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BILATERAL SPHENOPALATINE GANGLION BLOCKS FOR REFRACTORY BURNING MOUTH SYNDROME

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Background: Burning mouth syndrome (BMS) is a debilitating condition that produces a burning pain of the oral cavity and does not have identifiable inciting factors. There are no established treatments, but neuropathic medications have been trialed with variable efficacy. Utilizing targeted nerve blocks could provide therapeutic benefit when treating patients with refractory BMS symptoms.

Case Report: This case describes the treatment of a 61-year-old woman with BMS who presents with refractory symptoms despite maximum dose trials of neuropathic medications. She was successfully treated using bilateral sphenopalatine ganglion blocks. The patient reported a decrease in pain from 7/10 to 1/10 one day after the procedure and described a 90% benefit.

Conclusions: Bilateral sphenopalatine ganglion blocks should be considered to treat refractory BMS.

Key words: Refractory burning mouth syndrome, burning tongue, stomatodynia, nerve block, local anesthesia, pain management

BACKGROUND

Burning mouth syndrome (BMS) is a debilitating condition that affects many patients worldwide. The prevalence has been studied to be around 0.1% to 3.9%, with the highest prevalence occurring among postmenopausal women (1-3). The symptoms most often include a chronic, burning pain of the oral cavity with worsening throughout the day. Typically, the patient complains of mild-to-moderate bilateral pain that occurs daily for multiple hours (4). In some cases, BMS can be traced to a precipitating traumatic event or underlying condition, but usually does not have any obvious inciting factors and is considered an idiopathic condition (5).

While the pathophysiology of the syndrome is largely unknown, the presentation for the vast majority of patients with BMS is consistent with neuropathic pain (3). One commonly proposed mechanism is the development of trigeminal small fiber neuropathy in the intraoral mucosal epithelium. Xerostomia is frequently associated with BMS, likely due to this neuropathy as opposed to glandular damage. Tongue biopsies that have been performed in patients with BMS have shown decreased density of nerve endings and increased action of transient receptor potential cation channel subfamily vanilloid 1 (TRPV1) channels (4,6). These TRPV1 channels fibers are derived from the trigeminal nerve and have been found to conduct taste and nociceptive signals, with increased expression likely contributing to the pain sensation in BMS (3).

Diagnostic criteria for BMS have not been established, so it is currently a diagnosis of exclusion after other causes of oral pain have been ruled out. There are no established treatments currently, but most physicians will initiate neuropathic medications, such as gabapentin and amitriptyline, with variable effi-

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cacy. Previous studies (3,7) have attempted symptom relief with other methods, such as selective serotonin reuptake inhibitors, cognitive behavioral therapy, or benzodiazepines, with poor efficacy. A case report (8) in the Brazilian Journal of Anesthesiology showed a bilateral sphenopalatine ganglion block was effective for this condition. Accepted as management for trigeminal neuralgia, sphenopalatine ganglion neuralgia, acute migraines, and other chronic orofacial pain conditions, sphenopalatine ganglion blockade has shown promise as a solution for treatment-resistant neuropathic pain (9). The proposed mechanism for the block is the reduction of pain transmission via the maxillary nerve (V2) and mandibular nerve (V3) branches of the trigeminal nerve. Prognosis for patients with BMS has been variable. Some cases are transient and resolve after symptomatic treatment, while nearly half of the cases last several years or never fully resolve (10).

Here we describe a case of a patient with BMS refractory to medical management for multiple years. We utilized a bilateral sphenopalatine ganglion block to improve our patient's pain by 90% with significant improvement at the one-month follow-up.

CASE

The patient is a 61-year-old woman who presented to our clinic with BMS refractory to medical management for 3 years. Past medical history is significant for type 2 diabetes mellitus, hypertension, hyperlipidemia, esophageal reflux, and major depressive disorder. Past surgical history is significant for uvulopalatopharyngoplasty, tonsillectomy, and adenoidectomy in 2010. Allergies include cephalexin (rash), shellfish (hives), vancomycin (hives), and bupropion (dry mouth).

In March of 2019, the patient describes waking up one morning with constant, burning oral pain on the top of her tongue and feeling as if the sides of the tongue were being sliced by her teeth. The inner side of the lips and cheeks also had this pain. Her mouth felt dry and there were no identifiable triggers or alleviating factors. The pain would worsen with spicy foods and sugar. She also described an alteration in taste and intermittent white tongue, which was tested positive for candidiasis by her dentist. Patient denied any associated numbness, tingling, or recent oral procedures. She was diagnosed by an oral pathologist who started her on clonazepam and amitriptyline, which provided no relief. Since then, the patient attests to a daily pain scale ranging from a constant 3/10 to 6/10 with intermittent episodes of 8/10 pain.

