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REVIEW ARTICLE

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Worldwide prevalence estimates of burning mouth syndrome: A systematic review and meta-analysis

Shuangshuang Wu[1] | Wenqing Zhang[1] | Jingxian Yan[2] | Noboru Noma[3] | Andrew Young[4] | Zhimin Yan[1]

[1]Department of Oral Medicine, Peking University School and Hospital of Stomatology, Beijing, China

[2]Chinese Medical College, Tianjin University of Traditional Chinese Medicine, Tianjin, China

[3]Department of Oral Diagnostic Sciences, Nihon University School of Dentistry, Tokyo, Japan

[4]Department of Diagnostic Sciences, Arthur Dugoni School of Dentistry, University of the Pacific, Stockton, CA, USA

Correspondence
Zhimin Yan, Department of Oral Medicine, Peking University School and Hospital of Stomatology, Beijing, 100081 China.
Email: yzhimin96@163.com

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Abstract

Objectives: To evaluate the worldwide prevalence and epidemiology profile of burning mouth syndrome.

Material and Methods: A systematic review and meta-analysis was conducted. Search strategies were performed in PubMed, EMBASE, Web of Science, the Cochrane Library, China National Knowledge Infrastructure, and Wanfang database for studies published before January 31, 2021, for the prevalence of burning mouth syndrome.

Results: Eighteen articles were included. The overall pooled prevalence of burning mouth syndrome was 1.73% (95% CI = 0.176–0.351, n = 26,632) in general population, and 7.72% (95% CI = 0.434–0.691, n = 86,591) in clinical patients. The subgroup analysis by continent showed that among the population-based studies the prevalence in Asia (1.05%) lower than in Europe (5.58%) and North America (1.10%). The subgroup analysis by gender showed the prevalence of female (1.15%) was higher than male (0.38%) in general population. The subgroup analysis by age showed the prevalence was higher for people over 50 (3.31%) than under 50 (1.92%).

Conclusions: The pooled prevalence of burning mouth syndrome was relatively high in both general population and clinical patients, varies in different regions with the highest prevalence in Europe, and females over 50 years were the most susceptible group. More epidemiological surveys on the prevalence of burning mouth syndrome are needed.

KEYWORDS

burning mouth syndrome, epidemiology, meta-analysis, prevalence

1 | INTRODUCTION

Burning mouth syndrome (BMS) is a chronic pain disorder that mainly affects menopausal or postmenopausal women (Komiyama et al., 2013; Teruel & Patel, 2019). It was defined by the International Association for the Study of Pain (IASP) as a chronic intraoral burning sensation that has no identifiable cause either local or systemic condition or disease (Treede et al., 2019). The pain caused by BMS mainly involves tongue and palate, accompanying xerostomia and taste changes, and affects patients' daily activity as speaking and eating (Ariyawardana et al., 2019; Klasser et al., 2008). BMS has a rather long disease course with an average of 6 to 7 years, and as such a chronic orofacial pain disorder, it is frequently associated with psychological distress, anxiety, and depression (Ducasse et al., 2013). It inevitably brings a tremendous individual and societal impact, resulting in long-term sick leave and poor quality of life, and causing high socioeconomic costs and a relatively large burden on healthcare system (Pereira et al., 2021).

Population-based studies indicated that BMS affects a substantial proportion of adults. The prevalence of BMS is reported ranging widely from 0.7% to 15% in various races, populations, and settings

(Beneyto et al., 2008; Bergdahl et al., 1999; Coculescu et al., 2014). In a population-based study by Lou. et al., the prevalence of BMS was estimated at 1.38% of residents aged 17 to 92 years in Shanghai, China (Lou et al., 2016). Another cross-sectional study conducted in Sweden showed a prevalence of BMS of 3.7% among total of 1,427 adults between the ages of 20 and 69 (Bergdahl et al., 1999). While it raised up to 15% in a clinical-based retrospective study from Brazil (Fattori et al., 2019).

Although the studies assessed BMS distribution in the target population, the knowledge of the worldwide prevalence and epidemiology profile of BMS is still inadequate. Therefore, we conduct this systematic review and meta-analysis to qualitatively and quantitatively evaluate the best available scientific evidence on the global BMS prevalence profile in order to address the need for healthcare professionals, researchers, and policymakers.

2 | MATERIALS AND METHODS

This systematic review was reported in accordance with the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement) (Moher et al., 2009).

