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CASE REPORT

Case Report: Rescue of Relapsed Pain in a Patient with Complex Regional Pain Syndrome Type II by Adding Another Dorsal Root Ganglion Lead

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Abstract: We present on a patient with complex regional pain syndrome (CRPS) following ankle surgery. Pain was refractory to both conservative and surgical measures including neurotomies, ankle fusion, hardware removal, and spinal cord stimulation (SCS) trial. A dorsal root ganglion (DRG) stimulation trial with lead placements at L4, L5, and S1 provided significant pain and functional improvement. However, during the implantation, we were able to place only two DRG leads at L4 and L5 and not S1 due to difficulties with advancing the lead to the desired location. Nonetheless, the two DRG leads provided 90% pain relief and 75% functional improvement for 9 months. However, the patient experienced pain symptoms similar to that of pre-implant without a clear trigger after 9 months despite no DRG stimulator hardware malfunction or lead migration. A decision was made to re-try implanting the S1 DRG lead, which was successful and provided significant pain relief.

Keywords: DRG-stimulation, loss of efficacy, CRPS

Introduction

CRPS describes a painful condition that is associated with motor, sensory, skin, and autonomic changes such as mild to severe burning or throbbing pain with accompanying changes in skin color, temperature, and/or swelling with weakness of the affected limb.[1] The risk factors for CRPS are limb injury, prolonged immobilization, or surgery resulting in inflammatory or immune reaction in the central and peripheral nervous systems. The incidence of CRPS after an ankle trauma is as high as 15%.[2] Treatment options for CRPS have varying efficacy.[3] Neuromodulation has proven to be beneficial for providing pain relief and functional improvement in cases of treatment refractory CRPS.[4] A potential mechanism of action includes inhibition of painful stimuli at the dorsal horn neurons via continuous stimulation of Aβ fibers in the dorsal columns or DRG. Habituation and loss of efficacy from neuromodulation are common.[5] Additionally, pain generating pathways originating from a single DRG can influence dermatomal distributions above and below, suggesting a single dorsal horn ganglion stimulation (DRG-S) lead may impact several spinal levels.[6–8] In this case report, we describe relapsed pain refractory to DRG-S that was rescued with a new DRG lead placement in a level below the previous leads in a CRPS patient.

Case Presentation

A 39-year-old man presented to our institution for chronic right foot pain following right ankle fracture and subsequent surgeries. He had CRPS-like symptoms involving the right lower extremity on the dorsum aspect of the foot from the ankle to the toes. The pain was described as a constant stabbing quality and rated a 9/10 on the visual analog scale (VAS). He reported vasomotor symptoms of asymmetrical warmness and skin color changes along with trophic symptom of nail pitting. The pain had been refractory to open reduction and internal fixation, ankle fusion, hardware removal, US guided nerve blocks at the common peroneal nerve and sciatic nerve, deep and superficial peroneal neurectomies, stem cell

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injections, neuropathic pain medications, and opioids. The physical exam revealed edema and erythema of the right foot and trophic changes in the right first toenail compared to the left. A thermoregulatory sweat test showed anhidrosis in the right ankle and foot. Electromyography demonstrated electrodiagnostic evidence of right peroneal mononeuropathy at a level between the branch to the peroneus longus and the branch to the peroneus tertius. He was diagnosed with CRPS type II. We initially proceeded with a Medtronic SCS trial because the patient's insurance had denied DRG-S. Two eight contact Medtronic SCS trial leads were placed at the bottom of T11 and top of T12 (Figure 1). However, SCS failed to achieve greater than 50% pain relief and 50% functional improvement. He did not proceed to implant.

After two years, the patient's insurance approved a DRG-S trial. The DRG-S trial leads were placed at L4, L5, and S1 (Figure 1). During the 7-day trial period, the patient reported 95% pain relief and rated the pain a 0/10. His sleep increased from 4 hours per night to 8 hours per night, and his ability to sit for work without pain interference improved from 3 hours to 9 hours. During the trial, he was able to walk greater than 2 miles per day. During implantation, we were able place the L4 and L5 leads, but not the S1 lead due to difficulties with advancing the lead from the S1 foremen to the desired location (Figure 1). Despite this issue, at subsequent follow-ups, the patient continued to report a full night of sleep without pain interference. He was able to sit for greater than 8.5 hours per day and walk for more than 20 minutes at a time. He was able to taper off Amitriptyline, Oxycodone, and Pregabalin with pain rating of 1/10.