Subsequent workup was negative for hypothyroidism, iron, folate, and B12 deficiency. Over the years, she was seen by her primary care physician (PCP), ear, nose, and throat physician, oral pathologist, and psychiatrist and was prescribed nonsteroidal anti-inflammatory drugs, muscle relaxants, neuropathic medications, narcotics, and benzodiazepines without significant relief, even with increased doses. Because of this long history of failed medication trials and worsening symptoms, methylene blue oral rinses were attempted. Methylene blue oral rinses were shown in a case report to aid BMS symptoms (11). Unfortunately, this intervention did not offer relief and, in December of 2021, the patient applied for disability and quit her job as due to the intractable symptoms from her condition.

In May of 2022, we evaluated the medical literature for nontraditional methods of treatment and decided to perform a bilateral sphenopalatine ganglion block. This technique was described in a case report in the *Brazilian Journal of Anesthesiology* as an effective treatment for this condition (8). The proposed mechanism for the block is the reduction of pain transmission via the V2 and V3 branches of the trigeminal nerve.

On the day of the procedure, informed consent was obtained regarding the treatment and research related to the treatment. Risks and benefits of the procedure were conveyed and understood. A time-out was taken to identify the correct patient, procedure, and site prior to starting the procedure. The patient was placed in the supine position on the exam bed and positioned at 45°. The patient was relaxed and conversant. A cotton-tipped applicator was soaked in a solution filled with 1.5 mL of 0.5% bupivacaine and 5 mg of dexamethasone. The cotton-tipped applicator was then inserted into each of the patient's nostrils, oriented along the upper border of the inferior turbinate and directed backwards and upwards until the posterior wall of the nose was reached where they were left in place for 10 minutes. After 10 minutes, the applicators were removed and the patient was sat up and observed for 15 more minutes before discharge to monitor for signs of local anesthetic toxicity.

The results of this procedure were positive. Prior to treatment, the patient reported pain of 7/10. After the procedure, the patient reported pain of 1/10. She also described it as a 90% benefit and was ecstatic with the improvement. She continued to report 90% relief the day following the procedure and at a PCP visit one week later. At the one-month follow-up in our clinic,

the patient reported significant improvement in pain and a new pain baseline of 1/10 to 3/10 without speech or eating difficulties.

DISCUSSION

BMS can be a problematic diagnosis for the patient and presents problems to the physician due to limited treatment options and knowledge of the underlying pathophysiology and etiology. One proposed mechanism is neuropathic in nature as several studies (12,13) have shown a significant decrease in the density of epithelial and subpapillary nerve fibers of the tongue in patients with this condition. Other studies (3,14) have theorized the pathology involves abnormal interactions between the sensory functions of facial and trigeminal nerves, small and large fiber neuropathy with subsequent sensory dysfunction, and alterations in blood supply and the autonomic innervation. Due to lack of knowledge of underlying pathophysiology, treating the patient's symptoms is the primary goal.

The target of the sphenopalatine block is the sphenopalatine ganglion, located in the pterygopalatine fossa (8,9). This fossa is situated posterior to the maxilla and deep to the infratemporal fossa on both sides of the skull. The sphenopalatine ganglion has sensory fibers from some of the branches of the trigeminal nerve, including branches of the V2 and V3, which provide sensation for the gums, palate, and anterior two-thirds of the tongue. These are the typical areas affected in BMS and, as a result, make a desirable location for a nerve block to treat the condition.

In this case report, we provide a therapeutic option for patients with BMS refractory to medical management. Future goals will be aimed to repeat this block if symptoms recur with scheduled follow-ups. Our findings are limited by the lack of definitive data surrounding the treatment and pathophysiology of BMS, small sample size, and lack of long-term follow-up data. Further data collection and research contributions surrounding this topic are needed.

CONCLUSIONS

BMS symptoms refractory to medical management should prompt a trial of a bilateral sphenopalatine block, as demonstrated in this case report. This is a safe and effective method of controlling pain in a difficult-to-manage case of BMS. We report a case of significant symptomatic improvement in our patient with BMS after undergoing this procedure. While these results are both promising and encouraging, we will need to follow-up in the future to evaluate the longevity and effectiveness of the treatment. Further research regarding the safety profile and complications of this intervention for BMS is also warranted.

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