Educational case report

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A case report of wireless peripheral nerve stimulation for complex regional pain syndrome type-I of the upper extremity: 1 year follow up

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Abstract

Background: Complex regional pain syndrome (CRPS) is a chronic disabling painful disorder with limited options to achieve therapeutic relief. CRPS type I which follows trauma, may not show obvious damage to the nervous structures and remains dubious in its pathophysiology and also its response to conservative treatment or interventional pain management is elusive. Spinal cord and dorsal root ganglion stimulation (SCS, DRGS) provide good relief, mainly for causalgia or CRPS I of lower extremities but not very encouraging for upper extremity CRPS I. we reported earlier, a case of CRPS I of right arm treated successfully by wireless peripheral nerve stimulation (WPNS) with short term follow up. Here we present 1-year follow-up of this patient.

Objective: To present the first case of WPNS for CRPS I with a year follow up. The patient had minimally invasive peripheral nerve stimulation (PNS), without implantable pulse generator (IPG) or its accessories.

Case report: This was a case of refractory CRPS I after blunt trauma to the right forearm of a young female. She underwent placement of two Stimwave electrodes (Leads: FR4A-RCV-A0 with tines, Generation 1 and FR4A-RCV-B0 with tines, Generation 1) in her forearm under intraoperative electrophysiological and ultrasound guidance along radial and median nerves. This WPNS required no IPG. At high frequency (HF) stimulation (HF 10 kHz/32 μs, 2.0 mA), patient had shown remarkable relief in pain, allodynia and temperature impairment. At 5 months she started driving without opioid consumption, while allodynia disappeared. At 1 year follow up she was relieved of pain [visual analogue scale (VAS) score of 4 from 7] and Kapanji Index (Score) improved to 7–8. Both hands look similar in color and temperature. She never made unscheduled visits to the clinic or visited emergency room for any complications related to the WPNS.

Conclusions: CRPS I involving upper extremity remain difficult to manage with conventional SCS or DRGS because of equipment related adverse events. Minimally invasive WPNS in this case had shown consistent relief without any complications or side effects related to the wireless technology or the technique at the end of 1 year.

Implications: This is the first case illustration of WPNS for CRPS I, successfully treated and followed up for 1 year.

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

1 Introduction

According to the International Association for the Study of Pain (IASP), complex regional pain syndrome (CRPS) type I, a reflex sympathetic dystrophy, is defined by hyperalgesia, discoloration of skin, allodynia, abnormal sudomotor/vasomotor/motor functions and swelling of the extremity secondary to a noxious event without anatomical nerve damage, while the distribution of the symptomatology defies dermatomal topography or the degree of disability or suffering [1–3]. The exact etiology of CRPS still remains unknown and it is essentially a clinical diagnosis without any specific laboratory tests to clinch the entity. However findings on radiographic, electrophysiological and diagnostic nerve blocks provide some information pointing towards this disease [4].

As a result of enigmatic pathophysiology presenting with mixed somatic and autonomic symptoms, management of CRPS requires combination of medical, psychological, interventional and neuromodulation methods like spinal cord stimulation (SCS) and peripheral nerve stimulation (PNS); often recommending earlier intervention to

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reduce disability [5, 6]. Even then, patient selection for either SCS or PNS is not simple because of the immunological, neurological, psychological and genetic factors engaged in the etio-pathogenesis of CRPS [7–10].

Additionally, there is paucity of data on the long term effectiveness of traditional SCS in the treatment of CRPS [11, 12].

Hewitt NA and Cox P reported the hepatotoxicity of long term Ketamine infusion in CRPS [13].

We reported our experience with wireless PNS in the management of CRPS type I of the upper extremity, earlier [14]. The following is the outcome of this patient at 1 year follow up.

2 Case illustration

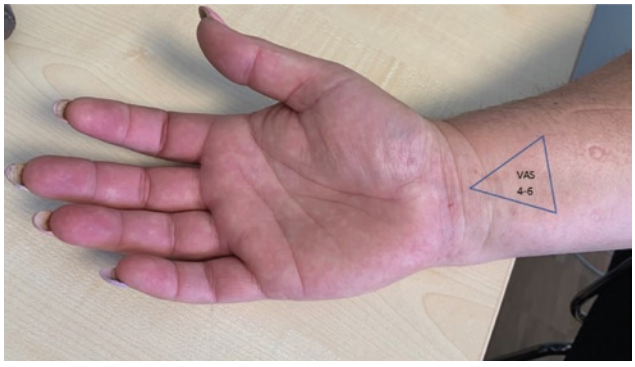
Earlier we described the presentation of this CRPS type I, in detail. This young female patient presented with relentless progression of symptoms following blunt trauma to her right hand 7 years back. She had sensory and motor impairment of her right hand along with impaired temperature sensations and vivid discoloration of hand. Preoperative Kapandji score was 4 and visual analogue scale (VAS) score was 7. Following failed conservative medical management, nerve blocks, ketamine infusion therapy and opioids she underwent wireless neuromodulation treatment.

