Case Report

A case of migratory stomatitis in a young male patient: Management and differential diagnosis

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Abstract

Migratory stomatitis (MS) is an uncommon inflammatory condition with unclarified etiology, which despite its benign nature, may raise concerns for patients and diagnostic difficulties for professionals. This case report aims to describe the clinical features of a patient who presented with MS in conjunction with benign migratory glossitis (BMG), and its diagnostic process and management. The patient, a 25-year-old man, sought diagnosis of an oral condition, with cyclic behavior, which had been causing him great discomfort for a year. The patient presented erythematous patches on his lower lips and right side of the buccal mucosa, surrounded by a slightly elevated halo with a concomitant classical picture of BMG. After analysis of his entire symptomatology, the diagnosis of MS associated with BMG was concluded. The patient received clear explanations and symptomatic treatment. The diagnosis of MS may be challenging, even to oral medicine practitioners, especially if it occurs alone. MS with concurrent manifestation of BMG may make the conditions easier to diagnose, but it does not exclude the need to apply a complete process of differential diagnosis to rule out other similar possibilities.

Keywords: Benign migratory glossitis, erythema chronicum migrans, geographic tongue, hypersensitivity, stomatitis

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INTRODUCTION

A wide range of inflammatory conditions, resulting from either local or systemic origins, may affect oral mucosa. Some of them may pose great difficulties to establish their diagnosis, while others may be clarified on a clinical basis only. Benign migratory glossitis (BMG) can be included in the latter category. Although relatively common, its etiology remains unknown and several patients still seek professional assistance when faced with the management of this condition.

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BMG, also known as geographic tongue, is a condition that usually affects the anterior two-third of the tongue dorsum, and often occurs simultaneously with fissured tongue (FT).[1,2] It received this name due to its characteristic of appearing in one place and migrating to another in a period of hours or weeks.[3] Clinically, the condition is characterized by irregular erythematous areas associated with atrophy of filiform papillae and sub-epithelial inflammation, surrounded by a white rim with an annular or circinate aspect that corresponds to hyperkeratosis and epithelial acanthosis.

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Most patients do not report any symptoms,[4] while others may complain of burning, dysgeusia or foreign-body feeling.[5]

A condition with characteristics similar to those of BMG may affect other sites of the oral mucosa, and may be described as erythema migrans or migratory stomatitis (MS), the etiopathogenesis of which is also unclear, but this condition is much rarer in comparison with BMG. MS may involve any site of the oral mucosa but more frequently affects the labial, buccal and soft palate mucosa tissues.

The diagnosis is based on clinical findings, along with the patient's report of multiple lesions in the mouth, varying in size and shape, which migrate from time to time. MS may be a challenging condition to diagnose, since the differential diagnosis may be composed of several pathological conditions, when professionals are faced with a clinical condition manifesting multiple irregular erythematous areas throughout the oral mucosa, such as candidiasis, psoriasis, Reiter's syndrome, lichen planus, heterogeneous leukoplakia, lupus erythematosus, syphilis, herpes simplex virus infection and drug reactions.[2]

The present case report aimed to describe the clinical, diagnostic and therapeutic characteristics of a patient manifesting MS in conjunction with BMG, who was treated at our Department of Oral Diagnosis, and to discuss the management strategies applied to such cases.

CASE REPORT

The patient, a 25-year-old man, sought assistance at the Oral Diagnosis Clinic, Stomatology Department of Sao Paulo University, complaining of a burning sensation in his lips and buccal mucosa. He reported that the symptoms had started a year ago with lesions in his lower labial mucosa, with a cyclic behavior, lasting a week or two, and after a few days, arising in a different area of the lower lip. In the last 6 months, his buccal mucosa, mainly on the right side, was also affected by similar lesions. These bouts presented symptoms of variable degrees ranging from mild discomfort to very annoying burning sensations.

The patient had previously consulted some health professionals and used several therapeutic products: Antiseptics, lanolin, topical corticosteroids and other anti-inflammatory drugs, without clarification of the diagnosis or adequate control of the symptomatology.

Intraoral examination revealed a reddish area on the right side of his lower lip, circumscribed by a slightly elevated red halo and a well-circumscribed reddish area with no elevated borders in the right buccal mucosa close to the upper fornix, as shown in Figure 1.

The patient also presented a classical condition of BMG that was completely asymptomatic and of which he was already aware. Figure 2 depicts two different occasions with an interval of 15 days between them, showing the characteristic mutational behavior of BMG. A mild, fissured tongue associated with the BMG condition was also noted.

The patient's medical history revealed multiple episodes of allergic dermatitis and a diagnosis of atopy. According to the patient, his deceased father was also a very allergic person.

