The Diagnosis of Burning Mouth Syndrome Represents a Challenge for Clinicians

Michele D. Mignogna, MD, DDS
Associate Professor

Stefano Fedele, DDS, PhD
Lecturer

Lucio Lo Russo, DDS, PhD
PhD Student

Stefania Leuci, DDS
Resident

Section of Oral Medicine
Department of Odontostomatological and Maxillofacial Sciences
University "Federico II" of Naples
Naples, Italy

Lorenzo Lo Muzio, MD, DDS
Associate Professor
Institute of Odontostomatology
University of Ancona, Italy

Correspondence to:
Dr Michele D. Mignogna
Department of Odontostomatological and Maxillofacial Sciences
University "Federico II" of Naples
Via Pansini n.5
80128 Naples, Italy
Fax: +39 081 7462197
E-mail: mdmig@tin.it or mignogna@unina.it

Aims: *To evaluate the occurrence of professional delay in the diagnosis, referral, and treatment of patients with burning mouth syndrome (BMS).* ***Methods:*** *Fifty-nine patients (51 women and 8 men; average age, 60.5 years; age range, 32 to 88 years) diagnosed with BMS at our institution were retrospectively studied. Data were collected about the onset of oral symptoms, consultations with medical and dental practitioners, and misdiagnosis before definitive BMS diagnosis and treatment.* ***Results:*** *The average delay from the onset of the symptoms to definitive diagnosis was 34 months (range, 1 to 348 months; median, 13 months). The average number of medical and dental practitioners consulted by each patient over this period and who initially misdiagnosed BMS was 3.1 (range, 0 to 12; median, 3). Candidiasis and aspecific stomatitis were the most frequent misinterpretations of the symptoms before appropriate referral. In about 30% of cases, no diagnosis of the oral symptoms was made or explanation given.* ***Conclusion:*** *Professional delay in diagnosing, referring, and appropriately managing BMS patients occurred frequently in the group studied. No significant differences were found in the number of medical and dental practitioners who were consulted. Emphasis must therefore be placed upon educational efforts to improve health care providers' awareness of BMS. This should increase the rate of recognition and appropriate referral or treatment of patients with chronic orofacial pain due to BMS.* J OROFAC PAIN 2005;19:168–173

Key words: burning mouth syndrome, diagnostic delay, orofacial pain, professional education

Burning mouth syndrome (BMS), also known as orodynia, oral dysesthesia, glossodynia, and stomatodynia, is a complex disorder of unknown etiopathogenesis.[1,2] Although many systemic and local disorders are able to cause a burning sensation localized at the oral mucosa, "true" idiopathic BMS is defined as a burning pain in the tongue and other oral mucosa membranes in the absence of clinical and laboratory abnormalities.[1–7] Some authors would classifiy BMS with the "dynias," a group of chronic focal pain syndromes with a predilection for the orocervical and urogenital regions and unknown pathogenesis.[7]

BMS pain is usually moderate to intense,[1,7,8] and like other chronic pains, it has frequently been associated with insomnia and mood changes, such as irritability, anxiety, and depression, thus finally leading to the disruption of patients' normal social relationships.[7,9–12] Although some researchers have suggested that the disorder may be a manifestation of somatization,[13–17] others have reported BMS to be more closely related to neuropathic pain than to psychosomatic syndromes.[18–20]

In the past few decades, several different treatments and management modalities, including benzodiazepines, tricyclic antidepressants, gabapentin, trazodone, selective serotonin reuptake inhibitors, amisulpride, topical capsaicin, alpha-lipoic acid (thioctic acid), and cognitive behavioral therapy,[2,21–24] have been tested with different and often discouraging outcomes. This has led to the assumption that BMS therapy is always difficult, often unsuccessful, and rarely resolvable.[25,26] However, some of the more recent therapies, although not curative, have been reported to be somewhat effective in alleviating symptoms in a significant percentage of patients.[22–25,27] These findings, although apparently in contradiction with previous data, might be explained by improved attention to the careful diagnosis of BMS. In fact it has been reported that the evaluation of BMS treatment outcomes from earlier studies was often problematic because of a failure to distinguish between patients with idiopathic BMS and patients with BMS symptoms due to local or systemic factors.[26] This aspect, in association with the low methodological quality of most existing studies,[2] has led some authors to conclude that to date there is little published research to provide a clear, conclusive demonstration of an effective intervention and treatment for BMS sufferers.[2] Nevertheless it should be stressed that "absence of evidence is not evidence of absence"[28] and that methodological flaws in the design of previous trials do not necessarily signify a true lack of effect.[2] Thus, findings from descriptive studies reporting that the oral pain characterizing BMS could actually be managed and controlled, leading to a significant improvement in patients' quality of life, cannot be ignored and suggest that further evaluation in the form of well-conducted randomized controlled trials are warranted.

