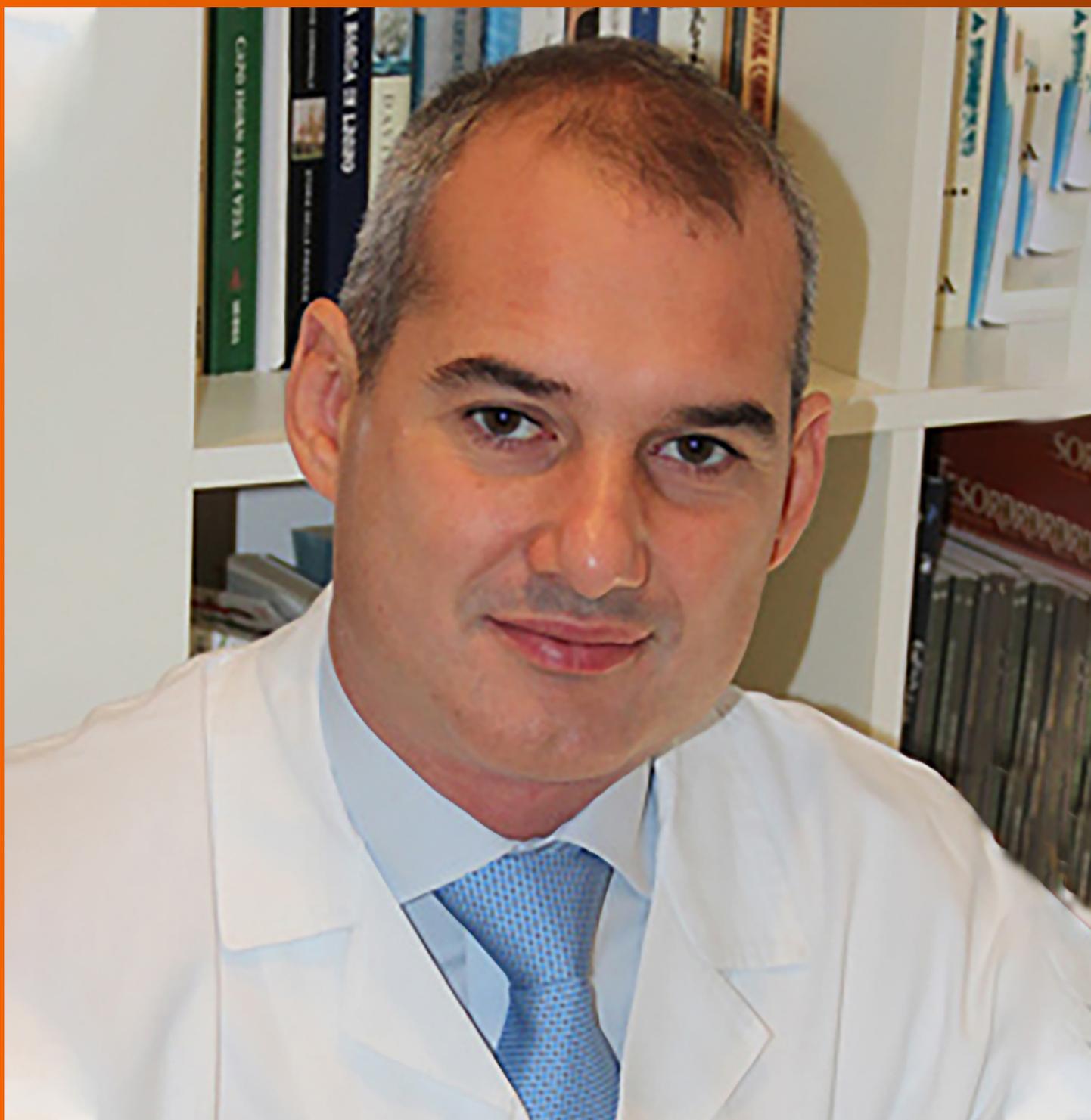


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**RESPONSIBLE EDITORS FOR THIS ISSUE**

Production Editor: *Jia-Hui Li*; Production Department Director: *Xiang Li*; Editorial Office Director: *Jin-Lei Wang*.

