Educational case report

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Wireless peripheral nerve stimulation for complex regional pain syndrome type I of the upper extremity: a case illustration introducing a novel technology

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Abstract

Background: Complex regional pain syndrome (CRPS) is a debilitating painful disorder, cryptic in its pathophysiology and refractory condition with limited therapeutic options. Type I CRPS with its variable relationship to trauma has often no discernible fractures or nerve injuries and remains enigmatic in its response to conservative treatment as well as the other limited interventional therapies. Neuromodulation in the form of spinal cord and dorsal root ganglion stimulation (SCS, DRGS) has shown encouraging results, especially of causalgia or CRPS I of lower extremities. Upper extremity CRPS I is far more difficult.

Objective: To report a case of upper extremity CRPS I treated by wireless peripheral nerve stimulation (WPNS) for its unique features and minimally invasive technique. The system does not involve implantation of battery or its connections.

Case report: A 47 year old female patient presented with refractory CRPS I following a blunt trauma to her right forearm. As interventional treatment in the form of local anesthetics (Anesthesia of peripheral branches of radial nerve) and combined infusions of ketamine/lidocaine failed to provide any significant relief she opted for WPNS treatment. Based on the topographic distribution, two electrodes (Stimwave Leads: FR4A-RCV-A0 with tines, Generation 1 and FR4A-RCV-B0 with tines, Generation 1), were placed along the course of radial and median nerves under ultrasonography monitoring and guided by intraoperative stimulation. This procedure did not involve implantation of extension cables or the power source. At a frequency of 60 Hz and 300 μs the stimulation induced paresthesia along the distribution of the nerves. Therapeutic relief was observed with high frequency (HF) stimulation (HF 10 kHz/32 μs, 2.0 mA) reducing her pain from a visual analogue scale (VAS) score of 7–4 postoperatively. Three HF stimulations programs were provided at the time of discharge, as she improved in her sensory impairment to touch, pressure and temperature at her first follow up visit. At 5-months she was able to drive, did not require opioids and allodynia disappeared.

Conclusions: In a case with difficult CRPS I involving upper extremity, a minimally invasive WPNS of radial and median nerves provided good symptomatic relief. The procedure was tolerated well and both electrodes remained in place without any adverse events.

Implications: In view of the very limited options currently available to manage CRPS, WPNS can be a promising therapeutic modality.

Keywords: neuromodulation; peripheral nerve stimulation; wireless; complex regional pain syndrome; upper extremity.

1 Introduction

Complex regional pain syndrome (CRPS) is a conglomeration of symptoms and signs involving autonomic and somatic nerve functions, to a debilitating extent, with unknown pathophysiology. The International Association for the Study of Pain (IASP), introduced the nomenclature and diagnostic criteria refining the descriptive terminology into type I and type II based on the nerve injury. The type I, also known as reflex sympathetic dystrophy, is characterized by disproportionate pain, hyperalgesia, allodynia, swelling, discoloration of skin, abnormal sudomotor, vasomotor and motor functions following a noxious event without injury to the nerve/s defying the dermatomal distribution or any other condition that correlates with the degree of disability and suffering [1–3].

CRPS, however, is still a clinical diagnosis and has no specific tests to define its presentation even though findings on radiography, electrodiagnostic tests, thermography,

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sweat analysis and nerve blocks provide information to define the disorder. Some of these cases show improvement without interventions, further complicating the management paradigms, leading to delays in diagnosis as well as treatment [4]. Combination of physical and psychological therapy combined with medical as well as interventional management followed by SCS and peripheral nerve stimulation (PNS) provide an acceptable protocol in CRPS I although nonresponders and delayed interventions would demand an earlier interventional procedures to reduce the disability with improved outcomes [5, 6].

However, in practice, selection of cases for neuromodulation is complicated by the overlying psychopathology intertwined in the origins of CRPS along with predisposing factors like genetic factors, that may jeopardize the response [7] autoimmune antibody-mediated neuronal and vascular tissue damage [8], autonomic nervous system and adrenergic receptor perturbations [8], psychological sequelae [9], and financial and socioeconomic implications secondary to both morbidity and disability from this syndrome [10]. CRPS with an incidence of 5.4–26.2 per 100,000 patient-years is estimated to affect 16,000–78,000 people every year with profound impact on quality of life [10–12]).

On the other hand, traditional SCS as a tertiary option in the management of CRPS lacks data on its effectiveness and mechanisms of efficacy [13].

2 Case illustration

A 47 year old female patient suffered an accident by falling on her hand, 6 years ago. There were no fractures of the long bones but she developed CRPS of her right hand in the form of weakness of right hand associated with tingling and numbness on her hand and forearm. She was extremely sensitive to pressure on forearm and complained of colder extremity.

