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Case Report

Staged Bilateral Nucleus Caudalis DREZ Lesioning for Persistent Idiopathic Facial Pain: Exceptional Case Report

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ABSTRACT

Persistent idiopathic facial pain (PIFP) is a rare but challenging type of facial pain. How to manipulate such a problem becomes more contentious in cases with bilateral complaints. In situations when conventional treatments fail to reduce the pain, some treatment options like nucleus caudalis dorsal root entry zone (DREZ) lesioning shine as valuable means of managing such intractable problems. Herein, we report an individual with bilateral PIFP in which the patient suffered from intractable pain and was the first to undergo bilateral nucleus caudalis DREZ operation for PIFP. The patient was followed up and the procedure resulted in immediate and complete pain relief.

KEYWORDS: Facial pain, Persistent pain, DREZ, Nucleus caudalis

■ INTRODUCTION

Persistent idiopathic facial pain (PIFP), previously designated as atypical facial pain, is a rare but challenging type of facial pain that does not have the characteristics of the cranial neuralgias and is not attributed to other disorders (16, 24). As was defined by the International Association for the Study of Pain and International Headache Society, PIFP occurs daily and persists for all or most of the day, is limited to an area on one side of face but is deep and poorly localized, is not associated with sensory loss or other neurological deficits, and persists without any demonstrable local cause (16, 24).

The recommended treatment for PIFP is generally tricyclic antidepressants and selective serotonin and norepinephrine reuptake inhibitors (37). In some cases or over time, pharmacological and noninvasive treatments fail to reduce the pain or the treatment itself ignores its efficacy considering the side effects. In these situations, some treatment options like tractotomy-nucleotomy and nucleus caudalis dorsal root entry zone (DREZ) lesioning shine as valuable means of managing such intractable problems (19, 43).

We report herein an individual with bilateral PIFP, rare in its nature, in which the patient suffered from intractable pain on her right side after being followed for more than nine years with a similar pain on the left side. She had been treated previously for the left-sided pain with nucleus caudalis DREZ lesioning, resulting in complete relief. For the newly formed right-sided pain, no satisfactory outcome could be achieved with all procedures and medications provided, and she was finally treated with nucleus caudalis DREZ lesioning, with immediate and complete pain relief.

■ CASE REPORT

History and Presentation

A 41-year-old woman who had been followed by the senior author (YK) with the diagnosis of left PIFP, complained of persistent intractable pain on the right side of her face that had increased gradually in terms of severity and intensity over the past nine months. The pain was described as piercing, throbbing and stabbing, with distribution to the ipsilateral side of the neck and occipital region. The pain was accompanied

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by diplopia, ipsilateral miosis, eye redness with minimal discharge, and nausea with occasional vomiting. The patient had ipsilateral numbness, tingling and weakness in her neck, shoulder, arm, and hand, and facial allodynia was also present. The magnetic resonance imaging (MRI) revealed no significant vascular compression or intracranial abnormalities. Her Visual Analogue Scale (VAS) score was 9.8.

Before the initiation of her complaints, she had experienced some psychological deterioration but the psychiatric consultations revealed no proven problems. The electroencephalogram demonstrated no abnormalities. Duloxetine was effective at first, but her pain severity increased gradually. Her medications included Duloxetine (CYMBALTA®, 30 mg, 2x1), a combination of paracetamol, caffeine and codeine phosphate (GERALGINE-K®, 2x1) and later, 2 doses daily of intravenous morphine sulfate, in the last two months. The pain responded mildly to oxygen supplementation. The pain was severe enough to prevent her from conducting daily activities and rendered her completely incapacitated. She stated her intent to commit suicide due to the intractable persistent pain and her deteriorated life quality.

Her medical history revealed onset of a similar pain on her left side 12 years before. She had been followed and treated by another team with the diagnosis of migraine with aura and cluster headache. No single or combination therapy of analgesics or interventional therapies provided satisfactory pain relief. Later, she was referred to the senior author and was diagnosed with PIFP. After a follow-up period of six months with medications, a left-sided computed tomography (CT)-guided

trigeminal tractotomy-nucleotomy (TR-NC) procedure was achieved with complete pain relief. Twenty-three months later, the same pain relapsed after a blunt facial trauma. A second left TR-NC procedure was performed with partial pain control. Pain intensity increased in eight months, and the patient was finally treated by the senior author with left nucleus caudalis DREZ lesioning, with complete pain relief over 80 months of follow-up.

