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A Population-Based Study of the Incidence of Burning Mouth Syndrome

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Abstract

Objective—To calculate the incidence of burning mouth syndrome (BMS) in Olmsted County, Minnesota, from 2000 to 2010.

Patients and Methods—Using the medical record linkage system of the Rochester Epidemiology Project, we identified newly diagnosed cases of BMS from January 1, 2000, through December 31, 2010. Diagnoses were confirmed through the presence of burning pain symptoms of the oral mucosa with normal oral examination findings and no associated clinical signs. Incidence was estimated using decennial census data for Olmsted County.

Results—In total, 169 incident cases were identified, representing an annual age- and sex-adjusted incidence of BMS of 11.4 per 100,000 person-years. Age-adjusted incidence was significantly higher in women than men (18.8 [95% CI, 16.4–22.9] vs 3.7 [95% CI, 2.6–5.7] per 100,000 person-years [$P<.001$]). Postmenopausal women aged 50 to 89 years had the highest disease incidence, with the maximal rate in women aged 70 to 79 years (70.3 per 100,000 person-years). After age 50 years, BMS incidence in men and women significantly increased across age-groups ($P=.02$). Olmsted County study participants were predominantly white, which is a study limitation. In addition, diagnostic criteria for identifying BMS in the present study may not apply for all situations because no diagnostic criteria are universally recognized for identifying BMS.

Conclusion—To our knowledge, this is the first population-based incidence study of BMS reported to date. The data show that BMS is an uncommon disease highly associated with female sex and advancing age.

Keywords

burning mouth syndrome; incidence; Olmsted County; population-based; Rochester Epidemiology Project

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Background

Burning mouth syndrome (BMS) is defined as a chronic burning pain of the oral mucosa with no associated clinical signs or apparent extraneous cause. Over the years, BMS has also been known as *orodynia*, *glossodynia*, *stomatodynia*, *oral dyesthesia*, *glossopyrosis*, *stomatopyrosis*, *sore mouth*, *sore tongue*, and *persistent idiopathic orofacial pain*. The burning discomfort is typically bilateral and can localize to any area of the oral cavity—most commonly, the tongue.^{1–3} BMS is associated with xerostomia, dysgeusia, and such medical conditions as diabetes mellitus, chronic pain, anxiety, and depression.^{4–6}

The pathogenesis of BMS is poorly understood. Psychological, hormonal, neurologic, and allergenic etiologic factors have been proposed.^{7–15} Tongue thrusting, lip sucking, depression, and smoking are risk factors for BMS.² Pharmacologic treatment of BMS relies on clonazepam, capsaicin, tricyclic antidepressants, and other medications used to treat neuropathic pain.^{16,17} Cognitive behavioral therapy for patients with psychiatric comorbidities may be beneficial.¹⁸ Other nonpharmacologic treatments have involved the recommendation of smoking cessation and biofeedback therapy.¹⁹

The diagnosis of BMS, according to the third edition of International Classification of Headache Disorders, requires a superficial, burning oral pain that recurs for more than 2 hours per day for more than 3 months with normal-appearing oral mucosa and no more-appropriate diagnosis.²⁰ Other causes of burning pain that must be excluded are salivary hypofunction, menopause, oral candidiasis, nutritional deficiencies, endocrinopathies, bruxism, medication adverse effects, dental trauma, mucosal irritation from dentures, and allergic contact stomatitis. Because of the challenging nature of the BMS diagnosis, the mean time from onset of symptoms to BMS diagnosis is more than 1 year. BMS is also misdiagnosed by an average of more than 3 physicians before the correct diagnosis is made.²¹

Perhaps owing to the difficulty of diagnosing BMS, limited epidemiologic data have been published (Table 1).^{1,5,22–31} Depending on the study, prevalence rates range from 1% to 40%. The disease is highly prevalent in middle-aged women.^{3,32,33} To our knowledge, there are no population-based data on the incidence of BMS. The aim of this study was to calculate the incidence of BMS in a single well-studied and well-characterized US county.