**NAME OF JOURNAL**

*World Journal of Clinical Cases*

**ISSN**

ISSN 2307-8960 (online)

**LAUNCH DATE**

April 16, 2013

**FREQUENCY**

Thrice Monthly

**EDITORS-IN-CHIEF**

Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

**EDITORIAL BOARD MEMBERS**

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

**PUBLICATION DATE**

August 16, 2021

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<https://www.wjgnet.com/bpg/gerinfo/208>

**ARTICLE PROCESSING CHARGE**

<https://www.wjgnet.com/bpg/gerinfo/242>

**STEPS FOR SUBMITTING MANUSCRIPTS**

<https://www.wjgnet.com/bpg/GerInfo/239>

**ONLINE SUBMISSION**

<https://www.f6publishing.com>

## Dopamine agonist responsive burning mouth syndrome: Report of eight cases

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**Author contributions:** Du QC and Wang WF were responsible for the data collection; Du QC and Ge YY drafted the manuscript and interpreted the results; Wang WF and Xiao WL designed the study and were the project supervisors; all authors reviewed and approved the final manuscript.

### Informed consent statement:

Written informed consent was obtained from the patients for the publication of this report and the accompanying images.

**Conflict-of-interest statement:** The authors declare no conflicts of interest for this manuscript.

### CARE Checklist (2016) statement:

The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

**Open-Access:** This article is an open-access article that was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution

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## Abstract

### BACKGROUND

Burning mouth syndrome (BMS) is characterized by burning sensation of the oral mucosa. There is a lack of effective treatment. In recent years, a special subtype of BMS has been reported, in which oral burning sensation is alleviated after chewing, speaking, or dopaminergic drug delivery. Currently, there are few reports about the subtype of BMS in China. This study was a retrospective analysis of the clinical data of BMS patients sensitive to dopamine agonist at our hospital, aiming to improve the recognition on this disease.

### CASE SUMMARY

Eight patients diagnosed with dopamine agonist responsive BMS at the Liaocheng People's Hospital from January 1, 2017 to June 30, 2020 were recruited. The clinical manifestations, treatment, and prognosis were retrospectively analyzed. There were three male and five females in the eight patients. The median age was 56 years (range, 46-65 years). All the eight patients showed burning pain in the mouth. The symptoms were mild in the morning and severe in the evening, and alleviated after chewing, talking, and other oral activities. Four patients were accompanied by restless legs syndrome (RLS). Family history of RLS was positive in two patients. All patients were treated with pramipexol, and symptoms were basically relieved after 2-8 wk.

### CONCLUSION

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**Manuscript source:** Unsolicited manuscript

**Specialty type:** Dentistry, oral surgery and medicine

**Country/Territory of origin:** China

**Peer-review report's scientific quality classification**

Grade A (Excellent): 0  
Grade B (Very good): 0  
Grade C (Good): C  
Grade D (Fair): 0  
Grade E (Poor): 0

**Received:** April 22, 2021

**Peer-review started:** April 22, 2021

**First decision:** May 24, 2021

**Revised:** June 4, 2021

**Accepted:** June 15, 2021

**Article in press:** June 15, 2021

**Published online:** August 16, 2021

**P-Reviewer:** Jeong SH

**S-Editor:** Ma YJ

**L-Editor:** Wang TQ

**P-Editor:** Li JH



Dopamine agonist responsive BMS is a special subtype of BMS, which is alleviated after oral activities. Dopamine receptor agonist is an effective treatment.

**Key Words:** Burning mouth syndrome; Restless legs syndrome; Dopamine receptor agonists; Chinese; Case report

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**Core Tip:** Dopamine agonist responsive burning mouth syndrome (BMS) is a special subtype of BMS. We retrospectively summarized the clinical data of eight patients with a diagnosis of dopamine agonist responsive BMS in China to improve the recognition on this disease. All patients showed burning pain in the mouth, which was mild in the morning and severe in the evening, and alleviated after chewing, talking, and other oral activities. Four patients were accompanied by restless legs syndrome (RLS). Family history of RLS was positive in two patients. All patients were treated with pramipexol, and symptoms were basically relieved after 2-8 wk.

