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Dorsal Root Ganglion Stimulation in a Refractory Case of Complex Regional Pain Syndrome Types I and II in an Isolated Extremity

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Background: The treatment of complex regional pain syndrome (CRPS) is a difficult endeavor. The advent of neuromodulation interventions has led to new therapeutic options for chronic pain syndromes. Coinciding neuromodulation devices, such as spinal cord stimulation (SCS) and dorsal root ganglion stimulation (DRGS), may have a part in refractory CRPS.

Case Report: The authors present a case of a 50-year-old man status post a traumatic skiing accident 3 years prior to presentation with residual CRPS symptoms after an SCS implant. Additional multilevel coverage for refractory chronic neuropathic pain was achieved with the implantation of multiple DRGS devices.

Discussion: This case provides support for the use of DRGS in patients needing supplementary neuropathic pain coverage. Outcomes were reflected in diminished pain scores, an enriched quality of life, and enhanced functional capacity measures.

Key words: Complex regional pain syndrome, dorsal root ganglion stimulator, dorsal root ganglion stimulation, neuromodulation, spinal cord stimulator, spinal cord stimulation, chronic pain, case report

BACKGROUND

Complex regional pain syndrome (CRPS) treatment strategies have historically limited neuromodulation implementation to a single device. In convoluted case presentations, a multidevice approach may be of utility in targeting various pain generators. Dorsal root ganglion stimulation (DRGS) has a role to play in patients who were once thought to be precluded by an existing spinal cord stimulation (SCS) device (1,2). DRGS is a relatively novel modality for the treatment of refractory neuropathic pain. The device acts on the dorsal root ganglia of the spinal nerves, which contain cell bodies for primary sensory neurons that relay sensory information from a defined region of the body. DRGS leads are implanted percutaneously in contact with the DRG in the intraforaminal space, in conjunction with a pulse generator implanted in the body cavity that provides the signal (3). The exact mechanism of action of DRGS is unknown. Current hypotheses include feedforward inhibition and electrical stimulation leading to the correction of abnormal neuronal activity and hyperexcitability. This is accomplished by the alteration of ion channels in primary sensory neurons causing hyperpolarization of the T-junction and subsequent low-pass filtering of action potentials (3).

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SCS is elicited through the insertion of electrodes into the dorsal epidural space. Conventional SCS acts by increasing cholinergic stimulation and inducing the release of a variety of neurotransmitters (i.e., gamma-aminobutyric acid, adenosine, 5-hydroxytryptamine, etc), while decreasing glutamate release. This generates a modulation of pain signals (4). Because of the suspected mechanisms of action of DRGS and SCS, these modalities have been used in the treatment of neuropathic pain conditions, such as CRPS (1,5). Neurostimulation normalizes pain signals present in painful neuropathic conditions like CRPS, emerging as a potential mainstay in management (2). The authors present a case of a patient after a traumatic left lower extremity injury causing obstinate areas of leg pain. This case advocates for the utility of DRGS in a patient with incomplete, yet beneficial coverage with SCS alone.

CASE PRESENTATION

A 50-year-old male ski patroller presented to the interventional pain management clinic for left medial lower extremity neuropathic pain and left, deep medial knee pain after a traumatic work-related accident 3 years prior to presentation. While working on ski patrol, his ski dislodged, piercing the medial aspect of his left upper leg. The accident caused significant soft tissue damage and neurological injury resulting in surgical soft tissue repair. At his initial new patient visit to the clinic, he reported severe, aching, 6/10 neuropathic pain on the Numeric Rating Scale located in the left superficial knee, leg, heel, and foot. He further described a deep, aching pain in the left distal medial thigh in the territory of his knee. He had tissue texture, color, and temperature changes with allodynia in his left knee, leg, and foot. The patient reported he was unemployed and had difficulties completing activities of daily living independently due to severe sleep disturbances.

Prior to presentation to the pain clinic, the patient had undergone extensive medical management with imperfect, temporary results. His prior workup consisted of a nerve conduction study/electromyography (NCS/EMG) with no sensory responses in the left saphenous nerve and mild-to-moderate injury to the left obturator nerve. He subsequently underwent left saphenous neurolysis after 2 series of confirmatory blocks followed by the implantation of a peripheral nerve stimulator over the left saphenous and obturator nerves with no pain relief. Eventually, a SCS was placed 16 months prior with burst stimulation and lead coverage to the T9 level. SCS initially provided the patient with 60% pain relief of superficial lower leg and foot neuropathic pain. Unfortunately, this benefit was temporary due to suspected habituation despite attempts to reprogram the device and the deep medial knee pain remained refractory. An attempt was made to target the deep medial knee pain with left intraarticular knee injection and left genicular nerve radiofrequency ablation after confirmatory blocks. This provided little long-term effect much like the other trialed interventions prior to presenting to the current pain clinic.

On physical examination, the patient had a mildly antalgic gait with decreased stance phase on the left. He had mild medial joint line tenderness extending into the posterior horn of the medial meniscus. He was nontender at the tibial tuberosity with no significant patellar ballottement and a firm endpoint on Lachman's test, varus, and valgus stress tests. Updated imaging, including x-ray and magnetic resonance imaging of the left knee and lumbar spine, were grossly unrevealing for a new acute pathology. His patient health questionnaire-9 (PHQ-9) revealed a score of 20, and his patient health questionnaire-2 (PHQ-2) had a score of 5. Pain psychology evaluated the patient and reported that he had a pain disorder with anxiety and depression. Further analysis unveiled the patient had somatic preoccupation, perceived disability, medical reactive depression, dysthymia, and kinesiophobia.

