Genc and Aydoseli
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CASE REPORT

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Computed tomography-guided trigeminal tractotomy-nucleotomy for refractory chronic craniofacial pain: a case report

Cetin Genc[1*] and Aydin Aydoseli[2]

Abstract

Background Refractory chronic craniofacial pain is a debilitating condition with limited treatment options. This case report presents the use of percutaneous computed tomography-guided trigeminal tractotomy-nucleotomy under general anesthesia in a patient with refractory trigeminal neuralgia and post-treatment anesthesia dolorosa. This case report expands the use of TR-NC to include treatment-resistant post-treatment anesthesia dolorosa and introduces real-time verification under anesthesia, potentially broadening access and improving precision, while highlighting the need for further research on long-term efficacy and patient selection for this promising technique.

Case presentation A 50-year-old woman with 12 years of chronic facial pain in the opthalmic and maxillary trigeminal branches despite multiple surgical and medical interventions underwent computed tomography-guided trigeminal tractotomy-nucleotomy under general anesthesia. Initial pain relief was significant, but symptoms gradually returned within 12 months. Transient mild ataxia was the only complication.

Conclusion This case suggests trigeminal tractotomy-nucleotomy under general anesthesia with real-time verification may be a valuable option for patients with refractory post-treatment anesthesia dolorosa. However, the transient nature of pain relief necessitates further investigation of long-term outcomes and optimal patient selection.

Keywords Neuroradiology, Neurosurgery, Radiofrequency ablation, Trigeminal neuralgia, Trigeminal tractotomy, Nucleotomy

Introduction

Recognizing that the pathway for facial pain extends to the medulla through the spinal trigeminal tract, intramedullary tractotomy has been identified as a method to alleviate facial pain while preserving touch sensation pathways to the pons [1]. Trigeminal tractotomy refers to the targeted destruction of the descending trigeminal tract, acknowledged as an effective treatment for various conditions, including malignancy-related facial pain, neuropathic facial pain, postherpetic neuralgia, glossopharyngeal neuralgia, geniculate neuralgia, and refractory trigeminal neuralgia (TN). This approach proves valuable not only for denervating trigeminal pain areas but also for addressing regions innervated by the 7th, 9th, and 10th cranial nerves [2, 3].

The initial stereotactic trigeminal tractotomies using radiofrequency (RF) thermocoagulation for TN treatment were conducted in 1967 and 1968 [4, 5]. Schvarcz further advanced this procedure, creating lesions primarily in the second-order neurons at the oral pole of the nucleus caudalis, specifically at the occipitocervical

*Correspondence:
Cetin Genc
cgenc@bwh.harvard.edu
[1] Department of Neurosurgery, Brigham and Women's Hospital, Harvard Medical School, Boston, MA, USA
[2] Department of Neurosurgery, Istanbul Medical Faculty, Istanbul University, Istanbul, Turkey

junction. This modified approach was termed trigeminal nucleotomy [6].

Our objective is to present a rare case involving the treatment of refractory TN through percutaneous trigeminal tractotomy-nucleotomy (TR-NC), guided by computed tomography (CT). This represents the first reported use of TR-NC for post-treatment anesthesia dolorosa under general anesthesia, potentially paving the way for a new treatment option for this challenging condition. Additionally, the utilization of real-time intraoperative electrode verification under general anesthesia provides a unique aspect to this case, potentially increasing the safety and accuracy of the procedure while making it more accessible to patients who may not tolerate awake procedures.

Case presentation

A 50-year-old woman was referred to our clinic due to a 12-year history of chronic, intense facial pain localized in the left ophthalmic (V1) and maxillary (V2) trigeminal branches. Despite a three-year trial of carbamazepine (max 1600 mg/day) and gabapentin (max 1200 mg/day), the patient remained unresponsive. Previous surgical interventions included microvascular decompression performed twice, balloon compression once, and gammaknife RF once. Temporary relief was achieved with botulinum toxin injections in the eye muscles. The pain, rated at 8 on the Visual Analog Scale (VAS), occurred over 5 times a day, exacerbated by fatigue and talking, and presented as throbbing, stabbing, with tingling sensations in the face and eyes.

Neurological examination revealed hypoesthesia and neuropathic pain distributed through the ophthalmic and maxillary nerve areas. Magnetic resonance imaging (MRI) showed a postoperative Teflon implant due to microvascular decompression in the perimesencephalic cisterns, under the trigeminal nerve tract, along with a 3-mm diameter craniectomy defect extending on the left retrosigmoid area. After 12 years of incapacitating headaches, the patient was referred to our neurosurgery clinic.