2.1 | Protocol

The protocol was registered a priori in PROSPERO international prospective register of systematic reviews (CRD42021229903).

2.2 | Search strategy

The two authors (SS W and WQ Z) independently searched for relevant studies in PubMed, EMBASE, Web of Science, the Cochrane Library, and two Chinese databases (China National Knowledge Infrastructure [CNKI], Wanfang database) published before January 31, 2021(Gray literature databases was not included). The terms and keywords we searched were as follows: (prevalence OR epidemiology OR epidemiological OR "cross-sectional survey" OR "cross-sectional study") AND ("burning mouth syndrome" OR stomatodynia OR stomatopyrosis OR glossopyrosis OR glossodynia OR "sore mouth" OR "sore tongue" OR "oral dysesthesia" OR glossalgia OR "chronic orofacial pain"). The references of relevant studies were also manually searched to avoid omitting any study related to our review.

2.3 | Eligibility criteria

The objective of this study was to evaluate the worldwide prevalence of BMS, meanwhile, analyze the association between the prevalence and possible related variables. The following eligibility criteria were based on the CoCoPop framework (Munn et al., 2015, 2018).

2.3.1 | Type of participants

Patients were diagnosed with BMS in population-based studies and clinical-based studies.

2.3.2 | Condition

Studies of the prevalence of BMS and with adequate data (e.g., sex and age distribution, geographic location, publication year) to get the prevalence of BMS and the epidemiology profile of BMS among different settings.

2.3.3 | Context

All studies conducted in countries around the world to investigate the prevalence of BMS in general population or clinical settings.

2.3.4 | Type of studies

Original studies written in English or Chinese, population or clinical-based, analyzing BMS prevalence with sufficient data. When the results were come from the same study population, we will include the most recently published or those providing more complete data. Moreover, review articles, meta-analyses, case reports, protocols, short communications, personal opinions, letters, conference abstracts, or laboratory research were excluded.

2.4 | Data extraction

SS W and WQ Z independently screened the full-text articles to determine inclusion. If the two authors had divergence, ZM Y, N N, A Y came to a consensus. Then, data were collected and analyzed using standardized forms. The following data were extracted from each included study: the first author, publication year, corresponding author affiliation, place and country, source of patients, recruitment period, study design, population sample size, number of BMS cases, diagnostic criteria, sex, age, and other characteristics (Table 1 and Table 2).

2.5 | Critical appraisal

SS W and WQ Z evaluated the quality of studies by the methods recommended for systematic reviews addressing disease prevalence questions (Joanna Briggs Institute, University of Adelaide) (Table S1). ZM Y, N N, A Y were involved in the discussion of uncertain events. This guidance consists of nine items and studies were categorized based on the percentage of yes answers as high quality (≤49%), moderate quality (50%–69%), or low quality (≥70%) (Munn et al., 2015).

TABLE 1 Characteristics of population-based studies of burning mouth syndrome

Study	Continent	Number of		Prevalence, %	Age, y	Study Period	Source of participants	Diagnostic method
		BMS	Total					
Bergdahl et al., 1999 (Sweden)	Europe	53 Male: 11 Female: 42	1,427 Male: 669 Female: 758	3.75	20–69	NR	The Public Dental Health Service Registers	C (NR)
Lou et al., 2016 (China)	Asia	9 Male: 3 Female: 6	653 Male: 337 Female: 316	1.46	17–92	2014	General Population	C (equivalent to IASP, 2013)
Tammiala-Salonen et al., 1993 (Finnish)	Europe	34	431 Male: 194 Female: 237	7.99	30–91	NR	General Population	C (NR)
Kohorst et al., 2015 (US)	North America	149 Male: 24 Female: 125	13,545	1.10	25–90	2000–2009	The Rochester Epidemiology Project (REP) Medical Records Linkage System	C (NR)
Cheng et al., 2007 (China)	Asia	79	1,154 Male: 519 Female: 635	6.88	>60	NR	Elderly Population	C (equivalent to IASP, 2013)
Wang et al., 2020 (China)	Asia	7 Male: 4 Female: 3	1,000 Male: 560 Female: 440	0.75	15–80	2018–2019	Rural Population	C (equivalent to IASP, 2013)
Zhang et al., 2016 (China)	Asia	21 Male: 0 Female: 21	3,756 Male: 1771 Female: 1985	0.57	35–74	2015–2016	General Population	C (equivalent to IASP, 2013)
Miao et al., 2009 (China)	Asia	4 Male: 2 Female: 2	3,168 Male: 1584 Female: 1584	0.14	5–74	NR	General Population	C (equivalent to IASP, 2013)
Wang, Sun, et al., 2018 (China)	Asia	1 Male: 0 Female: 1	1,498 Male: 720 Female: 778	0.10	2–89	NR	General Population	C (equivalent to IASP,2013)

