C A S E  R E P O R T

Epidural unilateral stimulation with “adaptive stim” option in treatment of type II CRPS

Armen Samvelovich Simonyan1 | Vladimir Mihaylovich Tyurnikov1 | Anna Dmitrievna Simonyan2 | Artem Olegovich Gushcha1

Abstract
CRPS is a type of severe pain syndrome and can be triggered by previous surgery or trauma. CRPS involves vasomotor changes such as changes in color and temperature of the skin, edema, increased sensitivity to touch, and a limited range of movement. Depending on the presence of nerve damage, CRPS is divided into two types. CRPS type II is associated with a confirmed peripheral nerve injury, while CRPS type I is not associated with an apparent peripheral nerve injury. Despite the ongoing therapy, sometimes, patients still have persistent, burning pain. Intractable CRPS that fail more conservative treatments may undergo neuromodulation. We want to present to your attention a case report of the successful treatment of a patient with CRPS type II using epidural unilateral stimulation. The 44-year-old woman came to us with complaints of burning pain and numbness of 1–3 fingers of the right hand, the lateral surface of the right wrist, and lower quarter of the forearm, and shooting pain in the projection of the right median nerve from the shoulder to the wrist. A clinical diagnosis was made—CRPS type II. During the stimulation trial, the most effective pain relief was obtained when the electrode was located in the right side of epidural space at the C4-Th1 level. The implantation of a pulse generator was performed, and the final selection of the stimulation parameters was carried out (Pulse width: 60 ms, Rate: 210 Hz, and Amplitude: 0.9–1.6 V). The severity of pain syndrome was measured using validated scales in the preoperative period (VAS: 8–9, Pain Detect: 22, NTSS-9: 4.62, and DN4: 8), in the early postoperative period (VAS: 0–1, Pain Detect: 6, NTSS −9: 0.66, and DN4: 1), and after 12 months (VAS: 0–2, Pain Detect: 6, NTSS-9: 0.99, and DN4: 1). Observation during 12 months showed that a stable analgesic effect of neurostimulation was achieved using standard neuromodulation regimens and the adaptive stim option. Unilateral stimulation is an effective type of SCS in the treatment of pain syndromes. adaptive stim is usually not applicable for lead implantation at the cervical level. Nevertheless, the rational use of stimulation at threshold values allowed our patient to use adaptive stim in a non-standard situation.

1Neurosurgery Department, Research Center of Neurology, Moscow, Russia
2Neurology Department, Burnasyan FMBC Research Center of FMBA of Russia, Moscow, Russia

Correspondence
Armen Samvelovich Simonyan, Neurosurgery Department, Research Center of Neurology, Moscow, Russia. Email: armenmed@mail.ru

Funding information
No funding to declare.
1 | INTRODUCTION

CRPS is a type of severe pain syndrome and can be triggered by previous surgery or trauma. CRPS involves vaso-motor changes such as changes in color and temperature of the skin, edema, increased sensitivity to touch, and a limited range of movement. CRPS type II is associated with a confirmed peripheral nerve injury, while CRPS type I is not associated with an apparent peripheral nerve injury. There are four diagnostic tools for CRPS in adult populations (Veldman criteria, IASP criteria, Budapest Criteria, and Budapest Research Criteria).

The complex treatment of CRPS includes pharmacotherapy, nerve blocks, physical and psychological measures, and rTMS. Despite the ongoing therapy, sometimes, patients still have persistent, burning pain. It leads to the disability of patients and a decrease in the quality of life. Also, the long-lasting, severe pain can result in psychological disorders such as depression and anxiety. Therefore, controlling CRPS-induced pain is a challenge in clinical practice. Intractable CRPS that fail more conservative treatments may undergo neuromodulation in the form of spinal cord stimulation (SCS), dorsal root ganglion stimulation (DRG), or peripheral nerve stimulation (PNS). Such factors will generally determine the choice of which modality is more suitable as pain localized to a specific nerve territory or pain that is felt mainly distal in an extremity.

Unilateral epidural stimulation and stimulation of the DREZ zone were described much less often in the literature. However, in our opinion, the effectiveness of unilateral stimulation and DREZ stimulation is not inferior and, in some cases, even exceeds destructive interventions.

2 | CASE DESCRIPTION

We want to present to your attention a case report of the successful treatment of a patient with CRPS type II using unilateral epidural stimulation. The 44-year-old woman came to us with complaints of burning pain and numbness of 1–3 fingers of the right hand, the lateral surface of the right wrist, and lower quarter of the forearm, and shooting pain in the projection of the right median nerve from the shoulder to the wrist. The patient had previously suffered an injury to the right hand with damage to the tendons and median nerve and underwent several reconstructive surgeries. After the injury, the appearance of a pronounced pain syndrome was noted. The patient underwent conservative therapy for 3 years. Various combinations of drugs were included in the therapy regimen (NSAIDs, anticonvulsants, antidepressants, opioids, and botulinum toxin). Physiotherapy was carried out along with medications. There was no significant improvement in the patient’s condition. Multiple surgical interventions, including radiofrequency ablation (RFA) and sympathectomy, with a short-term positive effect, were carried out. Taking into account the clinical picture, anamnesis, and using the Budapest criteria, a clinical diagnosis was made—CRPS type II. A decision was made to implant an epidural electrode to perform a stimulation trial. During the stimulation, the most effective pain relief was obtained when the electrode was located in the right side of epidural space at the C4-Th1 level (Figure 1A).