Surgical treatment: After obtaining informed written consent patient underwent placement of Stimwave leads (Stimwave Technologies, Fort Lauderdale, FL, USA) Two stimulating implantable electrodes (Leads: FR4A-RCV-A0 with tines, Generation 1 and FR4A-RCV-B0 with tines, Generation 1) were placed under ultrasonographic (USG) monitoring along median and radial nerve on the volar aspect of right forearm. At 60 Hz and 300 μs, intraoperative stimulation induced paresthesia along the nerve distribution. Once intraoperative radiography confirmed appropriate positioning, a stimulation protocol (high frequency [HF] 10 kHz/32 μs, 2.0 mA) was initiated for best therapeutic relief.

Soon after implantation, VAS score came down to 4 and her sensory symptoms improved. After 5 months, allodynia disappeared, allowing her to drive a car while opioid consumption was no longer required.

1-year follow-up:

She was examined after 1 year in detail. During this 1 year she never made any emergency calls or visits to the emergency room. Pain reduced to VAS score of 4, Kapandji Index improved to 7–8 with better movements of fingers. Color of the hand returned to normal with normal temperature sensation. There was a small residual painful area (Photograph 1) proximal to the electrodes on palmar aspect of the forearm (2×4 cm). Occasionally she used Ketamine nasal spray (5 mg/hub).



She used three programs during this time. With Program 1, she kept low power transmission with power index 24 (Fig. 1). Program II delivered the most comfortable relief at 1.5 kHz (Fig. 2), 3.0 mA and Program III with 10 kHz, 2.0 mA was intermittently applied (Fig. 3). She used stimulation at different times during the day depending upon the intensity of pain; mostly either during driving or in the night. The most acceptable component of the wireless peripheral nerve stimulation (WPNS) to her was the peripheral band, the wearable antenna, with a longer than usual (100 cm) cable that she can pull out through the sleeve, while the transmitter remained in the pocket. A stimulation with the StimPod (TM) for approx. five minutes at 8mAmp 2 Hz showed a significant reduction in the painful area for about 3 days.

At present, after 15 months, patient came with improved symptomatology and functional abilities. She was able to drive her car and perform daily activities without much impediment. However, a small area (about 2 cm×4 cm) of allodynia close to her wrist on the palmar aspect, unresponsive to stimulation. This required pregabalin (200 mg twice a day) and oxycontin + naloxone (twice a day) with intermittent Ketamine nasal spray on demand.

Follow up radiographs of the forearm and hand were obtained to verify the location of the implanted electrodes (Figs. 4 and 5). Both anterior-posterior (Fig. 4) and lateral (Fig. 5) views confirmed appropriate placement of two electrodes along the median and radial nerves.