A long period from the onset of the symptoms up to definitive diagnosis is commonly observed in daily practice,[29] leading to a delay in the appropriate management of the condition and the persistence of distressing and often invalidating oral symptoms over several months and, in certain cases, years. During this period most patients usually consult several clinicians, often without obtaining a clear explanation or appropriate diagnosis of their painful condition.

In this study, a group of BMS patients was analyzed retrospectively with regard to the onset of oral symptoms, consultations with medical and dental practitioners, and misdiagnosis before definitive BMS diagnosis. The aim of this study was to investigate the occurrence of professional delay in the diagnosis, referral, and treatment of patients with BMS and to find countermeasures worthy of further evaluation.

Materials and Methods

In total, the records of 59 consecutive patients with BMS diagnosed at our institution during the period from December 2002 to July 2003 were reviewed. The study sample comprised 51 women (86.5%) with an average age of 62.7 years (range, 32 to 88 years; median, 63.5 years; 90th and 10th percentiles, 78 years and 46 years, respectively) and 8 men (13.5%) with an average age of 53.2 years (range, 42 to 73 years; median, 50.5 years; 90th and 10th percentiles, 68.1 years and 42 years), mostly referred by their dentists or family physicians.

Diagnostic criteria included "all forms of burning sensation in the mouth, including complaints described as stinging sensation or pain, in association with an oral mucosa that appears clinically normal in the absence of local or systemic diseases or alterations."[2,6] The specific inclusion criteria were *(1)* symptoms of diffuse burning pain of the tongue and/or oral mucosa, associated or unassociated with subjective oral dryness or loss or alteration of taste or sensation; *(2)* burning pain almost every day; *(3)* normal-looking mucosa in the region of burning; and *(4)* absence of systemic disorders or laboratory alterations known to be associated with orofacial pain.

Exclusion criteria were *(1)* presence of specific local etiologic evidence for the burning (eg, disease of the oral mucosa, hyposalivation); *(2)* presence of specific systemic etiologic evidence for the burning (eg, diabetes, anemia); and *(3)* regular use of medications known to be associated with oral burning and/or alteration of taste or sensation.

To ensure that the patients met the inclusion criteria, they were checked by specialists in oral medicine for disorders relating to hard and soft oral tissues (eg, dental and jaw diseases, diseases of the oral mucosa) as well as organic systemic diseases through routine hematological screening (blood count and levels of glucose, iron, vitamin B_{12}, and folate) (Table 1). In addition, when a complaint of xerostomia was present and/or the clinical examination suggested the presence of hyposalivation, sialometry was performed and the patient was evaluated for diseases relating to salivary gland hypofunction (eg, Sjögren's syndrome, hepatitis C virus, HIV-1 infection) and drug-induced dry mouth. Furthermore, patients sus-

Table 1 Major Local and Systemic Organic Disorders Known to be Associated with Orofacial Pain

Systemic	Local
Diabetes	Dental and periodontal diseases
Anemia	Temporomandibular disorders
Cerebrovascular diseases (infarction and hemorrhage)	Diseases of the salivary glands (including hyposalivation)
Multiple sclerosis	Sinusitis
Nonmetastatic lung cancer	Candidiasis
Metastatic malignancies	Diseases of the oral mucosa (eg, oral lichen planus, aphthous stomatitis)
	Postherpetic neuralgia
	Trigeminal neuralgia (typical and atypical)

pected of being affected by "scalded mouth syndrome" caused by angiotensin-converting enzyme inhibitors were excluded.[30,31]

