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Cervical Spinal Dorsal Root Stimulation in Trigeminal Neuralgia

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Background: The treatment of trigeminal neuralgia is a challenge especially for cases refractory to the common standard of care. Neurostimulation for pain relief has been used over the years with different targets and modalities. Few reports exist about the stimulation of high cervical spinal dorsal roots to treat trigeminal pain.

Case Report: We report a case of a refractory secondary trigeminal neuralgia that was progressively resistant to various treatments. A trial for upper cervical spinal dorsal root stimulation provided immediate good facial pain relief, evoking paresthesias only in the cervical dermatomes. Positive results were obtained over 3 years with reduction of pain, drugs, and improvement in quality of life.

Discussion: Neurostimulation of the high cervical spinal dorsal roots with the activation of the trigeminocervical complex may be an effective and safe treatment for refractory trigeminal neuralgia.

Key words: Spinal dorsal root neurostimulation, trigeminal neuralgia, trigeminocervical complex

BACKGROUND

Trigeminal neuralgia (TN) is one of the most painful conditions that affects the trigeminal or fifth cranial nerve. The prevalence ranges from 0.03% to 0.3% of the population and the most affected branches are the maxillary (V2) and mandibular (V3) (1-3). In cases refractory to medical therapy, several surgical (microvascular decompression) or percutaneous (thermal, glycerol rhizotomy, balloon compression, radiosurgery or neurostimulation) therapies are recommended, but all lack long-term efficacy and pain can return even if the procedure is initially successful. Spinal cord stimulation of the cervical dorsal column can be an effective and advanced treatment with acceptable pain relief for TN as well as for facial pain and some types of headaches (4). This traditional stimulation can be inaccurate and result in unwanted areas of stimulation, especially for localized pain conditions, with loss of pain relief over time. Dorsal root ganglion (DRG) stimulation is more selective and allows stimulation of only the painful dermatomes with a single or multiple dedicated electrodes placed around each ganglion (5). An alternative is spinal dorsal root (SDR) stimulation, which is similar to DRG stimulation, but SDR stimulation allows for one electrode to cover multiple roots without the need for a specific material for implant (6). The dorsal or posterior is the afferent sensory root of the spinal nerve. It is one of 2 roots that emerges from the spinal cord and travels to the dorsal root ganglion; the other is the ventral or anterior or efferent motor root. The stimulation of the sensory dorsal root is based on a percutaneous lead placed in the posterior epidural space through a vertebral paramedian approach, with the tip parallel alongside the spinal cord, as in traditional stimulation, but more lateral to the dorsal column and proximal to the ganglion. SDR stimulation evokes sensory paresthesias as a tingling sensation, avoiding the motor stimulation of spinal ventral root. We report here the case of a patient with refractory secondary TN (V2-V3, left) treated with upper cervical SDR stimulation and followed over 3 years.

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CASE

A 51-year-old woman developed TN 3 months after a subtotal resection of a left cavernous sinus meningioma involving the sinus and the medial sphenoid wing, the orbit, and other areas of the middle fossa. At the first visit she complained of sudden, recurrent paroxysmal and brief daily pain attacks characterized by a sensation of electrical shocks radiating along the left V2-V3 areas, and brief facial spasms triggered by talking, eating, and touching. Magnetic resonance imaging (MRI) showed the residual postsurgical meningioma (Fig. 1). Over the following 11 months, the neuralgia became progressively resistant to 400 mg carbamazepine 3 times a day in addition to 100 mg lamotrigine twice a day and 200 mg tapentadol twice a day. She reported drowsiness, confusion, dizziness, and depression as side effects of the therapy and a progressive worsening of her quality of life with impacts on daily performance. During this period, she also underwent cyberknife radiosurgery and 2 treatments of percutaneous radiofrequency thermal rhizotomy with limited short-time clinical benefits from both. No areas of hypoanesthesia and no motor deficits were present at the clinical examination after all of these treatments. In June 2016 she agreed, after providing written informed consent, to be treated with upper cervical stimulation for a trial period to test its effects on pain control and tolerability before permanent implantation. An epidural percutaneous octopolar lead, fluoroscopically guided, was placed in the posterior epidural space through a vertebral paramedian approach at C7-T1, positioning the tip at the left upper C1-2 cervical levels, proximal to the medial interpedicular line and parallel to the spinal cord (Fig. 2). Stimulation-induced paresthesias were overlapped to the left C2-C3 dermatomes and provided immediate pain relief just after the start. Stimulation did not produce paresthesias in the V3 or V2 dermatomes but obtained facial pain relief by stimulating only the C2 and C3 dorsal spinal roots. We measured the outcomes using the Numeric Pain Rating Scale (0-100) and the Nottingham Health Profile for quality of life (primary outcomes) and reduction of drug therapy (secondary outcome). During the trial period of 3 weeks, the patient had good pain relief and all drugs were progressively tapered down to complete withdrawal. She was compliant with stimulation therapy and showed improvement in daily function, mood, and quality of life. Scores were significantly improved when compared with baseline values for all measures during follow-up (Table 1). Therefore, the permanent implant was performed. During the 3 years following implantation, the patient always reported recurrence of pain when stimulation was turned off via the self-managed personal programmer. A residual pain persisted on the first division of the fifth cranial nerve. A little lead migration was observed but without a deficit of stimulation-induced paresthesias covering the area of the left C2-C3 dermatomes and without loss of facial pain relief (Fig. 3). The foraminal fluoroscopic view allows for seeing the position of the electrode in the posterior epidural space and for the correct stimulation of SDRs confirmed by sensory-evoked paresthesias. Stimulation parameters currently reported are: amplitude 0.8 to 0.9 V, frequency 100 Hz, pulse duration 240 milliseconds, and electrode polarity: (0, -; 5, +). Over the 3 years of follow-up, we made minimal setting variations.