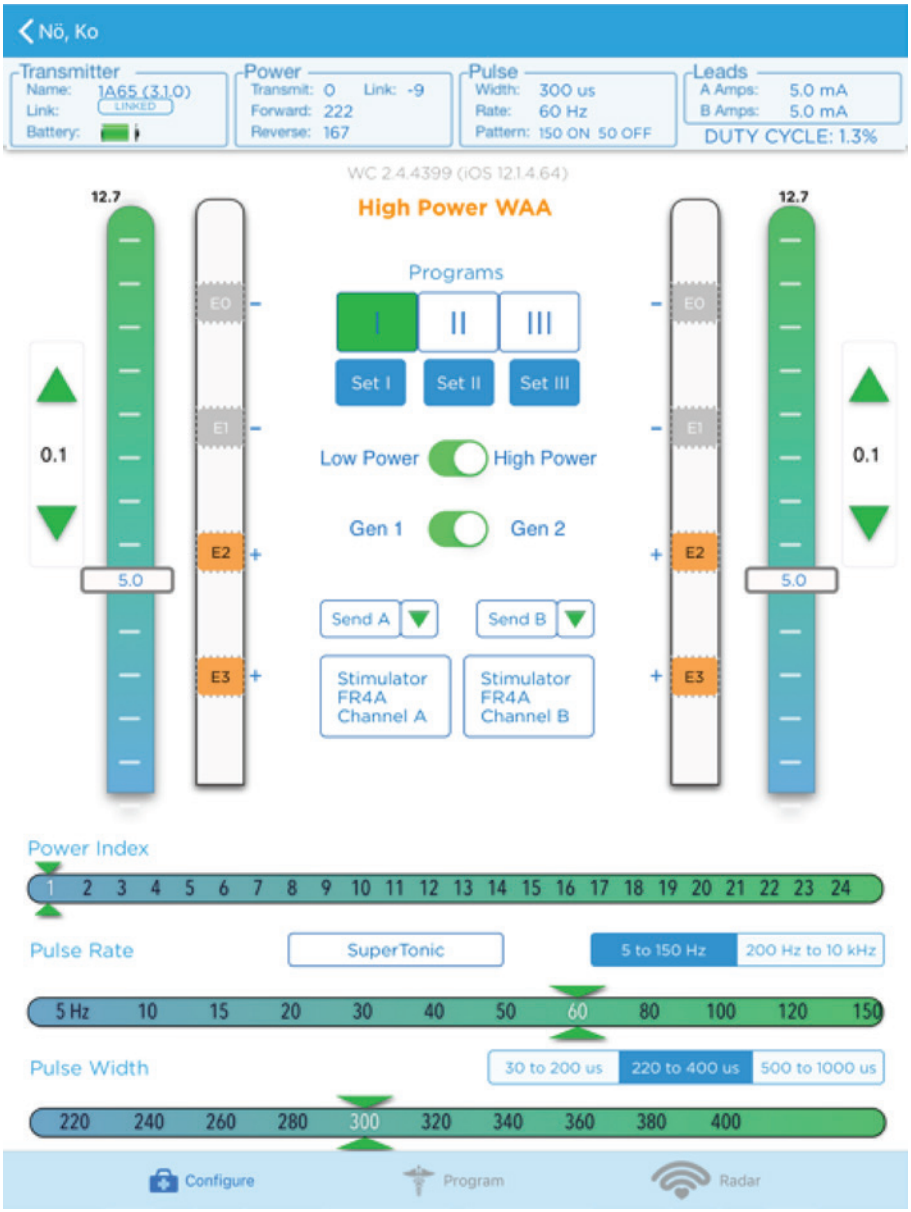


Fig. 1: Stimulation protocol-Program I.

3 Discussion

CRPS is an extremely disabling, refractory painful disorder affecting 16,000–78,000 people each year [10, 15, 16].

It has a challenging presentation for early diagnosis and effective treatment since the etiology remains obscure [4, 17]. Protocol for effective treatment includes patient education, physical therapy (PT) and interventional management [18–20], which was negatively influenced by diagnostic delay [21]. The goal of early neuromodulation would be restoration of motor function of the involved extremities for active rehabilitation [22, 23].

Upper extremity CRPS is particularly difficult to manage because of the anatomy of the sympathetic chain: the postganglionic fibers coming from the 2nd and 3rd sympathetic ganglia and the nerve of Kuntz, which requires SCS at both cervical and thoracic levels [24, 25]. In addition to this anatomical complexity, positional changes dampen the efficacy of traditional SCS [26].

Both SCS and dorsal root ganglion stimulation (DRGS) demonstrated efficacy when combined with PT in the initial follow up [5, 27, 28]. However, 5-year follow up results did not show better outcome with SCS and PT compared to PT alone; a complication rate of 38% was

