

CASE REPORT/CASO CLÍNICO

Burning Mouth Syndrome: Case Reports Síndrome da Boca Ardente: Casos Clínicos

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Abstract

Burning mouth syndrome (BMS) is a relatively common chronic disease of unclear etiopathogenesis. It is defined as a distinctive nosological entity that includes all forms of burning sensation in the mouth, including stinging sensation or pain, in association with an oral mucosa that appears clinically normal, in the absence of local or systemic diseases. It was hypothesized that psychological factors could influence its appearance, but also physiological factors, with some authors suggesting a neuropathic etiology.

Regarding the treatment of BMS, benefits were found in individual or group psychotherapy, as well as in psychopharmacological interventions, however its current therapeutic approach is not completely satisfactory. In this paper we describe two cases of BMS and discuss the possible etiopathogenesis and current therapeutic approaches.

Resumo

A síndrome da boca ardente (SBA) é uma doença crónica relativamente comum de etiopatogenia desconhecida. É definida como uma entidade nosológica distinta que inclui todas as formas de sensação de ardência bucal, incluindo sensação de picada ou dor, em associação com uma mucosa oral que aparenta ser clinicamente normal, na ausência de doenças locais ou sistémicas.

Foi colocada a hipótese de que fatores psicológicos pudessem influenciar o seu aparecimento, assim como fatores fisiológicos, com alguns autores a sugerir uma etiologia neuropática.

No que diz respeito ao tratamento da SBA foram encontrados benefícios em psicoterapia individual ou de grupo, assim como em intervenções psicofarmacológicas, no entanto a sua abordagem terapêutica atual não é totalmente satisfatória. Neste artigo descrevemos dois casos de SBA e discutimos a sua possível etiopatogenia e abordagens terapêuticas vigentes.

Keywords: Burning Mouth Syndrome/etiology; Burning Mouth Syndrome/therapy; Neuralgia

Palavras-chave: Síndrome da Boca Ardente/etiologia; Síndrome da Boca Ardente/tratamento; Neuralgia

Burning mouth syndrome (BMS) is a chronic disease of unclear etiopathogenesis,¹ also known as stomatodynia, oral dysaesthesia, glossodynia, glossopyrosis, and stomatopyrosis.² The International Association for the Study of Pain and International Headache Society defines it as a distinctive nosological entity that comprises all forms of burning

sensation in the mouth, including complaints described as stinging sensation or pain, in association with a normal oral mucosa, without the presence of local or systemic diseases.² It is a relatively common disease, especially amongst postmenopausal women, estimated to affect from 0.7% to 4.6% of the general population.¹

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The main complaint in BMS patients is the presence of an oral burning sensation, which may affect multiple sites of the oral cavity, with the tongue being the most frequently affected site,¹ especially the anterior 2/3 and the extremity, as well as the dorsum and the anterolateral margins. Often the anterior hard palate, the lip mucosa and the mandibular alveolar regions are also affected.³

The burning sensation is often bilateral and symmetrical and does not respect the anatomical distribution of a peripheral sensory nerve.³ It can be present during almost all day and often persists for many years.⁴ The pain may vary in its intensity and can improve with food intake.⁵ Xerostomia and dysgeusia are also frequent, and can constitute a diagnostic triad, although most cases present oligo or monosymptomatic.⁵

Although not consensual, it has been hypothesized that psychological factors could explain the origin of the symptoms in this syndrome, with several studies demonstrating a high frequency of comorbidity with psychiatric disorders. Depression appears to be the most prevalent psychiatric disease among patients with BMS, but anxious disorders, “cancerophobia” and hypochondriasis are also very common,¹ as well as personality disorders,³ which suggests that BMS may constitute a functional pain syndrome. A large percentage of these patients have a history of psychiatric hospitalization and are more likely to be under psychiatric treatment, often initiated before the onset of BMS symptoms.³ Increasing evidence also suggests that pain in idiopathic BMS may have a neuropathic etiology.^{3,6} It is important to emphasize that BMS is an exclusion diagnosis requiring the evaluation of other potential causes for the symptoms, such as salivary glands, endocrine and connective tissue diseases, gastroesophageal acid reflux, drug side effects (e.g. angiotensin-converting enzyme inhibitors, antibiotics, antiretrovirals), neurological changes, nutritional deficiencies, allergies, infections, climacteric, chemotherapy and radiotherapy, and mechanical factors (tongue trauma – poorly fitting dental prosthetics, piercings).^{4,7}

Regarding the treatment of BMS, although it is not completely satisfactory and total remission is unusual, benefits were found with both individual and group psychotherapy, as well as with psychopharmacological interventions,¹ including antidepressants (tricyclic, selective serotonin reuptake inhibitors - SSRI, serotonin noradrenaline reuptake inhibitors – SNRI); benzodiazepines (systemic use or local application of clonazepam); anticonvulsants; local capsaicin and alpha lipoic acid.⁶

In this paper we describe two cases of BMS occurring in patients with long history of depression, who received treatment for this disorder, but only improved their BMS symptoms after an approach that did not include antidepressants. This suggests that BMS may not be a mere somatic manifestation of depression or anxiety, although the comorbidity is frequent.

