

Burning mouth syndrome following damage to the olfactory system? An atypical case report *

Síndrome da ardência bucal após lesão do sistema olfativo? Um relato atípico de caso.

Cibele Nasri¹, Massako Okada², Gilberto Formigoni³, Manoel Jacobsen Teixeira⁴, José Tadeu Tesseroli de Siqueira⁵

* Received from Division of Dentistry and Department of Neurology Medical School, University of São Paulo, São Paulo, Brazil.

SUMMARY

BACKGROUND AND OBJECTIVES: Burning Mouth Syndrome (BMS) is a chronic pain condition in which the patient reports an intraoral burning sensation in absence of clinical and laboratory findings. Although its etiology is still unknown, recent studies suggests a neuropathic pain origin for this syndrome. This article reports a very unusual case of a patient who developed BMS and phantom taste after damage to the olfactory system exploring a possible association between BMS, gustatory and olfactory systems.

CASE REPORT: Male patient, 50-years-old had a chief complaint of burning sensation on the soft palate area for six years. He reported that the onset of burning was immediately after he had painted his house and slept overnight in a freshly painted room. He also reported worsening of his symptoms in the presence of strong aromas or

when he gets a cold, and a constant bitter taste. A clinical exam of the intra-oral mucosa was normal with no signs of lesions or any other pathology which could justify his complaint. All exams, serological and metabolic (glucose, iron, vitamin B₁₂, folate levels and CBC), did not show any significant findings. The final diagnosis was an atypical presentation of BMS.

CONCLUSION: Although the mechanisms involved are not clear, the case report a possible association between BMS, gustatory and olfactory systems. Future research is necessary to better understand the features involved.

Keywords: Burning mouth syndrome, Orofacial pain, Phantom taste.

RESUMO

JUSTIFICATIVA E OBJETIVOS: A Síndrome da Ardência Bucal (SAB) é uma condição de dor crônica em que os pacientes relatam uma queimação intraoral na ausência de achados clínicos e laboratoriais. Embora sua etiologia seja ainda desconhecida, estudos recentes sugerem uma origem de dor neuropática para tal síndrome. Este artigo relata um caso bastante raro de paciente que desenvolveu SAB e gosto fantasma após lesão do sistema olfatório, e explora uma possível associação entre SAB e sistemas gustativo e olfatório.

RELATO DO CASO: Paciente do sexo masculino, 50 anos, com queixa importante de queimação na área do palato mole por seis anos. Informou que o início da queimação foi imediatamente após ele haver pintado sua casa e ter dormido em um quarto recém-pintado. Também informou piora dos sintomas na presença de cheiros fortes ou quando pega um resfriado, além de um gosto amargo constante. O exame clínico da muco-

1. DDS, MSD Orofacial Pain Team, Dentistry Division, Central Institute, Hospital das Clínicas, Medical School, University of São Paulo, SP, Brazil.

2. MD, Multidisciplinary Pain Center, Division of Neurology, Central Institute, Hospital das Clínicas, Medical School, University of São Paulo, SP, Brazil.

3. MD, PhD, Department of Otolaryngology, Hospital das Clínicas, Medical School, University of São Paulo, SP, Brazil.

4. PhD, Director, Multidisciplinary Pain Center, Division of Neurology, Central Institute and Experimental Neurosurgery Division, Psychiatric Institute, Hospital das Clínicas, Medical School, University of São Paulo, SP, Brazil.

5. DDS, PhD, Supervisor, Orofacial Pain Clinic, Dentistry Division, Central Institute, Hospital das Clínicas, Medical School, University of São Paulo, SP, Brazil.

Correspondence author:

José Tadeu Tesseroli de Siqueira
Rua Maria Cândida, 135 – Vila Guilherme
02071-010 São Paulo, SP.
E-mail: jtts@uol.com.br

sa intraoral foi normal sem sinais de lesões ou outras patologias que pudessem justificar suas queixas. Nenhum exame sorológico e metabólico (glicose, ferro, vitamina B₁₂, níveis de folato e hemograma completo) apresentou achados importantes. O diagnóstico final foi uma apresentação atípica de SAB.