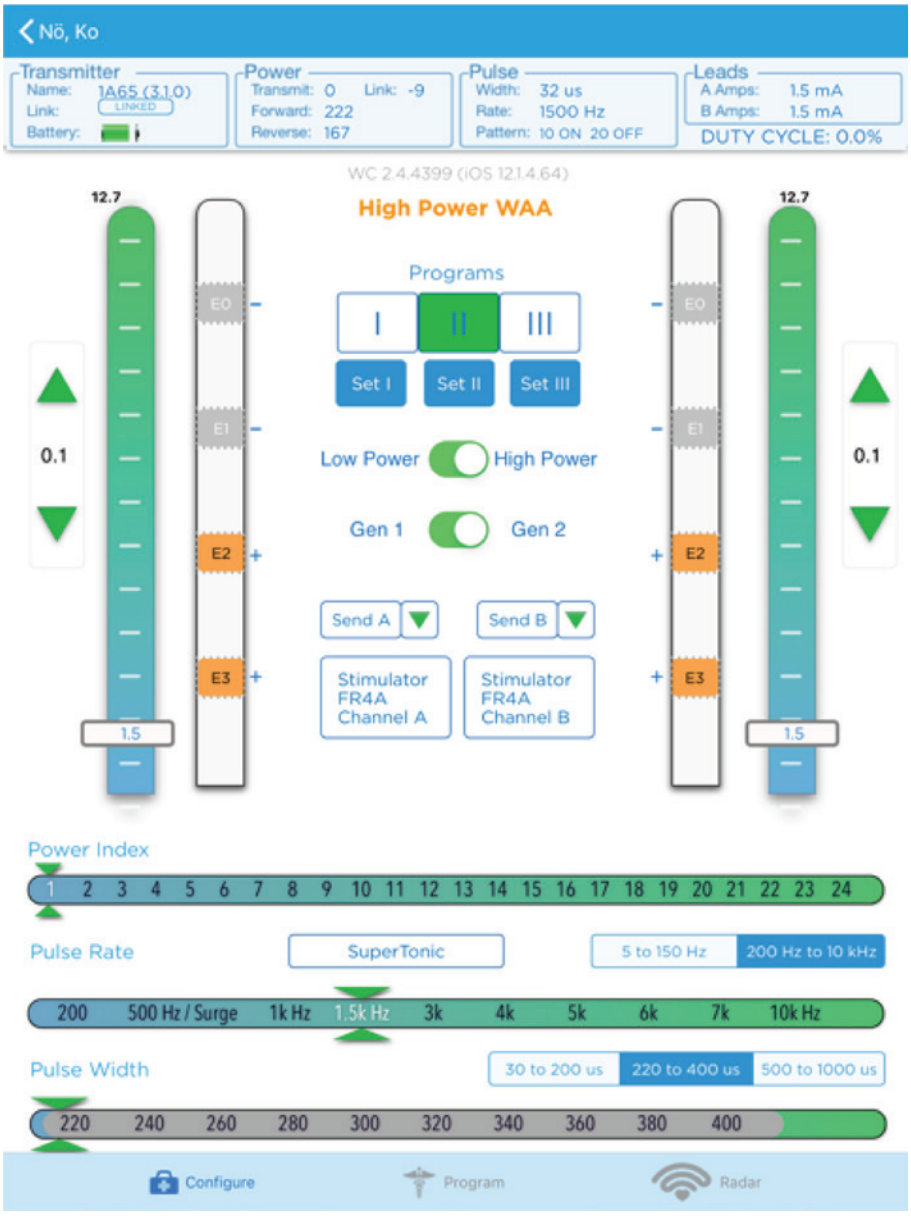


Fig. 2: Stimulation protocol-Program II.

reported with SCS after 2 years [29] while 9/24 patients underwent reoperation. Authors concluded that SCS "did not produce durable and statistically significant improvements in the pain from CRPS-I" [29].

Some authors proposed "stim vacation" to improve the outcomes but had very little success in reducing CRPS relapsing during SCS therapy [12].

Traditional SCS was also reported to have additional adverse events and revision surgeries due to implantable power generator (IPG) related adverse events and failures in 46% of CRPS cases. Revision of electrodes was needed in 25%, lead migrations in 46% and IPG pain indicated surgery for replacement in 33% [11]. DRGS also had similar number of adverse events and additional surgeries [30].

WPNS, on the other hand, requires implantation of the electrodes only without IPG. In a complicated disease like CRPS of upper extremity, WPNS is best suited since anatomically the leads could be implanted in close proximity to the peripheral nerves with ease of revision, if required. The wireless device has no tethering due to IPG or its accessories and additional anchors [31]. There have been successful case reports on wireless neuromodulation with short term follow up [14, 32, 33].

The present case illustrated the simplicity of the implant and good relief achieved with a novel wireless

