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Temporary Peripheral Nerve Stimulation for Complex Regional Pain Syndrome Type I in the Upper Extremity: A Case Report

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Background: Complex regional pain syndrome (CRPS) presents a significant treatment challenge, characterized by severe pain following trauma or surgery. This study examines CRPS management, emphasizing the role of temporary peripheral nerve stimulation (PNS) in alleviating subacute postoperative symptoms.

Case Report: Focusing on a case study of a 65-year-old woman with CRPS Type I, we explore the use of a temporary PNS system, highlighting its effectiveness in reducing pain by 80% and facilitating rehabilitation. The procedure involved ultrasound-guided percutaneous lead placement, showcasing a minimally invasive approach with substantial functional improvements for the patient.

Conclusions: This case underscores the potential of temporary PNS in CRPS treatment in the subacute setting, advocating for further research to validate its efficacy and expand its application in clinical practice. The manuscript calls for additional studies to explore the benefits of temporary PNS, aiming to enhance pain management strategies and improve quality of life for CRPS patients.

Key words: Peripheral nerve stimulation, complex regional pain syndrome, upper extremity pain, chronic pain, case report

BACKGROUND

Complex Regional Pain Syndrome

Complex regional pain syndrome (CRPS) is a chronic pain condition that can sometimes develop after trauma or surgery to an extremity, manifesting as hyperalgesia and allodynia (1). Formerly known as reflex sympathetic dystrophy (CRPS Type I) and causalgia (CRPS Type II), the 2 types differ in that Type I occurs in the absence of nerve damage and Type II occurs with known nerve damage (2). The pathophysiology of CRPS is not completely understood; however, it is believed to involve dysfunction of the central and peripheral nervous systems in response to tissue injury and inflammatory changes, as well as autonomic dysregulation (3).

CRPS Type 1 is generally recognized to have 2 clinical stages. The first is the acute "warm" phase, and the second is the chronic "cold" phase (4). During the acute "warm" phase, patients typically exhibit symptoms, such as inflammation, swelling, pain, and changes in skin temperature in the affected limb. Research suggests that the symptoms in the "warm" phase may result from an amplified innate immune response that causes skin cells, like keratinocytes, to release proinflammatory cytokines (5). The chronic phase is characterized by a decrease in inflammation, but ongoing pain. Although inflammation decreases in the "cold" phase, proinflammatory mediators remain elevated. Some symptoms of the chronic phase are due to elevated inflammatory markers from the acute phase, where cytokines, such as TNF-alpha, IL1B, and IL-17, activate osteoblasts and

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osteoclasts, leading to osteoporotic changes from rapid bone turnover. Neurogenic inflammation has a role in developing CRPS symptoms, like allodynia and hyperalgesia. Stimulation of peripheral nociceptive C fibers leads to the conduction of signals both toward the dorsal ganglia (afferently) and back to the affected tissue (efferently). This retrograde transmission results in the release of proinflammatory neuropeptides (4).

CRPS has an incidence of between 5 and 26 per 100,000 per year and it affects the female gender most commonly (6,7). While CRPS can occur at any age, the incidence peaks around age 40 to 70 years (6,7). Up to half of CRPS cases occur after fractures, and 30% to 40% result from surgical procedures or other injuries (7). The risk of CRPS increases with increasing severity of tissue damage, regardless, even minimal tissue injury can cause CRPS, such as injections, arthroscopy, or the use of tourniquets (7). There is a mixed consensus on the psychological factors that may play a role in the risk of CRPS (7).

CRPS is a clinical diagnosis based on the Budapest Criteria (8-10). The key feature of CRPS is debilitating pain that is disproportionate to the inciting event (9). Signs and symptoms can include changes in sensation, edema, decreased range of motion, and weakness of the extremity that usually occurs distal to the site of injury (1). For clinical diagnosis, it is important that the patient reports at least one symptom in 3 of the following categories: sensory, vasomotor, sudomotor, or motor/trophic (8-10). Additionally, the patient should present with at least one sign at the time of evaluation in at least 2 of the aforementioned categories (8-10).

