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SPINAL CORD STIMULATION IN SMALL FIBER NEUROPATHY: A CASE REPORT

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Background: Spinal cord stimulation (SCS) is a well-established pain treatment in several chronic neuropathic pain conditions refractory to conservative treatments. Neuropathic pain due to small fiber neuropathy (SFN) may represent a further indication.

Case Report: We treated with SCS a 22-year-old female patient with SFN and chronic neuropathic pain. After an initial effective trial targeting the lower limbs with improvement in pain and quality of life, a definitive implant was performed. Ten months later, a single cervical octopolar electrode was inserted to treat upper limb pain with the same protocol (i.e., initial effective trial and definitive implant). Also, in this case, the patient experienced a significant global reduction in upper limb pain.

Conclusions: To date, evidence for the SCS efficacy in refractory SFN-related pain is still limited to small case series or case reports. Our experience may help to validate this indication.

Key words: Small fiber neuropathy, spinal cord stimulation, neuropathic pain

BACKGROUND

Spinal cord stimulation (SCS) is a well-established neuromodulatory pain treatment whose aim is to deliver electrical impulses to neural structures adjacent to the epidural space (1). To date, SCS is widely used in the treatment of chronic refractory painful conditions, such as persistent spinal pain syndrome, neuropathic pain following radiculopathies, complex regional pain syndromes I and II, and chronic vascular pain (1). SCS showed promising results in the treatment of other neuropathic painful conditions, such as diabetic neuropathy, chemotherapy-induced neuropathies, and chronic low back pain, with a neuropathic component (2-5). Among the most recent indications, chronic neuropathic pain due to small fiber neuropathy (SFN) shows some interesting peculiarities. SFN is characterized by small delta and C-fiber damage mostly induced by either metabolic, immune-mediated, toxic, infectious, or genetic mechanisms, while, in some patients, SFN pathophysiology remains unknown (6). The small fiber damage may lead to somatosensory and autonomic impairment, and chronic neuropathic pain (6). The diagnosis of SFN cannot be performed by means of routine electrophysiological tests due to small nerve fibers' unique anatomophysiological characteristics: a skin biopsy seeking for signs of neural damage is necessary in order to confirm the diagnosis. To date, SFN treatment remains symptomatic only, a causal treatment is not available yet (7). Like in other neuropathic painful conditions refractory to conservative treatments, SCS may induce sensorial modifications over the painful area, thereby masking the patient's pain sensation. We report a case

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of a patient with refractory neuropathic pain from SFN successfully treated with SCS. The available literature regarding SCS treatment in SFN is also analyzed.

CASE PRESENTATION

Patient History

A 22-year-old female patient suffering from SFN was referred to our Pain Therapy Centre for refractory pain with a predominant neuropathic component. The past medical history included a second-degree obesity (body mass index 38.8) and polycystic ovary syndrome. In 2016, the patient started complaining of widespread pain, although favoring upper and lower limbs, and associated symptoms, such as allodynia (i.e., severe induced pain after extremely light touch), muscle weakness, prolonged rest stiffness, and low-grade fever. A mutation in tumor necrosis factor receptor superfamily member 1A-gene led to the initial diagnosis of recurrent fever syndrome, but this would not explain symptoms, such as burning pain in the feet, allodynia, reduced thermal and pinprick sensation in the painful areas, which our patient was experiencing. In 2019 and after several medical examinations, a skin biopsy revealed small nerve fiber damage hence suggesting the presence of an underlying SFN.

Initial Assessment

In 2021, at the time of our evaluation, the patient complained of chronic pain Visual Analog Scale (VAS, range 0-100) average score of 90, describing it as burning and stabbing with a Leeds Assessment of Neuropathic Symptoms and Signs (LANSS, range 0-24) score of 24 and a Douleur Neuropathique 4 Questions (DN4, range 0-10) score of 9. The patient reported a negative impact on daily activities and quality of life (QoL) with an Oswestry Disability Index (ODI, range 0-100) score of 80. Several pharmacological treatments, according to neuropathic pain guidelines, were attempted with initial pain relief, but severe, progressive pain worsened over the following weeks. Upon performing a careful psychological screening aiming to exclude the presence of any underlying psychiatric conditions, a trial of percutaneous SCS targeting the lower limbs innervation (i.e., single octopolar electrode, distal catheter tip at T7 level) was conducted (Fig. 1).

