

Burning mouth syndrome. A case report from diagnosis to cure

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ABSTRACT

Burning mouth syndrome (BMS) is a chronic disease of the oral mucosa characterized by persistent oral pain and no clinical lesions or abnormal biological investigations. This paper reports one case of BMS and discusses the main features of this disease's clinical and therapeutical management. A 60 years old female patient had burning sensation on the left lateral border of the tongue for 2.5 years. Local and systemic investigations found no abnormalities. The therapy included local and general medication and the symptoms were remitted. This case report highlights the heterogeneity of BMS, the associated comorbidities, and the evolution of symptoms from diagnosis to cure.

Keywords: Burning mouth syndrome, chronic oral pain, Visual Analog Scale, case report

INTRODUCTION

Burning mouth syndrome (BMS) is a chronic orofacial pain, burning or dysaesthetic sensation which manifest daily for at least 2 hours, more than 3 months and has no local or systemic cause [1,2]. An expert consensus on BMS analyzed the definition of the disease and diagnostic criteria and recommended changing the nomenclature from BMS to Burning Mouth Disorder as a syndrome includes a well defined group of features and in these patients oral burning or dysaesthesia is most frequent [3]. Oral symptoms reported by the patients show a range of variations from burning, pain, dysesthesia, subjective dry mouth, and dysgeusia [3]. The BMS etiology is multifactorial and carries psychological and psychiatric aspects with individual differences [4]. The neuropathic damage involved in BMS involves different levels of neuraxis such as peripheral, central or both overlapping [5]. The mechanisms described for the peripheral neuropathy are small fiber neuropathy or subclinical lingual, mandibular, or

trigeminal neuropathy or deficient dopaminergic top down inhibition in the basal ganglia [6].

The strategy to evaluate a BMS patient includes a complete anamnesis, a careful clinical examination, laboratory and allergy investigations [7]. The symptoms features include a description of the symptoms, type of the onset, triggering factors, duration and evolution, oral mucosa sites affected, symmetry.

DESCRIPTION OF THE CASE

A 60 years old female patient reported the main complaint of a burning sensation located on the left border of the tongue and soft palate. The patient previously consulted an ENT specialist and two dentists who found no clinical lesions. For the last 2,5 years this burning sensation (pepper-like) was continuously daily and started after 11 o'clock until 18 hours. The patient noted down that her husband died of lung cancer 6 months ago and that she endured this event with difficulty.

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Reviewing the medical history showed the presence of a mild essential hypertension treated with nebivolol 5 mgx2 daily (a beta blocking agent) and indapamide (1,5 mg). Also the patient reported a gastro-oesophageal reflux disease negative for *Helicobacter pylori* which was diagnosed and treated 5 years ago and currently she had no symptoms or treatment. Another diagnosed diseases were fibromatous uterus, dyslipidemia under treatment with atorvastatin (10 mg/daily) and controlled hypothyroidism treated 0.25 mcg daily.

The oral burning sensation was not present during food intake and at night. The burning sensation was located on the left border of the middle tongue. Its intensity has a slight variability during days but the patient could not correlate this variation to a factor. No other associated symptom (taste disturbance or xerostomia) was reported. The pain severity was assessed as the pain felt during the last week on a 0-10 Visual Analogue Scale numeric rating [VAS] (0 was defined as the absence of pain and 10 the maximal pain imaginable). On the first consultation the patient reported the burning sensation (VAS) at a value of 7. She also mentioned there were days with VAS value of 9.

On the oral clinical examination there were no detectable lesions of the mucosa. The dorsal tongue had the aspect of geographic tongue but the patient's complaints were not correlated with this aspect. The salivary flow was in normal range and the orthopantomogram evaluation showed no dental infection or lesions. The biological investigations (complete hemogram, hematologic examinations for anemia, vitamin B12, folate levels, glucose test, thyroid stimulating hormone, thyroxine, triiodothyronine and thyroid antibodies, optimum level serum of zinc) showed no pathological changes. *Candida* species were not detected by the mycological analysis and *Helicobacter pylori* test was negative.