Based on the clinical hypothesis of MS associated with BMG and the high level of discomfort reported by the patient, a corticosteroid mouthwash (0.05% clobetasol propionate) was prescribed for 2 weeks. Fourteen days after the first appointment, the symptomatology had subsided, and so had the clinical signs of both MS and BMG conditions, as shown in Figure 3.

The patient was shown to be a very anxious person and very concerned about his oral condition. He had a large number of self-taken pictures of his mouth showing the various aspects of his disease since its first occurrence (due to their poor quality the pictures have not been displayed) and had tried several over-the-counter medications before seeking assistance at our Institution. Once the diagnosis had been established and the patient had been provided with due clarification of the features of MS, he returned for a follow-up appointment 2 months later with no complaints and said he required no further consultations.

DISCUSSION

Both BMG and MS are considered inflammatory conditions of unknown etiology that present microscopic features consistent with psoriasiform dermatitis. MS mainly affects the labial and buccal mucosa, but any site can be involved.[6] Campana *et al*.,[7] found a large number of reports with palatal involvement – nearly 30%. There is also a frequent association of BMG with fissured tongue,[3] as occurred in our case.

According to the latest reports in the literature,[8,9] the prevalence of BMG among the general population is around 2%, with no gender predilection and it most commonly affects children and young adults.[9] This has allowed the assumption that the majority of cases of

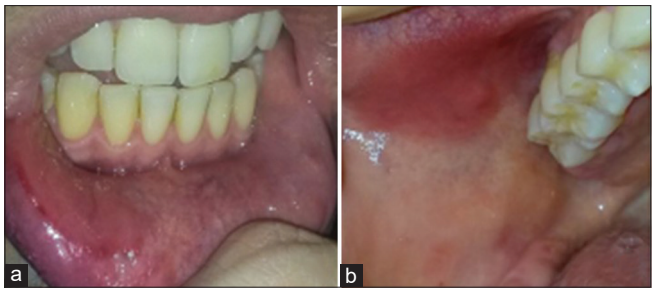

Figure 1: Erythematous lesions surrounded by elevated halo in labial mucosa (a) and well-circumscribed reddish area in right buccal mucosa (b)

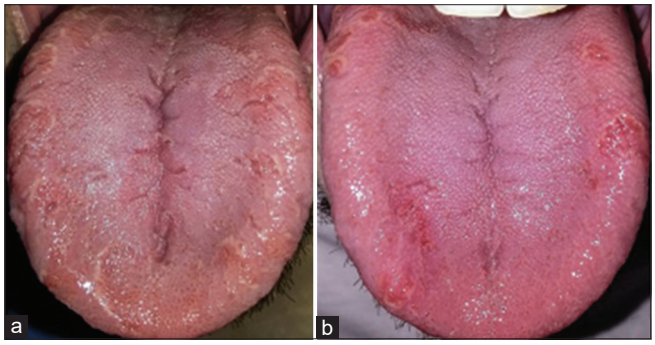

Figure 2: Benign migratory glossitis associated with fissured tongue showing areas of atrophic filiform papillae surrounded by whitish halos especially at the borders of the tongue dorsum (a) and the same site 15 days later showing a distinct pattern (b)

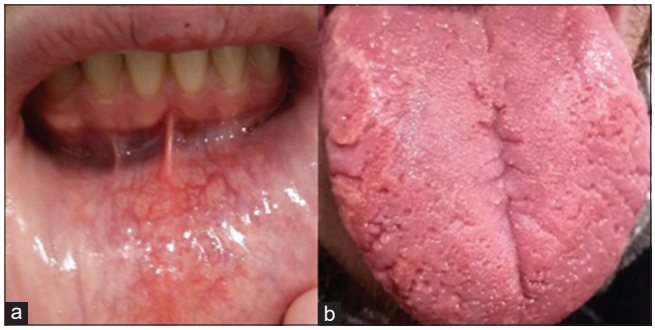

Figure 3: (a) Clinical appearance of migratory stomatitis and (b) benign migratory glossitis after 2-month follow-up demonstrating the cyclic nature of these conditions

BMG are diagnosed in young individuals, as soon as the patients themselves or their parents perceive the condition, thus explaining the epidemiological data. Adults usually seek assistance with the condition of BMG, only when it becomes symptomatic. MS is a much rarer occurrence although it is possible for misdiagnosis and underdiagnosis to take place. Campana *et al*.[7] identified only 64 cases of MS reported in the literature, 24 without association with BMG, in addition to two further reports with palatal involvement.

