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JADA 2013;144(10):1135-1142

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Defining and diagnosing burning mouth syndrome

Perceptions of directors of North American postgraduate oral medicine and orofacial pain programs

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The International Association for the Study of Pain¹ (IASP) defines burning mouth syndrome (BMS) as a burning pain in the tongue or other oral mucous membrane persisting for at least four months and associated with normal oral mucosa and normal laboratory findings.^{2,3} The IASP diagnostic criteria are as follows: burning sensation in the tongue or other parts of the oral mucosa, usually bilateral and associated with dysgeusia, dry mouth and denture intolerance. The International Headache Society⁴ (IHS) described BMS as an intraoral burning sensation for which no medical or dental cause can be found. The IHS further noted that pain may be confined to the tongue (glossodynia) with associated symptoms that include subjective dryness of the mouth (xerostomia), paresthesia and altered taste. The IHS provided the following diagnostic criteria for BMS: pain in the mouth present daily and persisting for most of the day, oral mucosa of normal appearance and exclusion of local and

ABSTRACT

Background. The authors conducted a study to ascertain participants' perceptions of and confidence in their responses regarding the definition and diagnosis of burning mouth syndrome (BMS).

Methods. The authors developed an eight-question questionnaire with input from several experienced clinicians in the fields of oral medicine (OM) and orofacial pain (OFP) and sent it to directors (n = 20; OM = 10; OFP = 10) of accredited postgraduate training programs in North America. They used descriptive statistics to analyze the results.

Results. The response rate was 65 percent (n = 13; OM = 6; OFP = 7). Participants reported a mean of 7.3 cases of BMS in any given three-month period, with 89 percent of these cases managed within the programs. They identified, with a high degree of confidence, overall criteria for establishing a definition and definitive diagnosis of BMS.

Conclusions. There were multiple similarities among participants' responses regarding the elements to be included in the definition and diagnosis of BMS.

Practical Implications. These data provide information on current status of definitions and diagnostic guidelines and may assist in development of future consensus statements on BMS that incorporate additional geographical representation and appropriate methodology.

Key Words. Burning mouth syndrome; diagnosis; respondents' perceptions; confidence rating scale.

JADA 2013;144(10):1135-1142.

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systemic diseases. In light of these somewhat imprecise definitions and descriptions, it is easy to understand the challenge facing health care practitioners when evaluating patients with BMS and the barriers to achieving an accurate and reliable diagnosis.

The prevalence of BMS is reported to be between 0.7 percent and 5.0 percent of the general population, depending on the methodology (survey or clinical assessment) being used in and the geographical setting of the study.⁵⁻⁸ BMS is reported most commonly by women in the fifth to seventh decade^{3,9,10} and usually manifests in a period between three years before and 12 years after the onset of menopause.³ It rarely manifests before the age of 30 years.^{11,12} Investigators report female-to-male ratios ranging from 3:1 to 16:1.^{3,8,13-15} The pain commonly occurs bilaterally, involving the anterior two-thirds of the tongue, followed by the dorsum and lateral borders of the tongue, the anterior aspect of the hard palate and the labial mucosa of the lips. The burning pain also may occur simultaneously at multiple sites.^{3,11,14-18} Other symptoms that manifest with the burning complaint include taste alterations,^{3,19} often described as the presence of a constant foul, bitter or metallic taste sensation, which may be equally as disturbing as or more disturbing than the oral burning pain itself.⁵ Intriguingly, there are conflicting objective data regarding decreased salivary flow rates in people with BMS.²⁰⁻²³ Nevertheless, findings in some studies have shown qualitative changes in salivary composition.^{21,23,24}

Several classification schemes have been proposed to assist in the diagnosis of BMS. One such classification, proposed by Lamey²⁵ and Lamey and Lewis,²⁶ contains three subtypes according to variations in pain intensity over 24 hours. This classification has not been validated and does not appear to be widely accepted by the scientific community. Jaaskelainen²⁷ proposed three distinct subclasses that were based on neurophysiological, psychophysical and functional imaging studies. Gremeau-Richard and colleagues,²⁸ using a double-masked crossover design, reported there to be two distinct groups of people with BMS, who could be classified on the basis of the location of neuropathic changes (that is, mediated by the peripheral or the central nervous system). A more pragmatic clinical approach is to separate BMS into two distinct categories: primary (essential or idiopathic) BMS, in which there is a lack of evidence of any other disease, and secondary BMS, in which an oral burning sensation is identified secondary to other clinical abnormalities or to systemic condi-

tions such as anemia, diabetes, thyroid disease or gastroesophageal reflux disorder. Therefore, the clinician bases a diagnosis of BMS on clinical presentation and on the exclusion of local and systemic factors.

