Case Report

Nucleus caudalis lesioning: Case report of chronic traumatic headache relief

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Abstract

Background: The nucleus caudalis dorsal root entry zone (DREZ) surgery is used to treat intractable central craniofacial pain. This is the first journal publication of DREZ lesioning used for the long-term relief of an intractable chronic traumatic headache.

Case Description: A 40-year-old female experienced new-onset bi-temporal headaches following a traumatic head injury. Despite medical treatment, her pain was severe on over 20 days per month, 3 years after the injury. The patient underwent trigeminal nucleus caudalis DREZ lesioning. Bilateral single-row lesions were made at 1-mm interval between the level of the obex and the C2 dorsal nerve roots, using angled radiofrequency electrodes, brought to 80°C for 15 seconds each, along a path 1 to 1.2 mm posterior to the accessory nerve rootlets. The headache improved, but gradually returned. Five years later, her headaches were severe on over 24 days per month. The DREZ surgery was then repeated. Her headaches improved and the relief has continued for 5 additional years. She has remained functional, with no limitation in instrumental activities of daily living.

Conclusions: The nucleus caudalis DREZ surgery brought long-term relief to a patient suffering from chronic traumatic headache.

Key Words: Dorsal root entry zone, headache, nucleus caudalis, trauma, trigeminal



INTRODUCTION

Every year, 1.7 million people in the United States suffer traumatic brain injuries.^[6] Following these injuries, headache is the most common symptom,^[2,22] affecting 57.8% of those injured.^[17] Even 3 years after a severe head injury, chronic headaches can persist for 23% of victims.^[4] Trauma-related headaches develop after falls and motor vehicle accidents,^[6] but are also described among military personnel following combat injuries.^[1,5,12,25,27] The pain may be unilateral and throbbing as in a migraine, bandlike and dull as in a tension headache, or a blend of these patterns.^[33] When severe, this type of headache can affect a person's ability to maintain employment and enjoy social relationships.^[2]

The prognosis of post-traumatic headache is generally good: many patients improve within months of their injury.^[29] Early on, post-traumatic headaches may be accompanied by other symptoms of the post-traumatic syndrome such as dizziness, fatigue, difficulty concentrating, and changes in personality.^[32] Among patients who experience the post-traumatic syndrome, about 80-90% can resume work duties within 2 years.^[21]

Treatment with tricyclic antidepressants and psychological counseling may aid recovery.^[22]

Although the majority of patients experience a tolerable recovery, some develop medically intractable and incapacitating headaches. Such patients may be referred for pain management consult. The evaluation may be obscured by psychological factors and issues of potential secondary gain. While recognizing the complexity and diverse etiology of post-traumatic headaches,^[11] we present what we believe is the first journal report of trigeminal nucleus caudalis dorsal root entry zone (DREZ) lesioning for the long-term relief of a chronic trauma-related headache.

CASE REPORT

A 40-year-old woman was referred to our clinic with a 3-year history of chronic bi-temporal headaches. Her headaches developed after she was ejected from a motor vehicle. The injury resulted in a temporary loss of consciousness, scalp laceration, and skull fracture without cerebrospinal fluid leak. She recovered after an 8-day admission at an outside hospital. Her memory of events was limited for 1 month, preventing her from recalling the headache's onset more precisely than "within a month of the injury." She had no prior history or family history of headaches. Her headache intensity was rated as 8-9/10, and these occurred on over 20 days per month. The headaches were exacerbated by bending forward, loud noise, and sunlight. She denied aura, visual symptoms, or food triggers. She had occasional nausea without vomiting. The pain character was described as throbbing, stabbing, and piercing. She felt a tightening pressure over a bi-temporal band-like distribution. She did not have any numbress, tingling or weakness in the face or extremities. She ambulated without difficulty and was otherwise neurologically intact. Her MRI did not show any significant intracranial abnormalities. Post-traumatic headaches must develop within seven days of an injury to meet criteria set by the International Classification of Headache Disorders,^[10] but considering our patient's memory limitation and 8-day admission with a skull fracture, her headache was reasonably classified as posttraumatic. A differential diagnosis would also include new-onset tension or migraine headache.