Methods

Study Setting

The present study was approved by the Mayo Clinic and Olmsted Medical Center institutional review boards. We assessed cases of BMS within the population of Olmsted County, Minnesota, made available through the Rochester Epidemiology Project (REP). REP is a medical records linkage system for the patient records of Olmsted County residents, which has accumulated all medical information on each patient since 1966. Practically the entire Olmsted County population receives care at a REP facility and is represented by the linkage system. In 2000 and 2012, 98% of this population agreed to participate in clinical research for at least 1 provider.³⁴ The population of Olmsted County is

largely white (86.5% in 2012) and socioeconomically and demographically similar to the white population in the United States. The county is largely isolated from a major metropolitan area, with a net in-migration.^{34–36}

Study Criteria

The REP was screened with the following 3 criteria: diagnosis or potential diagnosis of BMS through December 31, 2010; alive from January 1, 2000, through December 31, 2010; and date of last follow-up after January 1, 2000. The diagnosis codes for screening used the Hospital Adaptation of the International Classification of Diseases code “mouth burning” and the International Classification of Disease-9 code 528.9, “other and unspecified diseases of the oral soft tissues.”¹ Twenty-one patients without research authorization were excluded. The study was limited to incident cases; therefore, patients with a known diagnosis of BMS were excluded. All cases used in the study are residents of Olmsted County, received medical care at one of the REP facilities, and had a first diagnosis of BMS at a REP facility. The REP assigns a unique identifier for each person across all participating institutions. The earliest BMS incident was retained on the basis of the unique person identifier. Each chart was reviewed to confirm a specific BMS diagnosis. Diagnoses were confirmed through the presence of oral burning symptoms with a normal oral examination.

Data Collection

The objective of the study was to calculate incidence of BMS in Olmsted County. A retrospective review of the medical records documented date of birth, date of diagnosis, sex, smoking status, and race. The date of BMS diagnosis was the date when the patient received a first-ever diagnosis of BMS by either a dermatologist or a nondermatologist physician, provided the patient’s characteristics met BMS criteria. At each physician visit, the following data were collected: site of burning pain, intensity of pain, mediolateral and anteroposterior localization of symptoms, pattern of pain, temporal classification of pain, presence of dysgeusia, and presence of xerostomia. Intensity of pain was abstracted from either a 0-to-10 numeric pain scale or subjective description of mild, moderate, or severe pain. Abstracted data were entered into a database created for this particular study. With the assigned REP numbers, duplicate entries were identified and only the earliest BMS entries were retained.

Statistical Analyses

Data were analyzed with the SAS version 9.3 software package (SAS Institute Inc). The number of persons with a BMS diagnosis between 2000 and 2010 was used as the incidence rate numerator, while the decennial census data were used to determine the denominator for the incidence rate calculation. Age- and sex-specific incidence rates of BMS in Olmsted County during 2000 through 2010 were calculated overall and by each of the 11 years. With the total population structure in the United States in 2010, the sex- and age-adjusted rates were calculated. Between census years, linear interpolation was used. Poisson error distribution was used to compute the 95% confidence intervals. Patients were grouped by age with 0 to 40 years being the range of the youngest age-group, followed by age-groups 50 to 59, 60 to 69, 70 to 79, 80 to 89, and 90 years and older. All persons, including children

younger than 18 years, living in Olmsted County were assumed to be at risk for BMS during the incidence rate calculation.

Comparisons of incidence rates to sex, age-group, and time of diagnosis were assessed with a generalized linear model assuming a Poisson error structure. The observations used for the regression were crude incidence counts for all combinations of calendar year, age-group, and age, which were offset by the natural logarithm of the number of persons. The regression was used to determine statistically significant differences across age-groups and sex and to determine whether there was a statistically significant interaction between age-groups and sex. A similar regression was used to test for a linear trend over time periods. Likelihood ratio statistics with 1 degree of freedom were used to calculate statistical significance of linear trends in both time periods and age groupings. A dispersion parameter estimation was used to resolve the problem of overdispersion.³⁷

Results

Between January 1, 2000, and December 31, 2010, a total of 482 patients were identified as having a potential BMS diagnosis. Of the 482 patients, 191 were identified as having had a first-ever diagnosis of BMS as a resident of Olmsted County. Using our study criteria, we confirmed 169 incident cases. The majority of the 169 patients were female (84.0%), with a 5.2 ratio of women to men (Table 2). Most patients were white (93.5%). The mean age at diagnosis was 61.4 years (range, 19–98 years). At diagnosis, 11.2% of patients were current smokers and 26.0% were prior smokers.