SCS Trial

Several types of stimulation were tried (fast-acting subperception therapy, burst therapy [i.e., closely spaced train of high-frequency pulses followed by a plateau], 1,000 Hz, and tonic stimulations, all using current steering technology - Illumina™ 3D software [Illumina, San Diego, CA] - to map the pain areas). Of those, the tonic pattern shooting at 40 Hz and eliciting paresthesia in the target areas demonstrated the best results on pain relief and acceptability on the patient's part. After 8 days, due to the significant improvement in terms of pain and QoL, a definitive implantable pulse generator (IPG) implant was positioned (WaveWriter Alpha™ Spinal Cord Stimulator System, Boston Scientific®, Marlborough, MA).

Follow-up

After 3 months, the patient reported a further global improvement in terms of pain severity and QoL, which led to a sensitive reduction of painkillers consumption (Fig. 2). Due to these results, 10 months later, a single percutaneous cervical octopolar electrode was also inserted aiming to treat upper limb pain (i.e., single octopolar electrode, distal catheter tip at C3 level) (Fig. 3). Tonic stimulation at 40 Hz eliciting paresthesia in the target areas was again the modality of choice as patient-tailored best pattern for pain relief so far. After an effective 8-day trial, a definitive implant was performed connecting the new catheter to the IPG already in place. At 3 months of follow-up, the patient experienced a significant reduction in upper limb pain, too (VAS score from 90 to 40, DN4 score from 9 to 4, LANSS score from 16 to 6). At 15 months follow-up time, the patient reported no overall pain relapse or worsening.

DISCUSSION

The use of SCS is a rapidly expanding technique in chronic pain treatment, all the while being already widely accepted as a well-proven therapeutic option for some indications (1). SCS was also proposed to selected patients for whom the conventional pharmacological schedules have either proven ineffective or characterized by unbearable side effects (8). To date, SFN might represent a therapeutic target for SCS, nonetheless, this indication is still anecdotal, with little data available in the literature so far. The first available retrospective study reporting the use of SCS in SFN was performed by Hayek et al (9) in 2015. Among 345 patients submitted to SCS, 21 suffered from SFN. After a trial period, in 18 patients, a definitive implant was performed.

These patients displayed the highest implant-to-trial ratio (86%) when compared to those undergoing the procedure due to other indications. In 5 patients, an implant revision due to complications was reported, and in 2 patients the system was removed. Unfortunately, details regarding efficacy, complications, and technical notes (e.g., type of electrode, spinal levels, IPG characteristics, stimulation modalities, and removal motivations), were not available. In 2017, Eckmann et al (10) described a case of SCS in a 20-year-old SFN patient who underwent a contemporary trial in both the cervical (top of the C3 vertebral body) and thoracic regions (top of the T8 vertebral body). Due to the positive effect on pain, the patient was later implanted with 2 cervical and 2 thoracic octopolar leads connected to 2 IPGs. The stimulation was tonic and paresthesia based. After 6 months, the patient continued to report excellent pain relief with a complete abolition of pain in both upper and lower limbs. The patient's QoL also improved dramatically, as they experienced an increased proneness to daily activity and proceeded to spontaneously abolish any painkillers intake. Different neurostimulation techniques were attempted in SFN patients. In 2017, Maino et al (11) described the use of dorsal root ganglion (DRG) stimulation to treat localized pain in the left foot of a 74-year-old SFN patient. The patient reported uncontrolled burning and shooting pain in his left foot, which had been progressively worsening during the past 6 years and nonresponsive to multiple medications (e.g., gabapentinoids, serotonin-norepinephrine reuptake inhibitors, amitriptyline, mirtazapine, lidocaine patches, topical capsaicin 8%, and medical cannabis) and nonpharmacological interventions (e.g., transcutaneous electrical nerve stimulation, physical therapy, acupuncture, and local corticosteroids infiltration). A neuromodulation trial to the left L5 DRG was performed percutaneously with a quadripolar electrode (Axium™ Neurostimulator System, Spinal Modulation, Inc., Menlo Park, CA). After 10 days of effective stimulation (improvement of 62.5% in terms of pain reduction), the neurostimulator was then permanently implanted. Two months postimplantation, the patient still experienced significant pain reduction and improvement in all domains of the McGill Pain Questionnaire. Also, the degree of disability, as measured by the ODI scoring system, decreased substantially over said follow-up time. A comparable ODI score reduction trend was observed in the subsequent months.