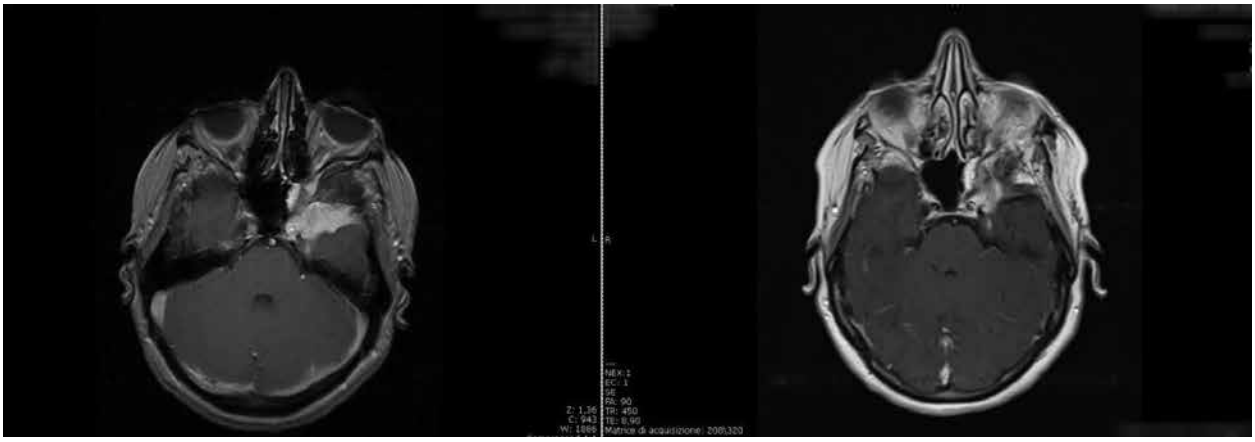

Fig. 1. MRI with pre- and post-surgical treatment of meningioma.
Abbreviations: MRI, magnetic resonance imaging