On examination, she had sensory impairment along the distribution of right radial and median nerves associated with motor deficit of right hand muscles. Kapandji score for the thumb opposition was 4.

There was impaired temperature sensation along with slight vivid discoloration of right hand, temperature differences between right and left hands were noted; right hand being cold compared to left. Along the right median nerve, area of allodynia could be mapped.

She received multiple treatment regimens in the form of regional anesthesia blocks, infusion therapy with lidocaine and ketamine, supplemented with pain medication including opioids, gabapentin and ketamine nasal spray. Right radial nerve block was positive- this was performed to see if a neuromodulation device like a peripheral nerve stimulator could reduce pain in the affected area, which was the case; landmark injection in the area of radial nerve with ropivacaine (0.75%; about 10 mL).

She opted for the minimally invasive wireless PNS as the pain remained resistant to all these interventions and medications. There was no reported nerve damage and hence nerve conduction studies were not performed.

3 Operative procedure

An informed consent was obtained prior to the operation, detailing the possible outcome including risks and complications. After thorough antiseptic preparation, the right forearm was draped and using aseptic precautions, intraoperative ultrasonographic (USG) recordings were obtained to map out the median and radial nerves on the anterior aspect (Fig. 1). A small 3–4 cm wide incision was given on the anterior surface of the forearm two fingerbreadths below the cubital fossa to facilitate introduction of the two Stimwave (Stimwave Technologies, Fort Lauderdale, FL, USA) electrodes (Leads: FR4A-RCV-A0 with tines, Generation 1 and FR4A-RCV-B0 with tines, Generation 1). The USG monitoring was continued during the procedure to keep the leads parallel to the peripheral nerves (Fig. 2). Intraoperative stimulation at a low frequency of 60 Hz and 300 µs induced paresthesia along the distribution of the nerves.

The position of the electrodes was confirmed by intraoperative radiographs (Fig. 3) and a stimulation protocol (HF 10 kHz/32 µs, 2.0 mA) provided therapeutic relief postoperatively. As paresthesia continued at low frequency,

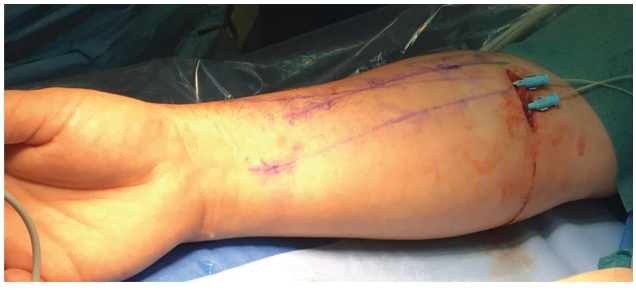

Figure 1: Intraoperative ultrasound guidance mapped out the location of the radial and median nerves on the anterior aspect of the forearm. Under local anesthesia (Lidocaine 1%; approx. 10 mL), two electrodes were introduced along the course of these nerves.

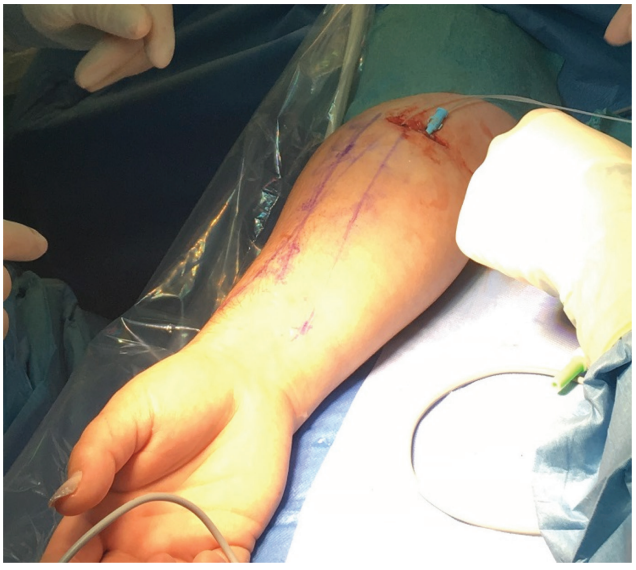

Figure 2: Following confirmation with intraoperative stimulation, the electrodes were secured in the forearm.