Against the background of the therapy, a significant decrease in the severity of the pain syndrome was noted, and the effective parameters of stimulation were
determined—Pulse width: 60 ms, Rate: 210 Hz, and Amplitude: 0.9–1.6 V (Table 1). On the next day after surgery, stable pain relief was achieved when the neurostimulator was turned on. The implantation of a pulse generator was performed after the stimulation trial (trial duration: 10 days). After the implantation (Figure 1B), a final selection of the stimulation parameters was carried out. Later, the adaptive stim option was activated, which, according to the stimulation parameters, was carried out. Later, neurostimulation very rationally. Stimulation is almost all the patient, greatly facilitates everyday life and reduces the time spent on programming the device. The patient uses neurostimulation very rationally. Stimulation is almost always carried out at the lower threshold of the therapeutic window. Movements in the craniocervical region (flexion, extension, rotation, etc.) change coverage area and paresthesias, but actions do not cause inconvenience to the patient since the stimulation strength does not reach the upper threshold of the therapeutic window, which allows applying adaptive stim. Observation during 12 months showed that a long-lasting analgesic effect of neurostimulation was achieved. The severity of pain syndrome was assessed using the scales VAS, NTSS−9, DN4, and Pain Detect (Table 1).

Compared with the early postoperative period (when complete regression of pain syndrome was noted), some decrease in efficiency is most likely due to “stimulation tolerance.”

### 3 | CONCLUSION

Most patients with CRPS I reported minor trauma prior to the development of symptoms, such as a sprain, fracture, fall, crush injury, burn, or soft tissue injury.² The pathogenesis of CRPS is not understood. However, evidence now emerging from many different fields suggests a multifactorial disorder triggered by an initial, sometimes relatively minor injury. There is then an aberrant response by the body with exaggerated immune response, maladaptive neuroplasticity, and abnormal vasomotor function within the tissues of the affected limb.⁵ The International Association for the Study of Pain (IASP) has endorsed the Budapest criteria for the diagnosis of CRPS. CRPS I is not associated with an identifiable nerve injury, whereas CRPS II is associated with a nerve injury.⁵ Physical and occupational therapy is a critical component of the rehabilitation process in patients with CRPS and is recommended as the first-line treatment.³ Historically, sympathectomy has been used to treat CRPS. This can now be performed using radiofrequency, chemicals, and surgery.⁶,⁷ Sympathectomy has a significant complication rate, including local anhydrosis and Horner’s syndrome.⁵ Ackerman showed that stellate ganglion blockade is effective for pain management in CRPS.⁸

A randomized study involving 24 patients with CRPS and SCS plus physical therapy (PT) reduced pain and improved health-related quality of life more than PT alone for up to 2 years.⁹ The potential that combination therapy with tonic-SCS and DRGS may be beneficial in patients with severe and refractory CRPS.¹⁰,¹¹ Data from the ACCURATE study suggest that DRGS could be used in patients suffering from chronic intractable pain conditions that are refractory to tonic-SCS.¹²

Unilateral epidural stimulation is an effective type of SCS in the treatment of pain syndromes. In our opinion, ablation (DREZ) is possible for patients with a relatively poor prognosis of survival for palliative purposes. In other cases, we consider neuromodulation primarily. The possibility of conducting a minimally invasive stimulation trial, the reversibility of the technique, and the ability to control the stimulation process, in our opinion, is an advantage over destructive interventions. Our clinical case confirms the possibility of using unilateral epidural stimulation with “adaptive stim” regimen to treat complex pain syndromes such as CRPS. Rational use of neuromodulation capabilities may, in rare cases, allow the use of adaptive stim in case of cervical epidural lead placement. Cases of migration of epidural leads have been reported in the literature, but improvements in implantation techniques have minimized this risk.¹³ The preoperative selection plays a crucial role in good results. If SCS effects do slowly diminish over time, DRG stimulation seems to be a treatment alternative.¹⁴ In our opinion, the rapidly developing

### TABLE 1  Dynamics of the severity of pain syndrome and stimulation parameters

|                              | Before SCS device implantation | Early postoperative period | Late postoperative period (after 6 months) |
|------------------------------|--------------------------------|-----------------------------|-------------------------------------------|
| VAS                          | 8–9                            | 0–1                         | 0–2                                       |
| Pain Detect                  | 22                             | 6                           | 6                                         |
| NTSS−9                       | 4.62                           | 0.66                        | 0.99                                      |
| DN4                          | 8                              | 1                           | 1                                         |
| Stimulation parameters       |                                |                             |                                           |
| Pulse width, Rate, Amplitude | 60 ms, 210 Hz                  | 60 ms, 210 Hz               | 20–80 ms, 210–240 Hz                      |
|                              | 0.9–1.6 V                      | 0.9–1.6 V                   | 0.9–1.6 V                                 |
neuromodulation technique opens up new possibilities in the treatment of pain syndromes.