**Citation:** Du QC, Ge YY, Xiao WL, Wang WF. Dopamine agonist responsive burning mouth syndrome: Report of eight cases. *World J Clin Cases* 2021; 9(23): 6916-6921

**URL:** <https://www.wjgnet.com/2307-8960/full/v9/i23/6916.htm>

**DOI:** <https://dx.doi.org/10.12998/wjcc.v9.i23.6916>

## INTRODUCTION

Burning mouth syndrome (BMS) is characterized by burning sensation of the oral mucosa in the absence of clinically apparent mucosal alterations, and it mainly affects the tongue[1]. According to recent studies, the prevalence of BMS ranges from 1.0% to 3.7%, and it is more common in women, with the highest prevalence in postmenopausal women[2]. BMS causes great harm to the physical and mental health of patients; however, there is a lack of rapid and effective treatment due to the unclear pathogenesis[3].

In recent years, a special subtype of BMS has been reported, in which oral burning is mild in the morning and severe in the evening, and alleviated or even relieved after chewing, speaking, and other oral activities[4]. Some patients also suffer from restless legs syndrome (RLS), and are sensitive to dopaminergic drugs[5]. The researchers concluded that the subtype of BMS was a variant of RLS after summarizing its characteristics and therapeutic effects[6]. Jung *et al*[7] reported that an elderly male patient without RLS only had oral discomfort, which was relieved after movement of the jaw and tongue, and treatment with pramipexole was effective, suggesting that the special subtype of BMS could exist independently of RLS. Currently, there are few reports about the subtype of BMS with a good response to dopaminergic drugs in China. This study was a retrospective analysis of the clinical data of BMS patients sensitive to dopamine receptor agonist at the Liaocheng People's Hospital, aiming to improve the recognition and prognosis of this disease.

## CASE PRESENTATION

### Chief complaints

Eight patients with BMS sensitive to dopamine receptor agonists were recruited from the Department of Stomatology or Department of Neurology of the Liaocheng People's Hospital from January 1, 2017 to June 30, 2020. All the patients met the following two standards: (1) Conformed to International Classification of Headache Disorders 3rd edition (ICHD-3) in the diagnosis of BMS standard[8], as follows: The pain form was burning and located in the mucosal surface; the duration of the disease was longer than 3 mo, and the pain lasted more than 2 h per day; there were no clinically apparent mucosal alterations; and the diagnosis of other diseases in the ICHD-3 classification was not met; and (2) Oral discomfort was mild in the morning and severe in the

evening, and alleviated when chewing or talking. Dopamine receptor agonist treatment was effective for reducing oral discomfort symptom.

### **History of present illness**

There were eight patients with BMSe sensitive to dopamine receptor agonists, including three males and five females, aged 46-65 years with a median age of 56 years. The course of disease was 12-42 mo. The involved sites were as follows: Anterior 2/3 of the tongue (8/8), hard palate (7/8), lip (5/8), buccal mucosa (4/8), soft palate (3/8), and larynx (1/8) (Table 1). All patients showed burning sensation in the oral cavity, seven (7/8) had accompanied dry mouth symptom, four (4/8) had accompanied numbness in the oral mucosa, two (2/8) had accompanied ant walking sensation in the oral cavity, and one (1/8) had accompanied taste disturbance (Table 1). The symptoms were mild in the morning and severe in the evening, and alleviated when chewing, speaking, and other oral activities.

### **History of past illness**

Four patients (cases 1, 2, 5, and 8) had accompanied restless legs syndrome, and all of them were female (Table 1). There was no history of iron deficiency, renal insufficiency, diabetes, thyroid disease, rheumatic immune system disease, *etc.* The patients also had no history of Parkinson's disease, multiple sclerosis, brain stem infarction, or other central nervous system diseases.

### **Personal and family history**

Two patients (cases 3 and 5) had a family history of RLS, and one patient (cases 6) had a family history of BMS (Table 1).

### **Physical examination**

General physical and neurological examinations were unremarkable. The intraoral inspection was normal.

### **Laboratory examinations**

Laboratory investigations, including routine blood tests, vitamins, iron, ferritin, creatinine, and thyroid hormones, revealed no abnormality.

