



## Case Report

# Percutaneous high cervical spinal cord stimulation for refractory trigeminal neuralgia

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## ABSTRACT

**Background:** Trigeminal neuralgia (TN) is a debilitating pain that affects the dermatomes associated with the trigeminal nerve (V1, V2, and V3). Unfortunately, many medical treatments and surgical procedures fail to sufficiently modulate the pain associated with this condition.

**Case Description:** This study presents two extreme cases of refractory TN (RTN) that progressed to atypical facial pain and describes successful mitigation of the neuralgia of said cases by percutaneous implantation of upper cervical spinal cord stimulation (SCS). The SCS was designed to target the descending spinal trigeminal tract.

**Conclusion:** Together, these cases collaborate with the limited literature and further delineate the use and potential advantages of SCS in the treatment of RTN.

**Keywords:** Descending spinal trigeminal tract, Facial pain, Neuromodulation, Spinal cord stimulation, Trigeminal neuralgia

## INTRODUCTION

Trigeminal neuralgia (TN) is a syndrome distinguished by unilateral, paroxysmal, and electric pain that is most often found in the V2 and V3 dermatomes of the cranial nerve five (CN V).<sup>[19]</sup> This pain is divided according to two common presentation schemes, type 1 which may present for brief instances followed by periods of complete remittance or type 2 which may present with constant intensity pain.<sup>[21]</sup> The etiology of TN is divided into classical and secondary TN. Classical TN is characterized by neurovascular compression of CN V in the prepontine cistern, and secondary TN is characterized by neurological disorders (e.g., multiple sclerosis).<sup>[5]</sup> The incidence rate of TN in the general population has been estimated at 28.9/100,000 person years with a higher prevalence in women and in those between the ages of 50 and 90.<sup>[20]</sup>

The first line of treatment for TN is pharmacological, with the most common treatment methods being carbamazepine or oxcarbazepine.<sup>[7]</sup> However, these drugs are often only capable of controlling pain in 75% of patients due to the presentation of significant side effects. Furthermore, up to 50% of patients who tolerate medical treatments have refractory TN (RTN).<sup>[13]</sup> For people with RTN, there are several surgical options available. Microvascular decompression in the

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presence of vascular compression is the most effective treatment with a recurrence rate of 10%,<sup>[3]</sup> while success for other procedures such as percutaneous rhizotomy and Gamma Knife radiosurgery can decrease over time to approximately 50% after 5 years.<sup>[6,11,17]</sup>

A line of therapy developing for RTN is based on neuromodulation. At present, there are few clinically reported methods of neuromodulation: Motor cortex stimulation,<sup>[8]</sup> deep brain stimulation,<sup>[18]</sup> gasserian ganglion/peripheral stimulation,<sup>[26]</sup> and upper cervical spinal cord stimulation (SCS).<sup>[27]</sup> However, these options had variable success rates and complications, and the exact mechanism by which RTN treatment by neuromodulation has not yet been determined.<sup>[4,12]</sup>

The selection criteria for the following patients was a RTN that progressed to trigeminal neuropathic pain, as a result of injury to trigeminal nerve by prior procedures, and described as unremitting throbbing and burning pain in the affected areas.<sup>[9]</sup>

## CASE PRESENTATION

### Case 1

A 76-year-old woman presented with RTN. The patient had a history of the right TN at the V1 and V2 distribution after a neurofibroma was resected from the pterygopalatine fossa at another institution. This was followed by a right balloon compression of the gasserian ganglion. Subsequently, the patient reported no improvement in pain. This was followed by a right retrosigmoid craniectomy for microvascular decompression, which also did not improve her pain. Retrosigmoid craniectomy was followed by a resection of V2 surgical scar soft-tissue mass after which the patient reported still no improvement. The patient also had a history of Botox injections that reduced pain for 2–3 months, as well as several nerve blocks that gave the patient only a temporary reprieve. She was followed by pain management physicians and headache neurologists with no improvement in pain (oxycodone, methadone, and lidocaine gel). The patient was later referred to our evaluation with a constant burning and paroxysmal stabbing right facial pain accompanied by nausea affecting the distal portion of V2 and V3. This pain was aggravated by talking, chewing, brushing teeth, smiling, and swallowing. The patient reported that pain prevented adequate sleep and social function with a constant severity of 10/10 on a visual analog scale (VAS). Due to the failure of these surgical and medical procedures to provide pain relief, the patient was recommended for a percutaneous trial of SCS with the intention of targeting the descending spinal trigeminal tract. The patient underwent a neuropsychological evaluation and was deemed an appropriate candidate for the trial (ruled out unresolved major psychiatry comorbidities,

