CASE REPORTS

Use of Topiramate for Glossodynia

Antonio Siniscalchi, MD,† Luca Gallelli, MD,†† Norma Maria Marigliano, MD,‡ Paola Orlando, MD,‡ and Giovambattista De Sarro, MD†

†Department of Neuroscience, Neurology Division, “Annunziata” Hospital, Cosenza; ‡Department of Experimental and Clinical Medicine, University Magna Grecia of Catanzaro, Clinical Pharmacology Unit, Mater Domini University Hospital, Catanzaro, Italy

ABSTRACT

Introduction. Glossodynia is a multifunctional disorder characterized by painful sensations in the mouth and throat and especially on the tongue. It is commonly cured by long-term therapy with systemic regimens of anxiolytics, antidepressants, and anticonvulsants.

Case. We report here the case of a 65-year-old woman with a 4-month history of glossodynia. Clinical and laboratory evaluations performed the diagnosis of idiopathic glossodynia, and several treatments with carbamazepine and then with gabapentin induced the development of serious adverse reaction. Only treatment with topiramate has been able to induce a complete improvement of symptoms.

Discussion. The pathogenesis of idiopathic glossodynia remains unclear, since it recently has been suggested as a possible neuropathic basis of burning mouth syndrome, demonstrating an altered excitability in the trigeminal nociceptive pathway at peripheral and/or central nervous system level. The various mechanisms of topiramate, which act at different neural transmission levels, blocking sodium and calcium channels, enhancing GABA concentration, and decreasing glutamate function at postsynaptic site, may explain the effects of topiramate in our patient.

Conclusion. Therefore, we suggest that topiramate could represent a useful therapeutic option in the treatment of glossodynia.

Key Words. Glossodynia; Carbamazepine; Gabapentin; Topiramate

Glossodynia is a multifunctional disorder characterized by painful sensations in the mouth and throat, and especially on the tongue. It has a prevalence of 4–5% in the general population [1]. Multiple etiological factors of local, systemic, and psychological origin have been suggested [2–5]. Although many drugs have been proposed for the treatment of glossodynia, the management of this manifestation is still not satisfactory [6,7]. It is commonly cured by a long-term therapy with systemic regimens of anxiolytics [8] and antidepressants [9,10]. In addition, it had recently been reported that gabapentin, an anticonvulsant drug, is effective in the treatment of glossodynia [11,12]. Topiramate is a new antiepileptic drug that has shown efficacy in intercostal [13] and trigeminal neuralgia [14]. We report here the case of a patient with glossodynia successfully treated with topiramate.

Reprint requests to: Dr. Luca Gallelli, MD, Department of Experimental and Clinical Medicine, University Magna Grecia of Catanzaro, Clinical Pharmacology Unit, Mater Domini Hospital, Via T. Campanella, 115, 88100 Catanzaro, Italy. Tel: +39-0961-712322; Fax: +39-0961-774424; E-mail: luca_gallelli@hotmail.com.

†Share the authorship.
A 65-year-old woman, with a 4-month history of glossodynia, was presented to the neurological division of Cosenza Hospital for consultation. Work-up revealed that oral pain occurred several times a day. A detailed anamnesis found that there was no history of alcohol or other drug abuse, there was no significant medical or surgical history, and she was not taking any medication. She denied symptoms consistent with psychiatric disorders. Physical examination revealed no underlying disorders at the time of our observation. Her blood pressure was 110/70 mm Hg, heart rate 85 beats/min, respiratory rate 22 breaths/min, and temperature 36.2°C. Oral mucosa and conjunctiva were pink and moist. The tongue appeared normal both before and after eating. The mandible was edentulous with lower complete denture, and the maxilla was partially edentulous. There were no evident signs of oral inflammations or infections. There was no angular cheilitis or lymphadenopathy.

A visual analog scale (VAS) was used to evaluate the perceived pain intensity, and a score of 6.2 was obtained. Dentist evaluation performed by a specialist excluded that oral pain was induced by denture factors. Oral culture excluded bacterial or fungal contaminations. Laboratory findings excluded systemic dysfunctions or malnutrition. Therefore, the diagnosis of idiopathic glossodynia was made, and carbamazepine 200 mg every 8 h was started. Two days later, the woman was discharged home on a regimen of carbamazepine 300 mg every 8 h.