She failed treatments with acetaminophen, butalbital, caffeine, celecoxib, clonazepam, dihydroergotamine, gabapentin, hydrocodone, nadolol, oxycodone, paroxetine, and sumatriptan. With each medicine trial, she was followed closely by her neurologist to reduce the risk of developing medication overuse headache. She tried bilateral C_2 ganglion blocks without relief. She achieved only temporary relief with botulinum toxin injections in the temporalis muscle. After 3 years of incapacitating chronic trauma-related headaches, and

with no pending litigation settlements, she was referred to our neurosurgery clinic for evaluation of her headache and consideration for bilateral radiofrequency trigeminal nucleus caudalis DREZ lesioning.

Surgical technique

The surgical exposure required a suboccipital craniectomy and C, hemilaminectomy. The dura was opened. Singlerow serial lesions were made at 1-mm intervals along the trigeminal nucleus caudalis using El-Naggar-Nashold angled radiofrequency thermocoupled DREZ electrodes (Cosman Medical®, Burlington, MA). The electrodes were brought to 80°C, for 15 s per lesion. The row began 1 mm below the level of the obex [Figure 1]. Using an electrode with 0.8 mm of proximal insulation and 1.8 mm of distal exposed tip, six serial lesions were made in the brainstem along a path that was 1 to 1.2 mm posterior to the exiting point of the accessory nerve rootlets. This entry point positioned the electrode near the center of the nucleus caudalis.^[26] Then, an electrode with 0.6 mm of insulation and 1.6 mm of exposed tip was used to make four more lesions along the same path, which proceeded toward the exiting position of the C₂ dorsal nerve roots. Then, three lesions were made with an electrode with 0.6 mm of insulation and 1.2 mm of exposed tip, followed by three more lesions made with an electrode with 0.6 mm of insulation and 0.8 mm of exposed tip. Because the symptoms were bilateral, lesions were also made on the opposite side.

Surgical outcome and postoperative neurological exam

Following surgery, her headache pain was immediately improved and she had no ataxia. Over the next 5 months, she had occasional discomfort behind her left eyebrow and along the left side of her nose. Both symptoms improved with gabapentin.



Figure 1: Schematic of bilateral single-row nucleus caudalis DREZ lesions. The electrodes have an exposed metal tip (light gray) and a portion of insulation (black). Electrode tip and insulation lengths decreased according to nucleus caudalis size and depth

Neurological exam at 5 months after surgery revealed normal sensation to touch along the face, but no sensation to pin prick in the V₁ distributions, bilaterally. Pin prick was sensed at half intensity along the V₂ distributions, and as 75% of normal in the V3 distributions. Perception of ice as cold was absent in the V1 distributions, less than normal in the V₂ distributions, and normal in the V₃ distributions. Perception of coffee as warm in the mouth and throat was normal bilaterally. Sensation to pin prick was normal at the earlobes and in the occipital C_2 and C_3 distributions. Vibration, temperature, and pin prick sensations were normal in all extremities. Position sense and motor strength were normal in all extremities. Her gait was normal with no difficulty walking quickly or turning. She felt her headache symptoms were 90% improved and she was glad that she had undergone surgery.

Two years after surgery, her headaches began to redevelop, especially around the left eye and forehead. Over the next 3 years, she was referred for neurology consultation to maximize nonsurgical options. Despite neurology-guided headache treatment, her headache intensity was rated as 9-10/10 on at least 24 days per month. At this time, she underwent a repeated bilateral nucleus caudalis DREZ procedure.

On her first 3 postoperative days, she had transient ataxia and mild dysmetria of the left arm. She was discharged on the fourth day after surgery with no headache, no facial pain, and only minimal ataxia. Three months after surgery, she denied frontal or parietal headache. She was weaned off all pain medication. Seven months after surgery, she was neurologically intact except for a lack of sensation to pin prick in the V₁ and V₂ distributions. Pin prick was perceived in the V₃ distributions. She could distinguish touch, hot, and cold in all trigeminal distributions. She had no imbalance in her gait.

She continued to do well until her last follow-up (nearly 5 years since her second surgery, 10 years since her first surgery, and 13 years since her injury). She recently reported only one or two short headaches per month, which were of an intensity of 1/10 and localized to the temple and occipital regions. She was not using any pain medication other than acetaminophen. In reflection, she is extremely glad that she underwent this surgery. She has remained very functional and can carry out instrumental activities of daily living without difficulty.