CONCLUSIONS

To date, evidence for the SCS efficacy in cases of refractory SFN-related pain is still limited to small case series or case reports, hence further data are necessary in order to validate this indication and standardize the

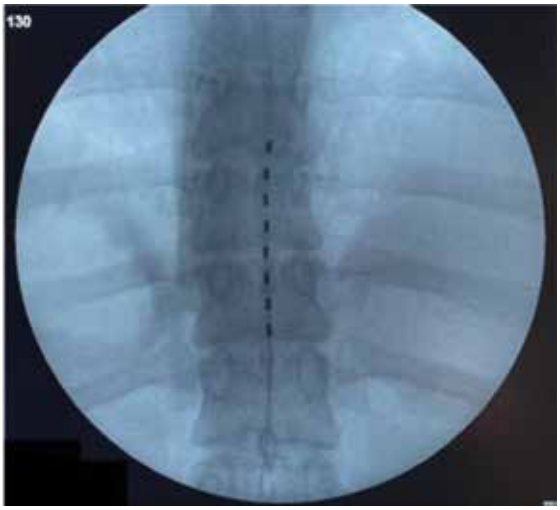

Fig. 1. Octopolar single thoracic electrode with the tip at T7 level.

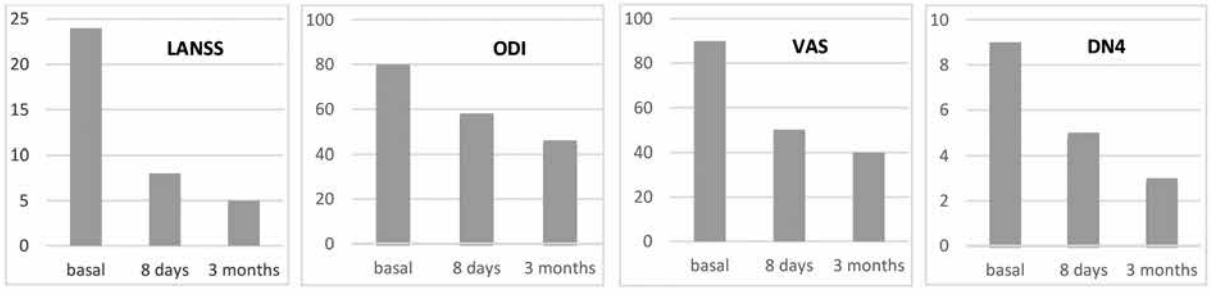

Fig. 2. Time course of LANSS, ODI, VAS, and DN4 during follow-up after thoracic catheter placement.
Abbreviations: LANSS, Leeds Assessment of Neuropathic Symptoms and Signs; ODI, Oswestry Disability Index; VAS, Visual Analog Scale; and DN4, Douleur Neuropathique 4 Questions.

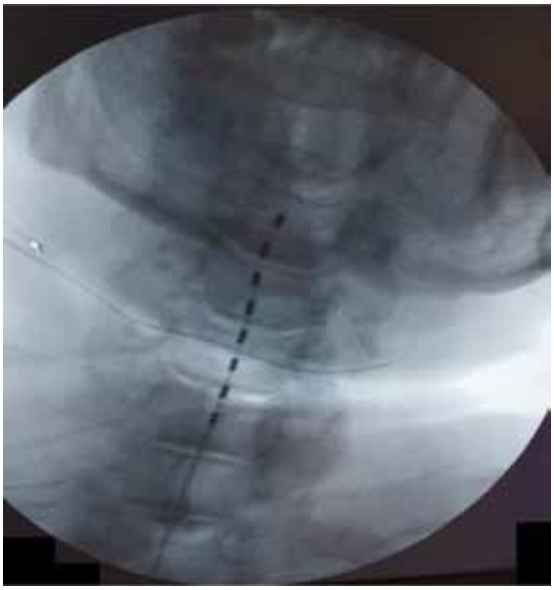

Fig. 3. Octopolar single cervical electrode with the tip at C3 level.

different stimulation techniques and the scores used to measure the outcomes, making them comparable. Nonetheless, we believe that advanced neuromodulation techniques, like SCS, should be always taken into consideration in patients with neuropathic pain due to SFN refractory to conservative treatments. The tonic stimulation proved to be effective compared to other patterns, although the small literature in SFN seems to suggest the use of high frequency, but this evidence needs to be better elucidated and correlated to the SFN pathophysiology. However, a careful patient selection regarding SFN diagnosis and exclusion criteria is mandatory prior to the start of any SCS trial. Moreover, the definitive implant should not occur without a preceding effective trial period (12,13). In conclusion, although the benefits of SCS are, in general, well-supported in neuropathic pain features, the application of this treatment may vary widely based on physician training and experience, clinical practice, and insurance coverage (13). For these reasons, starting from the clinical case descriptions in SFN patients, our experience can inspire the implementation of larger studies in the future, providing greater evidence.

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