CONCLUSÃO: Embora os mecanismos envolvidos não estejam claros, o caso relata uma possível associação entre SAB e sistemas gustativo e olfatório. São necessárias mais pesquisas para melhor entender as características envolvidas.

Descritores: Dor orofacial, Gosto fantasma, Síndrome da ardência bucal.

INTRODUCTION

According to the International Association for the Study of Pain, Burning Mouth Syndrome (BMS) is defined as a burning pain in the tongue and/or other oral mucous membrane in an absence of clinical signs or laboratory findings¹.

This condition affects predominantly middle-age women in the post menopausal period. Occurrence below the age of 30 is rare and the female-to-male ratio is approximately 7:1²⁻⁵. Multiple oral sites may be involved, but the anterior two-thirds and the tip of tongue are most commonly affected, followed by the palate^{6,7}.

The onset can be either gradual and spontaneous or sudden and related to a precipitating event. Patients usually report that the burning sensation presents its lowest intensity upon awakening, but reappears after the first meal of the day. Once begun, it is continuous, reaching the maximum intensity by late evening⁸.

The prognosis is poor and the burning sensation can last for many years. A complete spontaneous remission is rare and may occur in only 3% of the patients five years after the onset⁹.

It is important to distinguish between true BMS and symptomatic burning sensation. Burning mouth as a symptom occurs when the burning sensation is secondary to a local or systemic pathologic condition and BMS occurs when local or systemic causes cannot be identified; thus, the burning is the disease itself¹⁰. Although the BMS etiology is still unknown a growing body of evidence in the literature suggests that BMS is a neuropathic pain and might be related to the damage in the Gustatory System, particularly damage on the nerves that supply taste. According to Grushka and Bartoshuk¹¹, 2000 BMS would occur as a result of the disinhibition of nociception regulated by the interactions of the cranial nerves that supply taste VII, IX, X (Chorda Tympani,

Glossopharyngeal and Vagus nerves) and the Trigeminal nerve.¹¹ Soon after, Eliav et al.¹² demonstrated the hypo-function on the chorda tympani nerve in BMS patients reinforcing the theory of the involvement of the gustatory system in BMS patients. Moreover 70% of BMS patients report changes in taste perception (dysguesia or phantom taste), which reinforces the idea that BMS is associated with the Gustatory system⁴.

This article presents a very unusual case report where the patient developed BMS and taste phantom after damage in the olfactory system exploring a possible association of BMS, gustatory and olfactory systems.

CASE REPORT

Male patient, 50-years-old was referred to the Orofacial Pain Team, Division of Dentistry, Hospital das Clínicas, Medical School, University of São Paulo, with a chief complaint of burning sensation on the soft palate area for six years.

Reported that the onset of burning was immediately after he had painted his house and slept overnight in a freshly painted room. The next morning, when he awoke he experienced a burning sensation which increased gradually.

Since then it has been constant, mild in the morning and moderate/severe during the day until night. The patient reported worsening of his symptoms in the presence of strong aromas or when he gets a cold. The patient also reported a constant bitter taste.

His medical history includes Chagas' disease, hypertension and gastritis. He also reported to be "allergic to the glue smell" (SIC). Currently he is not on any medication and his hypertension is controlled by diet only.

Previously the patient was examined by a gastroenterologist and was diagnosed with gastritis. He was treated with no improvement of his burning mouth complaint.

The patient also was examined by a neurologist and an ear, nose and throat specialist with no findings related to his burning sensation.

A clinical exam of the intra-oral mucosa was normal with no signs of lesions or any other pathology which could justify his complaint.