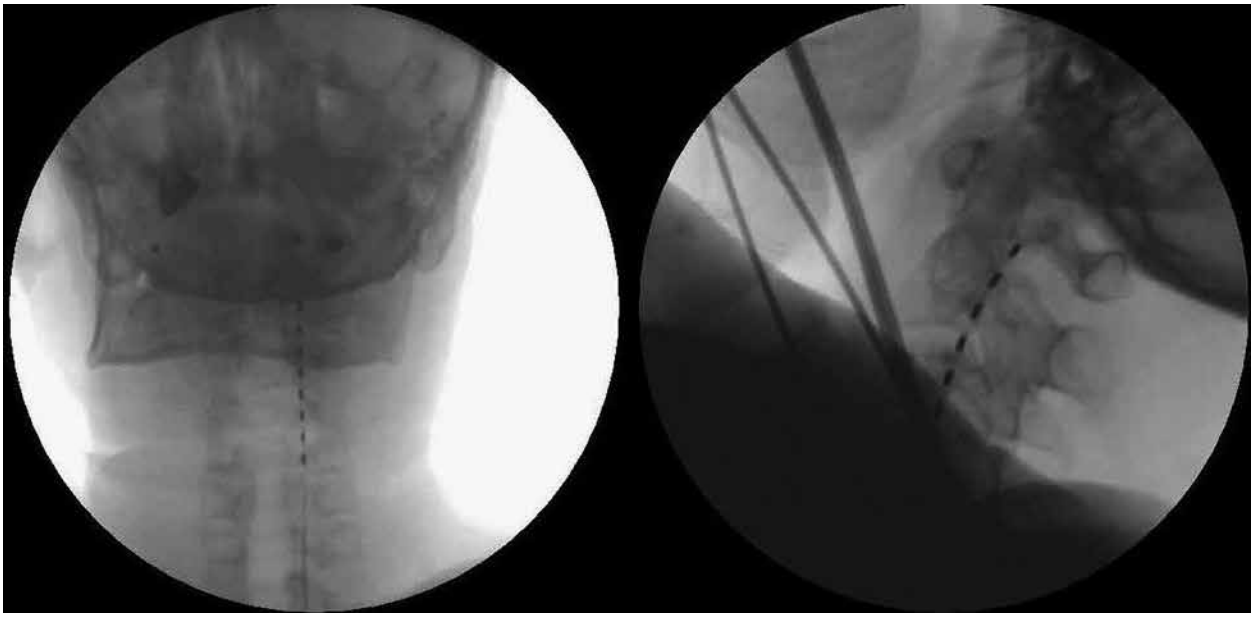

Fig. 2. Anteroposterior and lateral fluoroscopic views after trial implant.

DISCUSSION

TN is divided into primary, or classical, when there is a demonstration of neurovascular compression of the trigeminal nerve, and secondary, or symptomathic, if caused by several pathologic conditions such as intracranial space occupying lesions or multiple sclerosis (7). Medical therapy is the first-line treatment required for all of these patients (8), but refractory cases are a big challenge for physicians and the efficacy and safety of other therapies is still unclear (9). In our case report, at the time of neurostimulation choice, a second surgical treatment had been excluded and percutaneous treatments had failed. Among interventional pain procedures for trigeminal pain, neurostimulation has been reported with various nervous tissue targets such as peripheral branches, spinal cord and DRG, and with different results depending on the different modality of stimulation, conventional or high-frequency (10,11). The DRG is considered a probable mainstay of neuropathic pain and its direct stimulation can be more effective for localized pain syndromes (12). The primary sensory neurons are unique, since they have the cell body in the DRG, one centrifugal axon that lies within the peripheral nervous system, and a centripetal axon that extends into the central nervous system as a SDR and goes on toward the spinal cord, where it bifurcates into ascending and descending branches before entering at multiple levels in the dorsal horns (13). Selective stimulation of this centrally directed SDR is a suitable target to gain nociceptive modulation. We know that the dorsal column of the spinal cord mainly carries the large-diameter myelinated Aβ fibers and the non-nociceptive afferents, while the SDR also carries the thin myelinated Aδ fibers, the unmyelinated C fibers, and the nociceptive afferents that synapse in the dorsal horns and then form the contralateral spinothalamic tract. At the cervical spinal cord level there is the trigeminocervical complex (TCC) that extends from the pars caudalis of the inferior spinal trigeminal nucleus to the C1-4 spinal levels. It is an area with the convergence for nociceptive and temperature afferent inputs from the meninges and cervical structures, and

Table 1. Scores of one year of primary outcomes by a Numeric Pain Rating Scale, NPRS (0-100) and the Nottingham Health Profile for quality of life, NHP.

	Baseline	**Trial**	**2 Mos**	**1 Y**
NPRS (0-100)	100	25	30	20
NHP				
Energy Level	100	40.12	32.52	0
Pain	79.77	10.50	13.39	9.70
Emotional Reaction	84.4	43.25	30.21	21.77
Social Isolation	85.26	48.41	21.63	13.01
Sleeping	100	37.05	24.54	14.31
Physical Mobility	46.54	22.17	14.77	0
Total Score	*495.97*	*201.5*	*137.06*	*58.79*