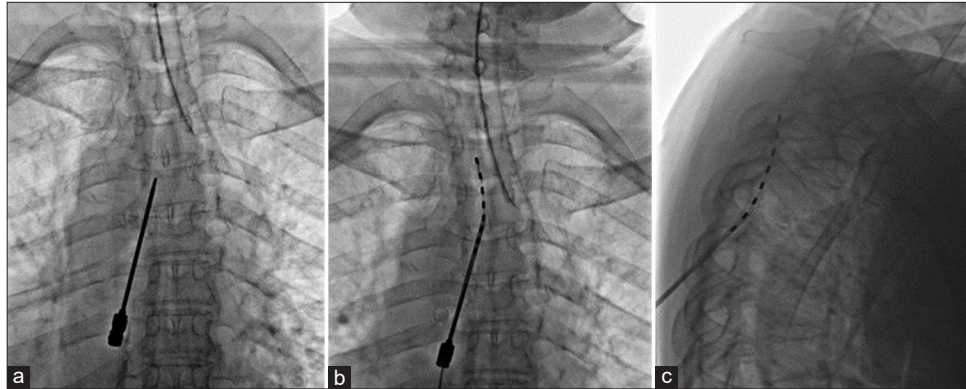
drug abuse, and unresolved secondary gain). She also had brain, cervical, and thoracic Magnetic resonance imaging (MRI) preoperatively. Under general anesthesia, the patient was placed in the prone position and fluoroscopy was used to identify the level of the thoracic entry point on the skin (T6) of the Tuohy needle and in the epidural space in T4 [Figure 1].

Then, a percutaneous lead of eight contacts (Medtronic 977A275 Lead Restore SureScan 75 mm compact 1 × 8) was advanced along the posterior epidural thoracic and cervical space under the guidance of fluoroscopy until the level of C1–C2 on the right side, at which point the lead was then secured in place. The needle was then removed and the lead was fixed in place using tape secured to the patient's skin. Then, X-rays were taken to determine the level of placement if the trial surgery was successful [Figure 2].

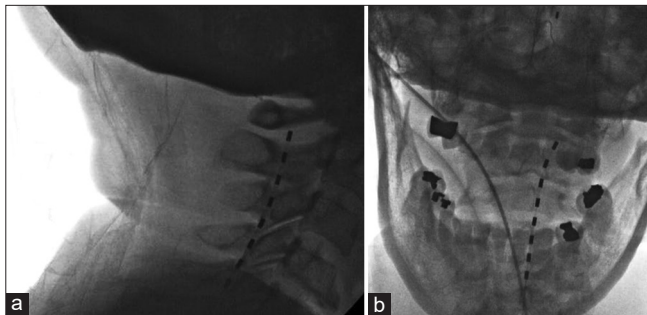
The patient was kept in a soft collar to avoid neck rotation and migration of stimulation, and the temporary lead was uneventfully removed 7 days later in the outpatient clinic. The patient reported that during the high-frequency stimulation period, she had a 60% reduction in pain. As such, the patient was then considered for the placement of a permanent SCS implant. The patient agreed to the permanent placement of the lead and as such was admitted back to the operating room. The patient was prepared for surgery and again with a similar technique to that of the trial implantation, replicating the placement of the cervical lead, except that she now had a lead anchored to the fascia in the thoracic area (requiring a 4 cm incision) and this was tunneled to the right posterior flank for the implantation of the subcutaneous battery (Medtronic 97715 Stim Medt Nerve Intellis). The incisions were then closed and properly covered and the patient recovered in stable conditions. Overall, with a short follow-up of 4 weeks (using two groups of stimulation, with 300 Hz, pulse width 170 ms, and varying intensities between 0.5 and 0.8 mA), the patient referred a significant improvement in her quality of life since her first stimulation session with 40% pain relief and referring to eating and talking more comfortably and rating her pain 6/10. No wound complications were encountered.

### Case 2

A 29-year-old woman had severe pain since 2006 on the right V1 and V2 distribution of the trigeminal nerve was referred to our clinic. The patient described this pain as initially a lacerating, electric, and shooting pain that lasted a few seconds to several minutes. She reported that this pain was triggered by wind, brushing teeth, stress, eating, cleaning her face, and bright light. The patient received several medical treatments including lamotrigine, phenytoin, gabapentin, topiramate, carbamazepine, clonazepam, celecoxib, nortriptyline, baclofen, hydroxyzine, sumatriptan, and rimegepant, all of which were ineffective in treating pain.