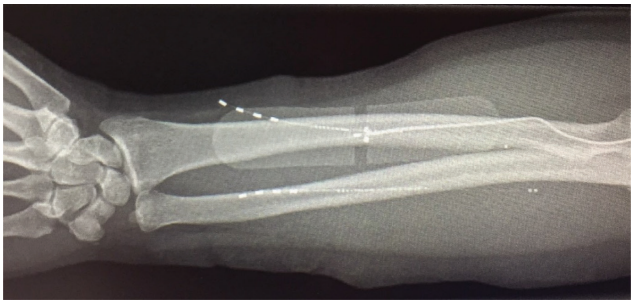

Figure 3: Final location of the electrodes as confirmed by intraoperative radiography. Electrode along the radial nerve could be seen along the ulna and the medical nerve electrode on the radius.

three HF programs were initiated at the time of the discharge (Figs. 4–6).

Postoperative course and evaluation: patient tolerated the procedure well. Visual analogue scale (VAS) score came down to 5–6 from a preoperative score of 6–7 soon after implantation. After a week, the VAS score further reduced to four keeping the patient happy about the pain relief. Prior to the implantation of the wireless stimulation device, she was very sensitive to pressure on forearm while this suffering went away after surgery. She could tolerate both touch and pressure while stimulation is on. Without stimulation, however, pain returned in approximately 1 h. During the follow up she had shown consistent improvement in all her symptoms. At 5 months there was significant reduction in pain (VAS < 4) along with improvement of motor skills. Allodynia disappeared and the patient was able to drive a car. Her medications were reduced and no more opioids were needed.

4 Discussion

CRPS is challenging in its presentation, course and response to treatment due to its unknown pathophysiology, which remains empirical at its best [14]. Early diagnosis and treatment protocol that includes education, physical, pharmacological and interventional options could be beneficial in providing better relief [15–17]. However, there is significant delay in making a diagnosis, nearly 6 months, contributing to unfavorable recovery [18]. Interventions to control pain in the form of nerve blocks followed by neuromodulation, could encourage the patients for active rehabilitation to restore limb movements and motor function [19, 20].

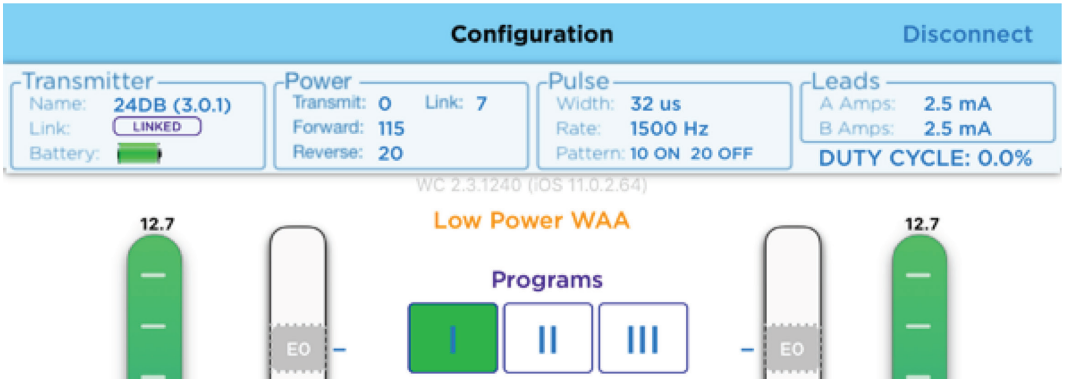

Figure 4: Program I.

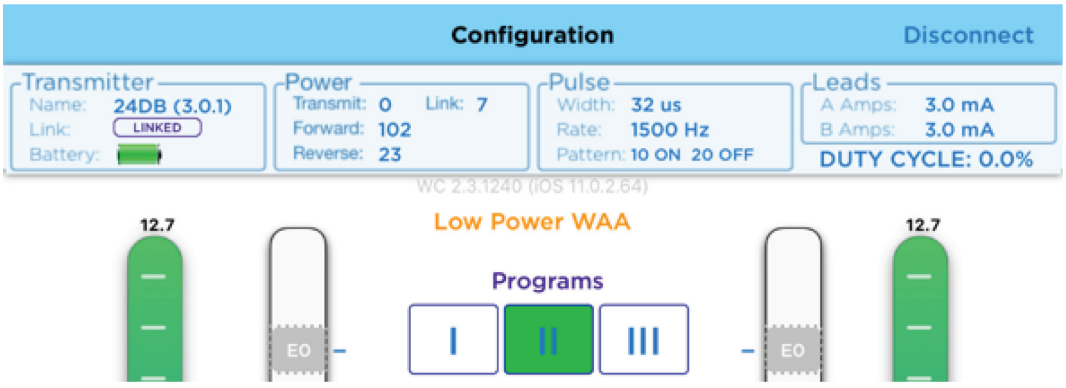

Figure 5: Program II.