To date, there is a lack of qualitative and quantitative analyses regarding clinicians' understanding of the diagnosis of BMS. We approached these limitations by soliciting opinions (via a confidence rating scale [CRS]) from experienced health care practitioners who treat BMS. This technique engages the resources of all participants and results in an enhanced decision-making ability among members of the group with regard to resolution of the clinical problem addressed.²⁹ This process has been used with an array of oral health issues, including decision support for diagnosis, and has led to improved outcomes for the conditions under investigation (such as outcomes assessment for periodontal therapy, referral criteria in pediatric dentistry and indications for use of radiography).³⁰

Our aim in this study was to gather data about the perceptions of a group of oral medicine and orofacial pain training program directors from the United States and Canada in terms of the definition of BMS and the various factors and variables used in, and assisting with, the determination of its definitive diagnosis.

METHODS

We designed a structured questionnaire with input from four experienced clinicians in oral medicine and orofacial pain (two from each field) who did not participate in the study directly. (The study protocol was approved by an accredited institutional review board.) Most questions were open-ended to facilitate variability of responses. This broad approach captured the most information regarding the respondent's opinion without limiting answers or leading him or her. The only question with designated response categories involved specific diagnostic testing for conditions associated with oral burning sensation that excluded BMS. We derived these categories from the current literature on BMS and the knowledge of the four experienced clinicians.

The questionnaire (Figure) contained eight questions pertaining to the diagnosis of BMS,

ABBREVIATION KEY. **BMS:** Burning mouth syndrome. **CRS:** Confidence rating scale. **IASP:** International Association for the Study of Pain. **IHS:** International Headache Society. **OFFP:** Orofacial pain. **OM:** Oral medicine.

and the responses to these questions were analyzed in this study. We addressed the following items: assessment of the respondents' clinical experience as it pertained to BMS; the most common characteristics to be used in a definition of BMS; the criteria necessary (including signs and symptoms) to make a definitive diagnosis of BMS while addressing local, systemic and psychological factors that need to be ruled out; diagnostic tests used to support a diagnosis of BMS; and perception regarding the etiopathogenesis of BMS. Because the majority of questions were in an open-ended format, thereby allowing multiple responses from each participant, we grouped responses according to consensus into broader, more inclusive categories for discussion purposes. All responses, with the exception of those related to the participants' clinical experience, were measured according to a CRS. The CRS is a Likert-like numeric rating scale with anchors of 1 (meaning "I am very uncertain") and 7 ("I am very certain"), and we included it to enhance the certainty of (strength of belief in) responses and reduce ambiguity (imprecision in the judged probabilities).³⁰⁻³² Respondents used this categorical scale to rate their level of confidence in their selection of each option, and we averaged all answers to produce a mean score with appropriate confidence intervals and standard deviations. We considered a high confidence rating as a 6.0 or greater.

We obtained information about directors of postgraduate oral medicine (OM) and orofacial pain (OFP) programs through our personal knowledge, as well as through consulting with the American Academy of Oral Medicine and the American Academy of Orofacial Pain. At the

1	Please list the mean number of patients with burning mouth syndrome (BMS) seen every three months.
2	Please list the most common characteristic(s) to be used in a definition of BMS.
3	Please list the criteria (including signs and symptoms) necessary for a definitive diagnosis of BMS.
4	Please list the local factors needing to be ruled out before a definitive diagnosis of BMS can be made.
5	Please list the systemic factors needing to be ruled out before a definitive diagnosis of BMS can be made.
6	Please list the psychological factors needing to be ruled out before a definitive diagnosis of BMS can be made.
7	What diagnostic tests are used to rule out local factors, systemic factors or both to support a definitive diagnosis of BMS? Please circle appropriate letter(s). a. salivary flow rates b. taste testing c. serologic studies d. soft-tissue biopsy e. microbiological cultures f. medication substitution
8	Please describe the etiopathogenesis of BMS.