Abbreviations: BMS, burning mouth syndrome; C, clinical diagnosis; IASP, International Association for the Study of Pain; NR, not reported.

TABLE 2 Characteristics of clinical-based studies of burning mouth syndrome

Study	Continent	Number of		Prevalence, %	Age, y	Study period	Source of participants	Diagnostic method
		BMS	Total					
Brailo et al., 2006 (Croatia)	Europe	150 Male: 27 Female: 123	1,399	10.75	23–88	2001	Department of Oral Medicine, School of Dentistry	C (NR)
Fattori et al., 2019 (Brazil)	South America	754 Male: 125 Female: 629	5,063	14.91	60–97	1977–2016	Stomatology and Prevention of Oral Maxillofacial Cancer	C (NR)
Netto et al., 2011 (Brazil)	South America	32 Male: 9 Female: 23	3,243	1.01	27–87	1999–2006	Oral Pathology Service at the School of Dentistry	C (NR)
Cardoso et al., 2016 (Portugal)	Europe	312 Male: 46 Female: 266	9,595 Male: 3,253 Female: 6,342	3.26	NR	2005–2016	A General Dental Clinic in Lisbon	C (NR)
Wang, He, et al., 2018 (China)	Asia	3,454 Male: 793 Female: 2,661	21,972 Male: 10,744 Female: 11,228	15.71	0–93	2013–2017	Department of Oral Medicine, Xiangya Hospital	C (equivalent to IASP, 2013)
Luo et al., 2020 (China)	Asia	395 Male: 144 Female: 251	4,435 Male: 2,166 Female: 2,269	8.90	1–86	2017–2019	Oral Mucosal Department of Stomatological Hospital	C (equivalent to IASP, 2013)
Liu et al., 2017(China)	Asia	112 Male: 18 Female: 94	1,500 Male: 678 Female: 822	7.50	0.3–85	2013–2014	Department of Oral Medicine, College of Stomatology	C (equivalent to IASP, 2013)
Li et al., 2015 (China)	Asia	158	2,308 Male: 1,038 Female: 1,270	6.86	NR	2015	Outpatient Department of Stomatology	C (equivalent to IASP, 2013)
Chai et al., 2014 (China)	Asia	2,577 Male: 295 Female: 2,282	37,076 Male: 12,911 Female: 24,165	6.96	0–90	2009–2013	Periodontal Mucosal Branch of Hospital	C (equivalent to IASP, 2013)

Abbreviations: BMS, burning mouth syndrome; C, clinical diagnosis; IASP, International Association for the Study of Pain; NR, not reported.

2.6 | Statistical analysis

STATA14.2 (Stata Corp) was applied to conduct data statistical analysis. Freeman–Tukey's double arcsine transformation method was adopted to convert the prevalence rates reported in all studies (Freeman & Tuckey, 1950). Then the back-transformation was performed to produce the pooled estimated prevalence and subgroup prevalence of BMS (with 95% CIs) (Nyaga et al., 2014). Q test and I^2 statistic were used to evaluating heterogeneity between studies and the forest plots were used to represent the results. When the heterogeneity of the studies was not significant ($I^2 < 50\%$ or $p > .1$), the fixed-effects model was selected. When the heterogeneity of studies was high ($I^2 > 50\%$ or $p < .1$), the random-effects model was selected. Egger's test was used to evaluate publication bias and the results were presented by funnel plots. Subgroup analysis was performed by continent, gender, and age, and $p < .05$ was considered with statistical significance.