Many patients with CRPS have a poor prognosis and greatly impacted quality of life. Early treatment is essential for slowing disease progression and restoring function (1). The cornerstone of treatment includes physical therapy (PT) and occupational therapy (OT) in addition to behavioral therapy (10). Pharmacotherapy for CRPS has limited evidence and consists mostly of the off-label use of drugs, such as anti-inflammatories, neuropathic agents (i.e., gabapentin, pregabalin, nortriptyline), glucocorticoids, and opioids (7). Interventional therapies are used for severe CRPS that have not responded to conservative measures. These can include a series of sympathetic nerve blocks, spinal cord stimulation (SCS), dorsal root ganglion stimulation (DRG-S), and peripheral nerve stimulation (PNS) (7). The goal of interventional therapies is to provide pain control that allows for participation in PT and OT to regain movement and strength in the affected extremity (10).

Peripheral Nerve Stimulation

PNS has been used for decades for the treatment of pain. In the past, PNS required surgical dissection to place leads and had concerning complications (11). Advancements in the past decade in PNS technology now allow for percutaneous placement with ultrasound guidance and a reduction in adverse events (11).

The proposed mechanism of action for PNS involves gate control theory (12). Electric neurostimulation signals are sent to nearby nonnociceptive nerve fibers, which then inhibit the A-delta and C fibers from transmitting nociceptive signals to the brain (12). Additionally, PNS is thought to directly inhibit the local neurotransmitters and inflammatory factors that may play a role in chronic pain (13).

PNS systems can either be fully implantable leads or temporary external leads. The US Food and Drug Administration has approved temporary PNS for up to 60 days for treatment of chronic pain, postsurgical pain, and posttraumatic pain of the back and/or extremities (11). PNS is also being used for pain that is centrally mediated, such as CRPS and phantom limb pain (11). The temporary PNS system is a device that can be used to treat these conditions. Studies (14) have shown that 60-day PNS with temporary externalized leads provides an analgesic benefit that can last up to 12 months in chronic pain following amputation. This case report presents a patient with CRPS Type 1 of the upper extremity who experienced clinically significant improvement using a temporary PNS system.

CASE REPORT

Our patient is a 65-year-old woman, who presented to pain management for right upper extremity pain following a right rotator cuff repair approximately one year prior. Following the surgery, she began to have significant swelling, pain, and stiffness affecting the entire right upper extremity, with the distal portion of her extremity affected more severely than the proximal. She described the pain as burning, sharp, and shooting. The reported pain score ranged from 5/10 on better days to 10/10 on the Visual Analog Scale (VAS) on the worst days, significantly impacting her range of motion and impeding activities of daily living. She reported severe swelling of the hand and forearm. She also experienced stiffness in the fingers, wrist, elbow, and shoulder. The patient reported periodic sensations of cold in her hand and arm, as well as allodynia. On physical examination, the right upper extremity was

grossly swollen compared to the left, particularly in her hand, fingers, and forearm. Splotchy discoloration was noted in the fingers of the right hand and in the hand itself. There was some degree of hyperalgesia to compression of the right hand and wrist. There was limited active range of motion of the right wrist, hand, and shoulder. The following signs and symptoms met the Budapest Criteria for CRPS: edema, hyperalgesia, allodynia, skin color changes, decreased range of motion, and temperature asymmetry. A diagnosis of CRPS Type 1 in the right upper extremity was established.

The patient did not experience relief of her symptoms with conservative treatments, including gabapentin, duloxetine, OT, and PT. Three right stellate ganglion blocks were performed with one-week intervals, resulting in a 50% reduction for 3 months, a repeat of the 3 right stellate ganglion blocks with one-week intervals was performed, resulting in another 3 months of 50% pain relief. Aggressive PT to enhance range of motion in her wrist and fingers was deemed necessary, but her sessions were limited by pain.

