



SCALDED MOUTH SYNDROME: A CHRONIC ORO- FACIAL PAIN DISORDER

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ABSTRACT

Burning Mouth Syndrome is a type of chronic oro- facial pain disorder of neuropathic/idiopathic origin. The term burning mouth syndrome is reserved for describing oral burning that has no detectable cause. It is accompanied by xerostomia and dysgeusia, and the tongue apex is the most commonly affected site. Patient reports intense burning as if the mouth or tongue were 'scalded or burnt. Hence, it has been termed as scalded mouth syndrome. The lack of understanding the cause and mechanism behind the syndrome adds to the difficulty in finding a therapeutic management program.

Clinical significance- Chronic oro-facial pain disorders are diagnoses of exclusion, often suffer from misdiagnosis or delayed diagnosis. Hence, a thorough understanding of the etiology and psychological impact of this disorder, combined with novel pharmacological interventions is required for better management.

KEYWORDS : Burning mouth syndrome, chronic oro-facial pain disorder, scalded mouth syndrome, Temporomandibular joint disorder

INTRODUCTION

Chronic orofacial pain (COFP) disorders, collectively, affect a large proportion of the population.^{1,2} They can involve dysfunction in multiple systems: musculoskeletal, vascular, neurovascular, neuropathic, idiopathic, and psychogenic.³ Burning Mouth Syndrome (BMS) is one of the less common neuropathic/idiopathic COFP disorders.¹ International Association of Pain and Headache Society defines BMS as a "distinctive nosological entity, including all forms of burning sensation of mouth, including complaints described as stinging sensation or pain in association with oral mucosa that appears clinically normal in the absence of local or systemic diseases or alterations."⁴ It is also known by various terminologies such as orofacial pain, stomatodynia, glossodynia, neuropathic pain, glossopyrosis, glossalgia ,stomatopyrosis, oral dysesthesia and scalded mouth syndrome^{5,6}.

Epidemiology

BMS is a disorder of middle aged and elderly individuals affecting an age group of 38-78 years.⁷ This condition has never been reported in children and adolescence and is extremely rare in patients under 30 years.⁸ Currently the prevalence of BMS in the general population is estimated to be 0.7% to 15%.⁹ Female predilection with a ratio between females and males varying from 3:1 to 16:1 has been documented in literature.^{10,11} Women are affected in average 7 times more than men and most commonly in the peri- and post-menopausal period.¹²

Clinical features

Burning mouth syndrome (BMS) is characterized by a spontaneous, chronic, continuous intense burning or stinging sensation of intraoral soft tissues, typically involving the tongue, with or without extension to the lips and oral mucosa.^{5,13} Intense burning is reported by the patient as if the mouth or tongue were 'scalded or burnt. Hence, it has been termed as scalded mouth syndrome.¹⁴ It is classically accompanied by gustatory disturbances like dysgeusia and parageusia and subjective xerostomia. Some patients may develop a single episode of burning sensation while some may show recurrent episodes that last for months or years. The most affected area is the tongue (tip and lateral borders), thus denominated as 'glossodynia' (painful tongue) and glossopyrosis

(burning tongue).¹⁵ Other associated symptoms: thirst, headache, pain in the temporomandibular joint (TMJ) tenderness/ pain in the masticatory and neck, shoulder, and suprahyoid muscles.¹⁶ The term 'burning mouth syndrome' was made acceptable because of the frequent association with other symptoms (xerostomia, taste alterations) and the complexity surrounding the condition of the patient.¹⁵

