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CASE REPORT

Sphenopalatine ganglion block – a new treatment for burning mouth syndrome?: a case report

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Abstract Burning mouth syndrome is a poorly understood entity for which current treatment modalities fail to provide effective relieve. Branches of the maxillary and mandibular nerves are responsible for the innervation of the affected area. These are also the nerves involved in trigeminal neuralgia, an entity where sphenopalatine block has proved to be effective. We present a case of a patient with burning mouth syndrome in whom a bilateral sphenopalatine ganglion block was successfully performed for pain treatment. It is an easy and safe technique that can be a valuable treatment option for these patients, although more studies are needed.

Introduction

Originally described by Fox in 1935, burning mouth syndrome (BMS) has generally being defined as a burning sensation in the oral mucosa for which no cause can be identified. Due to the absence of an uniform and commonly accepted definition, reports of its prevalence vary widely across studies, from 1% to 15%.[1] According to the International Association for the Study of Pain (IASP) Classification last reviewed in 2011, BMS is described as glossodynia and sore mouth, also known as Burning Tongue or Oral Dysesthesia, and is characterized by a burning pain in the tongue or other oral mucous membranes, often bilaterally. Xerostomia and dysgeusia are frequent accompanying symptoms. It affects predominantly post-menopausal women, mainly between 60–69 years of age.[2] Both the cause and pathophysiology of this syndrome are poorly understood. Several studies have reported changes in local peripheral small fibers, as well as in nociceptive channels and neuropeptides secreted, but it remains unclear if that is the starting point or a consequence of the syndrome. Current treatments available consist of

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medication, dietary supplements, psychological therapies, and topical treatments.

Sphenopalatine ganglion (SPG) was first described in 1749 by Johann Friedrich Meckel, and Sluder was the first to block SPG using a transnasal approach for the treatment of cluster headaches in 1908. Ever since, SPG block has been used to treat a variety of painful syndromes, such as trigeminal neuralgia, orofacial neuralgias, migraine, and headaches.[3]

We present a case of a patient with burning mouth syndrome in whom a SPG block was performed for pain treatment. To the knowledge of the authors, there is no description of SPG block being used for the treatment of BMS.

Case report

The patient provided her consent to report the case.

A 47-years-old female patient was referred to our pain unit due to oral pain with onset after excision of a radicular cyst located in the superior maxillary bone that lasted for 13 months prior to referral. The pain was described as an aching, burning pain in the palate, gums and superior incisors that worsened while eating and speaking and was accompanied by dysgeusia. She referred that this pain interfered with sleep, and due to its worsening with speech, also interfered with interpersonal relationships. She referred a maximal pain of 10/10 in the numerical pain rating scale (NPRS) and a minimal of 4/10, with pain of 8/10 being the standard during most of the day. No visible lesions were seen in the oral mucosa. She also suffered from fibromyalgia and depressive syndrome for which she was medicated with venlafaxine, topiramate, lorazepam, and cyclobenzaprine. Pregabalin and a combination of paracetamol with tramadol was added to the original medication.

After a month, the patient returned to our office and referred she had no benefit with the medication, maintaining a pain score of 9/10. Consequently, a SPG block was proposed. The block was performed using a nasal transmucosal approach with a long cotton-tipped applicator (Figure 1), previously soaked in a solution of 0.5% levobupivacaine and 4 mg of dexamethasone. The patient was positioned in the supine position with the head lifted approximately 45 degrees. The soaked cotton-tipped applicator was inserted in each nostril sequentially, orientated along the upper border of the inferior turbinate, directed backwards and upwards, until the upper posterior wall of the nose was reached, where it was left in place for 7 minutes in each side. After the procedure, she remained in observation with vital signs monitored for 15 minutes. At discharge, her pain score was 6/10. However, upon telephone control at 48 hours after the technique, the patient reported a substantial pain improvement, with NPRS of 3/10.

At 10 weeks after the procedure, she referred that a substantial improvement was felt for 6 weeks, during which the pain was 3/10 and her ability to speak and to eat were considerably enhanced. However, at that moment, her pain was back to being 8/10 at rest and 10/10 during food ingestion.

After 3 months the procedure was repeated. The same drug combination was used, and the long cotton-tipped applicator was left in place for twenty minutes in each nostril. Before the block, the patient described her pain as 6/10 at rest and 8/10 while eating. After the block, she reported pain relieve to 3/10 and improvements in the ability to speak. After 48 hours, she referred a ''considerable improvement that made all the difference'' since her pain had reduced to 1/10, even during meals. According to the Patients' Global Impression of Change scale, her condition was ''very much improved''. Upon telephone control at two weeks after the procedure, the patient mentioned absence of pain and an increased quality of life since talking and eating were no longer painful events.

