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ORIGINAL ARTICLE

Burning mouth syndrome: a retrospective study investigating spontaneous remission and response to treatments

A Sardella, G Lodi, F Demarosi, C Bez, S Cassano, A Carrassi

Unit of Oral Pathology and Medicine, School of Dentistry, University of Milan, Milan, Italy

OBJECTIVE: The aim of this investigation was to evaluate the spontaneous remission rate of burning mouth syndrome (BMS) in a group of subjects suffering from this syndrome.

SUBJECTS AND METHODS: The medical records of BMS patients attending the Unit of Oral Medicine (1995–2002) were reviewed. The patients with a follow-up period of at least 18 months were then contacted over phone and interviewed using a structured *ad hoc* questionnaire to record their current symptoms and data about their treatment responses to the therapies.

RESULTS: Forty-eight women and five men with a mean age of 67.7 years (range 33–82 years) were included in the study (mean duration of BMS 5.5 years, s.d. ±1.9 years, mean follow-up period of 56 months). As a consequence of different treatments, 26 patients (49.0%) reported no change in oral symptoms, 15 (28.3%) moderate improvement and 10 (18.9%) a worsening of oral complaints. Only two patients (3.7%) reported a complete spontaneous remission of oral symptoms without any treatment.

CONCLUSIONS: In this study, a complete spontaneous remission was observed in 3% of the patients within 5 years after the onset of BMS. A moderate improvement was obtained in <30% of the subjects.

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Keywords: burning mouth syndrome; treatment; remission; pain management

Introduction

Burning mouth syndrome (BMS) is considered to be a chronic idiopathic burning sensation or pain affecting the clinically normal oral mucosa for which dental or medical causes have been excluded. The term 'syndrome' implicates the simultaneous presence of several symptoms, most frequently a feeling of oral dryness and/or altered taste, in addition to a burning sensation in the oral mucosa (Grushka *et al*, 2002; Zakrzewska *et al*, 2005). BMS may have a negative impact on the subject's general and psychological well-being and can reduce the quality of life, even though it is not clear whether the psychopathologic distress is related to the syndrome or whether the former is a result of the chronic symptoms experienced by those patients (Al Quaran, 2004).

Despite many studies in the literature evaluating several aspects of BMS, its aetiology and pathogenesis are still unknown and, as a consequence, there is no causal and/or effective treatment (Zakrzewska *et al*, 2005). Furthermore, there is also a high variability in the prevalence data that vary from 0.7% to 15% (Pedersen *et al*, 2004) and reflects the lack of well-defined diagnostic criteria. On the contrary, very few studies on natural course or spontaneous remission in patients suffering from BMS are available (Gilpin, 1936; Gruskha *et al*, 1987; Danhauer *et al*, 2002).

The aim of this investigation was to evaluate the spontaneous remission rate of BMS in a group of subjects suffering from this syndrome.

Patients and methods

All patients with a provisional diagnosis of BMS attending the Unit of Oral Pathology and Medicine, University of Milan, received the definitive diagnosis of BMS after thorough clinical examination and a standard set of examinations including salivary flow rates measurements, laboratory evaluations (complete blood cell counts, blood glucose levels, serum iron and transferrin levels, serum vitamin B_{12} and folate levels) and isolation of *Candida* species from oral mucosal scrapes. Patch tests for contact allergy to partial denture materials were also performed when the condition was suspected. The results of these evaluations were recorded in medical files besides demographic and medical

Correspondence: Prof. A Sardella, Istituto di Odontoiatria, Via Beldiletto 1, I – 20142 Milan, Italy. Tel: +39 02 5031 9019, Fax: +39 02 5031 9041, E-mail: andrea.sardella@unimi.it
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Table 1 Burning mouth syndrome remission *ad hoc* questionnaire *(excerpt)*

Please, refer all of the treatments you have tried for your burning sensation

☐ benzodiazepines	☐ antidepressant drugs	☐ antifungal drugs	☐ anti-inflammatory drugs
☐ oral rinses	☐ topical steroids	☐ psychotherapy	☐ salivary substitutes
☐ vitamins	☐ other.....	☐ nothing	

Please, refer all of the specialists you have consulted for your burning sensation

☐ general physicians	☐ psychologists	☐ neurologists
☐ homeopaths	☐ other.....	☐ any

Are you currently using one of the following treatments? If yes, please refer

☐ benzodiazepines	☐ antidepressant drugs	☐ antifungal drugs	☐ anti-inflammatory drugs
☐ oral rinses	☐ topical steroids	☐ psychotherapy	☐ salivary substitutes
☐ vitamins	☐ other.....	☐ currently I don't use any treatment	

Do the medications/treatments used relieve or reduce your burning sensation?

