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CASE REPORT

Exploring burning mouth syndrome: Diagnosis and management of an unusual presentation: A case report

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ABSTRACT

Burning Mouth Syndrome (BMS) is a chronic pain condition of the oral mucosa without visible lesions and may be linked to systemic disorders. A 45-year-old male with anemia, gastroesophageal reflux disease (GERD), and suspected Crohn's disease presented with a persistent burning sensation, primarily on the tongue. Clinical findings included mucosal atrophy, gingival pigmentation, cobblestoning, and generalized chronic periodontitis. He was diagnosed with BMS Type 1, and systemic symptoms prompted further evaluation, raising suspicion for Crohn's disease. A multidisciplinary approach was initiated. This case highlights the rare association between BMS and Crohn's disease and emphasizes the role of oral signs in identifying systemic illness.

Keywords: Burning Mouth Syndrome, Crohn's Disease, Oral Manifestations, Management

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INTRODUCTION

Burning Mouth Syndrome (BMS) is a persistent neuropathic pain disease that affects the mouth. It is characterized by a burning or painful feeling in parts of the tongue, lips, gums, or palate that happens more than twice a day for more than three months without any lab abnormalities or obvious lesions.[1] Burning, stinging, tingling, itching, dry mouth (xerostomia), and changed taste are some of the symptoms.[2] While it can emerge spontaneously, possible factors include stress, new drugs, or past dental procedures.[3]

Recent research shows that psychological factors such as stress, anxiety, depression, and systemic conditions like hypertension, high cholesterol, hyperhomocysteinemia, irritable bowel syndrome (IBS), hypothyroidism, and gastroesophageal reflux disease (GERD) significantly contribute to the onset and persistence of BMS symptoms.[4]

A recent meta-analysis reported that the overall pooled prevalence of BMS is 1.73%, with a higher prevalence in females than males (1.15% vs. 0.38%). On subgroup analysis, the prevalence in Asian countries is 1.05%.[5]

BMS is typically classified into three types based on patterns of symptoms. Type 1, where symptoms develop usually during the day and worsen towards evening; Type 2, where symptoms are present upon waking and persist throughout the day; and Type 3, where symptoms are intermittent and can occur at any time.[6]

Crohn's disease (CD), a chronic inflammatory bowel disease, is known to have extraintestinal manifestations, including those in the oral cavity. Oral manifestations may precede gastrointestinal symptoms or occur simultaneously, complicating diagnosis and management. Among these, mucosal ulcerations, swelling, angular cheilitis, and a burning sensation have been reported.[7] However, the coexistence of BMS and Crohn's disease remains a rarely documented entity, warranting further clinical insight.

This case report highlights a unique presentation of BMS, emphasizing its rare association with Crohn's disease, aiming to underscore the importance of early identification and comprehensive management for improving patient outcomes in regions like Pakistan, where research and resources remain limited.

CASE REPORT

A 45-year-old male of lower middle-class socioeconomic status, as classified by the Modified Kuppuswamy Scale (2022), presented to the Dental outpatient department of Fatima Memorial Hospital,

Lahore, Pakistan in July,2024 with a 3-month history of burning sensation in the oral mucosa and tongue, aggravated by the intake of hot and spicy foods. The patient rated his discomfort as 7/10 on the Visual Analog Scale (VAS).

His medical history was significant for anemia and gastroesophageal reflux disease (GERD), accompanied by lethargy, poor appetite, diarrhea, and unintentional weight loss of 15 kg over four months, for which he was receiving treatment in the medicine department of Shalamar Hospital. The patient had a history of smoking and betel nut chewing for 12 years, both of which he had discontinued six years prior. Psychosocial assessment revealed low self-esteem and anxiety, contributing to poor compliance with previous medical care.

On intraoral examination, there was generalized gingival recession, pale pink oral mucosa with generalized gingival pigmentation (Figure 1), and cobblestone appearance of the buccal mucosa (Figure 2). A high mucosal frenum attachment was also noted. The tongue appeared bald and depapillated, with fissures on the posterior dorsum and a non-scrapable white keratotic patch on the left dorsal surface (Figure 3). Hard tissue examination showed generalized plaque, calculus, and extrinsic staining.
Extra-oral findings were unremarkable, with symmetrical facial features, no lymphadenopathy, a mouth opening of 35 mm within normal limits, and no temporomandibular joint tenderness. Other laboratory investigations are given in Table 1.

Figure 1: Gingival recession and buccal pigmentation

Figure 2: Cobblestoning in buccal mucosa

Figure 3: White keratotic patch

Table 1: Investigation reports of the patient with burning mouth syndrome

Complete Blood Count
Hemoglobin = 9.2 g/dL, microcytic hypochromic anemia, low MCV
Periodontal Charting
Mean probing depth=2.6 mm, mean attachment loss=3.9 mm, plaque score=24%, bleeding on probing=30%. Mobility in teeth # 46 and 26, furcation involvement in teeth # 17, 26, 27, 36, 37, 46, 48
Orthopantomogram (OPG)
Generalized horizontal bone loss; multiple carious lesions in teeth # 16, 17, 24, 25, 26, 27, 36, 37, 46 and 48. Irreversible pulpitis in tooth # 26. Taurodontism in tooth #37. No abscesses were present.
Endoscopy
Erosive esophagitis, evidence of acid reflux
Stool Examination
Presence of undigested food particles, occasional mucus, no ova/cysts
Abdominal Ultrasound
Mild fatty liver; no hepatosplenomegaly or structural abnormalities

A provisional diagnosis of burning mouth syndrome type 1 was made, associated with Crohn's disease. Other differentials included oral submucous fibrosis and leukoplakia. The periodontal diagnosis was generalized chronic periodontitis (stage 2, grade B) with smoking as a risk factor.

