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# NEUROMODULATION FOR TREATING ADOLESCENT COMPLEX REGIONAL PAIN SYNDROME: A CASE SERIES

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**Background:** Complex regional pain syndrome (CRPS) in adolescents is rare. When resistant to conservative treatment, therapeutic options are limited. One option is neuromodulation, but the evidence for its effectiveness is scarce.

**Case Reports:** We report 3 adolescent girls with refractory CRPS Type I who were successfully treated with neuromodulation. Two of them underwent dorsal root ganglion stimulation; the other, spinal cord stimulation. All 3 patients achieved > 50% pain reduction, restoration of function, and improved quality of life after failing multimodal conservative and minimally invasive injection management. Device-related complications, including lead migration and implantable pulse generator site pain, occurred in one patient but did not compromise her outcome.

**Conclusions:** Neuromodulation with dorsal root ganglion stimulation or spinal cord stimulation appears to be a safe and effective option for adolescents with refractory CRPS pain, improving pain and function when conservative therapies had failed.

**Key words:** Complex Regional Pain Syndrome, spinal cord stimulation, dorsal root ganglion stimulation, pediatric pain, pediatric neuromodulation, neuromodulation

## BACKGROUND

Complex Regional Pain Syndrome is characterized by disproportionate pain, sensory abnormalities, and autonomic dysfunction (1,2). The literature regarding pediatric patients who have CRPS and its treatment is limited to small retrospective studies; no large-scale randomized controlled trials have been conducted (1). The prevalence of CRPS in children has not been studied in the United States, however, in Scotland the prevalence has been reported to be 1.16 per 100,000 (2). In the literature, the mean age of presentation is 12.5 years; girls are affected more than boys. In 71% of these patients there is a clear preceding limb trauma with the lower limbs being affected in 76% of them. In 15% of these patients, a secondary site was reported with 5% ipsilateral to the primary injury site and 10% contralateral (3).

Since CRPS is a diagnosis of exclusion, the Budapest Criteria was created to reduce diagnostic ambiguity and improve accuracy in research studies (3). The pathogenesis of CRPS is also not well understood. It is believed there is a complex interaction between genetic susceptibility and an excessive inflammatory response to injury, followed by central nervous system sensitization (3).

The currently accepted treatments for CRPS include a multidisciplinary and multimodal approach starting with physical therapy, occupational therapy, and cognitive behavioral therapy. Medication options

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include gabapentinoids, N-Methyl-D-aspartic acid (NMDA) antagonists, antidepressants, antiepileptics, α-2 agonists, and both oral and intravenous analgesics. Minimally invasive options include sympathetic blocks, epidural catheter injections/infusions, regional anesthesia, surgical sympathectomy, spinal cord stimulation (SCS), and dorsal root ganglion (DRG) stimulation. Regarding SCS and DRG stimulation, there are no randomized controlled trials evaluating their efficacy and safety in pediatric patients, so their use in pediatrics is based on the results of studies in the adult population (2). The reports of neuromodulation being used for CRPS in pediatric patients is scarce; evidence is limited to case reports and small series. Here, we present 3 adolescent girls who were successfully treated with either SCS or DRG stimulation. Due to the retrospective nature of this chart review, informed consent was not obtained.

## CASE PRESENTATIONS

### Case One

A 14-year-old girl presented 6 months after a basketball-related injury to her left foot and ankle that occurred when she was 13 years old. Prior to the initial consult, her work-up included magnetic resonance imaging of the foot and ankle that showed no fracture or significant soft tissue injury. She was undergoing physical therapy and desensitization therapy. She reported severe neuropathic pain; allodynia; temperature and color changes; edema; and motor dysfunction, thus meeting the Budapest Criteria for CRPS Type I.

She underwent a lumbar sympathetic block but had minimal pain relief. Given her persistent, debilitating symptoms and the family's preference to avoid systemic medications, she underwent a DRG stimulation trial at left L4 and S1, resulting in a > 90% pain relief and restoration of function. This was followed by implanting a permanent DRG stimulator. This maintained excellent pain control and allowed her to return to normal activities including sports (golf, basketball) and showing animals. Over the subsequent year, she had 2 episodes of device-related complications: first, migration of the S1 lead and pain at the implantable pulse generator (IPG) site that required surgical revision and anchoring of the S1 lead. She later developed pain and allodynia over the revised IPG site after minor trauma which self-resolved. Throughout, her CRPS symptoms remained well controlled with DRG stimulation.