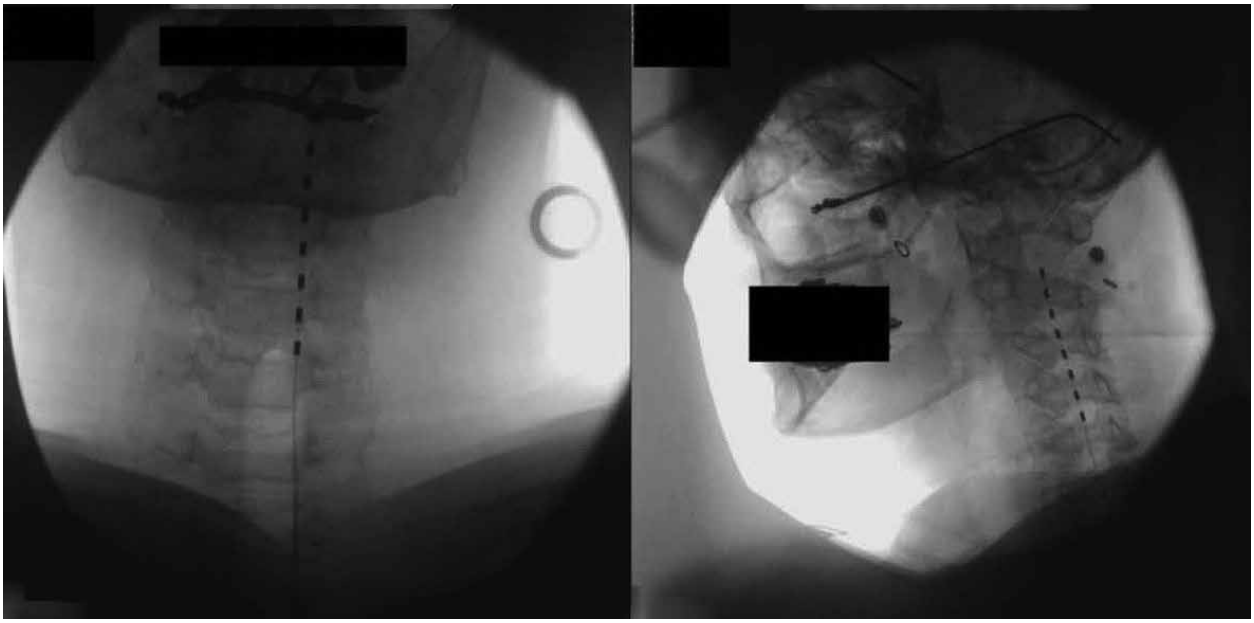

Fig. 3. Anteroposterior and foraminal fluoroscopic views after 3 years with a slight lowering of the lead tip.

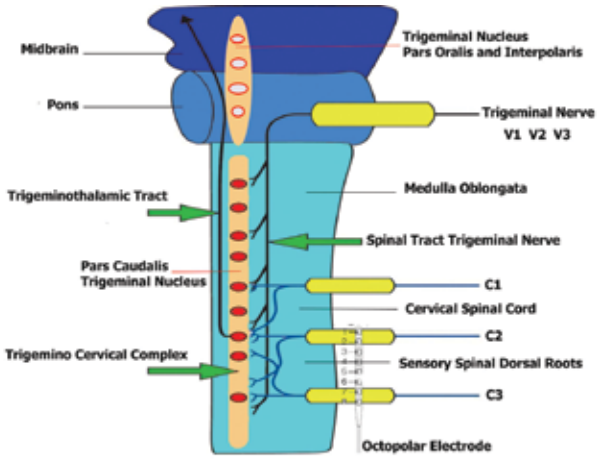

Fig. 4. Trigeminocervical complex schematic view with convergence of cervical and trigeminal nociceptive afferents.