Pathophysiology

BMS etiology remains obscure. Evidence of neuropathic origin with central and peripheral nerve system involvement has been documented. The possible theories behind the cause of BMS: 1. Abnormal interaction between the sensory functions of facial and trigeminal nerves. According to this theory, due to the high density of fungiform papilla present on the anterior aspect of tongue of certain individuals who are labeled as supertasters (mainly females) are at risk of developing burning pain. 2. Sensory dysfunction associated with small and/or large fiber neuropathy. 3. Centrally mediated alteration in the modulation of nociceptive processing. This theory explains the fact that there is a reduction in the nigrostriatal dopaminergic system resulting in reduced central pain suppression in BMS individuals. 4. Disturbances in the autonomic innervation and oral blood flow. 5. Chronic anxiety or stress results in the alteration of gonadal, adrenal and neuroactive steroid levels in skin and oral mucosa. Probably in a large percentage of patients BMS involves interactions among local, systemic and psychogenic factors.¹⁷⁻²¹ Local factors such as poorly fitting prosthesis, dental treatment, clenching, bruxism as parafunctional habits, allergic contact stomatitis, taste alterations, infections and xerostomia. Systemic factors such as endocrine (hypothyroidism, menopause, diabetes), deficiencies such as iron, vitamin B complex and zinc, anemia, medications, Sjogren syndrome and esophageal reflex. Psychologic causes such as anxiety, depression, cancerophobia and compulsive disorders are having relevance in association with secondary BMS.^{11,22,23}

Classification of BMS

On the basis of etiology, Scala et al.⁹ classified BMS into two categories: Primary or essential/idiopathic BMS, in which local or systemic causes cannot be identified, and involving peripheral or

central neuropathological pathways. "Secondary" BMS, resulting from local, systemic or psychological factors.^{11,15,16,22}

Based on symptoms Lamey & Lamb²⁴ classified BMS into 3 types:

1. Type 1 BMS (35% relative frequency): Patients have no symptoms upon waking but symptoms progress throughout the day reaching its peak intensity by evening. Night-time symptoms are variable. It is linked to systemic disorders like nutritional deficiency and diabetes.
2. Type 2 BMS (55% relative frequency): Patients have continuous symptoms throughout the day and are symptomatic at night resulting in sleepless nights. This type is associated with chronic anxiety due to altered sleep pattern and is related to use of antidepressant drugs, which cause xerostomia.
3. Type 3 BMS (10% relative frequency): Patients have intermittent symptoms throughout the day with symptom-free periods. Usually seen due to anxiety or allergic reactions especially to food allergens.^{9,16,22}

Diagnostic criteria

The diagnosis of BMS needs a careful analysis of the symptom pattern experienced by each patient. Diagnostic criteria developed by Scala^{9,25} -

Fundamental criteria: - Daily deep burning sensation of oral mucosa (bilateral), pain for at least 4–6 months, constant/increasing sensitivity during the day, characteristic symptoms not getting worse/sometimes improvement over ingestion of food or liquid, pain seldom interferes with sleep and never worsens, but may be relieved by eating and drinking.

Additional criteria: Other oral symptoms such as dysgeusia and xerostomia and the presence of sensory/chemo-sensory anomalies, psychological alterations /mood changes that translate patient's personality disorder.

In addition, Patients with BMS cannot have any signs of oral mucosal pathology, such as white lesion, erythema, atrophy, erosion, ulcer, and stomatitis. Candidal swab, oral galvanism measurement, salivary flow rate, gastric reflux tests to determine GERD and parafunctional habit investigations should be performed. Additionally, hematological screening (with special attention to serum ferritin, blood glucose levels and *Helicobacter pylori* antibodies) should be performed to identify possible underlying disturbances. Imaging modalities such as Magnetic resonance imaging (MRI), computed tomography (CT) scan or other imaging test can be performed to check for other health problems. Patch testing to check allergy to certain foods, additives or even denture materials. Biopsy of tongue or oral mucosa can also be done.^{5,26,27}

Then if any of these known local and systemic causes could not be confirmed a diagnosis of "true" burning mouth syndrome should be established.^{26,27}

Differential diagnosis

The most common conditions that may mimic BMS are: Atypical facial pain is a condition characterized by a burning, aching, or cramping sensation occurring on one side of the face, usually in the region of trigeminal nerve. Facial trauma, basal skull fracture with resultant injuries of any peripheral, or proximal branch of trigeminal nerve may cause this disorder. Atypical odontalgia is a painful toothache in a perfectly healthy tooth. The pain is continuous, usually burning, aching, or throbbing and most often occurs in maxillary molars. Diagnosis is based primarily on exclusion of other possible disorders. If a nerve block does not result in pain reduction, this diagnosis should be considered. Lingual nerve neuropathy may be due to complication of third molar extraction or dental anesthetic injection. It can result in anesthesia, paresthesia, or dysesthesia in the tongue and inner mucosa of lower lip, jaw, or chin. Neuralgias also present with burning pain that is similar to that perceived in BMS. Post-herpetic neuralgia (PHN) is the pain that lingers for 3 months after resolution of herpes zoster. It is caused by