3 | RESULTS

3.1 | Study selection and study characteristics

The following diagram (Figure 1) shows the results of article search and the studies selection process. A total of nine population-based studies ($n = 26,632$) from four different countries were included (Table 1). These nine studies all reported the prevalence rate of BMS, six studies from China, and the other three studies were from Finnish, USA, and Sweden. As to clinical-based studies ($n = 86,591$), there are nine from four different countries were included (Table 2),

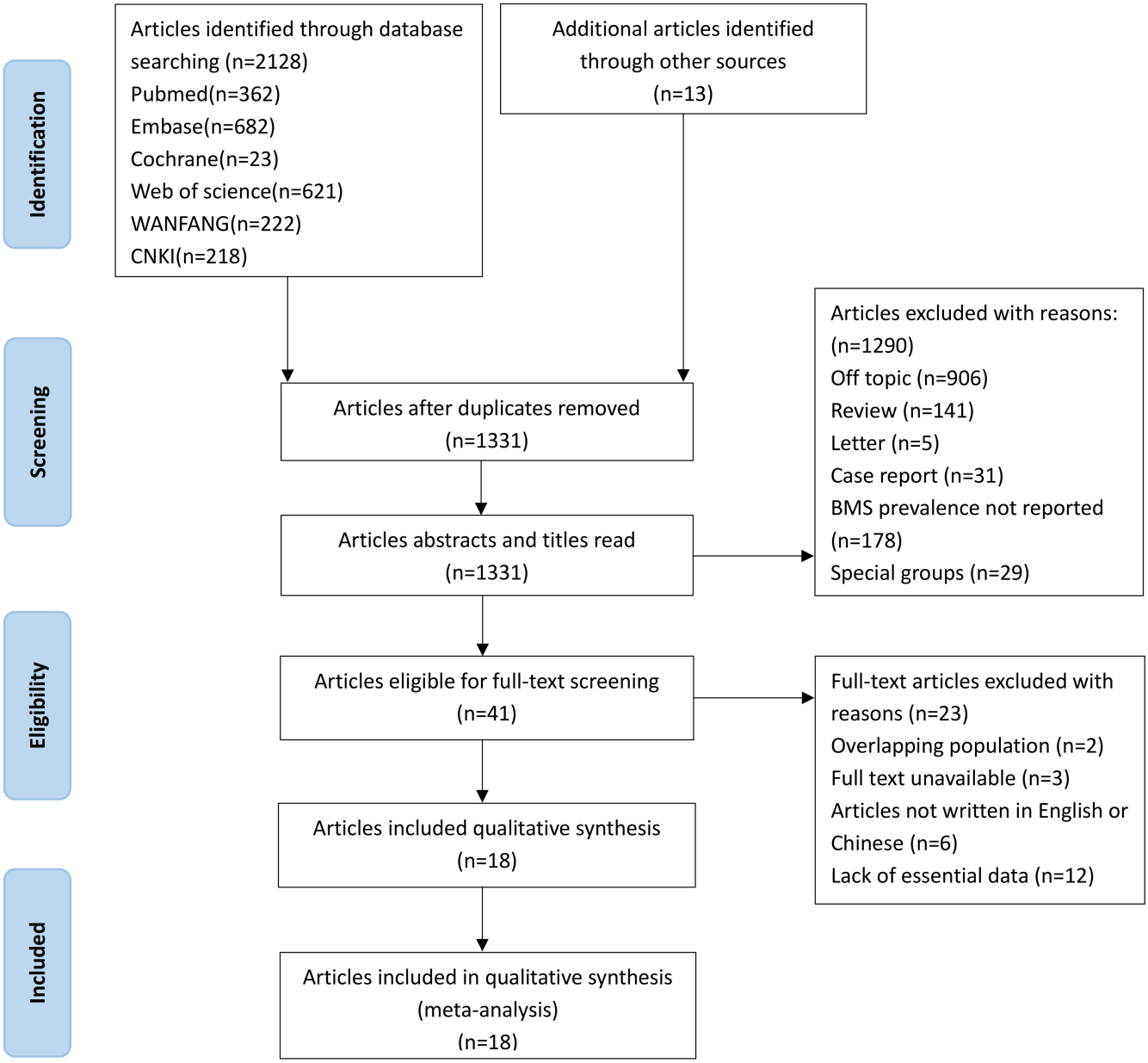

FIGURE 1 The flowchart of the literature screening process

with five studies conducted in China, two in Brazil, two studies in Europe, one from Portugal, and one from Croatia.