Related to the usual sleep habits, the patient reported that her sleep quality was good and she had 7 to 8 hours of sleep every night and free of oral burning symptoms. As the tongue symptoms were located on the left side, a neurological examination was done and detected no changes. An angio computer tomography of the brain revealed that there were no abnormalities or notable lesions.

The primary outcome was the decrease of pain intensity on VAS scale from 9 to 5 after 3 months of treatment with combined topical (homeopathic remedy) and systemic therapy (zinc supplements). The multifactorial etiology was explained to the patient. She declined the advice to use topical clonazepam at the recommendation of a psychiatrist. After the first 3 months, the general treatment was changed with a dietary supplement including a complex of B vitamins (B1, B3, B6, B12), folic acid,

and uridine monophosphate. After receiving treatment for three months, the patient's VAS score decreased to 3, improving her quality of life. The patient received no BMS treatment during the course of the following 12 months as the symptoms.

DISCUSSION

BMS is an idiopathic disease with a higher prevalence in postmenopausal women [8]. A significant emotional distress is the main feature of BMS [9]. The multifactorial etiology and heterogeneity of symptoms favor diagnostic delay as not all the medical specialists are familiarized with it. Recent studies consider that the neuropathic damage located in the central or peripheral nervous system or in both areas can trigger BMS [10].

The diagnostic criteria for BMS is described by the International Classification of Orofacial Pain as oral pain that is present daily for more than two hours for more than three months, has a burning quality, is felt superficially, with normal aspect of the oral mucosa, no local or systemic cause and does not fit another disease [2].

Regarding the physiological factors associated with BMS, depression and anxiety are the most common comorbidities [10]. The reported case associated stressful personal events but the patient was able to cope with it even without specialized treatment. The BMS was present long before the stressful event but an aggravation of the symptoms determined the patient to seek for diagnosis and treatment.

One common disorder associated with BMS is gastroesophageal reflux disease. Although there has been discussion of an association between these disorders, a substantial link was not detected. Russo et al. found a higher prevalence of BMS symptoms in gastroesophageal reflux patients compared to the general population, but a similar prevalence for chronic cough and pharyngitis [11].

As untreated hypothyroidism is a general factor that can determine a burning sensation in the oral mucosa, thyroid tests are included in the protocol of BMS diagnosis [12,13]. In the reported case the onset of symptoms and the controlled thyroid disease confirmed by blood tests confirm that there is no connection between both diseases.

Another comorbidity the reported patient presented was hypertension. Adamo et. al found that the prevalence of hypertension is higher in BMS patients when in healthy controls since there are common biological risk factors for both diseases [14].

The BMS treatment although comprise a large variety of options has limited effectiveness and include pharmacological treatment, psychosocial treatment and cognitive behavioral therapy [9]. Although some authors appreciate that zinc deficiency may be involved in BMS pathogenesis [15] and

other authors report symptomatic improvement of BMS with zinc intake [16] we administered 3 months of zinc supplementation with good outcome. Homeopathy is a possibility for BMS treatment [9]. Some authors report improvement of BMS symptoms after 4 years of individualized homeopathic treatment in patients who associated some psychological factors and neurological complaints [17]. The active ingredients contained in the topical treatment for the reported patient are nitum napellus, Belladonna, Calendula officinalis. The patient was advised to use these tablets when the symptoms were at high levels. Vitamin B therapy was demonstrated to bring benefits in BMS [18] and for our patient the 3 months intake of a complex of vitamin B reduced VAS value from 5 to 3.

Although a cognitive therapy was not applied, the empathic attitude of the attending physician

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contributed to the improvement of the symptomatology for the reported patient.

CONCLUSION

Burning mouth syndrome is a difficult to manage and diagnose disease, with idiopathic etiology and no standard treatment. A series of comorbidities are significantly associated with BMS and these can affect the outcome of the treatment.

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