Initial attempts at pain relief through motor cortex stimulation proved ineffective. Subsequently, an alternative approach, TR-NC, guided by CT was adopted.

TR-NC technique

30 min before inducing general anesthesia, 12 ml of 300 iohexol (Omnipaque, GE Health Care) contrast agent was injected into the subarachnoid space through lumbar puncture. Electrophysiological monitoring of the 9th, 10th, 11th, and 12th cranial nerves involved placing electrodes in all four extremities, the tongue, and the trapezius muscle to assess somatosensory and motor evoked potentials. The patient assumed a prone position with the head in flexion, and the neck was secured with a DORO (Pro Med Instruments) radiolucent skull clamp, allowing intraoperative CT scanning of the upper cervical spine.

Guided by a CereTom CT scanner (NeuroLogica Corp.), a 20 Gauge spinal needle was percutaneously inserted 2 cm lateral at the occipito-cervical region, visualized by CT (Fig. 1a). A thin, 1.25-mm-wide field-of-view CT scan measured the skin-to-dura distance and the diameters of the upper cervical spinal cord. The target, two-thirds of the distance from the midline to the equator of the spinal cord, was marked on the introducer metal cannula for the Nashold Thermocouple dorsal root entry zone (DREZ) electrode system (Cosman Medical). The radiofrequency needle electrode depth was set at 3 mm from the target. Correct electrode localization was confirmed in the subarachnoid space and

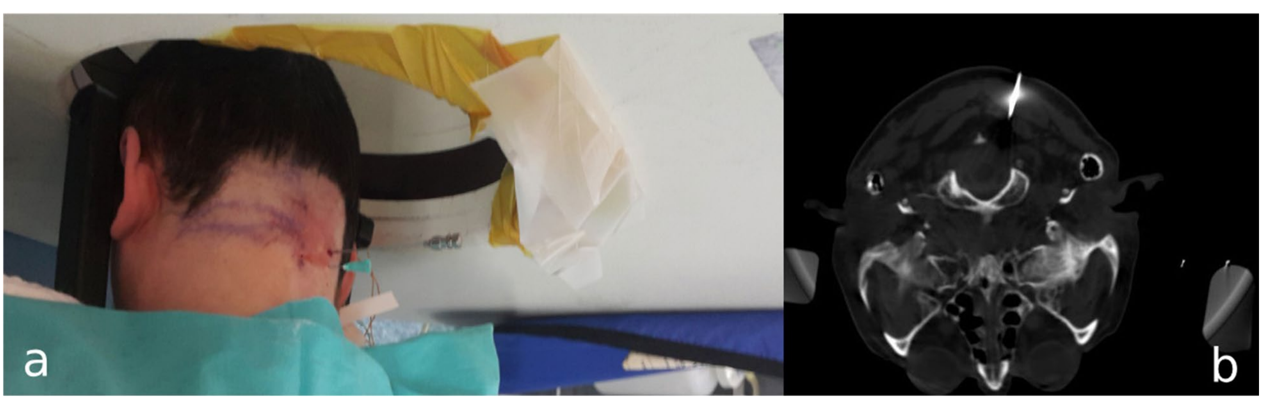

Fig. 1 **a** A 20-gauge spinal needle accurately placed through the into the subarachnoid space at the occipito-cervical region for percutaneous trigeminal tractotomy-nucleotomy. **b** Axial CT scan confirms the correct placement of the electrode within the targeted area of the spinal cord for trigeminal tractotomy-nucleotomy

spinal cord through impedance measurements using a Radionics RFG-3C generator (300–400 W and 1.4 KW, respectively). Axial CT sections and electrophysiological evaluations verified the accuracy of electrode positioning (Fig. 1b).

Functional confirmation of the target was achieved through electrical stimulation using the Levin-Cosman cordotomy electrode at approximately 0.1 V. After stimulation, a single classical RF-lesion was applied at 90 °C for 90 s. The patient was then awakened from general anesthesia and extubated without complications, although she did experience mild ataxia for seven days.

Postoperative MRI confirmed the hyperintense lesion on T2 sequence in the targeted area (Fig. 2). Initially, the patient reported significant pain relief, with her VAS score improving to 0 (no pain) in the V1 region and 2 (minimal pain) in the V2 region. However, the pain gradually returned to preoperative levels within 12 months.

