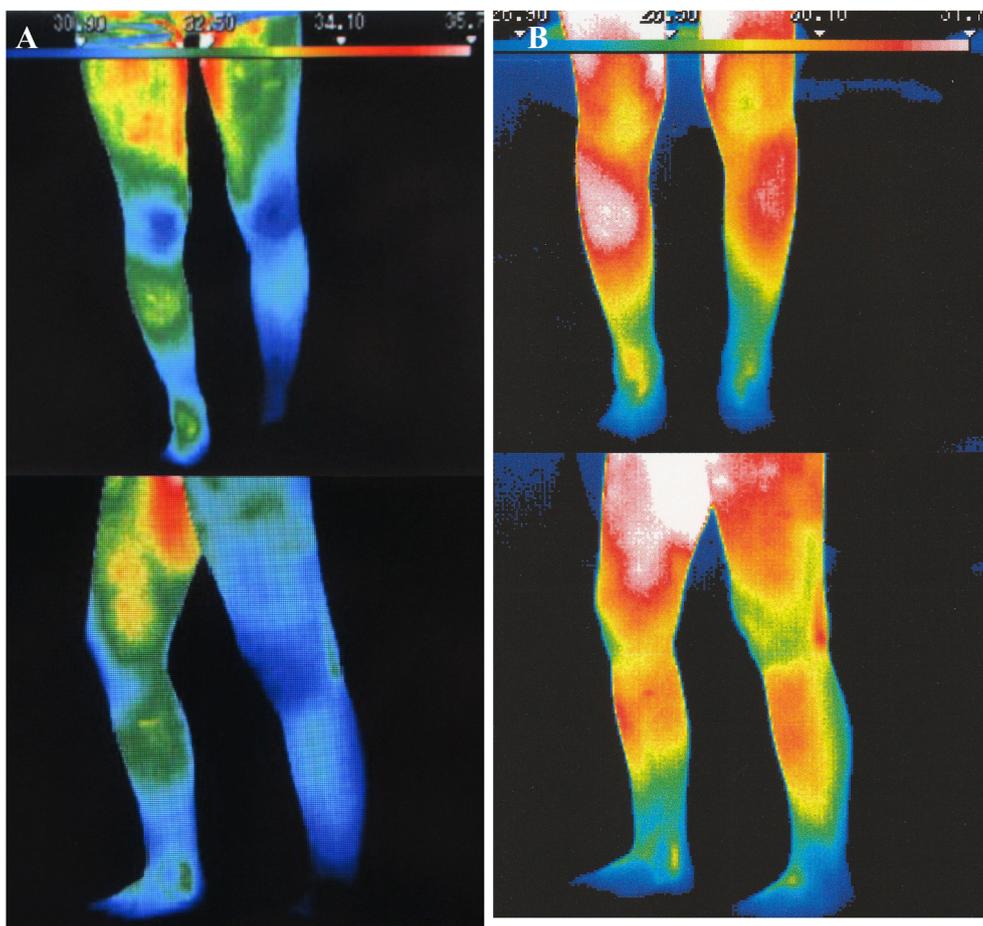


Fig. 3. (A) Thermographic images of both lower limbs. The skin temperature of the left lower leg was markedly lower than that of the right. Compared to the right, the temperature of the left fingers was slightly lower than that on the right. Central temperature was 32.5 degrees. The range was 6.4 degrees. (B) Postoperative thermography of the legs. There was no significant difference in skin temperature between the left and right legs. Central temperature was 28.5 degrees. The range was 6.4 degrees.

between the left and right lower legs (Fig. 3B). Two years after surgery, she returned to her former job as a part-time worker, without any complications.

4. Discussion

A previous randomized controlled trial [1] has revealed that a CRPS type I patient group that underwent SCS within two years after diagnosis showed more significant pain suppression, improved limb function, and better QOL than a group that received only physical therapy. Another study [2] has reported that earlier SCS intervention is important for pain control; SCS begun within two years for CRPS, achieved an analgesic effect in more than 85% of patients, whereas SCS begun at 15 years or later after onset improved pain in only 9%. In addition to severe pain, CRPS can cause joint contracture and deformity of the extremities, and thus early intervention should be considered for such patients.

In the present patient, we implanted the SCS system within two years after symptom onset, adopting conventional tonic stimulation with a frequency of 50 Hz, and obtained a favorable outcome. The stimulation conditions employed, including frequency, pulse width and intensity, depend on the range of paresthesia required. With regard to frequency, a setting higher than 30 Hz tends to widen the range of paresthesia [3]. In the present case, a frequency of 50 Hz was chosen according to the patient's subjective and objective symptoms, achieving a successful outcome. In clinical trials for CRPS, there was no significant difference between 40 Hz and high-frequency stimulation, with about half of patients preferring 40 Hz [4]. On the other hand, it has been reported that higher stimulation settings achieve a stronger analgesic effect [5].

Electrical stimulation of the dorsal cord evokes paresthesia in a large area below the stimulated segments, sometimes covering both sides

while stimulation of the dorsal root is likely to cover only one or a few segmental areas [6]. The combination of lead electrodes placed in the para-midline and midline will effectively evoke paresthesia sensation in the most painful segment and a wide range in the rest of the affected area, as shown in the present case.

It is reported that a history for allergy/hypersensitivity reactions is a predisposing condition for CRPS type I [7]. Our patient had also a history of multiple drug allergy, in accordance with this notion.

Thermography is a useful tool for displaying regional body temperature, graphically detecting infrared energy emitted from the human body surface. The cutaneous vasculature regulating heat dissipation is divided into capillaries existing in most skin and arteriovenous anastomosis existing only in the glabrous skin of the hands, feet and nose. The anastomoses directly connect arterioles and venules in a skin layer and are controlled by sympathetic nerves to conduct heat to the skin surface [8]. Thermoregulation of the skin is regulated mainly by the action of arteriovenous anastomoses, under the control of the hypothalamus via the sympathetic nerves. Hyperactivity of local sympathetic nerve fibers leads to a temperature decrease in the peripheral limbs. As effects of SCS on vascular symptoms of CRPS are thought to occur through two mechanisms: antidromic activation of spinal afferent neurons and inhibition of sympathetic efferents, leading cutaneous vasodilation that will increase skin temperature [9]. Thermography to measure the skin temperature of an affected extremity should be considered for positive visualization of CRPS [10].

In the present patient, monitoring of her skin temperature by thermography and MRA imaging of that could reflect a vasomotor reaction of her affected extremities were useful for early diagnosis and surgical intervention, leading to a successful outcome.

5. Conclusion

We have reported a young female patient suffering from severe pain and deformity of the leg caused by CRPS. Thermography was useful for both diagnosis and assessment of treatment efficacy, and early intervention using SCS was quite effective for improving her symptoms and activities of daily life.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.inat.2021.101297>.

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