

PERIPHERAL NEUROMODULATION FOR TREATMENT OF UPPER EXTREMITY COMPLEX REGIONAL PAIN SYNDROME FOLLOWING PERIPHERALLY INSERTED CENTRAL CATHETER PLACEMENT: A CASE REPORT

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- Background:** Complex regional pain syndrome (CRPS) is a chronic pain condition that typically affects distal extremities after an injury or noxious event, with symptoms disproportionate to the primary insult. We present the first successful use of a temporary peripheral nerve stimulator (PNS) followed by a permanent PNS system to treat CRPS Type 2 after failed responses to pharmacotherapies, trigger point injections, cervical epidural injections, and sympathetic nerve blocks.
- Case Report:** An 81-year-old patient developed CRPS Type 2 following a left antecubital peripherally inserted central catheter placement with left-hand paresthesia, edema, weakness, and skin color and temperature asymmetry. He achieved near-complete resolution of symptoms approximately 4 months after permanent PNS placement.
- Conclusions:** This case underscores the morbidity associated with iatrogenic complications, the challenges of managing CRPS when conventional treatments prove insufficient, and the necessity for further research to assess the effectiveness of PNS in improving both function and pain relief for CRPS patients.
- Key words:** Complex regional pain syndrome type 2, routine procedure, peripheral nerve stimulation, case report
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BACKGROUND

Complex regional pain syndrome (CRPS) is a chronic pain condition that typically affects distal extremities after an injury or noxious event, with symptoms disproportionate to the primary insult (1). Occurring 3-4 times more in women than men with a peak age of onset between 50 and 70 years old, these symptoms include sensory, vasomotor, sudomotor, and trophic changes based on the most recently revised International Association for the Study of Pain diagnostic criteria (2-5). The

clinical course may be highly variable, with a significant portion of patients having persistent pain and reduced function of the affected limb at one year (6). Fractures, blunt trauma, surgery, and carpal tunnel syndrome are common inciting events, although no identifiable event is seen in many cases (7). Reports of CRPS development following routine procedures, including vascular access and intramuscular injections, exist in the literature (8,9). Common treatment modalities for CRPS include physical therapy, such as graded motor therapy and

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mirror therapy, pharmacotherapy, such as gabapentin and bisphosphonates, and interventional therapies, such as sympathetic blocks and spinal cord stimulation (SCS). We present a case of an 81-year-old man who presented with CRPS (Type 2) following a peripherally inserted central catheter (PICC) that was successfully treated with peripheral nerve stimulation (PNS). The patient presented in this case report provided written consent for publication of results and Health Insurance Portability and Accountability Act authorization.

CASE PRESENTATION

The patient is an 81-year-old man, who presented to our practice for persistent left-hand pain following hospitalization for a pulmonary embolus 2 years prior. His medical history included atrial fibrillation, hypertension, asthma, osteoarthritis, and obesity, with a surgical history, including bilateral total knee arthroplasty and inferior vena cava filter placement. During hospitalization, the patient was treated with tissue plasminogen activator, and a left antecubital PICC was placed for fluid administration and lab draws. Shortly after placement, the patient experienced pain and swelling at the puncture site. The PICC line remained for 24 hours, and infusions/labs continued until the patient requested removal, which immediately reduced puncture site swelling. Over the next 48 hours, the patient endorsed pain along the antecubital fossa radiating to the shoulder and hand, accompanied by erythema, ecchymosis, and a persistent numbness and tenderness along the first 3 digits. Neurology was consulted, and gabapentin was started alongside supportive care. Outpatient electromyography and nerve conduction studies confirmed bilateral carpal tunnel syndrome.