Discussion

Lesioning of the descending trigeminal tractus and the nucleus caudalis is recommended for patients with anesthesia dolorosa, atypical facial pain, secondary neuropathy (post-traumatic or post-surgical), and malignancy-related facial pain [1, 4, 5, 7, 8]. Various surgical treatments, including peripheral neuromodulation, spinal cord stimulation, TR-NC, nucleus caudalis DREZ ablation, and motor cortex stimulation, have been explored for complex craniofacial pain.

TR-NC, involving lesioning the descending spinal trigeminal tracts in the medulla along with the nucleus caudalis, is advocated as a valid, highly efficient, less-invasive procedure. It has been suggested as a satisfactory treatment for medically and surgically failed, intractable TN [9]. The described case involves a patient with refractory TN who underwent repeated surgical and medical treatments without success, presenting severe neuropathic facial pain akin to anesthesia dolorosa.

So far, in the twenty-first century, only few papers have been published on the outcomes of trigeminal nucleotractotomy. The largest series of CT-guided tractotomy-nucleotomy procedures have been reported by Kanpolat et al. [8] and Raslan [11]. Moreover, to authors' knowledge, there is only one report about the preliminary results of general anesthesia for this procedure [7].

The percutaneous CT-guided TR-NC with a special electrode system was firstly described by Kanpolat et al. [10] in which the technique was comprehensively explained. They admitted the TR-NC with the awake patient as an applicable procedure under local anesthesia by intraoperative imaging and neurological confirmation. However, in our case, the procedure was unlikely to be performed under local anesthesia since patient declined such a procedure. Conscious sedation or monitored anesthesia are another options; yet, providing anatomical guidance by prone positioning and preventing any head movement which could cause complications such as hemiparesis or hemiplegia would add safety to the procedure. Since there are limited treatment options to reduce or relieve the debilitating pain caused by anesthesia dolorosa, clinicians may find TR-NC under general anesthesia as an efficient option to uncooperative patients. In addition, under general anesthesia, the real-time intraoperative verification of the electrode position before concluding the surgery is advantageous.

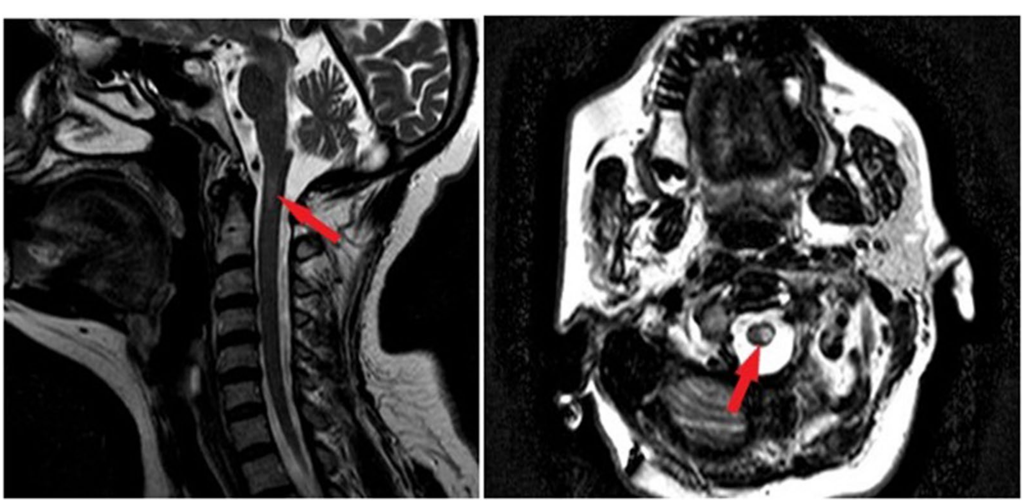

Fig. 2 T2-weighted MRI sequence shows a hyperintense lesion (red arrow) in the targeted area of the medulla oblongata, confirming successful electrode placement and lesion formation after trigeminal tractotomy-nucleotomy

"Increasingly, intraoperative CT technology is being embraced for its ability to minimize intra-patient transfers, streamline labor, and offer cost-effective solutions. Throughout this procedure, the numerous benefits of utilizing mobile intraoperative CT have become apparent, highlighting its value and efficiency. One of the pivotal advantages of employing intraoperative CT lies in its ability to facilitate surgical interventions while maintaining the patient's stability with a three-pin holder skull clamp. Intraoperative CT systems is the utilization of spiked heads specially designed to eliminate artifacts, particularly tailored for mobile CT setups. Consequently, this ensures that the surgical field remains free from obstruction, enabling the procedure to be conducted under sterile conditions with utmost clarity and accuracy. Unlike traditional CT devices, intraoperative CT obviates the need for patient transfer, streamlining the surgical process and mitigating the associated risks. The advent of portable CT devices has further amplified the benefits of intraoperative imaging, significantly optimizing both time and labor resources. In conclusıon, with its ability to provide real-time imaging guidance, mitigate complications, and minimize the need for patient transfers, intraoperative CT provide excellence in these similar medical procedure.