Data about the patients' age, sex, onset of oral symptoms, number of medical and dental practitioners consulted, and misdiagnosis before definitive BMS diagnosis and treatment were collected. Misdiagnosis was defined as failure to diagnose correctly BMS, either by providing an incorrect explanation of the symptoms or by providing no explanation, in patients who continued to suffer from the same symptoms and were eventually diagnosed with BMS at the authors' institution. Any earlier medical records that were available and interviews with patients were also used as data sources. The hormonal status of women was not checked, since hormonal replacement treatments in menopausal BMS patients have not supported previous theories about hormonal disturbances as a causative factor of BMS.[1]

Results

All the patients met the aforementioned criteria and were therefore diagnosed to be affected with BMS. Twenty-three patients (38.9%) reported a previous diagnosis of depression.

The average amount of time elapsed from the onset of the symptoms to definitive diagnosis (ie, the diagnostic delay) was 15.6 months for male subjects (range, 2 to 48 months; median, 14 months; 90th and 10th percentiles, 31.9 months and 2.7 months, respectively) and 36.8 months for female subjects (range, 1 to 348 months; median, 12 months; 90th and 10th percentiles, 72 months and 4 months, respectively). Considering the entire study group, the average diagnostic delay was 34 months (range, 1 to 348 months; median, 13 months, 90th and 10th percentiles, 72 months and 4 months, respectively). Eleven patients (18.6%) experienced a diagnostic delay of < 6 months; 18 patients (30.5%), 7 to 12 months; 8 (13.5%), 13 to 24 months; 8 (13.5%), 25 to 36 months; and 14 (23.7%), > 36 months. Thus, more than 50% of the BMS patients were characterized by a delay in appropriate diagnosis of at least 12 months.

The average number of medical and dental practitioners consulted by each patient during this period was 3.2 among female patients (range, 0 to 12; median value, 3; 90th and 10th percentiles, 5 and 1, respectively), and 2.5 among male patients (range, 1 to 5; median, 2; 90th and 10th percentiles, 4.3 and 1). Specifically, 42.3% of the patients consulted 0 to 2 doctors, a majority of patients (69.4%) consulted a maximum of 3 doctors, and 5 patients (8.5%) consulted 6 or more doctors (Table 2). Considering the entire study sample, the total number of clinicians consulted was 188, corresponding to 102 physicians and 86 dentists. While 100% of the dentists consulted were general practitioners, the physicians included 52 family physicians (50.9%), 10 ear, nose, and throat specialists (9.8%), 25 dermatologists (24.5%), and 15 maxillofacial surgeons (14.7%).

Candidiasis (12.5%) and aspecific stomatitis (described by doctors as "diffuse infection or inflammation of the oral mucosa") (15.3%) were the most frequent misdiagnoses of the symptoms. In about 30% of cases, no diagnosis or explanation of the oral symptoms was made. Other erroneous explanations of the oral symptoms included depres-

Table 2 Number of Physicians and Dentists Consulted Before Appropriate BMS Diagnosis

Number of health-care providers consulted before BMS diagnosis	Patients n (%)
0	1 (1.7)
1	8 (13.6)
2	16 (27.1)
3	16 (27.1)
4	7 (11.9)
5	6 (10.1)
6	2 (3.4)
8	1 (1.7)
10	1 (1.7)
12	1 (1.7)

Table 3 Type and Number of Misdiagnoses Before Appropriate BMS Diagnosis

Typology of misdiagnosis	Number (%) of misdiagnoses
No diagnosis	21 (29.2)
Aspecific stomatitis	11 (15.3)
Candidiasis	9 (12.5)
Depression	8 (11.1)
Allergic reaction to prosthesis	6 (8.3)
Hypovitaminosis	5 (6.9)
Hyposalivation/xerostomia	3 (4.1)
Trigeminal neuralgia	2 (2.8)
Viral hepatitis	2 (2.8)
Gastroesophageal reflux disease	2 (2.8)
Bruxism	1 (1.4)
Tracheitis	1 (1.4)
Food intolerance	1 (1.4)

sion, allergic reaction to dental prosthesis, hypovitaminosis, xerostomia, trigeminal neuralgia, viral hepatitis, gastroesophageal reflux disease, bruxism, tracheitis, and food intolerance (Table 3). Seventy-two different diagnoses were recorded, which suggests that a number of doctors consulted by the same patient gave 1 or more identical misdiagnoses.