ACKNOWLEDGEMENT
None.

CONFLICT OF INTEREST
All authors have no conflict of interest to report.

AUTHOR CONTRIBUTIONS
Armen Samvelovich Simonyan served as an idea author and involved in development of clinical aspects, article writing, and editing. Vladimir Mikhaylovich Tyurnikov involved in development of clinical aspects. Artem Olegovich Gushcha served as a scientific supervisor and involved in article editing. Anna Dmitrievna Simonyan involved in article editing.

ETHICAL APPROVAL
All performed procedures were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki Declaration and its later amendments.

CONSENT
Written informed consent was obtained from the patient to publish this report in accordance with the journal’s patient consent policy.

DATA AVAILABILITY STATEMENT
The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ORCID
Armen Samvelovich Simonyan https://orcid.org/0000-0001-8848-801X

REFERENCES
1. Chang MC, Kwak SG, Park D. The effect of rTMS in the management of pain associated with CRPS. Transl Neurosci. 2020;11(1):363-370. doi:10.1515/tmsci-2020-0120. PMID: 33335776; PMCID: PMC7711855.
2. Chmiela MA, Hendrickson M, Hale J, et al. Direct peripheral nerve stimulation for the treatment of complex regional pain syndrome: a 30-year review. Neuromodulation. 2021;24(6):971-982. doi:10.1111/ner.13295. Epub ahead of print. PMID: 33098229.
3. Goh EL, Chidambaram S, Ma D. Complex regional pain syndrome: a recent update. Burns Trauma. 2017;5. No Pagination Specified. doi:10.1186/s41038-016-0066-4
4. Mesaroli G, Hundert A, Birnie KA, Campbell F, Stinson J. Screening and diagnostic tools for complex regional pain syndrome: a systematic review. Pain. 2021;162(5):1295-1304. doi:10.1097/j.pain.0000000000002146. Epub ahead of print. PMID: 33230004.
5. Cutts S, Gangoo S, Srinivasan SH, Modi N, Pasapula C, Power D. Complex regional pain syndrome: an evolving perspective. Postgrad Med J. 2020;12(137808). doi:10.1136/postgradmedj-2020-137808. Epub ahead of print. PMID: 33184132.
6. Forouzanfar T, van Kleef M, Weber WE. Radiofrequency lesions of the stellate ganglion in chronic pain syndromes: retrospective analysis of clinical efficacy in 86 patients. Clin J Pain. 2000;16:164-168.
7. Nelson DV, Stacey BR. Interventional therapies in the management of complex regional pain syndrome. Clin J Pain. 2006;22:438-442.
8. Ackerman WE, Zhang JM. Efficacy of stellate ganglion blockade for the management of type 1 complex regional pain syndrome. South Med J. 2006;99:1084-1088.
9. Kemler MA, Barendse GA, van Kleef M, et al. Spinal cord stimulation in patients with chronic reflex sympathetic dystrophy. N Engl J Med. 2000;343(9):618-624. doi:10.1056/NEJM200008313430904. PMID: 10965008.
10. Ghosh P, Gungor S. Utilization of concurrent dorsal root ganglion stimulation and dorsal column spinal cord stimulation in complex regional pain syndrome. Neuromodulation. 2021;24(4):769-773. doi:10.1111/ner.13144. Epub ahead of print. PMID: 32162402.
11. Piedade GS, Vesper J, Slotty PJ. Synergetic efficacy of simultaneous DRG- and traditional spinal cord stimulation. Acta Neurochir (Wien). 2020;162(2):257-260. doi:10.1007/s00701-019-04166-y. Epub 2019 Dec 20 PMID: 31863300.
12. Deer TR, Levy RM, Kramer J, et al. Dorsal root ganglion stimulation yielded higher treatment success rate for complex regional pain syndrome and causalgia at 3 and 12 months: a randomized comparative trial. Pain. 2017;158(4):669-681. doi:10.1097/j.pain.0000000000008084. PMID: 28030470; PMCID: PMC5359787.
13. Hasoon J. Lead migration in spinal cord stimulation with loss of pain coverage in a CRPS patient. Saudi J Anaesth. 2020;14(2):280-281. doi:10.4103/sja.SJA_47_20. Epub 2020 Mar 5. PMID: 32317904; PMCID: PMC7164436.
14. Schwarm FP, Stein M, Uhl E, Maxeiner H, Kolodziej MA. Spinal cord stimulation for the treatment of complex regional pain syndrome leads to improvement of quality of life, reduction of pain and psychological distress: a retrospective case series with 24 months follow up. Scand J Pain. 2020;20(2):253-259. doi:10.1515/sjpain-2019-0081. PMID: 31743107.

How to cite this article: Simonyan AS, Tyurnikov VM, Simonyan AD, Gushcha AO. Epidural unilateral stimulation with “adaptive stim” option in treatment of type II CRPS. Clin Case Rep. 2022;10:e05305. doi:10.1002/ccr3.5305