At 9-month post-DRG-S implant, the patient reported 9/10 pain in the right foot in the same distributions to that of pre-DRG-S without any inciting event. He was restarted on Pregabalin without improvement. Six DRG-S reprogrammings/device interrogations were attempted without improvement. A lumbar spine X-ray was negative for lead migration or fracture. At this point, a decision was made to re-try implanting the DRG-S lead at S1, which was successful (Figure 1). At 1-month follow-up post S1 lead implantation, the patient reported significant pain relief with a pain rating

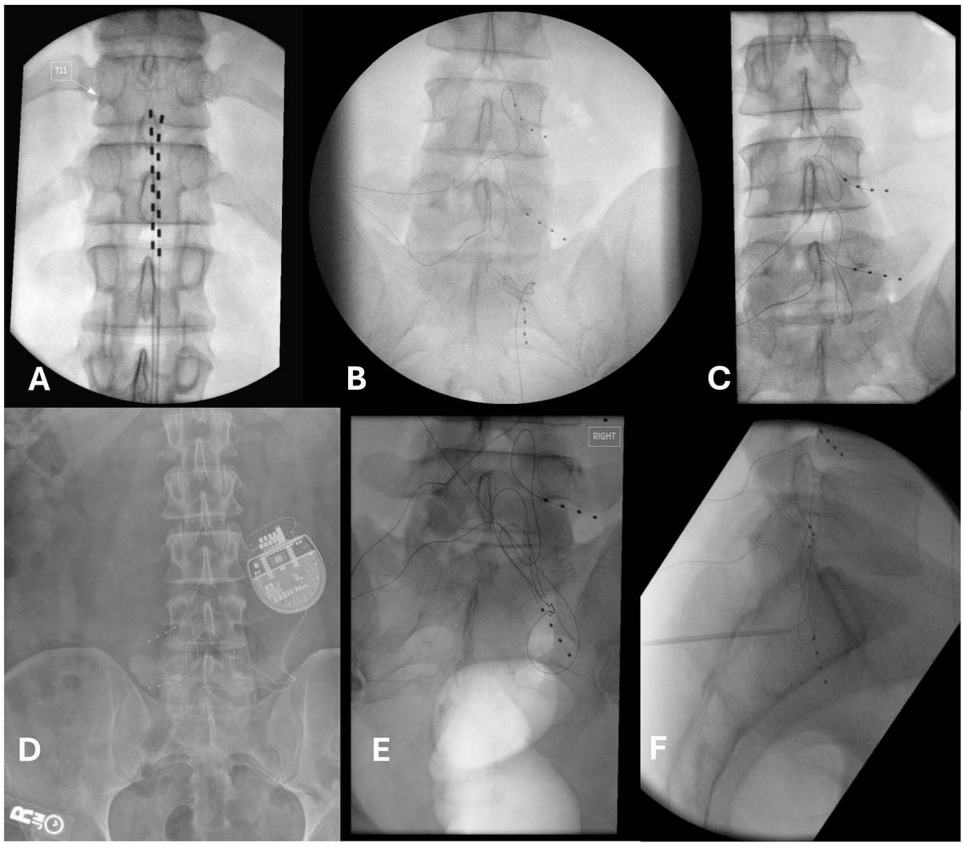

Figure 1 Fluoroscopy images of SCS and DRG stimulator lead placements. (**A**). SCS lead placements for the trial. (**B**). DRG lead placements for the trial. (**C**). DRG lead placements for the first implant. (**D**). Lumbar X-ray at 9 months after the first implantation. (**E**) and (**F**). Implantation of S1 lead.

Table 1 Time Course of Pain, Function, and Medications Outcomes with DRG-S Implant and Rescue

Time Frame	Pain Severity (VAS)	Functional Outcomes	Pain Medication(s)
Pre DRG-S	9/10	Sleep: 4 hrs/night Sitting: 3 hrs/day Walking: 3–4 min at one time	Amitriptyline, oxycodone, and pregabalin
Post DRG-S at L4 and L5	1/10	Sleep: 8 hrs/night Sitting: 9 hrs/day Walking: > 20 min at one time	none
LoE period	9/10	Sleep: 2–3 hrs before waking with pain	Pregabalin
S1 DRG-S rescue	2/10	Sleep: Improved Sitting: able to return to a desk job	Off pregabalin, started on oxycodone 10 q6 PRN by PCP for new back pain

of 2/10 on the VAS. He was able to return to work and was weaned off Pregabalin. At a 4-month follow-up visit, the patient continued to receive the same efficacy of pain relief and improvement of function (Table 1).