The patient was offered the SPRINT temporary PNS system (SPR Therapeutics, Cleveland, OH) to address her pain and assist her ability to participate in PT. The patient consented to undergo temporary PNS. She underwent an ultrasound-guided right upper extremity ulnar and median nerve block to determine the target nerves for PNS (Fig. 1).

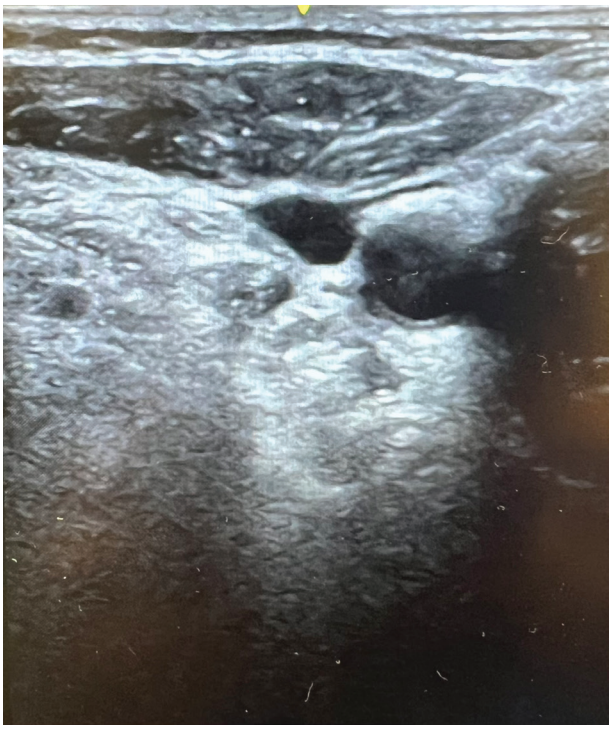

Fig. 1. Ultrasound image of peripheral nerve stimulator lead placement for ulnar and median nerve of the right upper extremity.

Procedure

The patient was positioned supine, and the right arm and hand were prepared and draped in a sterile manner. Utilizing ultrasound guidance, the leads were placed with direct observation as a conduction medium. First, the right median nerve was identified under ultrasound visualization, followed by insertion of a needle and placement of a guidewire along with a testing wire. Confirmation of the lead placement was achieved through local stimulation at extremely low sub-mA amplitude, coupled with the patient's report of stimulus-evoked sensations corresponding to regions of pain. Subsequently, the PNS lead was positioned adjacent to the right median nerve.

The right ulnar nerve was visualized using ultrasound guidance. Following needle insertion, a guidewire was introduced alongside the ulnar nerve under direct visualization of the ulnar artery and nerve. Confirmation of proper lead placement was obtained through local stimulation at extremely low sub-mA amplitude, after which the PNS lead was deployed adjacent to the right ulnar nerve.

All leads were confirmed to be subcutaneous and local stimulation was confirmed again (Fig. 2). Finally, both leads were connected to the external pulse generator and secured without complication. The patient was discharged to recovery in unchanged neurologic condition and instructed on the use of the external communicating generator. The SPRINT temporary PNS system program settings include a pulse rate of 12 to 100 Hz, an amplitude of 0 to 30 mA, and a duration of 10 to 200 microseconds (15).

Resolution

The temporary PNS system is designed to be in place for a duration of 60 days. The patient returned to the clinic 60 days postprocedure for lead removal (Fig. 3). She reported resolution of the swelling, cold sensation, and allodynia in her right hand. On physical examination, improvement in right-hand swelling was noted. The patient was still unable to fully close the right hand; however, grip strength was intact and improved. The

discoloration in the right hand had improved, and color was equal in both extremities. The patient was able to continue her PT sessions and observed improvements in her range of motion and right grip strength. The patient reported 80% pain relief and overall satisfaction with her outcome.

