Pulsed radiofrequency on the occipital nerve for treatment of short-lasting unilateral neuralgiform headache: A case report

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Abstract
Short-lasting unilateral neuralgiform headache (Short-lasting Unilateral Neuralgiform headache attacks with Conjunctival injection and Tearing (SUNCT)/Short-lasting Unilateral Neuralgiform headache attacks with Autonomic Symptoms) is a trigeminal autonomic cephalalgia with difficult treatment and its management is based on neuromodulator drugs and sometimes ablative procedures on the trigeminal nerve. A positive response to occipital anesthetic blocks and peripheral and deep neurostimulation has also been described. We present the case of a patient with criteria of left SUNCT and transient response to occipital anesthetic blocks, satisfactorily controlled with pulsed radiofrequency (PRF) of the occipital nerve. Upon examination, the patient had tenderness in the left greater occipital nerve (GON). Blockade was performed with anesthetic and corticosteroid, obtaining a highly positive but transient response. After several nerve blocks, the patient was referred to the Pain Unit where pulsed radiofrequency on the left GON was performed. After two sessions, more than 90% of reduction of pain was achieved, maintained for 12 months. There haven’t been found data in the literature on the use of GON PRF for the treatment of SUNCT, while there are descriptions for other types of cranial pain. The intention of our case is to make this procedure to be considered as an alternative for the treatment of this entity in patients who respond to anesthetic blocks.

Keywords
anesthetic blocks, greater occipital nerve, pulsed radiofrequency, SUNA, SUNCT, trigeminal autonomic cephalalgia

Background
Short-lasting unilateral neuralgiform headache is a type of trigeminal-autonomic cephalalgia characterized by attacks of unilateral pain, of moderate or severe intensity, located in the orbital, supraorbital, temporal, and/or other trigeminal distribution.1–3 Its initial description was made by Sjaastad et al.4 This syndrome contains two very similar phenotypes: SUNCT (Short-lasting Unilateral Neuralgiform headache attacks with Conjunctival injection and Tearing), when accompanied by ipsilateral conjunctival injection and tearing and SUNA (Short-lasting Unilateral Neuralgiform headache attacks with Autonomic Symptoms) when it occurs with only one or none of the above symptoms, although in both entities other autonomic manifestations, such as nasal congestion, rhinorrhea, eyelid edema, forehead and facial sweating, forehead and facial flushing, sensation of fullness in the ear, miosis, and ptosis, may also

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appear. The criteria currently used for the diagnosis of SUNCT and SUNA are listed in ICHD 3rd Edition.¹

So far, it hasn’t been found a completely effective therapy for SUNCT, which implies a significant associated morbidity. This report describes the case of a patient with SUNCT refractory to oral medications who sustained benefit from pulsed radiofrequency of greater occipital nerve (GON).

Case presentation
A 75-year-old woman with a history of dyslipidemia, had been diagnosed several years ago with chronic cluster headache, refractory to multiple preventive treatments and most of them poorly tolerated: verapamil at maximum dose of 120 mg twice a day (side effects of tachycardia and dizziness), lithium for short time (tremor and palpitations), topiramate 100 mg twice a day (weight loss and low mood), sodium valproate 500 mg daily (dizziness and tremor), gabapentin 1200 mg daily, lamotrigine (no more than 50 mg daily because of vomiting and light-headedness), and zonisamid 100 mg twice a day (loss of weight and depression). She had no response to corticosteroids (prednisone 60 mg daily), indomethacin at highest doses of tolerability (150 mg daily), and triptans (mild inconstant response to rizatriptan only); no response to supplemental oxygen either. The patient described daily pain bouts of unilateral stabbing in the left orbit and malar area, of slight intensity, separated by 3–4 months with good response as well, although the benefit tended to be shorter in time. Given the transient benefit of this procedure, we decided to refer her to the Pain Unit to perform pulsed radiofrequency (PRF) on the left GON. PRF, 120 s, two 42°C cycles with 22 G 6 cm needle, 5 mm CU (Cosman Medical, inc., Boston) active tip, was performed, obtaining excellent clinical response with subjective reduction of number and severity of pain by at least 80% after the first session. Eight months later a second session was held, with more than 90% of improvement. No side effects were objectified or notified by the patient after the sessions. The clinical response was maintained for 12 months, requiring over-the-counter analgesic (paracetamol + codeine) only once or twice per month for pain that seemed to be mild warnings of SUNCT.