3.2 | Overall prevalence of BMS

Through the meta-analysis, the prevalence of BMS was obtained from the population-based and clinical-based studies, respectively. The estimated prevalence of BMS in the population-based studies was 1.73% (95% CI = 0.176–0.351) by the random-effects model, and the heterogeneity between studies was high (I^2 = 97.6%; $p < .01$) (Figure 2). The estimated prevalence of BMS in the clinical-based studies was 7.72% (95% CI = 0.434–0.691) by the random-effects model, and the heterogeneity between studies was high (I^2 = 99.7%; $p < .01$) (Figure 3).

3.3 | By publication year

In order to find out if there was a correlation between the publication year and the prevalence of BMS, we conducted a correlation analysis (Figures S1 and S2). The population-based studies range from 7. 99% in 1993 to 0.75% in 2020 and the clinical-based studies range from 10.75% in 2006 to 8.90% in 2020. Scatter plot shows that there was no significant statistical correlation between the prevalence of BMS and the year of publication.

3.4 | By continent

The pooled estimated prevalence of BMS in the population-based studies was 1.05% (95% CI = 0.082–0.327) in Asia, 5.58% (95% CI = 0.297–0.656) in Europe, and 1.10% (95% CI = 0.194–0.227) in North America (Figure S3). The pooled estimated prevalence of BMS in the clinical-based studies was 8.96% (95% CI = 0.459–0.758) in Asia, 6.05% (95% CI = −0.084–1.077) in South America, and 6.46% (95% CI = 0.215–0.814) in Europe (Figure S4). Notably, the heterogeneity among the included studies were high.

3.5 | By gender

Six studies reported the prevalence of BMS by gender in the population-based studies, including 5,861 females (51.0%) and 5,641 males (49.0%) totally. The pooled prevalence of BMS for female was 1.15% (95% CI = 0.094–0.336), and for male was 0.38% (95% CI = 0.045–0.200) (Figure S5). Five studies reported the prevalence of BMS by gender in the clinical-based studies, including 44,826 females (60.1%) and 29,752 males (39.9%) in total. The pooled prevalence of BMS for female was 11.28% (95% CI = 0.463–0.907), and was 3.75% for male (95% CI = 0.252–0.528) (Figure S6). The heterogeneity among the included studies were high.