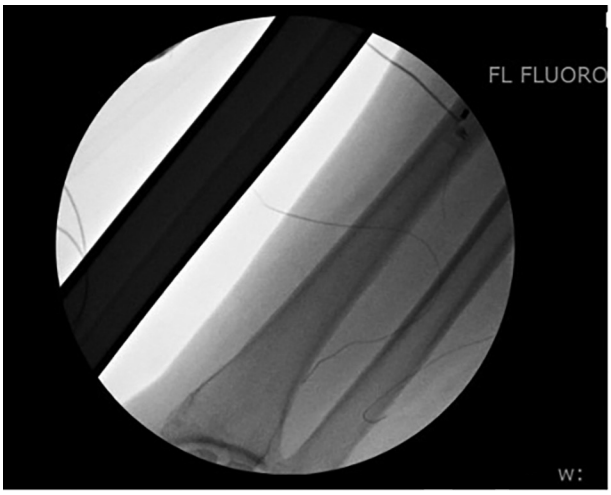

Fig. 2. Fluoroscopy image of peripheral nerve stimulator lead placement for ulnar and median nerve of right upper extremity.

Fig. 3. Patient returns for follow-up slightly over 60 days post-procedure for lead removal with reduced swelling, stronger grip, and improved range of motion.

DISCUSSION

CRPS can be a debilitating chronic pain condition that requires prompt diagnosis and intervention to restore quality of life (1). Interventional pain management options for CRPS include sympathetic nerve .blocks, SCS, DRG-S, and PNS (7). PNS may offer greater accuracy compared to SCS due to a more well-defined target (11). PNS has the advantage of using ultrasound guidance and percutaneous lead placement to offer a less invasive approach compared to other interventions (11).

The use of PNS for upper extremity CRPS using ultrasound-guided percutaneous lead placement has been described in a few studies and case reports. Bouche et al (16) used this technique to treat 16 patients with CRPS Type I or II, with leads implanted near the suprascapular nerve or cervical nerve roots of the brachial plexus. Results showed a 68% improvement in pain scores (16). Frederico et al (17) used an ultrasound-guided percutaneous technique for PNS of the brachial plexus in 10 patients with CRPS Type I or II and found a reduction in mean pain VAS scores from 8.9/10 to 3.8/10 at one-year follow-up. A case report (18) describes wireless PNS of the radial and median nerves for CRPS Type I of the upper extremity providing symptomatic relief and a decrease in opioid use. Another case report (19) showed success in ultrasound-guided percutaneous PNS of the cervical and upper thoracic sympathetic chain for CRPS Type I of the upper extremity.

In our case report, ultrasound guidance was used to percutaneously place 2 leads of a 60-day temporary PNS system to target the median and ulnar nerves. Temporary PNS systems have shown success in providing long-term pain relief at 12 months for chronic pain following amputation (14). This minimally invasive approach helps reduce complications, such as infection and lead migration (14). The risk of hardware malfunction is reduced due to the temporary nature of this method and the lack of internally placed hardware (14). Upon our review of the literature, there are a limited number of studies on the use of temporary PNS systems for CRPS of the upper extremity. Our patient's preprocedure symptoms affected the entire right upper extremity regionally, rather than in a particular nerve distribution. Postprocedure, she experienced improvement in swelling, color, sensation, and motor

function. While only the ulnar and median nerves were targeted, there was global improvement in her right hand. Our case report demonstrates a clinically significant improvement in symptoms without adverse effects using this technology.

CONCLUSIONS

This report highlights the necessity for additional research to further substantiate the efficacy and benefits of this therapy in treating individuals with CRPS, particularly in the subacute postoperative setting. The use of temporary PNS in both subacute and persistent postoperative contexts presents a unique treatment opportunity to enhance pain management and facilitate participation in rehabilitation for patients with CRPS and subacute or persistent postoperative pain.

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