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CASE REPORT

Low-dose aripiprazole for refractory burning mouth syndrome

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Abstract: We report a case of refractory burning mouth syndrome (BMS) ameliorated with low dose of aripiprazole. The patient was a 66-year-old female who had suffered from chronic burning pain in her tongue for 13 months. No abnormality associated with the burning sensation was detected in the laboratory tests and the oral findings. Considering the clinical feature and the history together, we diagnosed the burning sensation as BMS. The BMS pain was decreased by aripiprazole (powder) 1.0 mg/d, though no other antidepressants had satisfying pain relief. It could be supposed that the efficacy of aripiprazole is caused by dopamine stabilization in this case, and BMS might have a subtype that is reactive to aripiprazole. Further studies are needed to confirm the efficacy of aripiprazole for BMS.

Keywords: burning mouth syndrome, low-dose aripiprazole, chronic pain

Introduction

Burning mouth syndrome (BMS) is a burning or itching sensation in the normal oral mucosa. The pain is persistent, ranges from moderate to severe, and occurs particularly often in postmenopausal women. The International Association for the Study of Pain defines BMS, which is also called stomatodynia, glossodynia, oral dysesthesia, and persistent idiopathic orofacial pain, as "any form of burning or stinging sensation in the mouth in association with a normal mucosa in the absence of local or systemic disease".[1]

In the literature, antidepressants and benzodiazepines have shown beneficial effects in the treatment of patients with BMS.[2,3] However, there has been no report on treatment with a dopamine partial agonist for BMS. Herein, we report a case of treatment-resistant BMS successfully treated with low-dose aripiprazole. The Institutional Review Board of Tokyo Medical and Dental University approved this work. The patient provided written informed consent.

Case presentation

A 66-year-old female with no systemic disease or trauma history visited our clinic after being referred by her family physician. She complained of a chronic burning sensation in her tongue without taste alteration and dry mouth, which had lasted for 13 months.

The onset of the burning sensation was spontaneous. At first, she went to her family physician and was evaluated by laboratory screening tests. Hematological assessments of nutritional deficiencies and blood glucose levels were within normal limits. Though antibiotics, topical dexamethasone, and Kampo were administered at various clinics, the burning sensation was not ameliorated. Based on the medication effects, a candidal infection was excluded.

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At the first examination, she came to our clinic by herself. No orientation disturbance or any verbal fluency problem was observed. No abnormality associated with the burning sensation was observed in the oral findings. Her continuous pain developed as the day progressed and was improved by meals. Moreover, no mood disturbance was detected. Based on the clinical, laboratory, and anamnestic data, we established a diagnosis of BMS.

Because she was anxious about several side effects of antidepressants, she was started on escitalopram at 5 mg/d. However, the burning sensation was not reduced, even at 10 mg/d. Therefore, duloxetine was administered instead of escitalopram. Though duloxetine was initiated at 20 mg/d and gradually increased to 40 mg/d, her burning sensation was aggravated. Then, mirtazapine was administered instead of duloxetine. The burning sensation was partially reduced at 7.5 mg/d, but dose escalation was not effective. Therefore, low-dose aripiprazole powder (1.0 mg/d) was added to her treatment regimen. Two weeks later, she noticed an improvement in the burning sensation. A month later, she said that she almost always forgot about the burning sensation during the day. Six months later, the mirtazapine was stopped and there was no change for the worse. Two months later, the aripiprazole was also stopped, which resulted in recurrence of the burning sensation. Aripiprazole was restarted at 1.0 mg/d, and the burning sensation disappeared within a few days. With low-dose aripiprazole, she continued to live a healthy life without experiencing any side effect.

Discussion

We report a case of BMS that barely responded to antidepressants and was successfully treated with aripiprazole. Low-dose antidepressants, anticonvulsants, and benzodiazepines have been investigated and are the most accepted options for BMS. Tricyclic antidepressants are the most commonly prescribed drugs for BMS.[4] Additionally, it has been reported that serotonin–norepinephrine reuptake inhibitor[5,6] and selective serotonin reuptake inhibitor[7] are effective. However, some patients do not respond to any antidepressant. Recently, the effectiveness of atypical antipsychotics was reported in these patients.[8,9] To our knowledge, there is no report on the effectiveness of aripiprazole for BMS.