### Case 2

A 13-year-old girl developed CRPS Type I of the left lower extremity following a ballet-related hyperflexion injury. Her initial presentation was characterized by persistent left foot pain; swelling; color and temperature changes; and progressive functional decline, thus meeting Budapest Criteria for CRPS Type I. Her conservative management included physical therapy, desensitization, multiple medication trials (gabapentin, pregabalin, amitriptyline, low-dose naltrexone, ibuprofen), and topical therapies. She subsequently underwent a left lumbar sympathetic block, 2 separate left sciatic nerve peripheral nerve stimulator trials, and a 3-day continuous epidural infusion with ropivacaine, clonidine, and ketamine, none of which provided meaningful or sustained relief.

We started a DRG stimulation trial at left L4, L5, and S1, resulting in a > 80% reduction in pain, allodynia, swelling, and temperature asymmetry, as well as the restoration of function. She regained independent ambulation and resumed normal activities for the first time since her injury and subsequently underwent permanent implantation.

At her first postoperative follow-up, she reported approximately 65% sustained pain relief. She was able to ambulate without assistive devices, tolerate light touch, and resume household activities. She reported significantly improved sleep, reduced swelling, improved temperature regulation, and decreased muscle twitching in her left foot and ankle. She was able to transition back into normal shoes, which she had not worn in more than a year. Functionally, she was able to attend community activities with her friends and family. Encouraged by her progress, she expressed a desire to return to school earlier than the 6-week postoperative plan we had initially discussed with her.

### Case 3

A 14-year-old girl presented with a 9-month history of allodynia following an eversion injury of the left ankle sustained during a high school soccer match. She was evaluated by a fellowship-trained foot and ankle orthopedic surgeon who did not identify a structural etiology to explain her symptoms. Electromyography was nonfocal, and she was subsequently referred for further evaluation of suspected CRPS. At her initial visit, she met the Budapest Criteria for CRPS Type I. She had completed more than 6 months of intensive physical therapy, including desensitization, as well as

trials of neuropathic pain medications. Her mother, who served as her primary caregiver, was cautious regarding pharmacologic escalation due to a family history of medication misuse (not involving the patient or mother).

A lumbar sympathetic block was performed and provided transient relief; a second block produced similar temporary benefit. Given her persistent functional impairment, the care team discussed further interventional options. Referral for a peripheral nerve stimulation trial was completed; however, insurance coverage was denied since this therapy was deemed experimental for pediatric patients. After a discussion of the risks, benefits, and alternatives, permanent neuromodulation was recommended. While DRG stimulation was considered, concerns regarding the IPG's size relative to her age and body mass index (21 kg/m²) led to a decision to proceed with an SCS trial.

A 72-hour trial produced 75% improvement in pain and function, including improved sleep and the ability to attend school and ambulate in shoes without severe pain. Permanent implantation was performed 45 days later with a rechargeable device; there were no complications. At her 18-month follow-up, she continued to report > 50% pain relief, maintained functional gains, and required no further interventional procedures or controlled medications.

## CONCLUSION

These cases highlight the challenge of managing refractory CRPS pain in adolescents. In this case series we show that in 2 cases DRG stimulation and in one case SCS provided significant pain relief and improved function in patients that had failed more conservative management. This is consistent with the outcomes reported for adults with CRPS. Potential complications should be identified and discussed with the patient and family prior to any trial and implant. The risk of lead migration and IPG site pain are important considerations, especially for children and adolescents because they generally have higher activity levels than adults and the IPG's size is, proportionally, larger than it is in adults. Even though these complications occured in one of the cases, it did not affect the patient's pain relief. Limitations of our study include its retrospective nature and small sample size. Further prospective studies are needed to clarify patient selection, optimize techniques, and define long-term outcomes.

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