The patient also was referred for a serology, which included a basic metabolic panel (Glucose Serum level/Diabetes); iron serum levels/ferritin (iron deficiency anemia); vitamin B₁₂ and folate levels (vitamin B₁₂ and folate anemia respectively); and CBC (for general health status) as well, with no significant findings. Saliva fungal culture was negative.

Currently the patient wears a maxillary and mandible par-

tial denture which is in a good condition, as are the remaining teeth as well. Diagnosis: Burning Mouth Syndrome.

DISCUSSION

The burning mouth complaint not related to any clinical or laboratory findings, meets the current diagnostic criteria for BMS which is reached by the exclusion of any other condition that could justify the intra-oral burning sensation.

In this case all the possible local and systemic causes for his complaint were excluded. The fact that the patient presents with Chagas' disease does not contribute to his burning complaint. The hypertension has not been controlled with medication, just only by diet. Although gastritis is a common comorbidity in BMS patients it does not contribute to the burning complaint in this case as the patient was evaluated by a gastroenterologist who excluded the possibility of gastroesophageal reflux, a potential cause of burning mouth complaint¹³.

The location of the burning sensation on the palate and the fact that the intensity of the pain gradually increases during the day reaching the maximum intensity at night, is consistent with the general characteristics of BMS patients reported in the literature^{14,15}.

The interesting feature about this case is that the patient developed BMS on the following morning after he had slept in a fresh painted room suggesting a relationship between the two events. One possible explanation for this fact could be the damage of the olfactory system after exposure, by inhalation, to a chemical agent present in the paint causing nerve damage. It is known that accidental inhalation of paint solvents such as toluene, butyl acetate, benzene and others can be neurotoxic¹⁶. It is also known that an acute episode of neuritis-evoked neuropathic pain may contribute to the genesis of chronically painful peripheral neuropathies, and that a chronic focal neuritis might produce neuropathic pain in the absence of clinically detectable structural damage to the nerve¹⁷. The fact that the patient reports worsening of symptoms in the presence of strong aromas confirms the involvement of the olfactory system with his complaint.

But the fact that the patient report phantom taste (constant bitter taste), which is a disorder of the Gustatory system; also confirm the involvement of the gustatory system with his complaint. Taste phantoms, especially bitter and metallic are believed to be the result of disinhibition of the glossopharyngeal nerve following damage to the chorda tympani nerve^{18,19}. Moreover, primarily

bitter and metallic tastes are reported as a taste phantom, for 70% of BMS patients^{20,21}.

But the instigating question is how damage in the olfactory system could be associated to damage in the Gustatory System and BMS?

One possible explanation could be based in the sensorial interaction from Melzack and Wall where sensorial modalities as pain, smell and taste might interact²². Evidence shows the reduction of the olfactory threshold and changes of taste in patients with trigeminal neuralgia after been submitted to the compression of the trigeminal ganglion with a balloon as a treatment. This finding suggests a relationship between the damage of the trigeminal nerve, changes in taste and reduction of the olfactory threshold mediated by the trigeminal afferents (maxillary branch) reinforcing the Melzack and Wall theory of sensorial Interaction²³.

Functional (fMRI) studies confirm the projection of olfactory signals reaching the orbitofrontal cortex (mainly on the right side) which is a zone that integrates taste, vision, olfaction, and probably touch. It also show activation of the frontal operculum, insula and orbitofrontal cortices for both olfactory and taste stimulation²⁴.

It is also known that the olfactory and gustatory systems work together and are intimately related. Recently, Landis et al.²⁵ suggested that longstanding impaired olfactory function is associated with decreased gustatory function.

Certainly this case report has no intention to build up any theory about BMS etiology. But, as for the best of our knowledge there is no similar report in the literature, for sure has the intention to raise a possible new question for future research in this field.

CONCLUSION

Although the mechanisms involved are not clear, the case report a possible association between BMS, gustatory and olfactory systems. Future research is necessary to better understand the features involved.

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