The tongue was the most frequently involved anatomic location, affecting 52.7% of the 169 cases independently and 28.4% of cases in conjunction with other anatomic sites (Table 3). The lips (1.2%) and palate (0.6%) were rarely independently affected. However, the lips (20.8%), palate (10.1%), gingiva (6.6%), and buccal mucosa (6.0%) were often affected in conjunction with other anatomic sites. Intensity of reported pain had a broad range, but the majority had mild or moderate pain (69.8%). Disease was commonly symmetrical on both the mediolateral (88.8%) and anteroposterior (74.6%) axes. Pain was typically continuous (86.4%) and present throughout the day (69.2%). Dysgeusia of any type was present in 14.2% of cases (n=24), and 54% (n=13) of the 24 dysgeusia cases involved altered taste and 33% (n=8) a metallic taste. Xerostomia, a subjective report of oral dryness, was present in 33.7% of BMS patients.

The overall annual age- and sex-adjusted incidence of BMS in Olmsted County during the 11 years from 2000 through 2010 was 11.4 (95% CI, 10.6–14.4) per 100,000 person-years. The incidence of BMS in women exceeded the rate in men significantly: 18.8 (95% CI, 16.4–22.9) vs 3.7 (95% CI, 2.6–5.7), respectively, per 100,000 person-years when adjusted for age ($P<.001$). The highest incidence rate was in women aged 70 through 79 years (70.3 per 100,000 person-years); the highest incidence in men was also in the age-group 70 to 79 years (18.4 per 100,000 person-years) (Table 4). After age 50 years, the BMS incidence in both men and women significantly increased across age-groups ($P=.02$). No significant difference was found between the sexes in this increase ($P=.79$). In addition, no significant

linear trend from 2000 through 2010 was present in incidence rates of BMS ($P=.29$) (Table 5).

Discussion

The present population-based study of BMS calculated an overall annual age- and sex-adjusted incidence of BMS of 11.4 per 100,000 person-years in Olmsted County from 2000 through 2010. To our knowledge, no previously published study has reported incidence rates of BMS. Postmenopausal women aged 50 to 89 years had the highest incidence of disease, with the maximal rate seen in women aged 70 to 79 years. BMS incidence is associated with both advanced age and female sex. In addition, the mean age at diagnosis was 61.4 years. Overall, 37.2% of BMS patients had either a history of tobacco use or were currently using tobacco. The 11.2% rate of current tobacco use was comparable to the overall 12% rate of smoking in Olmsted County in 2014.³⁸ Most patients had mild or moderate burning pain (69.8%) with tongue involvement (81.1%) that was present throughout the day (69.2%).

The demographic characteristics of the patients in the present study are similar to those reported previously in the literature. The preponderance of women was consistent with other cohorts that reported a prevalence of 75% to 87%.^{2,5,39} As in this study sample, patient groups with the greatest BMS burden have been women of middle age and older.^{2,3,39} This finding has been attributed to both the frequency of psychological disorders in this group and the perimenopausal decline in estrogen and progesterone levels, which may lead to xerosis.⁴⁰ Increased use of hormone replacement therapy could also contribute to the high disease incidence in this group. The high disease occurrence in this group has also provided the basis for the proposal of hormonal pathogenesis of BMS.⁹

Higher rates of white patients (93.5%) were found in this BMS cohort than are observed in the general population of Olmsted County (90.3% white in 2000 and 85.7% white in 2010).³⁴ This finding may represent a predominance of BMS among white patients, but there are sparse reported data on BMS and racial demographic characteristics for comparison.