Case Reports

Case 1: 70-year-old female patient with 11-year psychiatric history of recurrent major depression, of mild to moderate

severity, without psychotic features and in full remission, and generalized anxiety disorder. She also underwent total thyroidectomy due to thyroid follicular carcinoma five years before, had a history of hypertension and dyslipidemia, and was medicated with levothyroxine, lisinopril + hydrochlorothiazide and simvastatin. She suddenly developed complaints of burning and pain sensations in the tongue’s dorsum that were persistent during the day but attenuated at mealtime. The onset of pain was temporally not compatible with medication changes. Because of these symptoms she appealed to several physicians of various specialties including gastroenterology, stomatology, endocrinology and neurology, who ruled out gastroesophageal acid reflux, salivary glands alterations, endocrine and connective tissue diseases, nutritional deficiencies, neurological changes, allergies and infectious causes. Initially, as doctors did not know what was causing the pain and suspected of pyrosis or tongue infection, multiple types of drugs were prescribed including proton pump inhibitor, sucralfate, antifungal and antiviral, but they were completely unsuccessful. She was finally referred to a psychiatry specialist and became apparent that the burning sensation seemed to be related with anxiety aggravation in the context of adverse life events. For the pain and anxious symptoms she was medicated with pregabalin, benzodiazepines and SSRI without total symptom relief. She began to reveal more significant improvements in BMS symptoms after introduction of carbamazepine 200 mg/day, chosen for being a recommended therapy for neuropathic pain.

Case 2: 58-year-old female patient who was being followed for two years in psychiatry and psychology consultations for recurrent major depressive disorder of mild to moderate severity, without psychotic features and with anxious distress, that had started 11 years before. She began to develop xerostomia and lingual burning complaints in a time of worsening depressed mood and increasing levels of anxiety in the context of general health concerns, having been diagnosed with adjustment disorder with mixed anxiety and depressed mood. She had a history of dyslipidemia and gastroesophageal reflux, for which she was successfully medicated for several years with simvastatin, proton pump inhibitor and sucralfate. The onset of the pain was not temporally compatible with medication changes. She was observed by various specialties including stomatology, endocrinology and gastroenterology, and salivary glands alterations, endocrine and connective tissue diseases, nutritional deficiencies, neurological changes, allergies and infectious causes were excluded, without indication for further investigation. After multiple therapeutic adjustments with SSRI and benzodiazepines, the pain complaints improved after introduction of topical application of 2 mg of clonazepam in the mouth, a recommended therapy for BMS.

Discussion

Both patients presented had clinical characteristics suggestive of BMS. They had a significant history of depressive

and anxious symptoms and reported worsening of pain complaints coincident with worsening mood, which suggests that BMS can have a psychogenic origin. However, on the other hand, given the favorable response to drugs indicated for the treatment of neuropathic pain (the first patient only experienced pain relief with carbamazepine, after unsuccessful antidepressant and anxiolytic prescriptions), this may indicate that BMS may not merely be a somatic manifestation of depression or anxiety but a result of a neurological disorder, although the comorbidity with psychogenic factors is high and they may in fact play a significant role in aggravation/maintenance of complaints. We also cannot neglect the hypothesis that the distress caused by these symptoms themselves may lead to depressive and

anxious syndromes, and that may raise the “chicken and the egg” dilemma in some cases.

The etiology and pathophysiological mechanisms of BMS are still unclear, as well as effective forms of treatment, since total remission is unusual. Some pharmacological approaches were described, but there is a lack of clinical practice guidelines to follow to treat or alleviate BMS symptoms. Regardless of whether it is cause or consequence, the comorbidity with depression or anxiety is frequent and must be correctly addressed by the psychiatrist with drugs approved for their treatment. Since tricyclic, SSRI and SNRI antidepressants, as well as clonazepam, seem to be useful in BSM symptoms we suggest that the pharmacological approach should preferably begin with a choice between these drugs.

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