

A Case Report of Burning Mouth Syndrome with Dry Mouth Managed by Kampo Medicine

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The patient was a 56-year-old woman who complained of chronic pain involving her tongue. We diagnosed her with burning mouth syndrome (BMS) based on exclusion of any local factors or systemic conditions. The patient not only had tongue pain but also had other signs and symptoms like scalloped tongue, dry mouth, and headache. To manage these additional issues, we used Goreisan, an herbal Kampo medicine, as a complementary alternative medicine (CAM) approach along with cognitive behavioral therapy (CBT). The patient's BMS was successfully managed with the combination of CAM and CBT, which may suggest that the pathophysiology for BMS might be nociplastic pain rather than purely nociceptive or neuropathic.

Key Words: Burning mouth syndrome; Psychogenic factor; Dry mouth; Kampo medicine; Nociplastic pain.

Burning mouth syndrome (BMS) is defined in the International Classification of Orofacial Pain (ICOP) as an intraoral burning or dysesthetic sensation, recurring daily for more than 2 hours per day for more than 3 months, without evident causative lesions on clinical examination and investigation.¹ Historically, BMS was classified as primary whenever a cause could not be found and secondary if caused by an underlying medical (local or systemic) condition. The latest available classification distinguishes between BMS with and without somatosensory changes based on quantitative sensory testing.¹

CASE PRESENTATION

A 56-year-old female patient (height 154 cm; weight 80 kg; body mass index 34 kg/m²) complained of chronic pain involving the left lateral part of her tongue (Table 1). Her medical history was significant for immuno-

globulin A nephropathy, obstructive sleep apnea (OSA), hypertension, and venous thrombosis, and she reported taking aldactone 25 mg/d, cilnidipine 10 mg/d, and aspirin 100 mg/d and using continuous positive airway pressure for her OSA. She described her tongue pain mostly as a burning sensation that continued all day. The patient's symptoms were exacerbated when talking but were reduced whenever she ate. Besides the tongue pain, she also complained of headaches and dry mouth, although a quantitative saliva test demonstrated results within normal limits (Saxon test: 3.94 g/2 min; normal ≥ 2.75 g/2 min). Extraoral and intraoral examinations revealed no obvious problems except a scalloped tongue (Figure). A panoramic radiograph was obtained, but it did not reveal any pathological findings.

To evaluate the patient's psychological factors, the Japanese version of the General Health Questionnaire-60 (GHQ-60) was used. It was classified into 4 subscales: (A) somatic symptoms, (B) anxiety/insomnia, (C) social dysfunction, and (D) severe depression. The patient's total score on the GHQ-60 and scores on its 4 subscales were high (total, 43/60; A, 5/7; B, 4/7; C, 6/7; D, 5/7).

The patient was diagnosed with BMS based on the exclusion of any local factors (eg, candidiasis, hyposalivation, mucosal lesions, trauma) or systemic conditions (eg, vitamin deficiency, diabetes, hypothyroidism, side effect of medications, autoimmune disorders). To manage her signs and symptoms (scalloped tongue,

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Table 1. Structured Pain Questionnaire Used to Assess Chronic Pain*

Questions	Patient's responses (signs and symptoms)
Location	Left lateral side of the tongue
Character	Burning
Intensity (NRS)	5
Duration	All day long
Frequency	Every day
Triggering or aggravating factors	Talking
Improving factors	Eating
Temporal pattern	Gradually increasing after waking
Associated signs and symptoms	Scalloped tongue, dry mouth, headache

* NRS, numerical rating scale (0–10).

dry mouth, and headache), we advised the patient use Goreisan, a traditional Japanese herbal Kampo medicine. After returning a few months later for follow-up, the patient reported that her quality of daily life had significantly improved and that her BMS had been successfully managed by counseling along with the use of the Kampo medicine.

DISCUSSION

Burning mouth syndrome is also known as glossodynia if the symptoms are confined to the tongue only.² Since the introduction of the ICOP,¹ the diagnosis of BMS is given only after ruling out all potential causes (Table 2). The etiology of BMS is still unclear. There is some evidence that BMS could be considered a neuropathic pain condition,³ but psychogenic factors are involved in

Figure. Scalloped Tongue

Clinical photograph demonstrating the patient's scalloped tongue. Teeth marks were observed on the lateral border of the tongue.

Table 2. Diagnostic Criteria for BMS According to the ICOP-1*

BMS diagnostic criteria
A. Oral pain fulfilling criteria B and C
B. Recurring daily for >2 hours per day for >3 months
C. Pain has both of the following characteristics: <ol style="list-style-type: none"> 1. Burning quality 2. Felt superficially in the oral mucosa
D. Oral mucosa is of normal appearance, and local or systemic causes have been excluded
E. Not better accounted for by another ICOP or ICHD-3 diagnosis

* BMS, burning mouth syndrome; ICHD-3, International Classification of Headache Disorders 3rd edition; ICOP-1, International Classification of Orofacial Pain 1st edition.

a large percentage of patients with BMS.⁴ Therefore, a thorough history, detailed physical examination, and psychological assessment are needed for a proper diagnosis. High scores on the psychometric test (GHQ-60) used in this case as well as carcinophobia (chronic fear of developing cancer), which is a common finding among patients with BMS, were noted with our patient.

The management for BMS also remains unestablished. Clonazepam has been used in clinical practice as a first-line medication for managing BMS.² Kampo medicine is one type of complementary alternative medicine (CAM) that utilizes herbal remedies. Goreisan is an herbal medicine consisting of ingredients such as Takusha (*Alismatis rhizoma*), Sojutsu (*Atractylodis lanceae rhizoma*), Chorei (*Polyporus*), Bukuryo (*Hoelen*), and Keihi (*Cinnamomi cortex*). It can be effective for symptoms such as edema, dizziness, diarrhea, headache, and dry mouth.

We believe that focusing not only on decreasing pain but also on reducing additional symptoms is important when managing BMS. As such, cognitive behavioral therapy (CBT) also may be helpful.² The patient's pain in the present case increased when talking but was reduced when eating. As such, we advised her to avoid aggravating factors and adopt improving factors in her daily life.

CONCLUSION

Considering that both CAM and CBT may be helpful for managing BMS and were effective in this case, the pathophysiology for BMS might be best considered a nociplastic pain that is neither purely nociceptive nor neuropathic.⁵

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