for mono and polysynaptic nociceptive afferents from the ipsilateral face that project in part to the trigeminal nucleus caudalis and in part to the Lamina I of the upper cervical dorsal horn. Therefore, nociceptive afferents of the cervical spinal nerves and the trigeminal nerve converge, generating a bidirectional exchange of sensory information and creating a substrate for referred pain and perhaps for pain inhibition by stimulation (Fig. 4). Sensory information can ascend or descend to 3 spinal segments in the dorsolateral tract and the substantia gelatinous of the spinal cord before entering the dorsal horn, allowing input from multiple cervical regions to converge with the TCC (14). Barolat et al (15) in 1988 implanted a 42-year-old man with advanced multiple sclerosis and left V2-V3 TN with 2 percutaneous epidural electrodes at the C1-2 level, positioning one in the midline and the other to the left of midline with a good result. In our case, we implanted during the trial only one electrode to stimulate the sensory afferent dorsal roots, based on our previous experiences by this target of stimulation applied to localized neuropathic painful conditions such as complex regional pain syndrome, postthoracotomy pain, and postherniorrhaphy pain. We thought that a second electrode to stimulate the dorsal columns could be added later if good pain control was not achieved with the first. During a cervical implant the final position of the tip of the electrode can vary at the beginning and over time for a primary or a secondary malposition. In our patient, the tip was initially positioned at the left C1-2 level; after 3 years it migrated down to C2, maintaining, however, the stimulation of the same C2-3 dermatomes with an efficacious inhibition of resistant left V2-V3 trigeminal pain, perhaps due to the large convergence of cervical nociceptive afferents to the TCC. Further studies are needed to better understand the role of cervical neurostimulation in the treatment of TN.

CONCLUSION

Our case report suggests that upper cervical SDR stimulation is promising, safe, and possibly effective for treating patients who experience refractory TN. Additional data are required to determine more exact considerations of the mechanism of action of cervical SDR stimulation.

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REFERENCES

1. De Toledo IP, Conti Réus J, Fernandes M, et al. Prevalence of trigeminal neuralgia: A systematic review. *J Am Dent Assoc* 2016; 147:570-576.
2. Mueller D, Obermann M, Yoon MS, et al. Prevalence of trigeminal neuralgia and persistent idiopathic facial pain: A population-based study. *Cephalalgia* 2011; 31:1542-1548.
3. Bendtsen L, Zakrzewska JM, Abbott J, et al. European Academy of Neurology guideline on trigeminal neuralgia. *Eur J Neurol* 2019; 26:831-849.
4. Velásquez C, Tambirajoo K, Franceschini P, Eldridge PR, Farah JO. Upper cervical spinal cord stimulation as an alternative treatment in trigeminal neuropathy. *World Neurosurg* 2018; 114:641-646.
5. Deer TR, Levy RM, Kramer J, et al. Dorsal root ganglion stimulation yielded higher treatment success rate for CRPS and causalgia at 3 and 12 months: Randomized comparative trial. *Pain* 2017; 158:669-681.
6. Levine AB, Steven DA, Parrent AG, MacDougall KW. Successful long-term nerve root stimulation for chronic neuropathic pain: A real world, single center Canadian experience. *Pain Physician* 2017; 20:95-106.
7. Headache Classification Committee of the International Headache Society (IHS). The International Classification of Headache Disorders, 3rd ed. *Cephalalgia* 2018; 38:1-211.
8. Cruccu G, Gronseth G, Alksne J, et al. American Academy of Neurology Society; European Federation of Neurological Society. AAN-EFNS guidelines on trigeminal neuralgia management. *Eur J Neurol* 2008; 15:1013-1028.
9. Taha JM, Tew JM Jr, Buncher CR. A prospective 15-year follow up of 154 consecutive patients with trigeminal neuralgia treated by percutaneous stereotactic radiofrequency thermal rhizotomy. *J Neurosurg* 1995; 83:989-993.
10. Finch P, Price L, Drummond P. High-frequency (10 kHz) electrical stimulation of peripheral nerves for treating chronic pain: A double-blind trial of presence vs absence of stimulation. *Neuromodulation* 2019; 22:529-536.
11. Van Buyten JP. Trigeminal ganglion stimulation. *Prog Neurol Surg* 2015; 29:76-82.
12. Mao Q, Yuan J, Xiong M, et al. Role of dorsal root ganglion K2P1.1 in peripheral nerve injury-induced neuropathic pain. *Mol Pain* 2017; 13:1-13.
13. Barolat G. Epidural spinal cord stimulation: Anatomical and electrical properties of the intraspinal structures relevant to spinal cord stimulation and clinical correlations. *Neuromodulation* 1998; 1:63-71.
14. Luz LL, Fernandes EC, Dora F, Lukoyanov NV, Szucs P, Safronov BV. Trigeminal Aδ- and C-afferent supply of lamina I neurons in the trigeminocervical complex. *Pain* 2019; 160:2612-2623.
15. Barolat G, Knobler RL, Lublin FD. Trigeminal neuralgia in a patient with multiple sclerosis treated with high cervical spinal cord stimulation. *Appl Neurophysiol* 1988; 51:333-337.