Figure 1 Cotton-tipped applicator used to perform the block.

Discussion

Sphenopalatine ganglion is located in the pterygopalatine fossa and represents one of the four parasympathetic ganglia of the head. Nevertheless, it also comprises sympathetic nerve fibers as well as some motor nerve fibers that accompany the parasympathetic nerves. SPG gives rise to the nasopalatine nerve, the greater and lesser palatine nerves, and branches of the pharyngeal branch of the maxillary nerve (V2 branch of the trigeminal nerve).

Due to its rich innervation, sphenopalatine ganglion block has been used for the treatment of chronic and acute pain, such as headaches, migraines, trigeminal and herpes zoster neuralgias, head and neck cancer pain, and even musculoskeletal pain, as well as for better hemodynamic control and analgesia in endoscopic nasal surgery, post-dural puncture headaches, and for hypertension treatment.

BMS is a syndrome whose cause remains unknow and is probably best explained as being multifactorial, involving biological (neurophysiologic mechanisms) and psychological factors. The pain is typically localized in the tongue, mostly in the anterior two-thirds, but can also be present in the anterior hard palate, mucosal aspect of the lip, and mandibular alveolar regions, whilst locations such as buc-

cal mucosa and floor of the mouth are rarely involved.[4] There seems to be an increased incidence and prevalence in post-menopausal women, and to be associated with other conditions such as fibromyalgia and depression, which is the case of the patient presented. Also, the patient reports initiation of symptoms after an invasive procedure, which is not uncommon in BMS.

A Cochrane review[5] on the interventions for treating burning mouth syndrome lastly updated in 2016 evidenced the lack of effective treatment options for BMS. The study demonstrated that short term relieve could be achieved with topical clonazepam, gabapentin and directed energy waves, and long term relieve with psychological therapy, capsaicin mouth rinse, and topical clonazepam.

The typical painful distribution of this syndrome includes an area that is mostly innervated by nerves that synapse in the SPG. The superior palate and teeth receive their sensitive innervation from branches of the maxillary branch (V2) of the trigeminal nerve, namely trough the lesser and greater palatine nerves and nasopalatine nerve. On the other hand, tongue sensory innervation is provided by a branch from the mandibular nerve (V3), namely the lingual nerve, and by a branch of the facial nerve – chorda tympani, with the most posterior part of the tongue being innervated by the glossopharyngeal, although this is not a typical pain location in BMS.

SPG block has been shown to provide an effective treatment for trigeminal neuralgia, which affects most commonly the V2 and V3 branches of the trigeminal nerve. The mechanism responsible for this pain amelioration is not completely understood, since only the V2 branch is clearly correlated with SPG, although it seems that SPG may contain dendrites of the neurons located in the trigeminal ganglion. Nevertheless, given the clear benefit demonstrated in previous studies for trigeminal neuralgia and given the sensory distribution of painful locations of BMS, the authors hypothesized that SPG block could be of value for this syndrome.

The authors decided for a nasal transmucosal approach with a cotton swab due to its tolerability. However, the block can also be performed using an infrazygomatic approach, or through the greater palatine foramen, but these are more invasive and painful techniques. The block can be repeated, or a more definitive technique can be offered to the patient, such as ablation, radiofrequency, or neurostimulation.

In conclusion, bilateral SPG block provides a possible new therapeutic target for these patients since it is an easy and safe technique that can provide effective relieve for a condition where complete symptom resolution is rarely obtained with the current treatment modalities. Nevertheless, this is a single case with a relatively short follow-up period and further studies are needed on the subject.

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Conflicts of interest

The authors declare no conflicts of interest.

References

1. Coculesco EC, Tovaru S, Coculesco BI. Epidemiological and etiological aspects of burning mouth syndrome. J Med Life. 2014;7:305–9.
2. Puhakka A, Forssell H, Soinila S, et al. Peripheral nervous system involvement in primary burning mouth syndrome - results of a pilot study. Oral Dis. 2016;22:338–44.
3. Piagkou M, Demesticha T, Troupis T, et al. The pterygopalatine ganglion and its role in various pain syndromes: from anatomy to clinical practice. Pain Pract. 2011:1–14.
4. Dym H, Lin S, Thakkar J. Neuropathic pain and burning mouth syndrome - an overview and current update. Dent Clin N Am. 2020;64:379–99.
5. Mcmillan R, Forssell H, Buchanan JAG, et al. Interventions for treating burning mouth syndrome (Review). Cochrane Database Syst Rev. 2016;11:CD002779.