Discussion

CRPS is a painful and debilitating medical condition. It is unknown why some injuries progress to CRPS while others do not. In this case, the ankle fracture and immobilization with a cast are risk factors to the development of CRPS.[9] Research suggests that aberrant immune-mediate inflammatory response to the ankle fracture may play a role in peripheral and central sensitization and sympathetically mediated vasomotor symptoms.[9] When pain persists despite appropriate trial of conservative measures such as physical/occupational therapy, medications, injections, and even surgery, neuromodulation is considered. In the last ten years, there has been numerous observational studies[10–14] and one randomized control trial[4] demonstrating that DRG-S improved pain and function in refractory CRPS. How DRG-S treats CRPS pain is not well understood. It is possible that inhibition of proinflammatory mediators produced by injured microglia and alteration of neuronal firing frequency contribute to symptom relief experienced by patients.[15,16]

Herein, we present a case of a 39-year-old male with a diagnosis of right lower extremity CRPS type II involving the peroneal nerve, who had a successful DRG-S trial with three leads placement at the right L4, L5, and S1 DRGs. Placement of the DRG leads at these locations was based on reports that the peroneal nerve receives segmental innervation from the nerve roots of L4 to S1.[17] Additionally, placement of DRG leads at L4 and L5 or L4 to S1 are in line with reported studies of DRG-S for the treatment of lower extremity CRPS.[18] During the implant, we were only able to place the L4 and L5 leads. Despite this, at follow-ups, the patient experienced significant pain and functional improvement for 9 months. A possible explanation is that only the L4 and L5 nerve roots portion of the peroneal nerve was affected by the CRPS. Therefore, this patient did not need a DRG lead at S1. An alternative explanation is that L4, L5 and S1 were affected by the CRPS. However, there is dual modulation from L5 and S1 DRGs. DRG stimulation at L5 provided pain relief at S1, which has been shown in the literature.[18,19]

This case may be an example of the crosstalk phenomenon between different DRG-S. It has been proposed that pain signal originating from dermatomal levels above and below the lead placement may be impacted by a single lead.[20] The sympathetic efferent ganglia located in the paravertebral ganglia are interconnected to different spinal levels, prevertebral ganglia, and effector organs. Thus, inhibition of sympathetic nerve fibers at one level via DRG stimulation may exert an inhibitory effect at the level of stimulation or the levels above and/or below via propriospinal pathways.[20] Moreover, the DRG stimulation at one lead can trigger endogenous opioids release and inhibit pain signal at the dorsal horn by binding to presynaptic and postsynaptic opioid receptors at the levels above and below.[21] This may explain why implantation of the right L4 and L5 DRG leads were able to provide similar results as the trial with three leads at the right L4, L5, and S1 DRGs. When the patient developed pain in the same area at 9 months post-implant, adding the right S1 DRG lead was able to adequately control pain signals from all three levels. Loss of efficacy of L4 and L5 DRG stimulation was rescued

with stimulation of S1 DRG. It has been shown in other studies that a single S1 DRG lead was able to provide significant pain relief and functional restoration to patients with CRPS of the foot.[18,19]

This case report highlights some key findings in the ACCURATE trial.[4] First, our patient received inferior pain relief with SCS compared to DRG-S. With the SCS trial, the patient reported only 20% pain relief without any significant functional improvement. However, with the DRG-S trial and implant, the patient had greater than 90% pain relief and improved sleeping quality, sitting duration, and walking distance. Second, during the SCS trial, the patient complained of unpleasant paresthesia with positional changes. This is a common occurrence with SCS that has not been observed in DRG-S patients. Lastly, during the SCS trial, the patient reported that the SCS failed to reach the area of his pain. DRG-S overcame the SCS's limitations of selective targeting capabilities.

Conclusion

In summary, three DRG-S leads trial at L4 to S1 provided significant pain relief and functional improvement for a patient with right foot CRPS. During the implant, only two leads were placed at L4 and L5 due to difficulties advancing the S1 lead. The patient had significant pain relief and functional improvement for 9 months until he experienced relapsed of symptoms without evidence of lead migration or device failure. After six failed reprogrammings, we decided to re-try implanting the S1 lead since the DRG-S trial was with three lead placements and he had significant pain relief. Additionally, there are reports that a single S1 lead was able to restore functional and improved pain in patients with CRPS in the foot.[19] Fortunately, we were able to rescue loss of efficacy by adding the S1 lead.

Abbreviations

CRPS, complex regional pain syndrome; DRG, dorsal root ganglion; DRG-S, dorsal root ganglion stimulator; SCS, spinal cord stimulator; VAS, visual analog scale.

Data Sharing Statement

Data sharing is not applicable to this manuscript.

Consent for Publication

This manuscript adheres to CARES guidelines. Written informed consent was gathered by the patient described in this case report and can be provided upon request. Institutional approval was not required to publish the case details.

Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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Disclosure

The authors have no competing interests to report in this work.

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