CLINICAL VIGNETTE

Unraveling Burning Mouth Syndrome

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Case

A 73-year-old patient presented with 4 months of intermittent burning oral pain. The pain was superficial involving the oral mucosa, tonsils and tongue and associated with dry mouth, without other alleviating or exacerbating factors. Prior medical history includes diabetes mellitus type 2, dyslipidemia, hypertension, coronary artery disease, pulmonary emphysema, obstructtive sleep apnea, insomnia, anxiety, orolabial herpes simplex virus infection, stimulant abuse disorder, opioid abuse disorder and current smoking. Examination of the oral cavity revealed normal, moist mucosa, without oral or orolabial lesions. Cranial nerve examination showed no focal deficits.

Laboratory testing included Hgb A1c 7.1%, serum iron 43 mcg/dL, iron binding capacity (TIBC) 400 mcg/dL, iron saturation 11% and ferritin 12 ng/mL. Despite low iron, there was no anemia or microcytosis. Hemoglobin was 13.1 g/dL, hematocrit 41.0% with MCV of 92.8 fL. Comprehensive metabolic panel, TSH, folic acid, zinc, thiamine, pyridoxine and cobalamin were all within normal limits. Testing for anti-SSA/SSB Ab, anti-transglutaminase Ab and anti-gliadin Ab was negative.

The patient was started on iron replacement and topical benzocaine lozenges as needed for pain. Dental evaluation was also advised to exclude dental causes.

The patient returned for follow up six weeks later and reported no change in symptoms. Their dentist reported no contributing dental disease. Physical examination of the oral cavity remained normal and the iron studies had improved to the low normal range with oral supplementation. The benzocaine lozenges provided temporary pain relief.

He was diagnosed with Burning Mouth Syndrome (BMS) and started on gabapentin 300mg twice daily and 600mg before bedtime for symptom control. At follow up, he reported improvement in symptoms, but still occasionally used benzocaine lozenges for breakthrough pain.

Discussion

Burning Mouth Syndrome is an intraoral painful burning sensation for which no other medical or dental cause of can be found. Associated symptoms may include subjective dry mouth, dysesthesia and/or altered taste. It is considered a diagnosis of exclusion. International Classification for Headache Disorders 3rd edition[1] diagnostic criteria are:

A. Oral pain fulfilling criteria B and C
B. Recurring daily for >2 hours/day for >3 months
C. Pain has both of the following characteristics:
    1. burning quality
    2. felt superficially in the oral mucosa
D. Oral mucosa is of normal appearance and clinical examination including sensory testing is normal
E. Not better accounted for by another ICHD-3 diagnosis.

The condition has a widely ranging reported prevalence from 0.1% to 40%.[2] It is more common in women, particularly post-menopausal women, and is highly associated with advancing age with a significant increase after age 50. One study reported a 2.58 increased risk above age 70, compared to younger than 50 years of age.[3] The pathophysiology of BMS remains unclear, however several mechanisms have been proposed. In 2005, Lauria et al reported decreased density of epithelial and subpapillary nerve fibers in anterior tongue biopsies in BMS patients.[4] They linked BMS to a peripheral small fiber neuropathy of the trigeminal nerve and chorda tympani branch of the facial nerve. Albuquerque et al reported changes in brain activation patterns on fMRI in patients with BMS, similar to other neuropathic pain conditions.[5] BMS has also been linked to dysfunction in the nigrostriatal dopaminergic pathways, which may be why it is more prevalent in patients with Parkinson's disease.[6,7]

BMS is highly associated with psychiatric conditions including depression, anxiety and personality disorders.[8,9] It has also been associated with other conditions including Parkinson's disease, diabetes mellitus, Sjorgren's syndrome, systemic lupus erythematosus, and nutritional deficiencies of thiamine, riboflavin, pyridoxine, cobalamin, folic acid and zinc.[10,11] All of these conditions are linked to small fiber neuropathies.

Management of BMS is similar to the management of other chronic neuropathies. Gabapentin alone was beneficial with symptoms reduction in 50% of patients. Addition of alpha-lipoic acid resulted in a 70% reduction as compared to 15% with placebo.[12] Three times a day topical clonazepam, described as

oral dissolution of 1mg tablets for 3 minutes with expectoration reduced pain intensity in 66% of patients.[13] Cognitive behavioral therapy once weekly for 3-4 months significantly reduced BMS symptoms as compared to the control group.[14] Other interventions with demonstrated benefit include: topical capsacin in xylocaine gel, selective serotonin reuptake inhibitors (sertraline and paroxetine), tricyclic antidepressants (amitriptyline and clomipramine), salivary substitutes and tongue protectors.[15,16]

Conclusion

Burning mouth syndrome can be a debilitating condition that affects nearly 1.3 million Americans.[15] It is associated with older age, female sex, menopause, psychiatric comorbidities and conditions associated with small fiber neuropathies. It is diagnosis of exclusion and—with the exception of appropriate dental evaluation—does not require evaluation or management by a medical specialist. While its etiology and pathophysiology remain elusive, it is important for primary care physicians to be able to recognize it and provide symptomatic treatment.

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