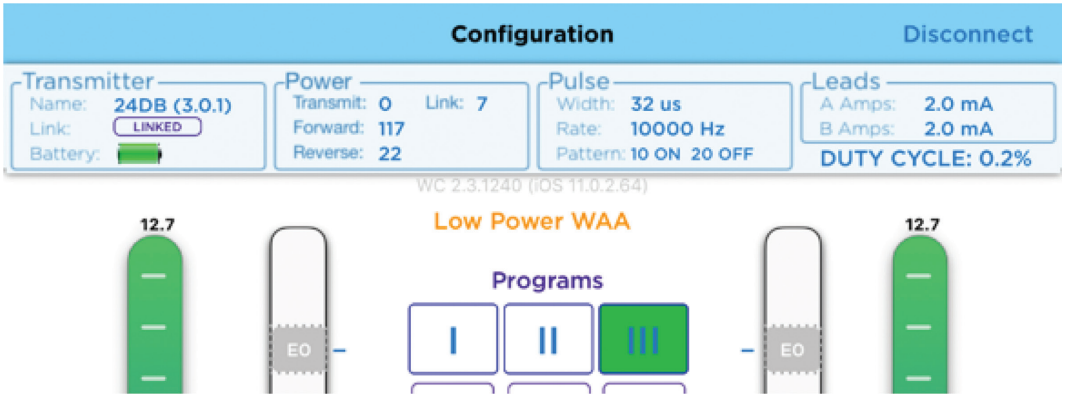

Figure 6: Program III.

However, there are only a limited number of neuromodulation devices currently applicable to these patients and PNS devices are yet to be approved by the United States Food and Drug Administration (FDA) for CRPS [21]. Additionally, upper extremity CRPS is extremely difficult to approach due to the anatomical location of the postganglionic sympathetic supply from the second and third thoracic sympathetic ganglia and the nerve of Kuntz. As many as 60% of CRPS might be having active participation of the fibers via the nerve of Kuntz, thus requiring stimulation of the cervical and the thoracic sympathetic fibers as well [22, 23].

SCS, although effective neuromodulation in pain management, fails to reach the target areas in the extremities and has set backs from positional changes in the stimulation apparatus [24]. DRGS, on the other hand, is more consistent in pain relief and is devoid of positional changes due to the location of the electrodes in the spinal canal with longer lasting stimulation advantages also [24, 25]. Earlier reports have demonstrated that SCS in combination with physical therapy (PT) was more effective than PT alone [26] and DRGS also yielded matching results though not better [5, 27].

Five year follow up results have shown that SCS combined with PT did not produce superior results compared to PT alone after 2 years [28]. Significant relapses with SCS over a period of time prompted other alternative treatment options for CRPS like “stim vacation” with only limited positive response [29].

Additionally, Visnjevac et al. in 2017 reported that 46% of CRPS patients with SCS required implantable power generator (IPG) replacement over 5 years and 25% had revision of electrodes for lead migrations in 46%. IPG site pain and discomfort necessitated revisions in 33% [13]. Although DRGS provided better relief [30], the equipment related complications remain owing to the implantable nature of the battery and the extension wires from the electrodes. In 36.8% patients treated with DRGS device related adverse events occurred and 46% procedural complications were reported in A Safety and Effectiveness Trial of Spinal Cord Stimulation of the Dorsal Root Ganglion for Chronic Lower Limb Pain (ACCURATE) study for

CRPS. Most of them were due to IPG surgery while lead migration in 10.8% resulted in loss of stimulation [31].

Wireless PNS (WPNS) technology: as described in the case illustration, the WPNS requires implantation of the electrodes with in-built receiver and is devoid of implantable power source or the additional surgical procedures. Noteworthy is the fact that the procedure is minimally invasive and performed under local anesthesia since there is no requirement for additional incisions to implant the extension wires or the IPG which would be located in the trunk, away from the extremity. For this anatomical lesion, WPNS is best suited for management of pain involving the upper or lower extremities. Additionally, in cases where the results are not as expected, revision procedures if required, would be far less invasive and explantation of the device would be much simpler compared to the traditional SCS or DRGS equipment. Miniature implantation devices with wireless access are promising since they are less invasive and do not require the power generators to be implanted and anchored to the connecting cables. Thus they also avoid the tethering effects of the cables [32]. The implant and device used in the present case belong to an advanced version of wireless technology, fully programmable from zero to 10,000 Hz. It met with all the demands of the present case and relieved that patient from the disabling pain, allodynia and weakness to bring her back to normal activities of daily living.

WPNS had demonstrated safety and feasibility in earlier reports and wireless technology yielded results comparable to the traditional implantable SCS equipment in several previous reports [33, 34] Further larger scale studies in multicenter randomized patient populations are required to establish the consistency and durability of this approach.

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