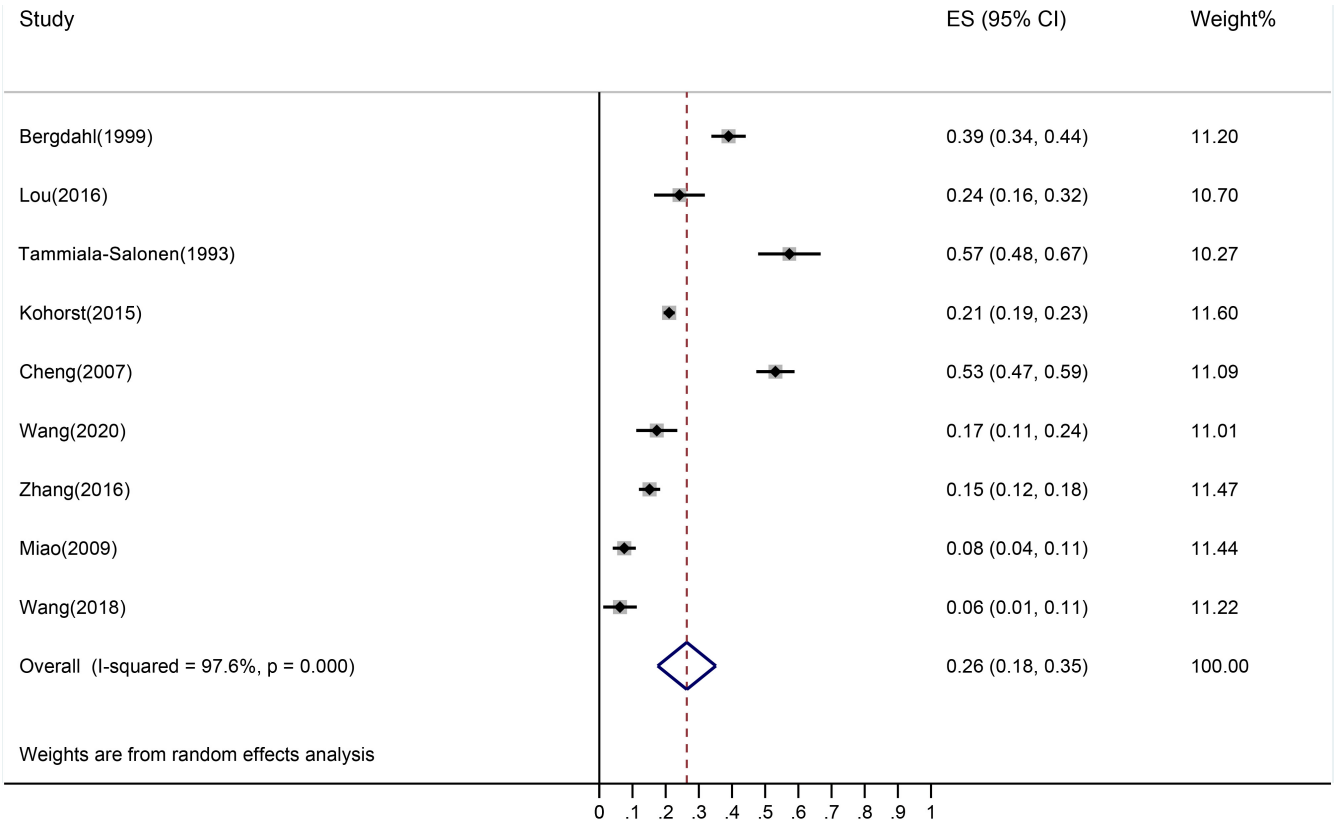

FIGURE 2 The forest plot of included population-based studies

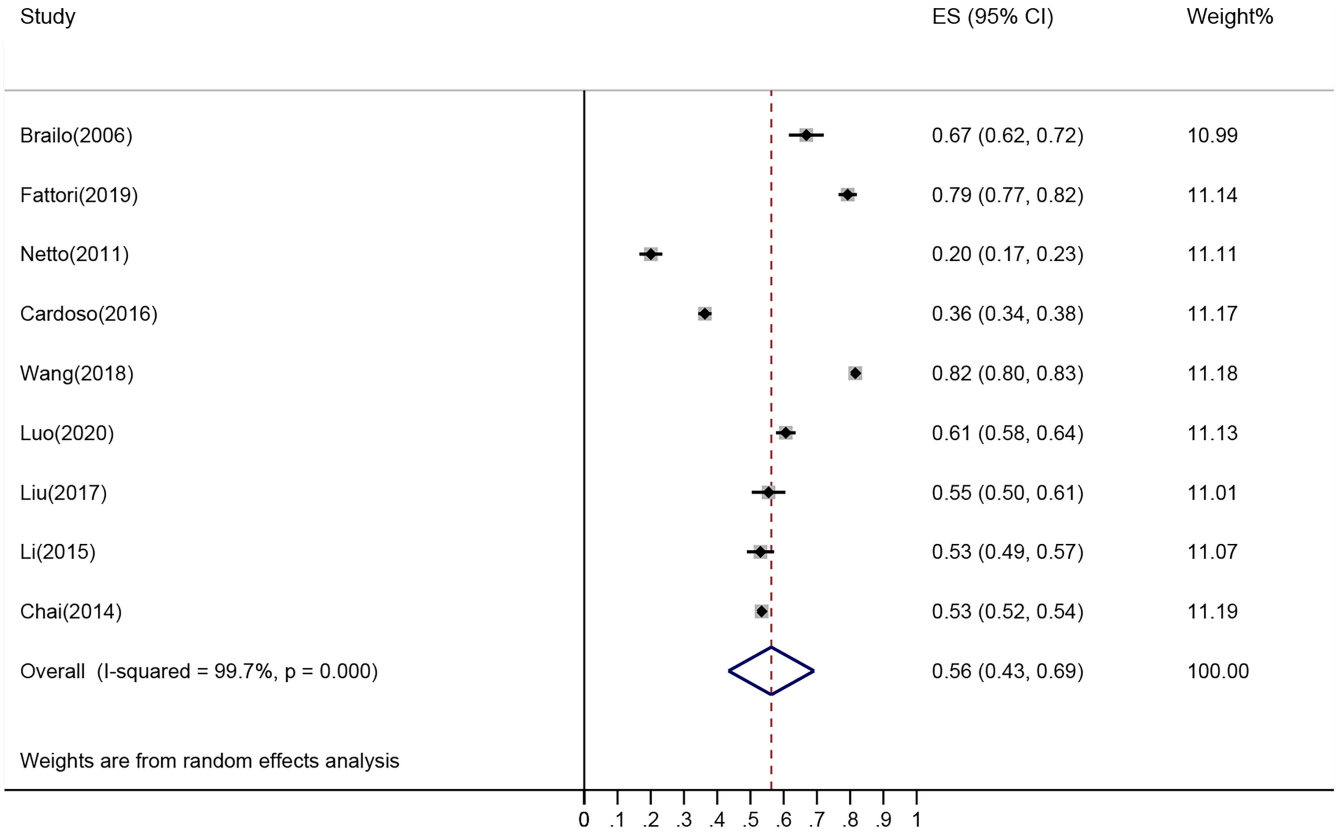

FIGURE 3 The forest plot of included clinical-based studies

3.6 | By age

Only six population-based studies provide data on the age distribution of the respondents. The overall pooled prevalence of BMS was 3.31% (95% CI = 0.043–0.689) among patients 50 years and 1.92% (95% CI = 0.134–0.423) in patients younger than 50 years and the heterogeneity among included studies were high (Figure S7).

3.7 | Publication bias

Egger's test showed that the publication bias of nine population-based studies and nine clinical-based studies were not statistically significant ($p > .05$) (Figure S8 and S9). Hence, the publication bias may not be the source of the high heterogeneity.

3.8 | Meta-regression

Meta-regression showed that continent was not the source of heterogeneity in both population-based studies and clinical-based studies ($p > .05$). Heterogeneity between the included studies (population-based studies; clinical-based studies) was also taken into account ($I^2 = 97.14\%$ and $\tau^2 = 0.02$; $I^2 = 99.69\%$ and $\tau^2 = 0.04$) (Table S2 and S3).

4 | DISCUSSION

BMS is a typical chronic orofacial pain disorder that involving the oral mucosal with unknown etiology and undergoing a rather long disease course (Ducasse et al., 2013). Meanwhile, it is prone to restrict the daily social activities of the patients, develop anxiety and depression, and cause the decline in quality of life (Gureje et al., 1998; Smith et al., 2001). Although dental professionals play a central role in the diagnosis, the management of BMS often benefits from multidisciplinary collaboration, for instance among dentistry, medicine, and physical therapy. Therefore, to provide sufficient knowledge of its epidemiology to dental specialties, medical professional and even the medical economists are essential, aiming at improving their knowledge of BMS and reducing the societal burden caused by BMS.