Figure. Questionnaire used in the study.

TABLE 1

Characteristics respondents most commonly chose to be used in a definition of burning mouth syndrome.			
CHARACTERISTIC	NO. OF RESPONDENTS (PERCENTAGE OF THE TOTAL*)	MEAN (SD)† CRS‡ SCORE	95 PERCENT CONFIDENCE INTERVAL
Burning in the Oral Mucosa	10 (77)	6.1 (1.10)	5.42-6.78
Absence of Clinical Findings	7 (54)	6.1 (1.07)	5.35-6.93
Burning in the Tongue	4 (31)	6.3 (0.96)	5.31-7.19
Chronic Pain (Burning Pain Lasting Longer Than Three Months)	4 (31)	6.8 (0.50)	6.26-7.24

* Total number of respondents = 13.
 † SD: Standard deviation.
 ‡ CRS: Confidence rating scale.

time of questionnaire distribution, there were 20 accredited postgraduate OM and OFP training programs in the United States and Canada (n = 20; OM = 10; OFP = 10). The questionnaire was delivered either electronically or via postal service to all training program directors in the United States and Canada. We made a second attempt to deliver the questionnaire to those who did not respond to the initial inquiry. We calculated and recorded descriptive statistics for the CRS ratings.

TABLE 2

Criteria respondents reported most frequently as necessary for a definitive diagnosis of burning mouth syndrome.

CRITERION	NO. OF RESPONDENTS*	MEAN (SD) [†] CRS [‡] SCORE	95 PERCENT CONFIDENCE INTERVAL
Subjective Sensation of Intraoral Altered Sensory Symptoms	10	6.2 (0.63)	5.81-6.59
Absence of Mucosal Pathology or Abnormalities	7	6.1 (0.69)	5.63-6.65
Absence of Dental Etiology	4	6.0 (0.82)	5.20-6.80
Presence of Burning Pain	4	6.0 (0.82)	5.20-6.80
Xerostomia (Dryness of the Mouth)	4	5.3 (1.70) [§]	3.58-6.92
Absence of Salivary Gland Hypofunction, Dysfunction or Both (Objective Lack of Salivary Flow Measured According to Weight or Volume)	3	6.3 (0.58)	5.68-6.98

* Total number of respondents = 13.
† SD: Standard deviation.
‡ CRS: Confidence rating scale.
§ Criterion with a mean CRS score < 6.0.

RESULTS

Thirteen (OM = 6; OFP = 7) of the 20 postgraduate directors responded, representing a response rate of 65 percent. The survey results showed that a mean of 7.3 cases of BMS had been diagnosed in each postgraduate program in any given three-month period (Figure, question 1), and approximately 89 percent of those cases had been managed within the postgraduate clinics. Table 1 describes the characteristics that respondents most commonly indicated should be used in a definition of BMS (Figure, question 2). Table 2 describes the criteria respondents reported most frequently as being necessary for a definitive diagnosis of BMS (Figure, question 3). The CRS results (Tables 1 and 2) demonstrated an overall elevated degree of confidence (6.0 or greater) among participants in their responses to the question about the most common characteristics to be used in a definition of BMS (Figure, question 2) and to the question regarding the criteria necessary for a definitive diagnosis of BMS (Figure, question 3), with the exception of xerostomia (Table 2).

Respondents reported various factors (Figure, questions 4-6) as needing to be ruled out before they could provide a definitive diagnosis of BMS (Table 3). The majority of respondents indicated measurement of salivary flow rates (n = 11; mean [standard deviation (SD)] CRS score, 6.2 [0.60], 95 percent confidence interval [CI], 5.82-6.54) as a diagnostic test to rule out a diagnosis of BMS (Figure, question 7). This was followed by serologic studies (n = 8; mean [SD] CRS score, 6.1 [0.64]; 95 percent CI, 5.69-6.57), medication substitution (n = 7; mean [SD] CRS score, 6.1 [0.69]; 95 percent CI, 5.63-6.65), microbiological cultures (n = 5; mean [SD] CRS score,