Operation

During preparation, an MRI scan was obtained in order to measure the diameters of the brainstem for the electrode arrangement (29). The patient's head was fixed with the Mayfield headholder and she was placed in the sitting position under general anesthesia. Preoperative corticoid (methylprednisolone) and intravenous (IV) antibiotic were administered. Neuromonitoring, stimulating and recording needle electrodes were placed on the skin. The incision was started at the midline just at the external occipital protuberance and extended below the spinous process of C2. Lamina and spinous process of C1 and left lamina of C2 were absent because of the previous operation on the left side; thus, the laminectomy of C2 was extended and the spinous process of C2 was removed with total laminectomy. Baseline evoked responses were measured 1 hour after induction from the right and left musculus trapezius, deltoideus, extensor digitorum communis, abductor pollicis brevis, and abductor hallucis, at 350 microvolts (mV). The dura was incised in a Y-shaped configuration using a surgical microscope, and the arachnoid was then opened. After a careful dissection and viewing of the important landmarks

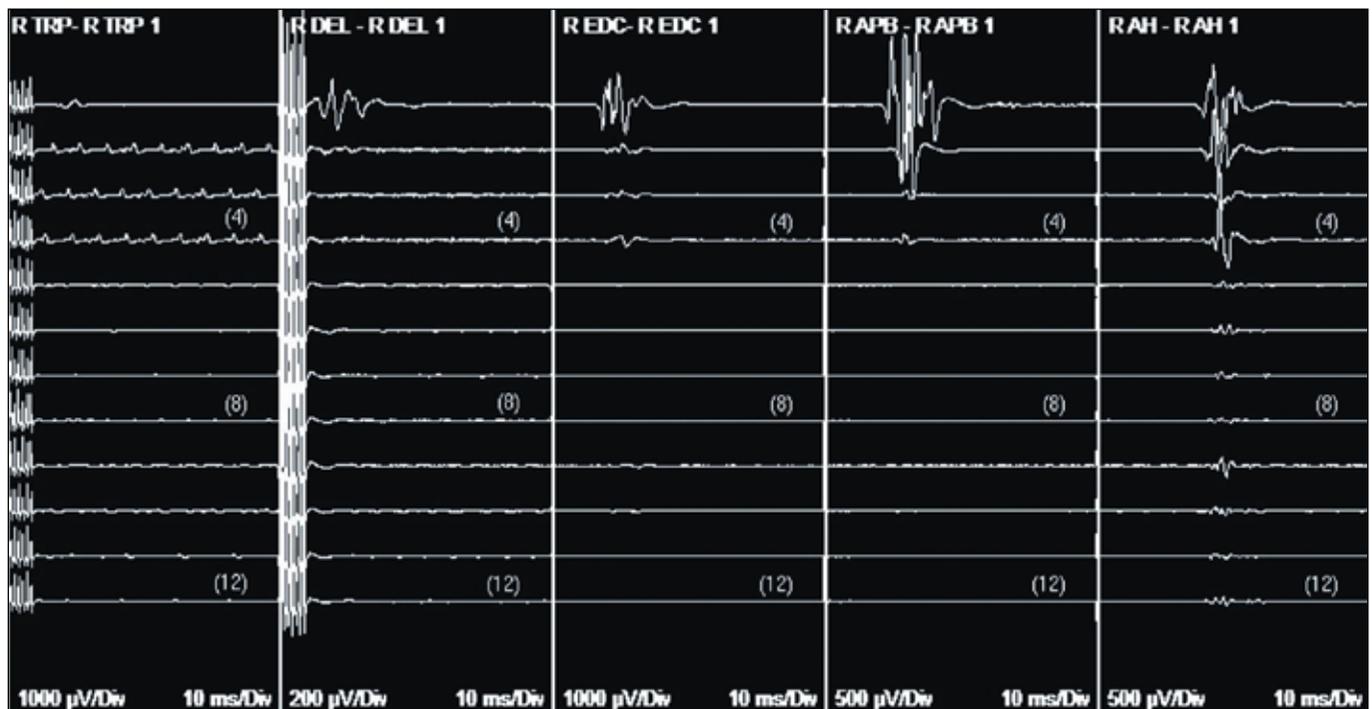


Figure 1: Motor-evoked potentials on the right at the different study recording periods. The five segments shown in the figure belong to measurements from the musculus trapezius (TRP), deltoideus (DEL), extensor digitorum communis (EDC), abductor pollicis brevis (APB), and abductor hallucis (AH).