### **Imaging examinations**

Periapical and panoramic X-ray examinations were performed without alterations. Magnetic resonance examinations of the brain were unremarkable.

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## **FINAL DIAGNOSIS**

The final diagnosis was a subtype of dopamine agonist responsive BMS based on clinical analysis.

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## **TREATMENT**

All the patients were treated with pramipexole hydrochloride tablets, which were taken 2-3 h before sleeping every day. The initial dose was 0.125 mg/d, adding up following the sequence of 0.25 mg/d, 0.375 mg/d, and 0.5 mg/d according to the improvement of the patient's symptoms until the maintenance dose of controlled symptoms was achieved.

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## **OUTCOME AND FOLLOW-UP**

The Visual Analog Scale (VAS) was adopted to quantify the pain degree of patients. The grading standard was[9]: 0 points, painless; < 3 points, mild pain, tolerable; 4-6 points, pain resulting in sleep disturbance, tolerable; and 7-10 points, gradually intense excruciating pain that interferes with appetite and sleep.

The criteria for efficacy evaluation were as follows[10]: Efficacy index = (VAS score before treatment - VAS score after treatment)/VAS score before treatment × 100%. Significant effect was regarded when pain was significantly relieved, efficacy index

**Table 1 Clinical data of eight patients with dopamine agonist responsive burning mouth syndrome**

Patient	Sex	Age (yr)	Duration of BMS (mo)	Involved sites	Clinical manifestation	VAS score	Concurrent RLS	Family history of BMS or RLS	Maintenance dose of pramipexole	Time required to achieve significant effect
1	F	53	24	Anterior 2/3 of the tongue, hard palate, lip	Oral burning pain, ant walking sensation, dry mouth symptom	6	Y	N	0.25 mg/d	2 wk
2	F	58	30	Anterior 2/3 of the tongue, hard palate, buccal mucosa	Oral burning pain, numbness, dry mouth symptom	8	Y	N	0.375 mg/d	4 wk
3	M	49	12	Anterior 2/3 of the tongue, hard palate, lip	Oral burning pain, dry mouth symptom	7	N	One brother with RLS	0.25 mg/d	4 wk
4	M	65	36	Anterior 2/3 of the tongue, hard palate, soft palate, buccal mucosa	Oral burning pain, ant walking sensation, dry mouth symptom, taste disturbance	8	N	N	0.5 mg/d	5 wk
5	F	62	42	Anterior 2/3 of the tongue, hard palate, soft palate, lip, buccal mucosa, larynx	Oral burning pain, numbness, dry mouth symptom	10	Y	One brother with RLS	0.5 mg/d	8 wk
6	M	46	24	Anterior 2/3 of the tongue, hard palate, lip	Oral burning pain, numbness	7	N	One sister with BMS	0.25 mg/d	4 wk
7	F	55	30	Anterior 2/3 of the tongue, hard palate, soft palate, buccal mucosa	Oral burning pain, dry mouth symptom	9	N	N	0.375 mg/d	6 wk
8	F	57	12	Anterior 2/3 of the tongue, lip, buccal mucosa	Oral burning pain, numbness, dry mouth symptom	7	Y	N	0.25 mg/d	3 wk

BMS: Burning mouth syndrome; RLS: Restless legs syndrome; VAS: Visual analog scale; F: Female; M: Male; Y: Yes; N: No.

was > 70%, and VAS score was  $\leq 3$  points. Effectiveness was regarded when the pain degree was reduced and efficacy index was  $\geq 30\%$  but  $\leq 70\%$ . No effect was regarded when there was no significant improvement in pain and efficacy index was  $< 30\%$ .

The VAS scores of eight patients ranged from 6 to 10 points. After 2-8 wk of dopamine receptor agonist dose titration, all patients reached the standard of significant effect, with significant reduction or even absence of burning pain, and the maintenance dose of pramipexole ranged from 0.25 mg to 0.5 mg per day (Table 1).