MS with absence of tongue involvement – hopefully rarer – may be more difficult to diagnose than cases such as the one presented here, in which the classical condition of BMG was present simultaneously. Nevertheless, even if associated with BMG, a careful investigation must be conducted to conclude a diagnosis of MS and rule out the presence of a different concomitant condition.[10]

Fungal infection caused by *Candida albicans*, denominated as oral candidiasis, when affecting the dorsum of the tongue is classically represented by rhomboid median glossitis and the clinical condition of our case at this site was typical of a BMG. The erythematous lesions on the lips and buccal mucosa could suggest candidiasis in cases of severe xerostomia or immunosuppression, which were not the case of our patient.

Psoriasis and Reiter's syndrome (reactive arthritis) were ruled out due to the absence of skin lesions, or oculo-genital lesions and arthritis, respectively.

Lichen planus was equally ruled out due to the absence of symmetrical, white striated mucosal lesions and short duration of the episodes, highlighted as mainly clinical features of this condition.

Heterogeneous leukoplakia was also not the case, since there was no hyperkeratotic reaction associated with the history of periodic recurrences and multiple migratory no white lesions. Erythroplakia would fit better if isolated lesions were present, but this diagnosis was also discarded in view of the history of multiple recurrent lesions.

Lupus erythematosus may produce oral mucosa lesions like lichen planus but is invariably accompanied by cutaneous lesions, photodermatitis and in the case of systemic disease, general symptomatology.

Syphilis may produce lesions such as those seen in the lips and buccal mucosa of our patient, but this disease does not behave clinically by presenting migratory lesions with such short duration.

Oral herpes infections – simplex or zoster – manifest in the mouth through a cluster of multiple tiny round ulcerations, preceded by vesicles, preferably in the attached gingiva or the hard palate in the case of herpes simplex, or following a nerve sheath path, unilaterally, in the case of zoster.

Drug or hypersensitivity reactions may resemble the clinical condition presented by our patient; however, these conditions are characterized by an acute flare-up, frequently preceded by bullae that produce pain and discomfort for a week or two. Although these reactions may recur if the

patient is exposed to antigens repeatedly, the lesions usually affect the same sites, adding others as recurrences take place. This did not match with the condition presented by our patient.

As previously mentioned, the presence of a classical condition of BMG and the detailed history reported by the patient made the diagnosis easier and more reliable.

MS as an isolated manifestation may make it more difficult to reach the final diagnosis and a biopsy procedure could be performed to rule out other possibilities of diseases such as some of those mentioned earlier on in this discussion. In the case reported here, our team decided to postpone the biopsy procedure for a while to allow a period of clinical follow-up, since MS was included as a main clinical diagnosis. The maneuver proved to be very useful to support the diagnosis and helped to reassure the patient and gain his adherence to a regular follow-up regimen.

Atopy has been included as a condition associated with BMG and MS, in addition to psoriasis and Reiter's syndrome.[6,11] Santos Netto *et al*.[12] suggested a classification of BMG and MS based on the distribution of lesions and medical history: Type 1– BMG; 2– MS without BMG; 3– MS with BMG; 4– BMG with cutaneous disease; 5– MS without BMG with cutaneous disease and 6– MS with BMG with cutaneous disease. Our patient reported atopy and would be classified as Type 6 in the classification proposed by Santos Netto.

The therapeutic management of symptomatic presentations of BMG or MS remains an issue of debate and there is no consensus in the literature. Several strategies such as the use of topical corticosteroids, local anesthetics, antihistamines, anxiolytic and anti-inflammatory agents[6,13] have been used up to the present time, most of them not supported by any adequate controlled studies. Our patient was given a corticosteroid mouthwash (0.05% propionate of clobetasol) for 2 weeks, which proved to be efficient to improve his symptomatology, but we do believe that providing the patient with proper information on the characteristics and behavior of the condition produced a more positive outcome in the patient than the medication itself.

CONCLUSION

BMG and MS are benign mucosal lesions with an etiology that is still undefined, which lesions can appear alone or simultaneously and generate discomfort to patients in some cases. If MS occurs alone, however, diagnosis may be a challenge even to oral medicine practitioners. MS with concurrent manifestation of a condition of BMG may make the conditions easier to diagnose, but it does not exclude the need to apply a complete process of differential diagnosis to rule out other similar possibilities.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understand that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Ethical approval

This report was exempted from requiring ethical approval because medical data records of the patient were used, without exposing his identity or disclosing photographs that identified his facial or other characteristics. We are including the Portuguese version of a document entitled "Patient Release Form" that consists of detailed explanations given to the patient, about further publication and about the Journal. This form was signed by the patient and represents his agreement with its content.

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Conflicts of interest

There are no conflicts of interest.

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