Discussion
Regarding the pathophysiology of SUNCT and the rest of trigeminal autonomic cephalalgias (TACs), it has been proposed that pain and autonomic phenomena are mainly due to the activation of the trigeminal nerve and the parasympathetic system due to the pathological disinhibition of the trigeminal-autonomic reflex.⁵,⁶ This is a reflex route that consists of a connection in the brainstem between the trigeminal nerve and the parasympathetic flow of the facial nerve through the superior salivary nucleus at the pontine level and the sphenopalatine ganglion. Postganglionic parasympathetic neurons contain nitric oxide synthase,⁷ vasoactive intestinal polypeptide, and pituitary adenylate cyclase activation polypeptide, which translate clinically into pain crisis in the trigeminal territory, accompanied by autonomic symptoms: rhinorrhea, tearing, and nasal congestion. Other symptoms such as ptosis or miosis appear to be secondary to sympathetic dysfunction; it is hypothesized that it is due to compression of the sympathetic plexus that surrounds the internal carotid artery, secondary to vasodilation due to parasympathetic activation.

It’s been considered that the brain structure involved in the disinhibition mechanism of trigeminal autonomic reflex is the posterior hypothalamus, which is known to have a modulating role in the nociceptive and autonomic pathways, specifically the trigeminal-vascular nociceptive pathways.⁵ Studies of functional neuroimaging have demonstrated the activation of the posterior hypothalamus during SUNCT/SUNA attacks,⁹–¹¹ supporting their primary role in these pathologies. However, it also appears that peripheral mechanisms may be involved in the complex pathophysiological mechanisms of this disorder,¹² such as demyelination of the trigeminal dorsal root, as occurs in trigeminal neuralgia.

Although TACs are primary headaches of central origin not related to GON lesions, the scientific evidence supporting the modulating role of the occipital nerves on the nociceptive afferents vehiculated by the trigeminal nerve is increasing. It has been described an anatomical convergence of the occipital nerves and the trigeminal nerve in the trigeminal nucleus caudalis at the brainstem level¹³ and in the upper segments of the cervical cord, where the sensory neurons corresponding to the occipital region are overlapped with the trigeminal spinal nucleus neurons. From the functional point of view, there is also a convergence of cervical and trigeminal fibers on the same second order neurons.¹⁴,¹⁵

The present treatment of SUNCT is based on preventive oral medications, being lamotrigine the most effective one,⁶,¹⁰ although gabapentin, topiramate, and carbamazepine, among others, may be useful.³,⁶,¹⁷,¹⁸ Intravenous lidocaine has been reported to produce a highly effective response but often short-lasting effect, therefore used as a transitional treatment.³,⁶,⁹,¹⁹ In refractory patients to them, other therapeutic alternatives are considered, that is, ablative procedures on the trigeminal nerve⁵ and deep
brain stimulation on the posterior hypothalamus, although it has been reported that few patients have substantial and persistent effect with this technique.\(^1\)

In patients with different types of unilateral headaches as migraine, cervicogenic headache, and TACs (including SUNCT) which often associate tenderness in the area innervated by the occipital nerve, different techniques such as anesthetic block\(^{19–22}\) and occipital nerve stimulation\(^{23}\) are applied. As it is suggested by Busch and colleagues, the occipital nerve probably has an excitatory influence on trigeminal circuits that can be reduced by selective inhibition of that nerve due to anesthetic blockade.\(^1\)

PRF is a minimally invasive intervention technique first described by Sluijter,\(^{24}\) which provides an effective treatment strategy for chronic neuropathic pain. The PRF treatment consists of short bursts of high frequency current followed by a heat dissipation interval of 480 ms, which ensures that the temperature of the tissue to be treated does not exceed 42°C. This avoids the appearance of damage to the nervous tissue\(^{25,26}\) producing only modulation of the nerve function\(^27\) and thus avoiding the disturbance of sensory perception after the treatment (great advantage to be highlighted regarding the thermocoagulation technique of conventional radiofrequency).

Based on the pathophysiological mechanism previously described and the clear response to anesthetic block in this patient, we considered the possibility of achieving longer benefit through another technique not used so far for this type of headache, such as PRF on the ipsilateral GON.

To date, multiple studies have been published confirming that PRF successfully modulates several types of peripheral neuropathic pain,\(^{28,29}\) including several types of cranial pain as occipital neuralgia, chronic migraine and cervicogenic headache (occipital nerve PRF),\(^{30,31}\) and refractory cluster headache (sphenopalatine ganglion).\(^{32}\) However, there are no data in the literature on the use of this technique for the control and reduction of pain in short-term unilateral neuralgiform headache, SUNCT.

**Conclusions**

Our report shows that PRF of the GON might be considered as a therapeutic option for the treatment of SUNCT in patients responding to anesthetic blocks of this nerve.

**Clinical implications**

- Short-lasting unilateral neuralgiform headache is a rare but disabling cephalalgia with no completely effective therapy.
- Some patients obtain transient relief with anesthetic blocks of the ipsilateral GON.
- PRF of GON may be a therapeutic option for the treatment of SUNCT in patients responding to anesthetic blocks of this nerve.

**Informed consent**

Written informed consent was obtained from the patient for publication of this case report.

**Declaration of conflicting interests**

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