Aripiprazole is an atypical antipsychotic that bears the properties of D2 and 5-HT_{1A} partial agonists and is a potent antagonist of the 5-HT_{2A} receptor.[10] Unlike other atypical antipsychotics that are dopamine D2 antagonists, aripiprazole has a unique effect on the dopamine system. Because of its D2 partial agonist activity, some studies have mentioned that aripiprazole has a role as a dopamine system stabilizer. This suggests that a lower dosage of aripiprazole increases dopamine activity.

Recently, neurotransmitter positron emission tomography data indicated that hypofunction of the dopaminergic system within the basal ganglia was related to BMS.[11] One possible explanation is that the efficacy of aripiprazole in treating BMS is partly caused by stabilization of these dopamine receptors within the basal ganglia. Especially as this case did not have any response to a selective serotonin reuptake inhibitor (escitalopram) or a serotonin–norepinephrine reuptake inhibitor (duloxetine), it is strongly suggested that the efficacy of aripiprazole is caused by dopamine stabilization. Considering this dopaminergic hypothesis, other treatments including other antipsychotic drugs and electroconvulsive therapy might be options of treatment for BMS.

Recently, the role played by the dopamine system of the brain in pain control has garnered attention. Kasahara et al[12] reported four cases of refractory chronic pain that improved with a low dose of aripiprazole. In their paper, they hypothesized that dopamine plays a role in pain processing via μ opioids. It is also believed that the dopamine and opioid systems interact in complex ways in the treatment of chronic pain.

Conclusion

This report suggests that low-dose aripiprazole may be effective for patients with treatment-resistant BMS. Further studies are needed to confirm the efficacy of aripiprazole for BMS.

Disclosure

The authors report no conflicts of interest in this work.

References

1. Merskey H, Bogduk N. *Classification of Chronic Pain*. 2nd ed. Seattle: IASP press. 1994.
2. Spanemberg JC, Rodríguez de Rivera Campillo E, Salas EJ, López López J. Burning mouth syndrome: update. *Oral Health Dent Manag*. 2014;13(2):418–424.
3. Zakrzewska JM, Forssell H, Glenny AM. Interventions for the treatment of burning mouth syndrome. *Cochrane Database Syst Rev*. 2005;1: CD002779.
4. López-Jornet P, Camacho-Alonso F, Leon-Espinosa S. Efficacy of alpha lipoic acid in burning mouth syndrome: a randomized, placebo-treatment study. *J Oral Rehabil*. 2009;36(1):52–57.
5. Kato Y, Sato T, Katagiri A, et al. Milnacipran dose-effect study in patients with burning mouth syndrome. *Clin Neuropharmacol*. 2011;34(4): 166–169.
6. Mignogna MD, Adamo D, Schiavone V, Ravel MG, Fortuna G. Burning mouth syndrome responsive to duloxetine: a case report. *Pain Med*. 2011; 12(3):466–469.

7. Yamazaki Y, Hata H, Kitamori S, Onodera M, Kitagawa Y. An open-label, noncomparative, dose escalation pilot study of the effect of paroxetine in treatment of burning mouth syndrome. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*. 2009;107(1):e6–e11.
8. Ueda N, Kodama Y, Hori H, et al. Two cases of burning mouth syndrome treated with olanzapine. *Psychiatry Clin Neurosci*. 2008;62(3):359–361.
9. Rodriguez-Cerdeira C, Sanchez-Blanco E. Treatment of burning mouth syndrome with amisulpride. *J Clin Med Res*. 2012;4(3):167–171.
10. Chen T-Y, Tzeng N-S. Aripiprazole: a dopamine modulator that mimics methylphenidate in producing faster antidepressant effects. *Med Hypotheses*. 2013;81(2):183–185.
11. Jääskeläinen SK. Pathophysiology of primary burning mouth syndrome. *Clin Neurophysiol*. 2012;123(1):71–77.
12. Kasahara S, Kunii Y, Mashiko H, Otani K, Konno S, Niwa S. Four cases of chronic pain that improved dramatically following low-dose aripiprazole administration. *Prim Care Companion CNS Disord*. 2011;13(2):PCC.10l01078.

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