6.6 [0.55]; 95 percent CI, 6.12-7.08), soft-tissue biopsy (n = 3; mean [SD] CRS score, 6.3 [0.58]; 95 percent CI, 5.68-6.98) and taste testing (n = 2; mean [SD] CRS score, 6.0 [0.0]). Table 4 illustrates that the majority of respondents (54.5 percent) associated BMS with a neuropathic etiopathogenesis (Figure, question 8). Interestingly, participants displayed uncertainty in their responses when defining the etiopathogenesis for BMS (Figure, question 8) as idiopathic or unknown or psychological or psychosocial (Table 4).

DISCUSSION

To our knowledge, this is the first study in which investigators have garnered clinicians' perceptions and described their degree of confidence in their responses regarding the diagnostic criteria for BMS across emerging diagnostic disciplines such as oral medicine and orofacial pain. The results of our study support the assumption that the 13 postgraduate program directors (OM = 6; OFP = 7) who responded to our survey had experience in the clinical management of BMS (for instance, a mean of 7.3 cases of BMS had been diagnosed in their clinics in any given three-month period), thereby justifying their participation as a sample of clinicians qualified to render perceptions regarding this condition. Participants displayed overall confidence in their responses and consistency with the current literature¹⁻⁴ with respect to the most common characteristics to be used in a definition of BMS (Table 1). Notably, only four of the 13 program directors reported a burning sensation in the tongue and chronic pain as characteristics that should be used in a definition of BMS. Perhaps many of

TABLE 3

Factors respondents indicated as needing to be ruled out before a definitive diagnosis of burning mouth syndrome could be made.*

FACTOR (TOTAL NO. OF RESPONSES)	NO. OF RESPONSES (PERCENTAGE OF TOTAL RESPONSES)	MEAN (SD) [†] CRS [‡] SCORE	95 PERCENT CONFIDENCE INTERVAL
Local (n = 55)			
Oral mucosal lesions (lichen planus, benign migratory glossitis, benign mucous membrane pemphigoid, pemphigus, oral ulcers, oral cancer, leukoplakia)	11 (20.0)	6.4 (0.67)	5.96-6.76
Microbial infection (fungal, [§] viral or bacterial)	10 (18.2)	6.2 (0.63)	5.81-6.59
Dental issues (poorly fitting dental prostheses, mechanical irritations, dental caries, periodontal or pulpal disease)	9 (16.4)	6.2 (0.44)	5.93-6.51
Salivary gland hypofunction/dysfunction	8 (14.5)	6.3 (0.71)	5.76-6.74
Contact allergy/mucosal contact irritants (contact allergy or allergy to dental materials, mouthwash, spices, food ingredients, additives or preservatives)	8 (14.5)	6.1 (0.35)	5.89-6.37
Parafunctional habits of tongue or lips	4 (7.3)	6.5 (0.58)	5.93-7.07
Other (nerve trauma or neuropathy, toxic environmental exposure, taste disorder)	5 (9.1)	6.4 (0.55)	5.92-6.88
Systemic (n = 31)			
Autoimmune disorders (Sjögren syndrome, connective tissue disease, systemic lupus erythematosus)	8 (25.8)	6.4 (0.52)	6.02-6.74
Vitamin or nutritional deficiencies	6 (19.4)	6.3 (0.52)	5.92-6.74
Endocrine disorders (diabetes, metabolic diseases)	6 (19.4)	6.3 (0.52)	5.92-6.74
Medication adverse effects	4 (12.9)	6.8 (0.50)	6.26-7.24
Other (hematologic disorder, gastrointestinal condition, cancer, psychopathology)	7 (22.6)	6.4 (0.79)	5.85-7.01
Psychological (n = 28)			
Anxiety	11 (39.3)	6.0 (0.77)	5.54-6.46
Depression	9 (32.1)	5.9 (0.78)	5.38-6.40
Somatoform disorder	4 (14.3)	6.0 (0.82)	5.20-6.80
Other (psychosis, obsessive-compulsive disorder)	4 (14.3)	6.5 (0.58)	5.93-7.07

* Multiple responses were allowed from a single participant.
[†] SD: Standard deviation.
[‡] CRS: Confidence rating scale.
[§] There were six responses (each with a CRS score = 6.0) endorsing fungal infection as a local factor, which composed 60 percent of the responses in the "microbial infection" category.