varicella zoster virus and is typically confined to a same dermatome. Differentiating PHN in cases of zoster sine herpete and BMS is further cumbersome. Trigeminal neuralgia (TN) -the pain characteristically has an electric shock-like quality, intense, shooting and stabbing type and is unilateral. Pain in TN is precipitated by a light touch on a "trigger zone" present on the skin or mucosa within the distribution of the involved nerve branch. Hypesthesia and autonomic symptoms seen. Local anesthetic blocks, which temporarily eliminate the trigger zone, may also be helpful in diagnosis. Glossopharyngeal neuralgia- pain occurs in deep throat, ear or pharynx characterized as sharp and stabbing pain which occurs in paroxysms. Pain is triggered by stimulating the pharyngeal mucosa during chewing, talking and swallowing. The application of a topical anesthetic to the pharyngeal mucosa eliminates glossopharyngeal nerve pain and can aid in distinguishing it from the pain of other neuralgias. Temporomandibular joint disorders is another differential for BMS because of sharp aching pain which occurs on chewing / bruxism/ jaw movement and is always associated by either clicking sound, deviated jaw, reduced mouth opening, tenderness of masticatory muscles, headache or wearing of teeth^{1,28}

Prognosis:

Improvement in cases of BMS has been cited in half to two-thirds of patients within 6 to 7 years of onset, however prognosis is poor as literature reports complete spontaneous remission in only 3% of the patients within 5 years after the onset.^{29,30}

Treatment and Management^{5,16,27,28}

The first step in the treatment of BMS was subject to the differentiation of primary from secondary form because in the presence of the latter, therapy was directed to treating the causal disease. Thus, in the presence of allergic contact reactions, the simple removal of the suspected allergen (e.g. the material/ dental alloy) determined the remission of the symptoms of BMS. In the case of idiopathic BMS, the therapeutic principles cover a triple purpose: improvement of symptoms, correction of biological and/or morphological disturbances and the therapy of psychoemotional changes. Therapeutic strategies included benzodiazepines (clonazepam), tricyclic antidepressants (amitriptyline), anticonvulsants (gabapentin), selective inhibitors of serotonin receptors (paroxetine and sertraline), capsaicin topical/ systemic, alpha-lipoic acid (neurological antioxidant), benzydamine hydrochloride at 0,15% or 3%, hormone replacement therapy, vitamins supplementation and/ or zinc, iron and psychocognitive therapy. As an adjunctive therapy method, acupuncture is referred to in the art as being beneficial for the relief of symptoms in patients with BMS. Thereby, the key to successful management is a good diagnostic work-up and coordination between the dental practitioners and appropriate physicians and psychologists.^{5,6}

CONCLUSION

Burning mouth syndrome (BMS), a chronic and intractable orofacial pain syndrome. It is characterized by the presence of burning sensation of the oral mucosa in the absence of specific oral lesion. BMS is a diagnosis of exclusion and so remains an important diagnostic challenge to clinicians. An interdisciplinary and systematic approach is required for better patient management.

Conflict of declaration - None

REFERENCES

1. Tait RC, Ferguson M, Herndon CM. Chronic Orofacial Pain: Burning Mouth Syndrome and Other Neuropathic Disorders. *J Pain Manage Med* 2017;3(1): 120.
2. Setty S, David J (2014) Classification and epidemiology of orofacial pain. In: Vadivelu N, Vadivelu A, Kay AD, eds. *Orofacial Pain: A Clinician's Guide*. Switzerland: Springer: 15-24.
3. Balasubramaniam R, Klasser GD, Delcanho R. Separating oral burning from burning mouth syndrome: unravelling a diagnostic enigma. *Aust Dent J* 2009; 54: 293-299.
4. Merskey H. International Association for the Study of Pain, Classification of chronic pain. 2nd ed. IASP task force on taxonomy. IASP press 1994;209-14.
5. Anuradha Sunil et al. An Overview of Burning Mouth Syndrome. *Indian Journal of Clinical Practice* 2012; 23(3):145-152.
6. Serrano O, Munerato M. Burning Mouth Syndrome – Latest update. *International Journal of Dentistry Research* 2016; 1(1): 14-23.