DISCUSSION

Anatomy of the trigeminal pain pathway

First-order trigeminal sensory neurons enter the brainstem at the pons [Figure 2]. Neurons carrying motor and touch signals connect to nuclei within the pons, but neurons that carry pain signals descend to the cervicomedullary junction as the spinal trigeminal tract.^[18] The first-order neurons of the spinal trigeminal tract synapse to the deeper second-order neurons of the nucleus caudalis [Figures 2 and 3]. These second-order neurons then cross the midline and carry pain signals to the ventroposteromedial nucleus of the thalamus via the ventral trigeminothalamic tract.^[23] The pain signals are then carried to the sensory cortex by third-order thalamocortical neurons.

History of dorsal root entry zone lesioning

The nucleus caudalis DREZ procedure is an extension of a concept pursued by Sjoqvist in 1937.^[8,31] At that time, trigeminal pain was treated by sectioning the trigeminal nerve, as popularized by Frazier.^[7] Sjoqvist wanted to



Figure 2: Trigeminal pain pathway. From the pons, trigeminal pain signals travel to the cervicomedullary junction via the spinal trigeminal tract, which synapses to the deeper second-order nucleus caudalis. The ventral trigeminothalamic tract carries signals to the contralateral thalamus



Figure 3: Axial brainstem schematic, 8 mm below obex, posterior shown upward.Trigeminal Tract (TT),Trigeminal Nucleus Caudalis (NC), Accessory Nerve rootlet (XI), Dorsal Spinocerebellar Tract (DSC), Fasciculus Cunneatus (FC), Pyramidal Decussation (PD), Corticospinal Tract (CS). DREZ electrode superimposed

block the trigeminal pain pathway without causing the loss of touch sensation, and subsequent corneal ulceration injury which could occur after sectioning the trigeminal nerve. Understanding that the facial pain pathway descended to the medulla as the spinal trigeminal tract, he used intramedullary tractotomy to relieve facial pain and spare touch sensation pathways to the pons.^[31]

Using trigeminal tractotomy, Kunc found that lesioning the descending trigeminal tract near the level of the obex interrupted the first-order afferents that supplied both the rostral and caudal portions of the nucleus caudalis. A rostral tractotomy would result in analgesia of the entire face, while more caudal lesions only resulted in analgesia of the periphery of the face. The finding correlated with the Déjerine "onion peel" model for the segmental mapping of the nucleus caudalis, in which rostral levels convey central face pain sensation and caudal levels convey peripheral face pain sensation.^[15]

Trigeminal tractotomy was further advanced by Crue and Hitchcock who independently designed stereotactic techniques to perform the tractotomy percutaneously.^[3,11] In their techniques, a single lesion was made by a radiofrequency electrode. While some patients received relief with the procedure, the overall surgical results were inconsistent.^[3]

In 1972, Sindou developed a DREZ lesioning technique for arm pain associated with Pancoast-Tobias syndrome.^[30] It was later used for deafferentation pain following brachial plexus avulsion injuries based upon the idea that denervation pain could develop from the secondorder neurons of the substantia gelatinosa.^[20] Schvarcz recognized the functional and structural similarity between the substantia gelatinosa of the spinal cord and the trigeminal nucleus caudalis of the cervicomedullary junction.^[28] Applying the principle of DREZ lesioning, Schvarcz began performing single lesion radiofrequency trigeminal nucleotomy.^[28] In 1982, Nashold used an open technique with radiofrequency DREZ electrodes to target the second-order neurons of the trigeminal nucleus caudalis and block the trigeminal pain pathway.^[8]

Nashold's technique differed from Schvarcz's in that he made a series of lesions to cover the length of the nucleus caudalis rather than a single percutaneous lesion. An advantage of Nashold's open technique was that brainstem landmarks could guide lesion placement. Additionally, a range of the nucleus could be targeted, addressing deafferentation pain signals that could be emitted from the nucleus caudalis at levels above or below where a percutaneous electrode could be placed.