Upon withdrawal of pramipexol, the symptoms of one patient (case 1) relapsed around 7 d after. Since then, she continued to take pramipexol (0.25 mg) 2-3 h before sleeping every day, with total control of the burning sensation. All other patients continued to take the maintenance dose of pramipexole and had no recurrence of symptoms in the follow-up period. There were no significant tolerability issues. All patients did not report side effects.

## DISCUSSION

BMS is a chronic disorder characterized by burning pain in the mouth in the absence of any visible oral or medical cause. Most patients with BMS are accompanied by anxiety, depression, and other mental disorders[11], which seriously affect the sleep and quality of life[12]. In recent years, the incidence of BMS has been increasing year by year, however, due to the complex etiology and pathogenesis, there is still no specific and effective treatment[3].

Prakash *et al*[6] reported five patients with BMS in whom levodopa was effective, of whom four suffered from RLS at the same time, and the other one patient's brother suffered from RLS. They found that the clinical symptoms of these patients were mild in the morning and severe in the evening, which were alleviated or even relieved after

chewing, speaking, and other oral activities. These characteristics are very similar to those of RLS, so they suggested that BMS with RLS or family history of RLS may be a variant of RLS, and dopaminergic drugs should be tried for this subtype of BMS. Jung *et al*[7] named this special subtype of BMS “restless mouth syndrome” for the first time. Up to now, there has been no report about dopamine responsive BMS in China. This study retrospectively analyzed the clinical data of eight patients with BMS who were sensitive to dopamine receptor agonist. The results showed that this subtype of BMS was more common in middle-aged and elderly women, and some patients could be accompanied by RLS. In this study, four patients with RLS were female, which suggested that female patients with dopamine agonist responsive BMS were more likely to be complicated with RLS. However, the significance of such obvious gender difference still needs to be further analyzed by expanding the sample size. Dopamine agonist responsive BMS usually involves the anterior 2/3 of the tongue, hard palate, lips, buccal mucosa, soft palate, and even the throat, which is consistent with the traditional BMS[13].

BMS that is sensitive to dopaminergic drugs has the clinical characteristics of both BMS and RLS. The underlying reason is that BMS and RLS share some of the same pathogenesis. In recent years, a number of studies have shown that a variety of oral discomfort in patients with BMS is related to central nervous system lesions, among which the deficiency of dopamine inhibition function plays a prominent role[14-16]. Bilateral responses at the end of blink reflex are absent in 25% to 36% of BMS patients, which is similar to Parkinson's patients[17,18]. In addition, positron emission computed tomography showed that the content of dopamine transmitter in presynaptic nerve endings and the function of postsynaptic D2 receptor decreased in patients with BMS[19]. The results of these two studies suggest that the loss of striatal dopamine system inhibition is related to the occurrence of BMS. Similarly, a large number of studies have shown that dopamine neurotransmitter reduction or dopamine receptor dysfunction play a key role in RLS[20,21]. The most powerful evidence is that patients with RLS have improved symptoms after taking a low dose of levodopa or dopamine receptor agonist[22]. Therefore, abnormal dopaminergic system plays an important role in the occurrence and development of both BMS and RLS.

Currently, there are still no specific and effective therapeutic methods for BMS. Some drugs with neuroprotective effect and acting on related analgesic targets can partially relieve BMS symptoms, but the efficacy is not satisfactory[23]. Based on the treatment guidelines for RLS and the treatment experience reported in previous studies[24], all patients in this study were treated with low-dose of pramipexole hydrochloride tablets, which were taken 2-3 h before sleeping every day, and gradually increased to the maintenance dose that could control the symptoms. All patients were basically in remission at 2-8 wk, suggesting that this treatment regimen had a significant effect, which was obviously different from traditional BMS.

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## CONCLUSION

In conclusion, dopamine agonist responsive BMS is not only a special subtype of BMS, but also a variant of RLS. It is mainly characterized by burning sensation in the mouth, mild in the morning and severe in the evening, alleviating or even relieving after chewing, speaking, and other oral activities, and is sensitive to dopamine receptor agonist. Improving the understanding of this subtype of BMS sensitive to dopamine agonist is helpful to give early appropriate treatment and improve the prognosis of patients.

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## ACKNOWLEDGEMENTS

We thank the patients and their family for their support.

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