☐ no, my oral symptoms remain the same
☐ no, my oral symptoms worsened
☐ yes, my oral symptoms partially improved
☐ yes, my oral symptoms completely disappeared

information, drugs history, the oral symptoms intensity assessed using a Visual Analogue Scale (from 0, no pain to 10, extreme pain) (Scott and Huskisson, 1976), the oral sites affected and the duration of the syndrome.

In this study, we reviewed the medical records of BMS patients referred to the Unit during the period January 1995–December 2002. A total of 108 BMS files were analysed and only the patients with a follow-up period of at least 18 months were contacted over phone and interviewed using a structured *ad hoc* questionnaire (Table 1).

The questionnaire allowed collection of information on the past and current treatments prescribed by the specialists of the Unit as well as therapies self-administered and/or prescribed by other specialists or general practitioners (GPs). Furthermore, present symptoms (i.e. current burning sensation), their localization, data about treatment responses to the therapies and other specialists eventually consulted were also recorded in the questionnaire.

Results

Fifty-three of 108 BMS patients who had had a follow-up longer than 18 months were included in the study. This group comprised 48 women and five men with a mean age of 67.7 years (range 33–82 years). The mean duration of BMS was 5.5 years (s.d. ± 1.9 years) and the mean follow-up period was 56 months (96–18 months). In Table 2, the clinical characteristics of these patients are summarized.

The following information was collected during the phone interviews.

Therapies
Regarding the therapies used for relief from burning oral symptoms, patients reported the use of several treatments, some prescribed after the definitive diagnosis and during the recall visits in our Unit and others self-administered and/or prescribed by other specialists. In particular (Table 2), patients used chlorhexidine oral rinses (20/53), benzodiazepines (15/53), antihistaminic local drugs (11/53), anti-inflammatory local and systemic drugs (8/53), antidepressant drugs (8/53), antifungal agents (6/53), vitamins (4/53), psychotherapy (2/53), salivary substitute (2/53), topical steroids (1/53), capsaicin (1/53) and various type of local injections (i.e. human placenta, 1/53). Nineteen patients (19/53) did not undergo any treatment.

Other specialities
Several patients had consulted other specialists for their oral problems. In particular, GPs (10%), psychologists (5%), neurologists (2%), homeopaths (2%) and rheumatologists (0.5%).

History of burning
As shown in Table 2, 26 patients (49.0%) reported no change in oral symptoms, 15 (28.3%) moderate improvement and 10 (18.9%) a worsening of oral complaints. Only two patients (3.7%) reported a complete remission of oral symptoms.

The complete remission of oral burning sensations (2/53, 3.7%) was seen in patients without any treatment (*spontaneous remission*). These two patients were women, with a mean duration of the syndrome of, respectively, 68 and 65 months. In the first case, a supportive reassuring care was effective in resolving oral burning. In the second case, the patient experienced the disappearing of the oral symptoms with a novel self-realization after taking care of the newborn grandchild.

Discussion

The cause of BMS and its pathogenic mechanisms are still unknown. Evidence of a possible relationship between BMS and psychogenic factors has not been proved. Recently, several papers have investigated the trigeminal somatosensory system in order to detect neurogenic abnormalities (Svensson *et al*, 1993; Jääskeläinen *et al*, 1997, 1999, 2000; Gao *et al*, 2000; Forssell *et al*, 2002). These studies suggest peripheral alterations in the function of the sensory trigeminal system with the presence of abnormal reflex (i.e. blink