The nucleus caudalis DREZ procedure is similar to spinal DREZ procedures, but it does not involve lesioning the pons at the location of the trigeminal nerve entry. Instead, a series of lesions are made below the level of

the fourth ventricle and extending to the C_2 dorsal nerve roots. Many variations of the DREZ lesioning technique have since been used, including using laser^[16] energy, using ultrasonic energy,^[9] and broadening the nucleus caudalis lesion width by using a double-row of lesions.^[24] Kanpolat innovated a CT guided percutaneous technique for single nucleotomy lesion placement.^[13] In 1994, El-Naggar modified the radiofrequency electrode design by angling electrodes that had proximal insulation.^[19]

Surgical indications for nucleus caudalis dorsal root entry zone lesioning

The nucleus caudalis DREZ surgery has primarily been used to improve deafferentation facial pain, as exists in post-herpetic neuralgia and anesthesia dolorosa.^[14,18] Nucleus caudalis lesioning has also been used for pain related to craniofacial and oral cancer.^[13] Because it was recognized that some headaches also follow the trigeminal pain pathway, the DREZ surgery has been offered to select patients with intractable migraine and cluster headaches. In 1994, Nashold reported a patient with migraine-cluster headaches who achieved good pain relief for several months, but then experienced a pain recurrence.^[19] In 1996, Nashold reported two patients with cluster headaches; one achieved pain relief for 6 years after surgery and was then lost to follow-up, the other had fair relief.^[18] In 2008, Kanpolat reported a patient's complete relief of cluster headache after percutaneous nucleotomy.^[13] Because the DREZ procedure is invasive and carries risks of neurological injury, all patients should first have maximized medical management options. Neuropsychiatric evaluation may also be indicated to enable multidisciplinary care and to guide prudent patient selection. The DREZ surgery can be considered only if a patient has uncontrolled, incapacitating, chronic pain despite less invasive treatments.

Surgical risks and technique considerations

Caudal to the obex, lesions are not expected to affect sensation to touch or affect trigeminal motor function. In older literature, the most common complication was ataxia, attributed to lesion extension to the dorsal spinocerebellar tract.^[8] Electrode design modifications added a segment of proximal insulation to spare the dorsal spinocerebellar tract. With these insulated electrodes, postoperative ataxia often resolves within a few days of surgery; however, permanent ataxia remains a risk that should be discussed with the patient before surgery.

The corticospinal tract is another neighboring structure that is vulnerable to injury [Figure 3]. Because the surface depth of the nucleus caudalis becomes more shallow at the level of C_2 , in comparison to the level of the obex,^[26] we use electrodes of decreasing length. Additionally, the electrodes are designed with a thickened hub to prevent

excessive penetration into the brainstem. Despite these safety measures, the risk of permanent weakness due to corticospinal tract injury exists.

We use the open DREZ technique because it allows visualization and protection of brainstem surface blood vessels and accessory nerve rootlets. These rootlets serve as an external landmark to localize the nucleus caudalis.^[26] The open technique may increase the potential risk for cerebrospinal fluid leak or wound infection, but we believe that the visualization of these structures adds safety for the placement of the electrode.

The patient must be cautioned of the possibility of pain recurrence after surgery. If pain recurs, the median time for recurrence is 3 months after surgery.^[18] As demonstrated by this case example, a repeated surgery for patients with recurrent pain may re-establish pain relief. Because of the risk of damage to neighboring structures, we were cautious not to make too large of an initial lesion path; we used a single-row of lesions, made with a series of progressively shorter electrodes. While this caution may have limited the long-term effectiveness of the first surgery, we believed that the safety afforded was worthwhile. The second surgery more completely blocked the trigeminal pain pathway, although it was performed in the same fashion as the first.

CONCLUSIONS

After medical management failed to relieve our patient's trauma-related headaches, relief was achieved with the nucleus caudalis DREZ procedure. Pain relief after DREZ lesioning can be significant and long lasting. This procedure is not recommended for all patients with headaches, but in the literature, and in our experience, it has been effective for other patients that suffered from incapacitating and intractable cluster and migraine headaches.^[18,19] Because of the potential risk of neurologic injury during the surgery, it should only be performed by neurosurgeons who have trained under others experienced with the surgery. Although now rare, the real risks of permanent limb ataxia and weakness, lifethreatening risks associated with a major operation, and risk of infection must be carefully explained to patients so that they can decide if their pain is severe enough to undergo this invasive procedure.

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