The patient was diagnosed with CRPS type I (CRPS I) of the left medial knee in the setting of CRPS type II (CRPS II) in the left lower extremity and foot. Proper SCS T9 lead tip placement was confirmed on x-ray imaging. Due to the dampening of efficacy with the SCS covering the left superficial knee, and left medial leg from habituation, and his refractory deep knee pain, the patient elected to undergo DRGS. As a result, the patient underwent a DRGS trial targeting L3-L4 and L4-L5 DRGs with 50% relief of his total pain. After the trial, he had a mild residual area of pain in the bottom of the heel and lateral calf. He proceeded with a DRGS implant with leads placed at L3-L4, L4-L5, and an additional S1 lead to cover the aforementioned residual pain. The patient tolerated the procedure well without complications. After the DRGS implant, the patient reported near complete relief of his symptoms, especially relating to the medial aspect of his lower leg and foot. He reported decreased knee pain with enhanced ambulation distance. At the 6-week follow-up after reprogramming his DRGS and SCS, he reported marked relief in his overall

symptoms with an improved PHQ-9 of 16 and PHQ-2 of 4. At the 3-month follow-up, the patient indicated a 60% improvement in his pain with a warm sensation returning in his left knee. At the 10-month follow-up, he turned off his DRGS. In doing so, his prior pain returned and the efficacy of the DRGS was recognized by the patient. He elected to try ketamine infusions during the DRGS holiday with some relief in his symptoms. At the 21-month follow-up, he stated he was doing "better than ever" with a combination of DRGS, SCS, physical therapy, and scheduled ketamine infusions.

DISCUSSION

Delineating between CRPS I and CPRS II can be challenging diagnostically, especially in cases where the 2 types are superimposed in the same extremity. The approach in cases of arduous neuropathic pain conditions needs to be undertaken with ingenuity and caution. Given that CRPS is a diagnosis of exclusion, this case illustrates a linear, stepwise progression to exclude an assembly of possible central and peripheral pain generators. Though initially enigmatic in presentation, teasing out the 2 types of CRPS can be done systematically. If the etiology of the neuropathic pain is derived from peripheral nerve damage, as was the case for the patient's left lower extremity and foot pain in the setting of previously identified NCS/EMG findings, CRPS II can be attributed. Whereas, in CRPS I, nociceptive pain is found on the exam or supportive testing (5). The Budapest Criteria guidelines, adopted in 2004, unravel the differentiation between the 2 types. Early patient care for those with CRPS is critical for long-term symptom management (6). Ultimately, a swift, definitive diagnosis can be made through clinical judgment when putting together the patient's history, physical exam, and diagnostic tests.

A multidisciplinary treatment strategy is essential for those with implanted neuromodulatory devices. Close follow-up with neurostimulation industry teams is a prerequisite for reprogramming in sustaining pain relief. Other modalities, such as maintenance physical therapy, ketamine (7), and pain psychology, can provide lasting benefits to the patient. Ketamine infusions provide favorable pain control vs placebo for 12 weeks when used alone to manage pain associated with CRPS (8). Studies evaluating pain scores when using ketamine concurrently with a DRGS in CRPS have not been performed and would be a valuable area of research for future investigators.

Centralized pain syndrome refers to a neurological condition in which the central nervous system is implicated as the site of primary dysfunction and can explain how pain persists despite the lack of peripheral structural abnormalities (5,9). It is hypothesized that this occurs due to increased nociceptive afferent stimulation at the time of injury, causing persistent activity of primary nociceptive neurons. This postulate aids in educating those inflicted with persistent, unrelenting pain due to CRPS (9). Furthermore, it is critical to address breakthrough pain due to habituation from SCS, especially in patients with centralized pain syndromes (10). A 2-device approach in CRPS with SCS and DRGS has been displayed to decrease pain measures and improve function (11). This has been demonstrated previously, and in the case currently presented. The authors suspect that in our case, SCS and DRGS were working harmoniously in the left superficial knee, medial leg, and foot/heel. It is difficult to ascertain which device predominated the majority of pain coverage for the patient, though it is clear that he reported improvement in pain measures and a decrease in PHQ-2 and PHQ-9 scores following the DRGS placement that were not previously seen. Additionally, he had a near-complete return of symptoms after turning the DRGS off at the 10-month follow-up. An argument could be made for DRGS providing more benefit to the patient and suggests this intervention should be considered early in the treatment of CRPS moving forward.

The implementation of pain psychology principles and practices is synergistic when fused with the above pain management interventions (12). Coping strategies imparted with the assistance of an experienced pain management clinical psychologist are crucial to patient outcomes. However, research aimed at developing a standardized, systematic approach to psychological CRPS management is limited. The aim of all CRPS treatment is centered around the optimization of lifestyle and function, as a totality of pain relief from this approach may not be achieved.

CONCLUSIONS

This case exhibits a nonclassical presentation of CRPS I and CRPS II. Utilizing an amalgamation of DRGS and SCS may support augmentation of quality of life and functional outcomes measures in patients with contrarious CRPS. Furthermore, our case highlights a collaborative, interdisciplinary methodology for patient care. The distinctiveness of our patient presentation would be

rare to replicate in the future. In cases of general CRPS, DRGS should be considered as a treatment option early in the CRPS course. Although the bounds of current guidelines may not be altered, this case can serve as a future pillar in the art of ingenuitive treatment strategies using neuromodulation in refractory cases of CRPS.

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