Neurología. 2017;xxx:xxx—xxx

SOCIEDAD ESPAÑOLA DE NEUROLOGIA

NEUROLOGÍA

www.elsevier.es/neurologia

ORIGINAL ARTICLE

Burning mouth syndrome: clinical description, pathophysiological approach, and a new therapeutic option☆

A. Cárcamo Fonfría[a,*], L. Gómez-Vicente[b], M.I. Pedraza[c], M.L. Cuadrado-Pérez[b], A.L. Guerrero Peral[c], J. Porta-Etessam[b]

[a] *Servicio de Neurología, Hospital Universitario Fundación Jiménez Díaz, Madrid, Spain*
[b] *Servicio de Neurología, Hospital Universitario Clínico San Carlos, Madrid, Spain*
[c] *Servicio de Neurología, Hospital Clínico Universitario de Valladolid, Valladolid, Spain*

Received 29 April 2015; accepted 25 October 2015

KEYWORDS
Burning mouth;
Glossopyrosis;
Disabling;
Treatment;
Dopamine agonists

Abstract
Introduction: Burning mouth syndrome is defined as scorching sensation in the mouth in the absence of any local lesions or systemic disease that would explain that complaint. The condition responds poorly to commonly used treatments and it may become very disabling.
Methods: We prospectively analysed the clinical and demographic characteristics and response to treatment in 6 cases of burning mouth syndrome, diagnosed at 2 tertiary hospital headache units.
Results: Six female patients between the ages of 34 and 82 years reported symptoms compatible with burning mouth syndrome. In 5 of them, burning worsened at the end of the day; 4 reported symptom relief with tongue movements. Neurological examinations and laboratory findings were normal in all patients and their dental examinations revealed no buccal lesions. Each patient had previously received conventional treatments without amelioration. Pramipexol was initiated in doses between 0.36 mg and 1.05 mg per day, resulting in clear improvement of symptoms in all cases, a situation which continues after a 4-year follow up period.
Conclusions: Burning mouth syndrome is a condition of unknown aetiology that shares certain clinical patterns and treatment responses with restless leg syndrome. Dopamine agonists should be regarded as first line treatment for this entity.

☆ Please cite this article as: Cárcamo Fonfría A, Gómez-Vicente L, Pedraza MI, Cuadrado-Pérez ML, Guerrero Peral AL, Porta-Etessam J. Síndrome de boca ardiente: descripción clínica, planteamiento fisiopatológico y una nueva opción terapéutica. Neurología. 2017. http://dx.doi.org/10.1016/j.nrl.2015.10.008
* Corresponding author.
E-mail address: albacarcamof@gmail.com (A. Cárcamo Fonfría).

PALABRAS CLAVE
Boca ardiente;
Glosopirosis;
Discapacitante;
Tratamiento;
Agonistas dopaminérgicos

Síndrome de boca ardiente: descripción clínica, planteamiento fisiopatológico y una nueva opción terapéutica

Resumen
Introducción: El síndrome de la boca ardiente se define como sensación de ardor intrabucal, en ausencia de lesiones locales o patología sistémica que lo justifique. Se trata de una entidad con pobre respuesta a los tratamientos comúnmente utilizados, que puede resultar muy discapacitante.
Métodos: Analizamos prospectivamente las características clínicas, demográficas y la respuesta a tratamiento de 6 casos de síndrome de la boca ardiente diagnosticados en las consultas de cefaleas de 2 hospitales de tercer nivel.
Resultados: Se trata de 6 pacientes de sexo femenino, con edades entre 34 y 82 años, que referían síntomas compatibles con síndrome de la boca ardiente. En 5 pacientes, las molestias empeoraban a última hora del día y 4 referían mejoría de los síntomas con los movimientos linguales. En todos los casos la exploración neurológica fue normal, los estudios analíticos no mostraron alteraciones que justificaran los síntomas y en el examen odontológico no se evidenciaron lesiones intrabucales. Todas las pacientes habían sido tratadas previamente con los tratamientos convencionales, sin mejoría. Se instauró pramipexol a dosis entre 0,36 mg y 1,05 mg al día, con lo que se consiguió mejoría evidente en todos los casos, que persiste tras una media de 4 años de seguimiento.
Conclusiones: El síndrome de la boca ardiente sigue siendo una entidad de etiología desconocida, que comparte ciertos patrones clínicos y respuesta al tratamiento con el síndrome de piernas inquietas. Los agonistas dopaminérgicos deberían considerarse como tratamiento de primera línea en esta entidad.

Introduction

Burning mouth syndrome (BMS) or glossodynia is defined as intraoral burning or dysaesthetic sensation recurring daily during more than 2 hours over a period exceeding 3 months, without clinically evident causative lesions or any other possible causes.[1] It is a complex and probably underdiagnosed syndrome with no specific treatment. Patients with BMS frequently visit several specialists who may be unable to alleviate the condition or answer their questions. We present a series of patients with BMS who shared several clinical characteristics and displayed a similar response to treatment with a dopaminergic agonist.

Patients and methods

We prospectively analysed 6 patients with BMS who had been diagnosed with the disease at the headache units of 2 tertiary hospitals and describe their demographic and clinical characteristics and response to treatment. Case 1: 72-year-old woman with no relevant medical history. Over the course of several years, she had been experiencing an intraoral burning sensation which worsened in the evening. Pain affected the entire mouth, was severe in intensity, and caused discomfort. A neurological examination and a complete analysis (including ferritin, vitamin B_{12}, and thyroid hormone measurements) yielded normal results. The patient had previously visited several dentists who had not detected any significant alterations. Clonazepam mouthwash had failed to provide relief. Pramipexole dosed at 0.18 mg and administered in the afternoons and at night led to substantial improvement; the patient remained nearly asymptomatic 4 weeks after dose escalation. Case 2: 71-year-old woman with no relevant medical history. She was a frequent consumer of clebopride and simeticone. She reported an intraoral burning sensation that intensified over the course of the day. Pain affected the tongue in particular and subsided with tongue movement. Thorough neurological and maxillofacial examinations and a complete laboratory analysis revealed no abnormalities. Clonazepam and nystatin mouthwashes achieved no improvement. Our patient was treated with increasing doses of pramipexole. At 4 weeks (with doses of 0.18 mg in the afternoon and 0.36 mg at night), our patient experienced significant relief, which remains to date, after nearly 4 years of follow-up. Case 3: 37-year-old woman with a family history of glossodynia. Our patient had experienced symptoms compatible with BMS for several years. Symptoms fluctuated, with pain intensifying later in the day and subsiding while speaking or eating. A complete laboratory analysis and a neurological examination revealed no abnormal findings. A dentist's assessment had ruled out local lesions. Symptoms did not respond to clonazepam or amitriptyline. We initiated treatment with increasing doses of pramipexole until achieving significant improvement at 6 weeks from treatment onset (0.35 mg in the afternoon and 0.70 mg at