Tobacco use is not associated with BMS in the present cohort because the rate of use was equivalent to the rate in the general population of Olmsted County (12%, 2014).³⁸ Prior studies have found significant associations of smoking with BMS, citing odds ratios greater than 12,^{2,41} whereas a separate study found recent smoking cessation to be a risk factor for BMS development.² Some investigators have proposed that there may be a link between smoking and BMS since smoking causes a taste disturbance in some persons.³¹

Symptom characteristics in the present cohort were analogous to those cited in the literature. Many patients from this sample had symmetrical, bilateral mild burning pain of the tongue that was continuous throughout the day. The tongue has been described as the most frequently involved anatomic location,^{2,3} a claim supported by our study. Patients described their tongue symptoms as being more commonly anterior than posterior, which has been described previously.⁸ The present study also supports the typical bilateral distribution of BMS.^{3,39} Prior studies have cited scarce involvement of the lips, palate, buccal mucosa, and

gingiva.^{2,3} In contrast, we found frequent involvement in other anatomic sites, particularly the lips (20.8%) and palate (10.1%). Pain intensity in this population was milder than in other studies, which reported moderate to severe pain at the time of BMS diagnosis.^{3,39}

Findings of the present study support the claim that the pattern of symptoms in BMS is usually continuous, rather than paroxysmal.⁴² Lamey and Lewis⁴³ proposed 3 temporal types for BMS. Types 1, 2, and 3 involve, respectively, pain absent on waking and developing during the day, pain present throughout the day, and intermittent pain with pain-free days. Our patient population was most commonly classified as having type 2 BMS. Interestingly, 2 patients noted that their symptoms were present at night rather than during the day, a temporal classification that to our knowledge had been unreported previously. The prevalence of dysgeusia in our cohort was less than typically observed. In prior studies, as many as 60% of a BMS cohort reported a bitter or metallic taste and 35% reported altered taste.³ Xerostomia was present in 33.7% of patients, which is less common than other previously reported values.^{3,27,39} Only 1 patient reported sialorrhea, which has been similarly rare in previous BMS studies. Some investigators have suggested that sialorrhea may be a misinterpretation of xerostomia.⁴⁴ Xerostomia, the subjective report of oral dryness and a common symptom of BMS, must be understood as a separate entity from salivary hypofunction, which is a potential cause of oral burning.

The data show that BMS is highly associated with female sex and advancing age. In contrast, Olmsted County was only 51.1% female with 13.3% of the population older than 65 years in 2012. There was a decrease in female incident cases in 2009. Review of coding methods performed by the REP from 2000 through 2010 showed this finding to be accurate and not the result of missed cases. The analysis also showed incidence rates that increased significantly after age 50 years across age-groups but did not show statistically different trends in age between the sexes. The lack of sex difference may be owing to the low incident rates in male patients. As shown in Table 1, the reported prevalence rates of BMS differ widely, from 1.0% to 40%.^{1,5,22–31} Most studies report a prevalence of about 1% to 6%, with a significantly higher prevalence in women. Our study is suggestive that BMS is an uncommon disease. Confounding factors such as xerostomia may also be problematic. Xerostomia has been proposed to cause oral burning.^{9,39} One study found that in patients with xerostomia and BMS, 37% reported a reduction or alleviation of their burning symptoms after the treatment of xerostomia alone.³⁹ In addition, the use of diverse study methods in earlier studies—including surveys, questionnaires, and retrospective reviews—influences results and complicates comparison.^{5,22,23}

Lack of consistent BMS diagnostic criteria across studies also has contributed to variability. Many studies evaluated the symptom of oral burning rather than BMS itself.^{24,26,42} A particular study, by Tammiala-Salonen et al,¹ found that after the treatment of all possible organic causes in their cohort of patients with suspected BMS, the prevalence rate decreased from 15% to 1%.

The present study has several limitations. Pain intensity was not recorded consistently but was either documented numerically on the 0-to-10 numeric pain scale or subjectively described as mild, moderate, or severe pain, which could introduce variability into the

interpretation of these data. The population of Olmsted County used in this study is largely white (86.5% in 2012) and most patients in this cohort (white race, 93.5%) were reflective of the population. On the basis of racial demographic characteristics in this county, the epidemiological findings may be less applicable to more diverse populations. Of note, the third edition of the International Classification of Headache Disorders was published in 2013,²⁰ and patient data in the present study were from 2000 through 2010. By comparison with the criteria of the second edition, a BMS diagnosis described in the third edition requires recurrent daily pain for more than 2 hours per day for greater than 3 months, with a burning quality that is felt superficially in the oral mucosa.²⁰ Though these criteria were not used explicitly as diagnostic criteria in the present cohort, it is unlikely that many cases of BMS were included erroneously in this study. The present study did not analyze the medications and concurrent diseases that could be contributing to the symptoms of burning mouth, xerostomia, dysgeusia, and altered taste in this patient group.