It was found in this first meta-analysis of its kind that the overall prevalence of BMS was 1.73% in the general population and 7.72% in the clinical settings of dental practice. This study included clinical-based studies and population-based studies. The clinical patients, we included mainly came from the department of oral medicine, department of oral pathology and maxillofacial surgery, and oral medicine was indicated as optimal when compared to other dental specialties (Brailo et al., 2006; Fattori et al., 2019; Li et al., 2015; Netto et al., 2011). The data in clinical settings revealed that BMS patients had a high demand for medical consultation and further management (Xiao et al., 2020). In addition to dental department, a retrospective study found that BMS patients also sought help from

multiple clinics and other medical specialties such as otorhinolaryngology, dermatology, neurosurgery, and anesthesia departments (Freilich et al., 2020; Xiao et al., 2020).

Racial/ethnic disparities related to chronic pain have been studied widely. However, there is no such report on BMS. The subgroup analysis in this study implied that in general population, the prevalence of BMS was higher in Europe (5.58%) and North America (1.10%), and lower in Asia (1.05%). In contrast, in the clinical settings, the prevalence in Asia (8.96%) was higher than that in South America (6.05%) and Europe (6.46%). Previous studies on chronic orofacial pain had shown that the prevalence varies by ethnic group. A biracial cohort indicated that the prevalence of facial pain was significantly higher in Caucasians than African–Americans (Plesh et al., 2012). Another study showed that Native Americans had lower cold-pressor pain thresholds and tolerances than non-Hispanic Whites (Rhudy et al., 2020). These studies may help explain the racial differences this study revealed in the prevalence of BMS. We also found the prevalence of BMS age group varied from different countries, this may relate to the differences in socioeconomic and medical conditions among different regions.