Given the complex nature of the case, a multidisciplinary approach was planned. Phase 1, comprising the "Emergency phase," focused on patient education regarding smoking cessation and improving oral hygiene. Dietary counseling was given to avoid spicy foods and increase water intake. Nutritional supplements, (Surbex-Z, Fefol Vit capsules, Vitamin E) were prescribed to manage the patient's anemia.

The "Disease control phase", Phase 2, involved scaling and polishing, composite restoration in 37, root canal treatment for tooth 26, and extractions of non-restorable teeth i.e. 18, 27 & 46. The patient was also referred to a gastroenterologist for the diagnosis and management of suspected Crohn's disease and a dietitian for nutritional management. The patient also consulted Shalamar Hospital, Lahore, Pakistan, for 2nd opinion for suspected Crohn's disease.

Phase 3 included re-evaluation after two months to assess the persistence of the white keratotic patch (Figure 4) and cobblestoning on the buccal mucosa (Figure 5a, 5b). In Phase 4, definitive treatment involved crowns for root canal-treated teeth and prosthetic rehabilitation for missing teeth.

Figure 4: White keratotic patch after2 months

Figure 5a: Cobblestoning after 2 months

Figure 5b: Cobblestoning after 1 month

DISCUSSION

In addition to the well-known gastro intestinal symptoms, one important area for diagnosing Crohn's disease is the mouth region. Dental professionals are therefore essential in early identification.[7]

In this case, the patient described a burning feeling that was brought on by spicy food and persisted for several minutes. This is consistent with Type 1 BMS, in which symptoms usually get worse during the day and are frequently linked to systemic illnesses like GERD and Anemia.[7] Also, the intraoral examination of the patient revealed that he was having a cobblestone appearance, keratotic patches, and a depapillated tongue consistent with BMS, along with the diagnosis of periodontitis. Systemic symptoms of weight loss, anemia, loss of appetite, and diarrhea point towards the presence of Crohn's disease after complete laboratory investigations.

According to recent research, the most common oral symptoms of Crohn's disease are muco-gingivitis, granulomatous cheilitis (usually of the lower lip), mucosal cobblestoning, and linear ulcers. Histological analysis usually shows granulomatous inflammation, which is comparable to intestinal and oral lesions and is typified by Langhan-7 type giant cells, noncaseating granulomas, and mucosal fissuring.[7]

Additional findings include mucosal tags, deep ulcers with hyperplastic borders (mainly in labial, buccal, and retromolar areas), and indurated, polypoid lesions in the vestibule and retromolar region. Swelling, granulation, and hyperplastic gingiva and alveolar mucosa—sometimes with ulcers—may contribute to BMS symptoms.[8] Although oral symptoms of Crohn's disease are not unusual, co-occurring BMS is rarely documented in the literature, especially in South Asia; hence, our case report adds significantly to the scant data from this area.

Compared to the general population, Crohn's disease patients had a higher incidence of gum disease and dental decay. Increased susceptibility to periodontitis is a result of a change in the oral microbiota, which is marked by a decline in beneficial species like Streptococcus mitis and an increase in periodontal pathogens like Prevotella nigrescens and Prevotella intermedia.[9]

Anemia is a common extraintestinal symptom of IBD with oral signs. Iron deficiency causes angular cheilitis, atrophic glossitis, and pale mucosa. Crohn's related B12 deficiency leads to burning, ulcers, atrophy, and taste changes.[10]

Reduced bone mineral density is a result of inadequate calcium and vitamin D absorption, which frequently happens as a result of IBD. Furthermore, there is a correlation between low vitamin D levels and an increased risk of dental caries, tooth loss, and gum illnesses such as gingivitis and periodontitis.[12] So, our patient's diagnosis of periodontitis and its association with Crohn's disease is also being supported by the literature.[11]

A significant risk factor that most likely made the patient's periodontal disease and oral symptoms worse was his history of smoking and betel nut consumption. Because smoking can irritate the oral mucosa and trigger systemic inflammatory responses, it has been related to both BMS and periodontal disease.[3] Quitting smoking is vital for better periodontal health and may ease BMS symptoms, complicating treatment. This case highlights the importance of an interdisciplinary approach addressing both systemic and local factors.

Given the patient's anemia and weight loss, common in Crohn's and linked to oral burning, treatment included dietary changes and supplements. Topical steroids managed inflammation, and miconazole gel addressed possible fungal infections. Referrals were made to a psychiatrist (for anxiety and smoking), a nutritionist, and a gastroenterologist to manage systemic and local factors contributing to symptoms.

CONCLUSION

This case underscores the multifactorial origins of oral mucosal changes, influenced by lifestyle factors such as smoking and the betel nut use, alongside nutritional deficiencies likely stemming from Crohn's disease-related malabsorption. Clinical features like the cobblestoning and the burning sensations call for

comprehensive management strategy that integrates systemic health, nutritional support, and oral hygiene. Timely intervention and patient adherence played a crucial role in successful treatment.

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- **MZ:** Diagnosis, clinical management of oral condition, data acquisition and manuscript drafting
- **MD:** Managing the Crohn's disease in Shalamar Hospital, data acquisition ,provided input on correlation between oral findings and Crohns disease, critical review
- **NZ:** Data acquisition during internal medicine rotation in Shalamar Hospital, literature review, manuscript writing and final editing

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