Conclusion

We report the first population-based incidence study of BMS. The data show BMS is an uncommon disease, highly associated with both advancing age and female sex. Typical symptom characteristics show mild, symmetrical, bilateral burning pain of the tongue that is continuous throughout the day. Our findings provide a foundation for further studies of BMS epidemiology. Further studies should explore the potential influence of factors such as concurrent disease and medication use on the incidence of BMS on population groups.

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Abbreviations

BMS	burning mouth syndrome
REP	Rochester Epidemiology Project

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Table 1

Epidemiological Studies of Burning Mouth Syndrome

Reference	Incidence, ^a %	Prevalence, %	Study Method
Kohorst et al, present report	12.6, total; 4.3, male; 19.8, female		Population-based retrospective review of Olmsted County, Minnesota
Dangore-Khasbage et al, 2012 ²²		20.7	Cross-sectional study of 150 psychiatric patients at a single hospital in India
Netto et al, 2011 ²³		1.0	Retrospective review of 3,243 patients referred to oral pathology department at a single hospital in Brazil over 7 y
Suzuki et al, 2010 ²⁴		3.0, total; 2.8, male; 3.2, female	Questionnaire of 2,599 dental patients at 2 offices in Japan
Rabiei et al, 2010 ²⁵		16.9	Cross-sectional study of 216 nursing-home residents in Iran
Moore et al, 2007 ²⁶		2.1	Cross-sectional study of 216 volunteer participants in Pittsburgh, Pennsylvania
Brailo et al, 2006 ²⁷		5.4	Retrospective review of 1,399 patients referred to oral medicine department in Croatia in 2001
Bergdahl and Bergdahl, 1999 ⁵		3.7, total; 1.6, male; 5.5, female	Survey of 1,427 Swedish volunteers (age, 20–69 y)
Riley et al, 1998 ²⁸		1.7	Telephone survey of 1,636 volunteers from North Florida (age, >65 y)
Hakeberg et al, 1997 ²⁹		4.6	1,017 middle-aged and elderly women in Sweden
Ben Aryeh et al, 1996 ³⁰		10–40	154 women presenting with menopausal symptoms to menopause clinic
Thorstensson and Hugoson, 1996 ³¹		3.4	Survey of 533 Swedish volunteers (age, 20–70 y)
Tammiala-Salonen et al 1993 ¹		14.8	431 Finnish volunteers

^aIncidence from January 1, 2000, through December 31, 2010.

Table 2

Demographic Characteristics of Patients With Burning Mouth Syndrome

Characteristic	Total ^a (N=169)
Sex	
Female	142 (84.0)
Male	27 (16.0)
Age at BMS diagnosis, y	
Mean (SD)	61.4 (15.7)
Range	19–98
Race	
White	158 (93.5)
Nonwhite	11 (6.5)
Smoking status	
Current smoker	19 (11.2)
Former smoker	44 (26.0)
Never smoked	106 (62.7)

^aValues are presented as number and percentage of patients unless specified otherwise.