Two years after symptom onset, the patient continued to experience persistent burning left-hand pain along with periodic cold sensations, swelling, discoloration, and weakness. His pain flares were described as a 7-10/10 on the Numeric Rating Scale, severely limiting his range of motion and participation in physical therapy (PT). With disproportionate continuing pain with paresthesia, edema, weakness, and skin color and temperature asymmetry, a diagnosis of CRPS was established. The patient found minimal to no relief from therapies over these 2 years, including cervical epidural injections, cervical medial branch blocks, trigger point injections, and topical capsaicin. A series of stellate ganglion and median nerve blocks provided initial, but diminishing relief. Conservative measures, including gabapentin

and duloxetine, and PT to assist with contractures from immobility also offered minimal relief. Therefore, the decision was made to proceed with a temporary Sprint PNS (SPR Therapeutics, Cleveland, OH) for 60 days.

Procedure

The temporary PNS was placed under ultrasound (US) guidance using an in-plane approach toward the median nerve (Fig. 1). Throughout the 60 days postprocedure, PNS provided 50% pain relief and improved upper extremity function. However, after removal of the temporary stimulator, the patient complained of recurring symptoms and was interested in a permanent stimulator.

Thus, a PNS StimRouter implant (Bioventus, Durham, NC) of the left median nerve was placed (Fig. 2). The left median nerve was identified using US guidance in a short-axis view. Anesthesia of skin and subcutaneous tissue was performed with 2 mL of 1% lidocaine with epinephrine mixed with bupivacaine 0.25% using a 27G needle. An 11-blade was used to make a stab incision, and an 18G needle was used to locate the nerve. The test electrode was then inserted through the 18G needle and tested. An introducer probe (Bioventus, Durham, NC) was inserted and advanced under US guidance using an out-of-plane approach while keeping the needle perpendicular to the beam until reaching the nerve. There was no evidence of blood or paresthesia. Stimulation was seen with 1.5 V and was concordant with the patient's site of chronic pain after repositioning. The dilator was placed over the probe, and the stylet and probe were removed. The lead introducer was passed through the dilator sheath to the target nerve. Stimulation was seen with 2.0 V and was concordant with the patient's site of chronic pain. The sheath was removed, and the lead was tested again with concordant paresthesia. Using the same anesthetic, 2 mL was injected into the tunneling tract from the target nerve. The tunneling stylet was inserted to create a tract, followed by the dilator sheath. The lead was then placed inside the sheath, which was then divided and removed, followed by burying the lead. The incision was closed with sutures and secured with an adhesive dressing (Fig. 3).

Outcome

The patient was followed regularly postprocedure. At one week, pain relief was 20% to 30%, increasing to 90% to 95% relief at one to four months with periods of complete relief. At 14 months postimplant, pain

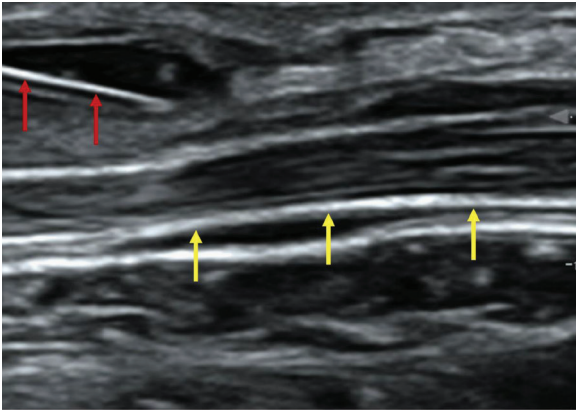


Fig. 1. US image of placement of temporary PNS lead (red arrows) toward median nerve (yellow arrows) using an in-plane approach. US, ultrasound; PNS, peripheral nerve stimulator.

relief remains > 90%. When therapy was turned off, numbness and pain gradually returned over several hours. Stimulation enabled the patient to consistently participate in mobility and functional exercises. Before neuromodulation, the patient was unable to clench his hand due to pain, which nearly resolved postimplant. The patient was able to log and record the intensity and combinations of different programs to a level that provided him with the most significant relief. The patient's optimum program was 100 μ s, 120 Hz, and 11 mA.