Table 2 Summary of patient conditions as recorded during the phone interview

Modification of oral symptoms	*n* (%)	*Localization*	*Reported treatments*	*Mean duration of BMS (months)*
No change	26 (49)	Tongue 20, lip 5, palate 2, gingiva 1, floor 1, buccal mucosa 1	Chlorhexidine 11, benzodiazepins 7, antihistaminic drugs 5, anti-inflammatory 4, antidepressant 5, antifungal agents 3, other 8, no treatments 7	62.5
Moderate improvement	15 (28.3)	Tongue 13, lip 4, palate –, gingiva 3, floor 2, buccal mucosa 2	Chlorhexidine 5, benzodiazepins 4, antihistaminic drugs 5, anti-inflammatory 4, antidepressant 1, antifungal agents 2, other 2, no treatments 5	69
Worsening	10 (18.9)	Tongue 9, lip 3, palate 4, gingiva 2, floor 1, buccal mucosa 1	Chlorhexidine 4, benzodiazepins 4, antihistaminic drugs 1, anti-inflammatory –, antidepressant 2, antifungal agents 1, other –, no treatments 5	67
Complete remission	2 (3.7)	Tongue 2, lip –, palate –, gingiva –, floor –, buccal mucosa –	No treatments 2	66.5

reflex) (Jääskeläinen *et al*, 1997). Support for this interpretation may be found in recent papers in which neuroprotective/neurotropic drugs such as alpha-lipoic acid (Femiano and Scully, 2002) or clonazepam (Gremeau-Richard *et al*, 2004) seem to improve the symptoms of BMS patients.

Besides these neuroprotective drugs, a large variety of medicines and therapies have been proposed in BMS but the treatment management of this syndrome is still not satisfactory (Scala *et al*, 2003; Zakrzewska *et al*, 2005). A weak placebo effect is reported in several controlled studies on systemic and local treatments (Bergdhal *et al*, 1995; Sardella *et al*, 1999; Tammiala-Salonen and Forssell, 1999). Unfortunately, very few studies on natural course or spontaneous remission in patients suffering from BMS are available (Gilpin, 1936; Gruskha *et al*, 1987; Danhauer *et al*, 2002).

The aim of this investigation was to evaluate the spontaneous remission rate of BMS in a group of subjects suffering from this syndrome.

Gilpin (1936) reported about 25 patients with BMS followed-up for an undefined period of time. In this group of patients, eight (32%) reported a spontaneous complete recovery, eight (32%) a moderate improvement and nine (36%) no improvement and/or worsening.

Gruskha *et al* (1987) considered 43 patients suffering from BMS with a follow-up period of nearly 6 years. The patients (mean age 68 years, s.d. $\pm$9.3 years, 81% female) were contacted and asked in a structured interview about their current pain. Twenty-three patients (54%) reported 'no change' (mean duration of BMS 6.5 years, s.d. $\pm$3.4 years), 13 patients reported 'complete remission' (mean duration of BMS 6.2 years, s.d. $\pm$2.8 years) and seven patients (16%) reported 'partial' remission (mean duration of BMS 7.1 years, s.d. $\pm$3.4 years). Of the 13 patients with a complete remission, nine could not recall any specific therapy or favourable factor. No significant differences were observed between the three groups for age, gender, duration of disease and distribution of burning sites. If we considered that, nine of 43 patients experienced a spontaneous remission, this rate was equal to 20% within 7 years after the onset of BMS.

More recently, Danhauer *et al* (2002) in a paper aimed at determining characteristics which were able to uniquely define the BMS patient, reported one spontaneous remission of oral symptoms out of 26 subjects (3.8%). The study group was composed of 26 patients (mean age 59.08 years, s.d. $\pm$12.4 years) with a BMS mean duration of 2.27 years (s.d. $\pm$3.81 years).

The results of the present study suggested that a complete spontaneous remission may be expected in only a small proportion of patients within 5 years after the onset of BMS. Furthermore, only a moderate improvement may be obtained, with different treatments, in $<30\%$ of subjects. The surprisingly wide different remission rate reported in other groups of BMS

patients (Gilpin, 1936; Gruskha *et al*, 1987) could be perhaps explained through a longer follow-up period.

This study shows that over 70% of BMS patients search and/or need treatment and continue to be high consumers of healthcare resources. As a consequence, it is important to deepen our understanding of the aetiopathogenetic mechanism of BMS and to develop new, safe and efficacious therapeutic approaches.

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