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Table 3

Symptom Characteristics With Burning Mouth Syndrome

Characteristic	Patients, No. (%) (N=169)
Site of burning pain	
Tongue only	89 (52.7)
Palate only	1 (0.6)
Lips only	2 (1.2)
Several sites, including tongue	48 (28.4)
Several sites with tongue unspecified	29 (17.2)
Intensity	
Mild (1–3 on pain-intensity scale)	71 (42.0)
Moderate (4–7 on pain-intensity scale)	47 (27.8)
Severe (8–10 on pain-intensity scale)	22 (13.0)
Variable	15 (8.9)
Data missing	14 (8.3)
Mediolateral localization	
Unilateral	9 (5.3)
Bilateral	150 (88.8)
Data missing	10 (5.9)
Anteroposterior localization	
Anterior	28 (16.6)
Posterior	4 (2.4)
Midline	126 (74.6)
Data missing	11 (6.5)
Pattern	
Continuous	146 (86.4)
Paroxysmal	9 (5.3)
Data missing	14 (8.3)
Temporal classification ⁴⁴	
Type 1 (pain absent on waking and developing during the day)	20 (11.8)
Type 2 (pain present throughout the day)	117 (69.2)
Type 3 (intermittent pain with pain-free days)	15 (8.9)
Pain at night, not during the day	2 (1.2)
Data missing	15 (8.9)
Dysgeusia	
Persistent taste	1 (0.6)
Altered taste	13 (7.7)
Metallic taste	8 (4.7)
Bitter taste	2 (1.2)
Not present	145 (85.8)
Xerostomia	
Present	57 (33.7)

Characteristic	Patients, No. (%) (N=169)
Not present	111 (65.7)
Sialorrhea	1 (0.6)

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Table 4
Incidence of Burning Mouth Syndrome per 100,000 Person-years by Age and Sex in Olmsted County, Minnesota, 2000–2010

Age-group, y	Women			Men			Both Sexes		
	No. of Cases	Rate per 100,000 Person-years	No. of Cases	Rate per 100,000 Person-years	No. of Cases	Rate per 100,000 Person-years	No. of Cases	Rate per 100,000 Person-years	
0–49	26	4.9	9	1.7	35	3.3			
50–59	35	37.0	7	7.8	42	22.8			
60–69	32	54.3	3	5.6	35	31.0			
70–79	28	70.3	6	18.4	34	46.9			
80–89	18	68.4	2	13.1	20	48.1			
90+	3	40.6	0	0.0	3	30.6			
Total rate (95% CI)		18.8 (16.4–22.9) ^a		3.7 (2.6–5.7) ^a		11.4 (10.6–14.4) ^b			

^a Incidence per 100,000 person-years is age-adjusted directly to the population structure of the US total population in 2010.

^b Incidence per 100,000 person-years is age- and sex-adjusted directly to the population structure of the US total population in 2010.

Table 5
Incidence of Burning Mouth Syndrome per 100,000 Person-years Over 11 Years in Olmsted County, Minnesota, 2000–2010

Year	Women			Men			Both Sexes		
	No. of Cases	Incidence Rate per 100,000 Person-years (95% CI) ^a	No. of Cases	Incidence Rate per 100,000 Person-years (95% CI) ^a	No. of Cases	Incidence Rate per 100,000 Person-years (95% CI) ^b			
2000	9	14.2 (6.5–27.0)	5	8.2 (2.7–19.1)	14	11.3 (6.2–18.9)			
2001	14	21.8 (11.9–36.6)	2	3.2 (0.4–11.6)	16	12.7 (7.2–20.6)			
2002	16	24.5 (14.0–39.8)	2	3.2 (0.4–11.5)	18	14.0 (8.3–22.2)			
2003	9	13.6 (6.2–25.7)	1	1.6 (0.04–8.7)	10	7.7 (3.7–14.1)			
2004	14	20.8 (11.4–34.8)	2	3.1 (0.4–11.1)	16	12.1 (6.9–19.6)			
2005	17	24.8 (14.5–39.7)	3	4.6 (0.9–13.3)	20	14.9 (9.1–23.0)			
2006	17	24.4 (14.2–39.1)	2	3.0 (0.4–10.8)	19	13.9 (8.4–21.8)			
2007	23	32.6 (20.7–48.9)	3	4.4 (0.9–13.0)	26	18.8 (12.3–27.6)			
2008	11	15.4 (7.7–27.5)	1	1.5 (0.04–8.1)	12	8.6 (4.4–14.9)			
2009	4	5.5 (1.5–14.1)	4	5.8 (1.6–14.7)	8	5.6 (2.4–11.1)			
2010	8	10.8 (4.7–21.4)	2	2.8 (0.3–10.3)	10	6.9 (3.3–12.7)			

^aIncidence per 100,000 person-years is directly age-adjusted to the population structure of the US total population in 2010.

^bIncidence per 100,000 person-years is directly age- and sex-adjusted to the population structure of the US total population in 2010.