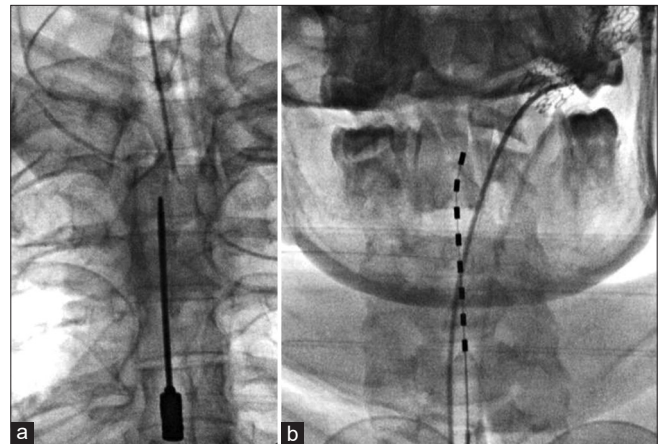
**Figure 1:** Fluoroscopy view of the percutaneous thoracic spine needle access (a); later the eight contact lead (dashed line), partially in the epidural space on anterior-posterior view (b); and lateral view (c) showing entire lead (dashed line image) within the thoracic epidural space.



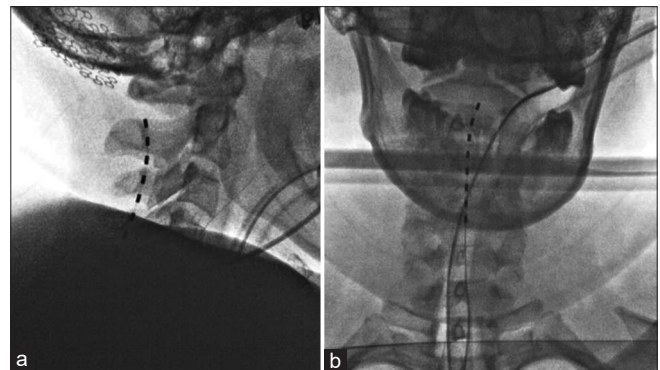
**Figure 2:** Lateral fluoroscopy view after the placement of the spinal cord stimulation (SCS) trial electrode (dashed line image), advanced to the upper cervical spine just below posterior arc of C1 level in the dorsal epidural space (a); Anterior-posterior fluoroscopy view showing the electrode (dashed line image) towards right side of the epidural space in the upper cervical spine (b).

She was then treated with microvascular decompression in 2017 which was effective at treating her pain for 2 years. Subsequent recurrence of pain along the V1 and V2 dermatomes brought the patient back for further treatment. The patient's RTN was treated with Gamma Knife surgery which had minimal effects on her pain and later with balloon compression, also unsuccessful. Her pain then became atypical with constant stabbing and burning sensation and allodynia. At this point, the patient reported that the pain was constant and interrupted her sleep with VAS 10/10 severity of the pain and was referred for our evaluation. She underwent neuropsychological evaluation, and brain, cervical, and thoracic magnetic resonance imaging, and was considered a candidate for the percutaneous upper cervical stimulation trial.

The patient was then prepared for the trial surgery in the prone position and under general anesthesia. With fluoroscopy guidance, a Tuohy needle was inserted into the epidural space at the T3–T4 level. From this point on, the percutaneous SCS lead advanced to the level of C2–C4

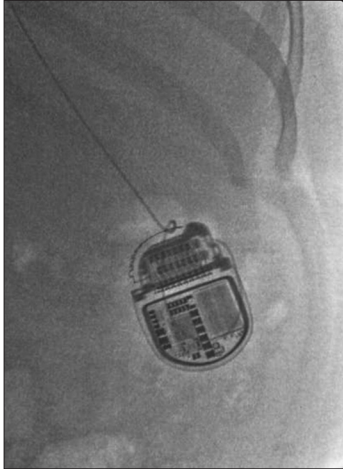


**Figure 3:** Spinal cord stimulation trial lead placement with Tuohy needle inserted at level T3-4 (a), and advancement of the eight contact lead (dashed line image) on anterior-posterior fluoroscopy view from cervical level C2 to C4 (b).



**Figure 4:** Lateral fluoroscopy view of the placement of the spinal cord stimulator lead (dashed line image) with upper contact between cervical level C1-C2 (a); on anterior-posterior view, the tip of the eight contact percutaneous lead (dashed line image) is directed to patient's right side (b).

that has resistance to further advancement [Figure 3]. The lead (Medtronic 977A275 Lead Restore SureScan 75 mm



**Figure 5:** Placement of the battery for the spinal cord stimulation lead in the right lower lumbar region.

compact 1 × 8) was then anchored and connected to an external battery.