DISCUSSION

We present a rare case of CRPS Type 2 following a left antecubital PICC placement with a 2-year history of left-hand paresthesia, edema, weakness, and skin color and temperature asymmetry. We report the first successful use of a temporary PNS followed by a permanent PNS system to treat upper extremity CRPS Type 2. Furthermore, PNS for this patient achieved near-complete resolution after failed responses to pharmacotherapies, trigger point injections, cervical epidural injections, and sympathetic nerve blocks.

Common complications with PICC placement include deep vein thrombosis, local infection, occlusion, and malposition, while neurologic complications are exceedingly rare and typically resolve over time (10-14). Although CRPS has been reported following repeated vascular access during transradial coronary interventions, intramuscular injections, and intravascular placements, no cases were found following PICC placement (8,9).

Existing literature for PNS and CRPS is also limited and includes one retrospective chart review from a

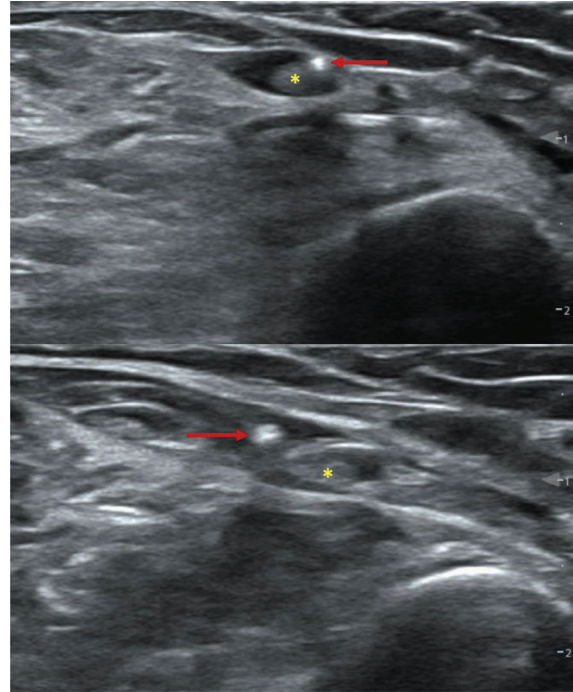


Fig. 2. US image of placement of permanent PNS lead (red arrow) proximal to the median nerve (yellow asterisk) using an out-of-plane approach. US, ultrasound; PNS, peripheral nerve stimulator.



Fig. 3. Picture illustrating position of device placement in left upper extremity.

single center where 25 of 240 patients with CRPS Type 2 showed significantly decreased pain scores, and case reports of PNS successfully reducing pain in CRPS Type 1 (15-22). One study (23) from Germany evaluated 11 patients with CRPS Type 2 and demonstrated significant pain reduction and improved functionality after 12 months.

Neuropathic pain, whether central or peripheral, is also challenging to treat with limited options for patients unresponsive to traditional therapeutic modalities. Neuromodulation techniques, such as SCS and PNS, based on Melzack et al's (24) gate theory, inhibit ascending noxious stimuli from nociceptors to the central cortex. While SCS and PNS have shown variable success in CRPS treatment, current evidence primarily supports SCS for neuropathic pain (25-27). Although less studied, PNS may offer advantages over SCS as it is less invasive with safer placement, reduced adverse events, greater selectivity, and greater accuracy (28). Thus, the effectiveness of PNS in this case was likely due to the precise identification of mononeuropathy and targeted placement of the device over the median nerve. Early

recognition of symptoms and prompt treatment can also prevent long-term loss of function, halting the natural progression and improving long-term morbidity with CRPS (29).

In summary, this rare case of CRPS Type 2 following routine intravenous placement underscores the morbidity associated with iatrogenic complications and sheds light on the challenges of managing CRPS when conventional treatments prove insufficient. It highlights PNS as a valuable alternative in improving both function and pain relief, while emphasizing the necessity for further research to assess the effectiveness for CRPS Type 2.

CONCLUSIONS

PNS represents a valuable therapeutic option for refractory CRPS. This case underscores the morbidity associated with iatrogenic complications, the challenges of managing CRPS when conventional treatments prove insufficient, and the necessity for further research to assess the effectiveness of PNS in improving both function and pain relief for CRPS patients.

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