About 18 days after discharge, she was readmitted because of the persistence of glossodynia and for the development of visual disturbance. Neurological examination revealed the presence of diplopia and ataxia, while pharmacological blood evaluation demonstrated high levels of carbamazepine (14 µg/mL; normal values < 12 µg/mL).

Therefore, carbamazepine was promptly discontinued, and gabapentin 300 mg every 8 h was initiated. Five days later, she was discharged with a complete remission of neurological adverse events. Ambulatory evaluation performed 4 days later revealed the persistence of oral pain, inducing an increase in gabapentin dosage to 900 mg every 12 h. About 4 weeks following the beginning of gabapentin, a new ambulatory evaluation revealed the appearance of dizziness and somnolence, which prompted discontinuation of gabapentin with the beginning of topiramate treatment (50 mg every 12 h). After 4 weeks with no adverse events, the topiramate dosage was increased to 100 mg every 12 h and then, 2 weeks later, to 150 mg every 12 h. Ambulatory evaluation revealed a VAS score of 2.5 with full resolution of symptoms, and she was complaint free at 1- and 3-month follow-ups without dose adjustments of topiramate or even drug substitution.

At present, the patient has not shown glossodynia, and no adverse event associated with topiramate has been recorded.

**Discussion**

Glossodynia is defined as a spontaneous syndrome of burning sensation, discomfort, pain irritation, or rawness of the tongue, lips, or oral cavity that frequently affects women in the 5th–7th decades [15]. It has previously been reported that there are two main category of glossodynia, one with organic systemic factors and the other with psychogenic factors [16].

Several systemic factors may influence the prevalence, development, and severity of burning mouth syndrome [17,18]. The most significant systemic factors predisposing conditions for burning mouth syndrome are menopausal disorders, diabetes, anemia, inflammatory bowel syndrome or other gastrointestinal disturbances, and xerostomia. Moreover, the location of the pain in the oral mucosa excludes diseases such as atypical facial pain, atypical odontalgia, and idiopathic facial arthromyalgias, which affect bones, teeth, muscles, and articulation, respectively. In contrast, in our patient, specialist evaluation performed by the dentist excluded that pain could be induced both by denture factors and by oral mucosal lesions such as traumatic lesions, infections (e.g., candidiasis), and chronic erosive/ulcerative stomatitis (aphthous stomatitis, erosive lichen planus, pemphigoid, pemphigus, etc.). Many drugs could be able to induce burning mouth [19,20], but we excluded this possibility because our patient was not taking any medication. Moreover, anamnesis also excluded psychological disorders, while laboratory findings did not reveal any nutritional deficiencies. Therefore, a diagnosis of idiopathic glossodynia was made. The pathogenesis of idiopathic glossodynia remains unclear, as it recently has been suggested as a possible neuropathic basis of burning mouth syndrome demonstrating an altered excitability in...
the trigeminal nociceptive pathway at peripheral and/or central nervous system level [15,21,22]. Many articles reported that carbamazepine and gabapentin were effective in the treatment of neuropathic pain syndrome [23,24]. In our patient, the treatment with carbamazepine induced the development of diplopia and ataxia. Moreover, it was reported that these adverse effects represents the common side effects induced by carbamazepine [25]. However, gabapentin treatment also induced the development of dizziness and somnolence as demonstrated in diabetic patients affected by painful neuropathy [26]. In addition, it recently has been demonstrated that topiramate could be used in the treatment of neuropathic pain [23], as well as in both idiopathic [27] and symptomatic trigeminal neuralgia [14,28]. In our patient, topiramate induced a dose-dependent improvement of oral pain symptoms. The various mechanisms of topiramate, which act at different neural transmission levels, blocking sodium and calcium channels, enhancing gamma-aminobutyric acid (GABA) concentration, and decreasing glutamate function at postsynaptic site [29], may explain the effects of topiramate in our patient. Other advantages of topiramate include the lack of relevant adverse effects and few interactions with other drugs. In conclusion, the present findings suggest that topiramate could represent a useful therapeutic option in the treatment of glossodynia.

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