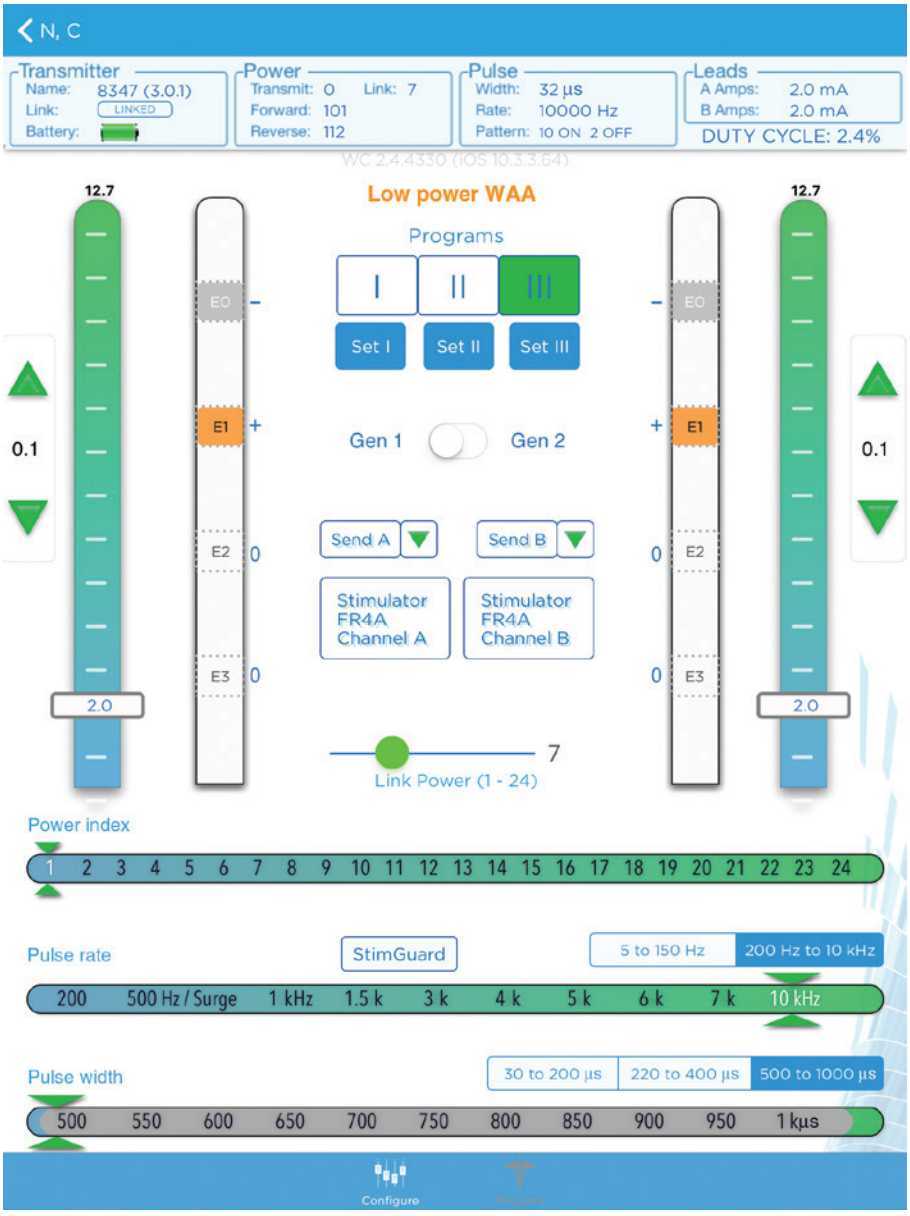


Fig. 3: Stimulation protocol-Program III.

Fig. 4: Anterior-posterior X-ray of the forearm demonstrating the location of the implanted electrodes *in-situ*.

Fig. 5: Lateral radiograph of the forearm disclosing the position of the implants at the required anatomical site.

technology in PNS. After the implantation patient had the option to use three programs viz. 60 Hz, 10 kHz, and 1.5 kHz during the trial phase. She perceived comfort at 10 kHz frequency during the initial period. However, at the subsequent sessions for programing, according to her, the best result was provided by 1.5 kHz stimulation for pain relief. At present, she has 1.5 kHz program along with another LF 60 Hz program available for use (which she uses mainly for checking the system functionality but not for therapy).

The patient had no adverse events or complications related to the technique or the technology and was tolerated very well by the patient. She never made any emergency call or any unscheduled visits to the emergency room or the clinic. At the end of 1 year and 3 months, she remains relieved of the disabling symptoms of CRPS, leading normal life, driving her car. Patient also enjoys the simplicity and flexibility of the frequencies available for pain relief.

Abbreviations: ACCURATE, a safety and effectiveness trial of spinal cord stimulation of the dorsal root ganglion for chronic lower limb pain; CRPS, complex regional pain syndrome; DRGS, dorsal root ganglion stimulation; FDA, Food and Drug Administration; HF, high frequency; IASP, International Association for the Study of Pain; IPG, implantable power generator; PNS, peripheral nerve stimulation; PT, physical therapy; SCS, spinal cord stimulation; VAS, Visual Analogue Scale; WPNS, wireless peripheral nerve stimulation.

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