TABLE 4

Etiopathogenetic theories for burning mouth syndrome.

NATURE OF HYPOTHETICAL ETIOPATHOGENESIS*	NO. OF RESPONSES (PERCENTAGE OF TOTAL RESPONSES [N = 33])	MEAN (SD) [†] CRS [‡] SCORE	95 PERCENT CONFIDENCE INTERVAL
Neuropathic (Peripheral or Central Neuropathies or Sensitization)	18 (54.5)	6.1 (0.87)	5.66-6.46
Idiopathic or Unknown	5 (15.2)	5.6 (1.10) [§]	4.60-6.60
Psychological or Psychosocial	3 (9.1)	5.3 (0.58) [§]	4.68-5.98
Hormonal	2 (6.1)	6.0 (0.0)	NA [¶]
Other (Taste Dysfunction or Dysregulation, Nutritional Deficiency, Chronic Ulceration)	5 (15.2)	6.0 (0.0)	NA

* Multiple responses were allowed from a single participant.
[†] SD: Standard deviation.
[‡] CRS: Confidence rating scale.
[§] Hypotheses with a mean CRS score < 6.0.
[¶] NA: Not applicable.

the respondents believed the definition of BMS should be more encompassing, including the entire oral mucosa (n = 10) rather than being restricted to the tongue region (n = 4). Certainly, the presence of a burning sensation in the entire

oral mucosa as compared with that only in the tongue region is more consistent with the clinical findings described in the literature.^{3,14,16-18} It is possible the majority of respondents did not endorse the inclusion of chronic pain (n = 4) in

the definition of BMS because they considered that an acute onset of an oral burning sensation would be an acceptable criterion to be included in a definition of BMS. Alternatively, they may have believed that the concept of chronic pain, as defined in our study (pain lasting longer than three months), was not consistent with their perception of chronic pain, which possibly involved a longer period (for instance, six months). The nature of these responses may reflect, in part, the nature of the referral network and the delay in patients' referral to OM and OFP clinics and programs.³³ We did not investigate further the issue of the temporal component regarding chronic pain in this study.

To assist the health care practitioner in providing a definitive diagnosis and to assist with consistency among clinical trials, it is important to standardize diagnostic criteria. Overall, participants in this study were confident in their responses in this regard in identifying the key diagnostic criteria, and they reported criteria similar to those published in the literature (Table 2) as being necessary for a definitive diagnosis of BMS.^{5,34-39}

It is of paramount importance for the health care practitioner to understand that BMS is a diagnosis supported by the nature of the symptomatic complaint and the exclusion of various local and systemic factors (Table 3). A detailed discussion of these factors is beyond the scope of this article; however, reviews regarding them appear in the literature.³⁶⁻⁴¹

The results of our study (Table 3) indicate that participants' responses were consistent with the current literature regarding the factors that need to be ruled out in the diagnosis of BMS. These were reported with a moderate to high level of confidence (range of mean CRS scores, 5.9-6.8; 95 percent CI, 5.20-7.24). The emphasis on the need to rule out fungal infection may be due to the often associated elevated prevalence of *Candida* species reported in people with BMS.⁴²⁻⁴⁷ The presence of a fungal infection—often associated with a bitter or metallic taste (a symptom also commonly reported by patients with BMS) and clinical findings of erythema (erythematous candidiasis) or pseudomembranes (pseudomembranous candidiasis) on the oral mucosa—often represents the true source of oral burning pain.⁴⁵ Patients with these symptoms may report increased pain on eating, likely because of irritation of the mucosa.^{48,49} The importance of ruling out the presence of a fungal infection cannot be understated—and if such an infection is identified, a diagnosis of secondary BMS would be appropriate, with

symptoms managed accordingly for an infection of this type.

Before diagnosing BMS, the health care practitioner would be prudent to obtain a thorough history and perform a comprehensive examination involving the use of adjunctive tests, imaging or both when deemed necessary. The use of certain diagnostic tests assists in ruling out factors that may be responsible for the oral burning symptoms, thus distinguishing primary from secondary BMS.