The patient reported a 90% pain relief with high-frequency stimulation (rate 300 Hz, pulse width 170 ms, and varying intensity from 1.8 to 2.2 mA) with a significant improvement in quality of life. With the success of the trial procedure, the patient was then readmitted to the OR replicating the initial trial procedure [Figure 4], placing an eight contact lead which was anchored to the thoracic fascia and tunneled to the right flank as described on the previous patient for battery implantation (Medtronic 97715 Stim Medt Nerve Intellis) [Figure 5]. The incisions were closed and dressed. The patient recovered without complications.

At 6 months after surgery, the patient reported between 60% and 98% reduction in both severity of pain and reduction in frequency of attacks (2-3 times per week), rather than lasting about 10 days. The patient was consequentially able to resume more normal daily activities and take significantly less pain medications.

## DISCUSSION

The techniques applied in each of the presented cases utilize high-frequency stimulation of the upper cervical spinal cord with the intention of targeting the descending trigeminal spinal nucleus that receives input from V3 branch. This anatomical distribution does not cover all the anatomical basis for ophthalmic nerve (V1) and mandibular nerve (V2) pain, although is able to offer partial coverage on the pain distribution. These two case examples are extreme cases of RTN with subsequent transformation to atypical facial pain after multiple invasive procedures. It is important to select appropriate patient candidates with realistic goals of treatment. Formal neuropsychological testing is required

to identify problematic emotional reactions, maladaptive thinking and behavior, and social issues that can contribute to pain and disability that can affect the surgical outcomes.<sup>[2]</sup> Given the multiple prior surgical interventions, our decision was to minimize the surgical procedure with a minimal invasive approach using only percutaneous implantation of the leads and using high-frequency stimulation. Both procedures were found to have significant improvements in patient condition and quality of life and no associated complications or surgical morbidity.

Additional evidence presented by Velasquez *et al.* suggested that the dorsal horn islet may convey the therapeutic effects of SCS.<sup>[25]</sup> The descending nucleus of the trigeminal nerve extends from the lateral medulla to the upper cervical cord, and the subnucleus lateral to the dorsal columns is the primary afferent site of mandibular nociceptive neurons as a target for SCS.<sup>[14]</sup> Although the exact mechanism is not fully determined, it is likely the functionality neuromodulation through SCS rests in the principles of gate control theory. Through stimulation of large diameter non-nociceptive Aβ fibers, it is likely that the thin C fibers are inhibited through inhibitory interneurons, thereby diminishing the signals associated with chronic pain.<sup>[16]</sup> Consequentially, SCS is not necessarily diminishing the firing of nociceptive neurons themselves but rather providing an additional stimulus resulting in the prevention of transmission through integration of other hyperpolarizing signals produced in the interaction between Aβ fibers and inhibitory interneurons. Research by Peirs *et al.* on mice populations provided limited evidence of such neural networks within the dorsal horn laminae and suggested that the nociceptive fibers were likely to terminate in superficial laminae I and II, while non-nociceptive sensory neurons were likely to innervate the deep laminae III-IV.<sup>[23]</sup>

Only eight other case reports<sup>[1,10,15,22,24,25,28]</sup> have described pain relief of at least 50% in a total of 28 patients associated with RTN SCS treatment; furthermore, only two reports detailed permanent implants placed by percutaneous technique with pain relief estimated at about 70%.<sup>[10,26]</sup> This minimally invasive procedure is not associated with high rates of infections, lead migrations, or skin erosions as reported on gasserian ganglion/peripheral stimulation (26.4%, 17.6%, and 29.4%, respectively),<sup>[26]</sup> and does not require an upper cervical or suboccipital incision with the expected surgical site pain as in paddle lead placement and associated morbidity. The reports of these case series collaborate with evidence given.

## CONCLUSION

Our study supports the use of SCS as a viable and safe treatment option for individuals with RTN, and highlights the necessity of conducting randomized controlled trials

on the treatment of RTN through neuromodulation of the descending trigeminal spinal nucleus.

### Statement of ethics

Ethical approval and patient's consent is not required for retrospective case report studies without identifiable information in accordance with Loma Linda University Institutional Review Board guidelines.

### Declaration of patient consent

Patients' consent not required as patients' identities were not disclosed or compromised.

### Financial support and sponsorship

Nil.

### Conflicts of interest

There are no conflicts of interest.

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