such the obex, rootlets, sulcus intermediolateralis, and cranial roots of the accessory nerve, DREZ lesions were made using the El-Naggar-Nashold nucleus caudalis DREZ electrodes (3 mm in length, 0.25 mm in diameter). Lesions were created just above the sensory rootlets of the C2 within the intermediolateral sulcus immediately above the obex behind the cranial roots of the accessory nerve. Fifteen standard radiofrequency (RF) lesions were made (70°C, 15 seconds for each lesion). Intraoperative electromyography (EMG) and motor-evoked potentials (MEP) were obtained by experienced staff after each three-four lesions to alert surgeons to possible permanent injury, providing more selectivity and controllability. There was no change regarding the MEPs on the left, but changes were observed during the last three lesions on the right while ascending the spinal cord. The baseline MEPs values on the right for the deltoideus, extensor digitorum communis, abductor pollicis brevis, and abductor hallucis were 128.32 mV, 839.16 mV, 907.08 mV, and 365.04 mV, respectively. All the measures were stable until the last three lesions, when the mentioned potentials were measured as 0 mV, 243.36 mV, 110.62 mV and 110.62 mV, respectively. After approximately 5 minutes, the second and third parameters were measured as 0 mV. No additional lesions were made above C1 rootlets level. Finally, the dura was sutured watertight with atraumatic silk, and closure was completed in order of anatomic layers.

Postoperative Course

The patient was observed in the intensive care unit for one day postoperatively. She was evaluated primarily regarding pain status and neurological functions. She was pain-free immediately with normal corneal sensation and no major limb dysfunction. On the second day, she was transferred to the general ward. Her VAS score for facial pain was 0. A mild ipsilateral upper limb ataxia and dysmetria were observed accompanying mild ipsilateral lower extremity ataxia, but she was able to walk with minimal support. On the 5th day, she was discharged and referred to a physical therapy center. The next day she was admitted with complaints of (pre) syncope, deteriorated consciousness, excessive sweating, tremor, and nausea. On examination, she was neurologically stable but hypotensive and bradycardic. The incision line was clean in terms of redness, discharge or any signs of infection. Her situation was regarded as opiate withdrawal. She was hospitalized, her vital parameters were monitored, and two diminished doses daily of morphine sulfate were administered. Her situation improved gradually and she was discharged on the 6th day. She had minimal ipsilateral ataxia, which improved with physical therapy. She has been followed for 13 months and still has complete pain relief without any motor or sensation problems.

■ DISCUSSION

History of the development of DREZ lesioning dates back to the 1930s, when Sjoqvist used intramedullary tractotomy to relieve facial pain and spare the touch sensation pathways (43). In 1968, Loeser et al. reported the possibility of an association between neuropathic pain and hyperactivity of dorsal horn neurons in the human spinal cord (21). Later, Schvarcz

reported the functional similarity between the substantia gelatinosa of the spinal cord and the trigeminal nucleus caudalis of the cervicomedullary junction (38). The main principle of DREZotomy is based on destroying the mentioned hyperactive neurons of the dorsal horn and the excitatory portion of Lissauer's tract (39). DREZ microcoagulation was performed first by Nashold and Ost Dahl for brachial plexus avulsion pain (30). It was the Bernard-Nashold group that first described the nucleus caudalis DREZ procedure (3). Different techniques, including microsurgical, RF, ultrasonic, and laser ablation, have been used to perform DREZ lesioning (7, 10, 30, 39, 40, 45). Later, in 2008, the senior author (YK) innovated a CT-guided percutaneous technique to perform nucleotomy (18).