7. Aggarwal A, Panat SR. Burning mouth syndrome: A diagnostic and therapeutic dilemma. *J Clin Exp Dent.* 2012;4(3):e180-5.
8. Grushka M, Epstein JB, Gorsky M. Burning mouth syndrome: differential diagnosis. *Dermatol Ther.* 2002;15:287-291.
9. Scala A et al. Update on burning mouth syndrome: overview and patient management. *Crit Rev Oral Biol Med* 2003;14(4):275–291.
10. Bergdahl M, Bergdahl J. Burning mouth syndrome: prevalence and associated factors. *J Oral Pathol Med.* 1999;28:350-354.
11. Abdullah M. Burning Mouth Syndrome: A Brief Review. *IOSR Journal of Dental and Medical Sciences* 2015; 14(6):76-81.
12. Burning Mouth Syndrome 2016 International Association for the Study of Pain.
13. Fedele S et al. Burning mouth syndrome (stomatodynia). *QJM* 2007;100(8):527-30.
14. Klasser GD, Fischer DJ, Epstein JB. Burning mouth syndrome: recognition, understanding, and management. *Oral Maxillofac Surg Clin North Am.* 2008;20:255-71.
15. López-Jornet P et al. Burning mouth syndrome: Update. *Med Oral Patol Oral Cir Bucal.* 2010;15 (4):e562-8.
16. Coculescu EC, Radu A, Coculescu BI. Burning mouth syndrome: a review on diagnosis and treatment. *Journal of Medicine and Life* 2014;7(4):512-515.
17. Bartoshuk LM et al. Taste damage: Previously unsuspected consequences. *Chem Senses.* 2005;30(Suppl 1):i218–9.
18. Jaaskelainen SK et al. Role of the dopaminergic system in chronic pain – A fluorodopa-PET study. *Pain.* 2001;90:257–60.
19. Heckmann SM et al. Oral mucosal blood flow in patients with burning mouth syndrome. *Pain.* 2001;90:281–6.
20. Woda A, Dao T, Gremeau-Richard C. Steroid dysregulation and stomatodynia (burning mouth syndrome). *J Orofac Pain.* 2009;23:202–10.
21. R. Aravindhan, Vidyalakshmi S, V S Prasad. Burning mouth syndrome: A review on its diagnostic and therapeutic approach. *J Pharm Bioallied Sci.* 2014 ; 6(Suppl 1): S21–S25.
22. Maltzman A, Moricca P, Niv D. Burning mouth syndrome: will better understanding yield better management? *Pain Pract* 2007;7(2):151-62.
23. L. Feller et al. Burning Mouth Syndrome: Aetiopathogenesis and Principles of Management. *Pain research and management.* 2017 <https://doi.org/10.1155/2017/1926269>.
24. Lamey PJ, Lamb AB et al. Type 3 burning mouth syndrome: psychological and allergic aspects. *J Oral Pathol Med.* 1994;23:216-9.
25. Mínguez-Sanz MP, Salort-Llorca C, Silvestre-Donat FJ. Etiology of burning mouth syndrome: a review and update. *Med Oral Patol Oral Cir Bucal.* 2011;16:E1448.
26. Brailo V et al. Oral burning symptoms and burning mouth syndrome-significance of different variables in 150 patients. *Med Oral Patol Oral Cir Bucal* 2006;11:E252-5.
27. Andy Sun et al. Burning mouth syndrome: a review and update. *J Oral Pathol Med* 2013;42:649–655.
28. Patil RG et al. Burning mouth syndrome: A review. *J Indian Acad Oral Med Radiol* 2017;29:25-9.
29. Sardella A LG et al. A Burning Mouth syndrome: a retrospective study investigating spontaneous remission and response to treatments. *Oral Dis* 2006;12:152-155.
30. Campillo E R, Lopez-Lopez J. Evaluation of the response to treatment and clinical evolution in patients with burning mouth syndrome. *Medicina oral, patologia oral y cirugía bucal.* 2013;18(3):e403-410