Discussion

In spite of the ongoing interest in and increasing knowledge about chronic orofacial pain-related disorders, the exact etiology and pathogenesis of BMS remains an enigma.[1,2] This in turn has led to confusion regarding the clinical management of patients, including diagnosis and treatment. In particular, the diagnosis of BMS seems to represent a challenge for clinicians. It is a common experience in clinics focusing on orofacial pain to see BMS patients who have been referred from one health-care professional to another without appropriate diagnosis and management.[23] Furthermore, a wide spectrum of misdiagnoses, misinterpretations, and consequently empirical treatments of the patients' symptoms by health-care providers is also frequently observed.[31] At the authors' clinic, the "typical" patient affected by BMS generally presents with a bag full of mouthwashes, antifungine drugs, antibiotics, vitamins, topical steroids, and other medications, together with a history of long-lasting persistent oral pain.

Delays in diagnosing, referring, and managing BMS patients appropriately not only may cause the oral pain to interfere chronically with a normal daily lifestyle and sleep pattern, but also could have a significant emotional impact on patients, who are sometimes suspected of imagining or exaggerating their symptoms.[23] Therefore, given the chronic nature of BMS, the need for early and appropriate identification of the disorder and commencement of an effective mode of treatment for sufferers is vital.[1] The results of our retrospective study have clearly shown that BMS patients frequently have a long history of treatment for their symptoms by inadequate empirical procedures due to incorrect diagnoses. These findings are in accordance with similar data previously reported by other authors.[32] This suggests that in a considerable number of BMS cases, clinicians did not suspect the real nature of oral symptoms.

All the patients in the present study suffered from their oral pain and discomfort for a long period, passing through a number of health-care providers ranging from 0 to 12 (average, 3.1; median, 3). It is not clear whether this situation is the consequence of the complex and largely unknown nature of BMS or the expression of inadequate knowledge among physicians and oral health-care providers about nondental orofacial pain syndromes. It is likely that both causes have contributed. Several papers focusing on the need to enhance the teaching of temporomandibular disorders and orofacial pain to predoctoral and postdoctoral dental students, as well as to participants in continuing education programs, have been published over the past few years.[33–35] At the same time, it should be stressed that the nature and the etiology of BMS, as well as other chronic pain syndromes, are still largely unknown, complicating their diagnosis and management. It has been suggested that, before a patient is diagnosed to be affected by BMS, the oral symptoms should have been present for at least 6 months.[36] Although the majority of the patients in the present study met this inclusion criterion (81.2%; n = 48), the authors do not completely agree with it. Some patients in the present study exhibited the typical features of BMS and thus were included in the study group, although they reported the oral pain to be present for fewer than 6 months. It is the authors' belief that, even when there are few doubts about the nature of patients' oral pain, clinicians should not delay in providing appropriate diagnosis and treatment.

Furthermore, another point highlighted by our study is the high percentage of patients (about 30%) who did not receive any explanation for their pain symptoms. It has been reported that reassurance, together with counseling, is vital for chronic pain sufferers. They can have a potent therapeutic action and can contribute strongly to an improvement in patients' quality of life, especially with respect to patients who complain that they do not receive enough information.[1] However, our findings have shown that many health-care providers do not provide the necessary caring and supportive approach to BMS patients.

In conclusion, despite the general assumption that BMS therapy is always difficult, it is important to emphasize that in many cases patients continue to suffer because BMS is usually misdiagnosed, incorrectly treated, and often not recognized.

Therefore, it appears clear that emphasis should be placed upon educational efforts to improve health-care providers' awareness of BMS. This should increase the rate of recognition and appropriate referral or treatment of patients with chronic orofacial pain due to BMS. Furthermore, given the high proportion of BMS patients lacking adequate explanation for their symptoms, special efforts should be directed toward providing patients with information about their condition, since this is regarded as a fundamental step in chronic pain management.[37,38]

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