Management of PIFP is a challenging problem even from the diagnosis phase in terms of definition and classification. The prevalence of orofacial pain was reported to range between 15-25%, and 10% of patients have chronic pain (20, 22, 34). Up to 30% of orofacial pain cannot be classified (46). PIFP crosses to the other side at a rate of 35% (13, 33, 35). The subjective nature of the pain, with no confirmatory laboratory test or imaging technique, renders the problem more complex for both the patient and physician and makes vital an effective communication. Treatment options, even multiple, may be disappointing to the patient when they fail to provide relief. It is also well recognized that no single treatment option will address all patients' problems (9).

Chronic pain is highly associated with depression due to its enduring burden (2, 32). It was reported that 95% of patients with PIFP complain of other symptoms like headache, neck and backache, and other systemic complaints (12). Madland et al. recommended evaluating patients regarding their disability in terms of psychological discomfort and psychological and social disability rather than confining the evaluation to pain intensity alone (23). The patient presented herein expressed a suicidal intent if the DREZ lesioning operation was not made available to her as an opportunity for cure. This reflects her satisfaction following the first left-sided nucleus caudalis DREZ operation and also the untreatable and intense nature of her disability on the right side.

The main issue regarding PIFP is the lack of a consensus regarding the treatment strategy and patient manipulation. Also how to manipulate such a problem becomes more contentious in cases with bilateral complaints. A multidisciplinary evaluation is essential for this population (11). Even controversial, recommended analgesics and other pain modulators may fail to achieve pain relief. Neurosurgical procedures like nucleus caudalis DREZ lesioning should be considered as valuable options when the classic treatments fail to reduce pain. There are reports in the literature of response to nucleus caudalis DREZ lesioning in the treatment of anesthesia dolorosa, postherpetic neuralgia, cerebral infarction, PIFP, multiple sclerosis, pain secondary to head trauma, craniofacial surgery, and neoplasm (3-5, 14, 25, 26, 28, 29). Gorecki et al. (14) reported limited response to the operation among PIFP patients and a small number of patients that maintained their pain-free status in the long-term. Nashold et al. reported

that less than one-fourth of patients remained pain-free one year after the operation (14, 27). Sindou et al. concluded that paroxysmal shooting pain was significantly more responsive to the procedure than pain described as a continuous burning sensation (41, 42). The literature reports a pain reduction that varies between 50% and 87% after DREZ lesioning for brachial plexus injury (6, 36, 42). Awad et al. (1) reported that 84% of their patients sought to undergo repeat DREZ lesioning when it was presented as a choice. In our reported case, a pain-free status of 80 months was achieved on the left, the formerly operated side, which seems to be beyond the reported pain-free survival range. In our previously reported series, 69% of patients reported satisfactory pain relief one year after DREZ lesioning (19). Furthermore, in two of our patients who were diagnosed with complex regional pain syndrome type I, a pain syndrome with no reported curative treatment, the DREZ operation was curative in one (71 months' follow-up) and provided satisfactory results in the other (44 months' follow-up) (17).

DREZ lesions in general are not expected to affect sensation to touch or trigeminal motor function, but ataxia, usually transient, was found to be the most common complication (14). Maximum attention should be given in cases with bilateral facial pain. Determining the real necessity for an additional lesioning to the other side and evaluating all other treatment options are essential. Delgado-López et al. (8) found that using a midline approach, preoperative steroids, a third-generation electrode, double-row coagulations, and sufficient rostral extension to be crucial for improved results. Intraoperative neuromonitoring is becoming the standard of care in many spinal surgeries. Intraoperative studies have shown that changes in MEPs correlate strongly with postoperative neurological status (31, 44). Usage of neurophysiological monitoring technologies may help the surgeon to locate and identify the targets as well as possible variant anatomy, thus avoiding such complications (15).

■ CONCLUSION

To our knowledge, the case reported herein is the first to undergo bilateral nucleus caudalis DREZ operation for PIFP. A multidisciplinary approach paying maximum attention to identifying and diagnosing pain syndromes correctly and in a timely manner is essential for satisfactory results when addressing such problems. The nucleus caudalis DREZ lesioning operation should be considered as a last but potentially curative treatment option in incapacitated cases that show poor or no response to the other treatment alternatives.

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