Burning Mouth Syndrome: Case Report

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FIGURE 1. Histiocyte showing phagocytosis of numerous erythrocytes. Color image is available online only at www.psychopharmacology.com.

The patient received only supportive care during his admission. Interestingly, halving of ferritin levels was observed in 1 day after cessation of lamotrigine, which corresponds to the half-life of ferritin,2 clearly supporting HLH as serious adverse event of lamotrigine. Venlafaxine was restarted on the second day after admission. Six days after admission, the patient had made a full clinical recovery, and his biochemical parameters had largely normalized and continued to do so during the next few weeks of outpatient follow-up.

Hemophagocytic lymphohistiocytosis has sporadically been described after initiation of lamotrigine. With an increasing number of reports surfacing in recent years, the association with this nontrivial condition of lamotrigine has largely normalized and continued to do so during the next few weeks of outpatient follow-up. Venlafaxine was restarted available online only at www.psychopharmacology.com.

AUTHOR DISCLOSURE INFORMATION
The authors declare no conflicts of interest.

Received November 10, 2020; accepted after revision February 8, 2021.

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Burning Mouth Syndrome
Case Report

To the Editor:
Burning mouth syndrome (BMS), also known as glossodynia or stomatodynia, is characterized by burning oral pain in the absence of an identifiable lesion.1 The prevalence of BMS is estimated to be between 0.7% and 4.5%.2-4 This condition is more common in postmenopausal women2 and often co-occurs with other symptoms, such as parageusia and xerostomia.3

There are no laboratory tests to establish a definitive diagnosis of BMS.6 As BMS is associated with somatization, anxiety, depression, dissociation, and personality disorders,7 a patient’s description of these symptoms may be overlooked. Herein, we present the case of a patient with bipolar disorder who developed BMS after the initiation of quetiapine.

CASE PRESENTATION

The patient is a 65-year-old White woman with a longstanding history of bipolar I disorder with mixed features. The patient’s symptoms were relatively controlled with 10-mg aripiprazole and 0.25-mg clonazepam daily. In May 2020, she reported increasing jaw tightness, and therefore, aripiprazole was cross-titrated with quetiapine. Concurrently, clonazepam was discontinued. By June 2020, the cross-titration was completed, and she was no longer taking aripiprazole, which was replaced with 300-mg quetiapine nightly. She reported resolution of her facial extrapyramidal symptoms and denied any adverse effects from quetiapine.
Throughout July 2020, she began to experience worsening xerostomia. She used sugarless gum and lemon drops/mints, but these did not alleviate symptoms. At this time, she demonstrated a weakly positive antinuclear antibody titer of 1:320. August 2020, her symptoms had significantly intensified; she reported a constant burning sensation on her hard palate accompanied by a sore throat. The symptoms were severe enough to cause her distress during the day and interfere with sleep.

Subsequently, the patient underwent an extensive medical workup. Electrolytes were within normal limits, as were renal and liver functions. Anti-Sjögren syndrome A/B–anti–Sjögren syndrome B–anti–double-stranded DNA, C3 and C4 complement, and extractable nuclear antigen/anti-Smith/antibisphosphatase antibodies were all negative. Complete blood cell count with differential, iron level, and hemoglobin/hematocrit level were within normal limits. Co-occurrence of pain symptoms and somatoform disorders may mimic BMS symptoms, which potentially may have confounded her presentation. There may also exist some correlations between iron deficiency states and BMS, however, our patient’s iron level was normal.

To the best of our knowledge, there are no other reported cases of BMS secondary to quetiapine. Although our patient responded well to olanzapine as has been described previously, treatment options for BMS are limited and are largely based on cessation of the presumed offending agents. Further literature on psychopathological explanations for BMS, along with randomized controlled trials for potential therapeutics, would be welcomed contributions.

AUTHOR DISCLOSURE INFORMATION
The authors declare no conflicts of interest.

Received January 8, 2021; accepted after revision March 25, 2021.

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