Trigeminal neuralgia induced by brainstem infarction treated with pontine descending tractotomy: illustrative case

Rachyl M. Shanker, MD,1 Miri Kim, MD, PhD,1 Chloe Verducci, BA,1 Elhaum G. Rezaii, MPH,1 Kerry Steed, BA,1 Atul K. Mallik, MD, PhD,2 and Douglas E. Anderson, MD1

Departments of 1Neurological Surgery and 2Radiology, Stritch School of Medicine, Loyola University Medical Center, Maywood, Illinois

BACKGROUND While cases of trigeminal neuralgia induced by a brainstem infarct have been reported, the neurosurgical literature lacks clear treatment recommendations in this subpopulation.

OBSERVATIONS The authors present the first case report of infarct-related trigeminal neuralgia treated with pontine descending tractotomy that resulted in durable pain relief after multiple failed surgical interventions, including previous microvascular decompressions and stereotactic radiosurgery.

A neuronavigated pontine descending tractotomy of the spinal trigeminal tract was performed and resulted in successful pain relief for a 50-month follow-up period.

LESSONS While many cases of ischemic brainstem lesions are caused by acute stroke, the authors assert that cerebral small vessel disease also plays a role in certain cases and that the relationship between these chronic ischemic brainstem lesions and trigeminal neuralgia is more likely to be overlooked. Furthermore, neurovascular compression may obscure the causative mechanism of infarct-related trigeminal neuralgia, leading to unsuccessful decompressive surgeries in cases in which neurovascular compression may be noncontributory to pain symptomatology. Pontine descending tractotomy may be beneficial in select patients and can be performed either alone or concurrently with microvascular decompression in cases in which the interplay between ischemic lesion and neurovascular compression in the pathophysiology of disease is not clear.

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KEYWORDS trigeminal neuralgia; neuropathic pain; brainstem; cerebral small vessel disease; stroke; ischemia; infarct

Trigeminal neuralgia (TN) most commonly presents as neuropathic pain secondary to neurovascular compression (NVC) at the nerve root entry zone (NREZ)1,2 or as a result of a space-occupying lesion impacting the trigeminal nerve.3 However, there exists a subset of patients in whom trigeminal pain is induced by brainstem ischemia, interrupting the NREZ, spinal trigeminal nucleus, or descending spinal trigeminal tract (SpTV).

While cases of TN induced by a brainstem infarct have been reported, there are no clear treatment recommendations for this subpopulation. Here, we present a case of infarct-related TN treated with pontine descending tractotomy (PDT) that resulted in durable pain relief following multiple failed surgical interventions.

Illustrative Case

Presentation

The patient was a 61-year-old man with a medical history of hypertension; remote mandibular fracture 45 years earlier; and a 30-year history of medically refractory, debilitating right-sided facial pain in the V2 and V3 distributions. He presented with type 1 pain, triggered by light touch, chewing, and swallowing, for which conservative management with carbamazepine, gabapentin, and duloxetine had failed. His preoperative Barrow Neurological Institute (BNI) pain score was V. He had undergone three prior interventions, including microvascular decompression (MVD) for identified NVC in 1998, MVD with PDT in 2003, and stereotactic radiosurgery in 2015 with only temporary pain relief.

Abbreviations

BNI = Barrow Neurological Institute; CSVD = cerebral small vessel disease; MRI = magnetic resonance imaging; MVD = microvascular decompression; NREZ = nerve root entry zone; NVC = neurovascular compression; PDT = pontine descending tractotomy; SpTV = spinal trigeminal tract; TN = trigeminal neuralgia.

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Operative Intervention

With the patient under general anesthesia, a left-sided curvilinear ret-
roofaicular incision was made. The dura was opened, and the trigeminal
nerve was visualized without evidence of NVC. A PDT was performed
just inferior to the NREZ using a stereotactically guided 7-mm right angle
probe. The right-angled microdissector fitted with neuronavigation was
oriented in the plane parallel to the trigeminal nerve as it entered the brain-
stem and was advanced dorsomedially to the main trigeminal sensory
nucleus to a depth of 7–9 mm with a 3-mm axial movement of the guided
microdissector along the interior intrapontine trigeminal nerve pathway
(Fig. 2), disrupting the SpTV superior to its descent as a compact tract.\(^4\)

Postoperative Course

The patient tolerated the procedure well without any postopera-
tive complications and was discharged to home on postoperative
day 3 with complete resolution of his trigeminal pain. At his first fol-
low-up, he had a BNI pain intensity score of 1 with lasting durable
pain management at 50 months postoperatively.