Eleven respondents endorsed the measurement of salivary flow rates as an important diagnostic test to determine salivary gland hypofunction or dysfunction. Although there is controversy among clinicians regarding the role of salivary flow in BMS, our data suggest that health care practitioners should incorporate into their diagnostic armamentarium and decision-making processes a means of objectively measuring salivary flow rates and methods of ruling out salivary conditions before they provide a definitive diagnosis of BMS. Participants did not indicate the need for use of imaging (dental or medical) in the diagnosis of BMS. This was most likely because we did not provide participants with the option of endorsing this diagnostic test, having deliberately excluded it from the designated response categories (Figure, question 7). There may have been additional diagnostic testing alternatives that we overlooked and did not include in the questionnaire. Furthermore, owing to the nature of the methodology, by which we forced respondents to choose a response from a prescribed menu, we were not able to elicit these other possibilities (we provided no space in which they could record diagnostic tests not present in the menu supplied), thereby introducing the potential for bias to the responses. It is interesting that three participants endorsed the use of soft-tissue biopsy as an important diagnostic test to rule out a diagnosis of BMS. Although there is no established consensus regarding the sampling of soft tissue (for example, to rule out mucosal disease or to observe small-fiber axonal degeneration in the tongue) for a definitive diagnosis of BMS, it is possible these participants responded in this manner because they suspected other soft-tissue diseases⁷ (Table 3) causing oral burning sensations that could be misconstrued as BMS.

Although the precise etiology and pathophysiology of BMS still is elusive (Table 4), the results of this survey are representative of the current published literature regarding BMS involving both central¹⁵⁰⁻⁵³ and peripheral^{10,54-56} neuropathic mechanisms. More than one-half

of the total number (n = 33) of responses (54.5 percent) supported the concept of BMS' having a neuropathic etiopathogenesis. Additionally, theories involving psychological and psychosocial issues^{25,57-63} and hormonal factors^{3,8,64,65} reported in the literature also were reported by the respondents. Generally, in comparison with other responses regarding the various topics addressed in the study, participants displayed a lower level of confidence in their responses in this category. This may be explained by the lack of strong scientific evidence supporting any one particular theory for the etiopathogenesis of BMS (perhaps with the exception of the neuropathic component).

Limitations. This study was limited by being based on self-reports gathered via a mainly open-ended survey. The open-ended format required categorization of responses by the investigators, which may have introduced misclassification bias. Furthermore, because the number of people surveyed was limited by the number of accredited programs (that is, 20 programs), the actual number of participants (n = 13, providing a response rate of 65 percent) was small. Although we did not conduct a formal assessment of each participant's knowledge regarding BMS and did not control for variability in participants' education, these individuals had considerable clinical education and experience in the diagnosis and management of BMS and were active in caring for patients with these symptoms. The CRS technique allowed us to gauge their level of confidence in their responses. Involving a greater number of participants by expanding the survey beyond North America or recruiting a greater number of experienced clinicians in the disciplines of OM and OFP and applying nominal techniques for consensus building would be of great benefit and should be a consideration in future studies. We did not perform a formal sample-size calculation, as our intent in this study was to capture the universe of all training program directors in North America. Furthermore, the reliability of the CRS technique, owing to its cross-sectional nature, could not be demonstrated in this study.

CONCLUSIONS

The findings in this study present an initial exploration of the perceptions of program directors of OM and OFP postgraduate programs in North America regarding diagnostic paradigms, clinical presentations and etiologic and pathophysiological theories regarding BMS. The participants were postgraduate program directors who were clinicians experienced in the diagnosis

and management of BMS. We observed similarities among respondents who had a high degree of confidence regarding variables associated with the diagnosis of BMS, such as neuropathic etiopathogenesis and objective assessment of salivary flow. Global standardization of the defining criteria for BMS and necessary diagnostic algorithms can be formulated by convening leaders in the field from different geographical regions. This worthwhile exercise, in addition to following strict methods for the development of a comprehensive consensus statement that expands the current definitions of BMS described earlier in this article, certainly would aid in the advancement of our knowledge of this condition to the benefit of both health care practitioners and the patients they serve. ■

Disclosure. None of the authors reported any disclosures.

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