Burning mouth syndrome secondary to pregabaline in a patient with mild frontal lobes atrophy

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We recently read with great interest the burning mouth syndrome (BMS) systematic review in your journal by Silvestre et al [1]. This paper reminds us of a very particular patient that we would like to discuss here.

A 71-year-old-male patient was seen in outpatient psychiatric consultation for spontaneous pain in the tip of tongue and burning sensation of gingival mucosa, complicated by suicidal ideation. His symptoms had started one year before, right after treatment with pregabaline for generalized anxiety disorder. There was also depressive mood with feeling of helplessness, hopelessness, social isolation and negative impact in his social life, all secondary to somatic complains. He had been searching for help in many dentistry, stomatology and neurology specialists but, whatever the diagnosis, his symptoms never responded to treatment. Oral cavity pathology was excluded and no changes were found on blood and urine samples. EEG showed multifocal slowing in frontal lobes, in relation with mild frontal lobes atrophy revealed by brain CT scan. Nonetheless, neuropsychological evaluation revealed normal intelligence without signs of dementia. The patient had been previously treated with many different drugs for dysthymia, hypothyroidism, Parkinson disease, benign prostatic hyperplasia, chronic gastritis and arterial hypertension. However, both patient and spouse were very certain that pregabaline was the only responsible for the symptoms. His daughter had 'migraine and depression' and his cousin had an undisclosed 'mental disorder'. There was no other relevant personal or familial previous history. Successful management of dysthymia was achieved with daily amitriptyline 100 mg plus paroxetine 20 mg, while parkinsonism and hypothyroidism were treated, respectively, with daily ropinirol 4 mg and levothyroxine 0.15 mg. After comorbidity treatment the patient was still complaining of his initial symptoms, so BMS (classic type 1) diagnosis was made and started on clonazepam 1 mg thrice a day, with full remission after 2 years of treatment. Following BMS relapse, and after almost 4 years of follow up, the patient abandoned consultation, hoping for better treatment in the oral maxillofacial surgery department of another hospital.

BMS is classified among the central causes of facial pain, and is defined as a spontaneous burning and painful sensation manifesting on the oral mucosa in the absence of exploratory findings or other identifiable local or systemic causes [2]. BMS cause is uncertain but some neuroimaging [3] and neurophysiologic [4] studies suggested some frontal lobes changes. Although pregabaline has already been proposed for the treatment of BMS [5] this case report alerts us for the possibility of a secondary BMS after the implementation of any kind of drug. All these patients shall be carefully studied in order to understand their symptoms and possible causes (e.g. iatrogenic and\or systemic). We believe that the combining effect of previous frontal lobes dysfunction plus concomitant iatrogenic drug may contribute for the complex physiopathology of some of the patients suffering of this syndrome. This hypothesis should be object of further studies, for better understanding and treatment of BMS.

References

1. Silvestre FJ, Silvestre-Rangil J, López-Jornet P. Síndrome de boca ardiente: revisión y puesta al día. Rev Neurol 2015; 60: 457-63.
2. Headache Classification Subcommittee of the International Headache Society. The International Classification of Headache Disorders: 2nd edition. Cephalalgia 2004; 24 (Suppl 1): S9-160.
3. Albuquerque RJ, De Leeuw R, Carlson CR, Okeson JP, Miller CS, Andersen AH. Cerebral activation during thermal stimulation of patients who have burning mouth disorder: an fMRI study. Pain 2006; 122: 223-34.
4. Eliav E. Altered structure and function in hippocampus and medial frontal cortex in patients with burning mouth syndrome. Pain 2014; 155: 1424-5.
5. López V, Alonso V, Martí N, Calduch L, Jordá E. Marked response of burning mouth syndrome to pregabalin treatment. Clin Exp Dermatol 2009; 34: e449-50.