The optimal efficacy of the DREZ operation manifests predominantly in patients grappling with deafferentation pain, typified by conditions like brachial plexus avulsion and spinal trauma. Conversely, individuals afflicted with peripheral nerve pain, such as those experiencing postrhizotomy syndrome and stump pain, tend to exhibit suboptimal outcomes. The theoretical underpinning suggests that deafferentation pain implicates a perturbation in the secondary sensory neurons constituting central pain pathways, precisely the neural entities targeted and eradicated by DREZ radiofrequency lesions. In the realm of facial pain alleviation, distinct patterns emerge. Pain emanating from trigeminal dysesthesia and peripheral facial injury, emblematic of peripheral involvement, demonstrates a less pronounced amelioration compared to pain rooted centrally—specifically, in pathological lesions of the brain stem (e.g., multiple sclerosis or thrombosis) [11–13]. Gurbani et al. reported a case involving multiple interventions, including gamma knife radiotherapy, percutaneous balloon trigeminal rhizotomy, and microvascular decompression. The patient returned to baseline pain levels at 1 month post-tractotomy [14]. Anisimov et al. presented a case with minimal pain reduction (VAS reduction from 10 to 8) after percutaneous nucleotractotomy for deafferentiation pain following a surgical attempt to treat trigeminal neuralgia [15]. While Kanpolat et al. reported an 80% success rate based on VAS scores in their series of failure trigeminal neuralgia patients, the long-term outcomes were not specified. In contrast, Raslan et al., in their extensive study involving 17 patients, noted 7 cases of failure and recurrence occurring approximately 5.6 months post-treatment [8, 16]. Our patient experienced only a temporary improvement following tractotomy. Despite initial relief, the symptoms recurred after a short period, highlighting the complexities and limitations associated with addressing postrhizotomy syndrome through tractotomy.

The occurrence of transient mild ataxia in our patient, which resolved within the second day postoperatively, raises considerations regarding the potential complications associated with trigeminal medullary tractotomy. Ataxia, resulting from an injury to the spinocerebellar tract and dorsal columns, is a recognized complication of this procedure, with a reported incidence ranging from 30 to 39% for ataxia involving both upper and lower extremities and 17% to 61% for ataxia limited to the upper extremities [17]. Although our patient experienced a relatively mild form of ataxia, its transient nature and resolution within a short period are encouraging aspects.

The recovery of the patient's complaints after the operation but recurring symptoms at six months post-surgery raise questions about the long-term benefits and necessitate a consideration of pros and cons. Future studies with larger cohorts and longer follow-ups are needed to evaluate the long-term efficacy and safety of TR-NC under general anesthesia for diverse facial pain types. Additionally, understanding factors influencing treatment response and developing better prognostic indicators could optimize patient selection and treatment outcomes.

Conclusion

The informative features of this case among the others are the efficacy of the procedure in relieving post-treatment anesthesia dolorosa and, the provision of real-time intraoperative verification of the electrode position with the anesthetized patient. On the basis of our case, it would appear reasonable to conclude that trigeminal tractotomy-nucleotomy, utilizing intraoperative CT guide, in fully anesthetized patients and under neuromonitorization may increase the safety of the TR-NC treatment to manage refractory trigeminal neuralgia and deserve further investigation. Given the restricted availability of clinical studies and the essential requirement for extended follow-up data on TR-NC outcomes in existing literature, each new case assumes significance in contributing to the documentation of similar situations.

Abbrevations

TR-NC	Trigeminal tractotomy-nucleotomy
TN	Trigeminal neuralgia
RF	Radiofrequency

VAS Visual analog scale
MRI Magnetic resonance imaging
DREZ Dorsal root entry zone

Supplementary Information

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Supplementary Material 1

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CG: Data collection, data analysis, manuscript writing. AA: Data collection, data analysis, manuscript review.

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Declarations

Ethics approval and consent to participate
Written informed consent for participation and publication was obtained from the patient.

Consent for publication
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Competing interests
The authors declare that they have no competing interests.

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