Discussion

Observations

Retrospective review of this patient’s imaging studies demonstrated a
dorsolateral pontomedullary hyperintensity that aligned with the descend-
ing spinal nucleus of the trigeminal nerve.\(^5\) While the patient was found to
have NVC during his first two MVDs, it is clear that decompression alone
did not resolve his pain, which allows us to question the role of NVC in his
symptomatology. Furthermore, NVC was not identified at his most recent
right-side craniotomy for PDT, even though there was persistence of pain. The
ischemic lesion likely affects the spinal nucleus of the trigeminal nerve,
and possibly the SpTV, contributing to his symptomatology. PDT disrupts
the descending SpTV, which successfully managed the pain symptoms
of this patient. While the earlier PDT did not result in lasting pain relief,
we identified several factors to explain a successful second attempt:
refinement of the operative technique, improved neuronavigation, and
the implementation of diffusion tensor imaging and tractography all
improved the likelihood of success for this patient at the time of his sec-
ond PDT.

We must also consider the possibility that this ischemic lesion was
not present upon his initial presentation and was instead caused by
perforator occlusion during one of his previous microsurgical interven-
tions. While possible, we find this less likely for the following reasons.
First, the hyperintensity was visualized near the level of the pontome-
dullary junction, quite inferior to the NREZ and previously manipulated
superior cerebellar artery. Second, this patient had a history of hyper-
tension and magnetic resonance imaging (MRI) evidence of supra-
and infratentorial CSVD. While this does not ensure causality, it sup-
ports the likelihood of CSVD as the underlying etiology.

Lessons

As seen in this study, CSVD is often an incidental finding of neuroim-
aging for alternative indications, and we argue it may be an overlooked
contributor to trigeminal neuropathic pain. Our preliminary institutional ret-
rospective data on the rates of CSVD among patients who underwent
microsurgical management of TN revealed that 30 (31%) of 97 patients
had evidence of CSVD on preoperative MRI as determined by neuroradi-
ologists. Consequently, we question whether CSVD with white matter
ischemic lesions affecting the SpTV or thalamocortical pathways contrib-
utes to TN symptomatology, either in combination with NVC or alone,
more often than traditionally thought.
In cases in which patients demonstrate evidence of brainstem ischemic lesions along the trigeminal pathway in addition to NVC, the degree to which either contributes to pain symptomatology may be questioned. PDT is a viable option to treat lesional brainstem causes of TN and can be performed concurrently with an MVD, allowing the surgeon to sever the descending nociceptive fibers of the SpTV during an open craniotomy. We previously reported that 37 (74%) of 50 patients, including those with both typical and atypical trigeminal pain and those with multiple sclerosis, had successful outcomes at the latest follow-up (BNI I–IIIA).4

In conclusion, we urge authors to routinely assess for brainstem lesions along the trigeminal pathway, even when potentially causative NVC is readily identified. CSVD is a common condition and can result in infarcted lesions less obvious than those of an acute stroke. PDT may be an effective treatment option in patients with combined lesional pathology and NVC because MVD and PDT can be performed during the same craniotomy. Alternatively, PDT can be performed alone at the discretion of the surgeon.

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Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions
Conception and design: Anderson, Shanker. Acquisition of data: Anderson, Shanker. Analysis and interpretation of data: Anderson, Shanker, Kim, Mallik. Drafting the article: Anderson, Shanker, Kim, Verducci, Steed. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Anderson. Administrative/technical/material support: Anderson, Mallik. Study supervision: Anderson. Figure design and revision: Mallik.

Correspondence
Douglas E. Anderson: Loyola University Medical Center, Maywood, IL. dander1@lumc.edu.