Gender differences in the prevalence of chronic pain have been indicated in many studies. A meta-analysis from the UK showed that chronic pain was more common in female than male (Fayaz et al., 2016). However, another study in China found that male had higher rates of chronic pain than female (Zheng et al., 2020). The previous study has shown that BMS is frequently seen in the perimenopausal and menopausal female and the female/male ratio for BMS was 7:1 (Nasri-Heir et al., 2015). It turned out in our study that the average female/male ratio for BMS was 3:1 in both general population and clinical patients, with the general higher trend in female than male. The gender differences might be related to physiological and behavioral differences. According to Maurer et al., male and female may have different pain thresholds due to different levels of hormone (Maurer et al., 2016). Moreover, females are more prone to anxiety and more willing to report health-related symptoms such as pain (Asher, Asnaani, & Aderka., 2017; Steel et al., 2014).

The economic burden was imposed on the patients and on the healthcare system should be seriously considered based on the global epidemiological data. Unfortunately, the economic parameters could not be extracted from the included BMS studies. Thus the financial burden of this disease could not be easily described. It was reported the cost of correct diagnosis was 1,570.7$ for patients with atypical odontalgia and 1,316.8$ for those who had persistent idiopathic facial pain (Xiao et al., 2018). Breckons's study showed that chronic orofacial pain patients' out-of-pocket costs were 463.5$ and indirect costs were 1,728.7$ for medical care every six months (Breckons et al., 2018). Since BMS is generally regarded as an exclusive diagnosis, it is as challenging to establishing the diagnosis as other persistent idiopathic facial pain disorders (Freilich et al., 2020). Therefore, it is important for health professionals to have a better understanding of the clinical features, diagnostic criteria as well as physical and financial burden based on the epidemiological data.

One of the most controversial aspects of BMS concerns the criteria used for diagnosis. Different diagnostic criteria have been used over the decades (IASP, 2013; IASP 2016; IHS, 2018; ICOP, 2020). Six population-based studies and five clinical-based studies we have included used the diagnostic criteria which equivalent to IASP (2013) (Table 1 and Table 2). According to the criteria, a diagnosis of BMS is made only after ruling out other causes of burning sensation. The most recent diagnostic criteria for BMS is an intraoral burning sensation that recurring daily for more than 2 hr over more than 3 months and without clinically evident causative lesions, meanwhile, has no identifiable cause either local or systemic condition or disease (ICOP, 2020). For now, the diagnosis criteria of BMS varies in clinical studies and cross-sectional studies, and the unified diagnostic criteria is very significant in the subsequent relevant studies (Brailo et al., 2006; Cheng, X, H., & Zhu, A, L., 2007; Liu et al., 2017).

This study has some limitations. Firstly, our analysis only included studies that written in English and Chinese. We did not access the studies of other languages, which may ignore some epidemiological information about BMS in other regions. Secondly, the numbers of studies we have included were limited and not all the target data from each included study could be extracted. Last but not the least, there was a high degree of heterogeneity among the studies due to different research designs, such as different research populations, sampling methods, sample sizes, and professional specialty of the investigators, so the pooled prevalence estimates reported in this meta-analysis should be taken with caution. In the future, in order to further understand the epidemiology profile of BMS, more high-quality cross-sectional surveys which using uniformed diagnostic criteria, standard sampling methods are required.

5 | CONCLUSIONS

The pooled prevalence of BMS was relatively high both in general population and clinical patients, confirming that merits attention of dental and healthcare professionals. Moreover, the results showed that the prevalence of BMS was varied in different regions, higher in female than male and higher in people over the age of 50. More high-quality, extensive, and using uniformed diagnostic criteria epidemiological surveys of BMS are needed, to determine the global prevalence of BMS more accurately.

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CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

AUTHOR CONTRIBUTIONS

Shuangshuang shuang Wu: Conceptualization; Data curation; Formal analysis; Investigation; Writing-original draft. **Wenqing**

Zhang: Data curation; Methodology; Validation. **Jingxian Yan:** Data curation; Investigation. **Noboru Noma:** Resources; Supervision; Validation. **Andrew Young:** Supervision; Validation. **Zhimin Yan:** Funding acquisition; Project administration; Resources; Supervision; Validation; Writing-review & editing.

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DATA AVAILABILITY STATEMENT

Data available on request from the authors.

ORCID

Shuangshuang Wu https://orcid.org/0000-0001-7759-0772
Noboru Noma https://orcid.org/0000-0001-5405-0407
Zhimin Yan https://orcid.org/0000-0002-0751-7374

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section.

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