



Hereditary Benign Migratory Glossitis in a Young Child: A Case Report

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Abstract

Aim: To present a rare case of hereditary benign migratory glossitis.

Background: The clinical presentation of the lesion may mimic other mucosal conditions, allergic reactions, deficiency states.

Case Description: A four year old child presented with pain in teeth and well circumscribed irregular areas of denudation on dorsal surface of tongue. Thorough history revealed similar asymptomatic condition in tongue of father and sibling of child. Other oral conditions were ruled out and diagnosed as hereditary benign migratory glossitis.

Conclusion and Clinical Significance: The pediatric dentist may come across such cases and should be aware of its clinical presentation, evaluation and management so as to prevent unnecessary investigations and medications.

Keywords: Geographic Tongue; Benign Migratory Glossitis; Hereditary Benign Migratory Glossitis

Introduction

Benign Migratory Glossitis (BMG), an inflammatory condition of the tongue [1] is also referred as wandering rash of tongue, geographic tongue, lingua geographica, erythema migrans, transitory benign plaques of the tongue, glossitis areata migrans, marginal exfoliative glossitis, pityriasis linguae and exfoliation areata lingual [2,3]. The term migratory is due to the change in site and its irregular presentation. The presence of concurrent epithelial desquamation at one site and proliferation at another is attributed to its clinical presentation [4].

The etiology of BMG is still an enigma. It is considered to be a congenital anomaly by some, while others believe it to be an

inflammatory reaction [2]. Various systemic conditions have also been associated with BMG, such as Reiter's syndrome, down's syndrome, psoriasis [5], anemia, diabetes, nutritional disturbances, lichen planus, candidiasis, hormonal imbalance [3], psychological upsets [5] and allergies [3]. The occurrence of BMG in parents and sibling combinations indicates polygenic mode of inheritance with increased tissue type HLA-B15 [6,7]. There are very few reports of hereditary BMG from the Indian subcontinent, particularly in children.

Case Report

A 4 year old boy reported to the department of Pedodontics and Preventive dentistry, with a chief complaint of pain in lower right

posterior tooth region of jaw since 3 days. The patient’s medical history was non-contributory. There was a history of dental trauma to upper anterior tooth one year ago, which did not present any symptoms. Intraoral examination revealed a non vital teeth with abscess w.r.t primary mandibular right second molar, dentinal caries with primary maxillary right first molar, primary mandibular left second molar and discolored right primary maxillary incisor. The dorsum surface of the tongue showed multiple, well-circumscribed areas devoid of filiform papillae, forming an irregular pattern on tongue (Figure 1). It was asymptomatic and not associated with any burning sensation. The patient did not give history of allergy to any medications or environmental factors. Hemogram was advised and the child was referred to a nutritionist. The nutritional status was found to be satisfactory. Following clinical examination and radiographic investigations a final diagnosis of dentoalveolar abscess was made w.r.t primary mandibular right second molar and Ellis class 9 fracture w.r.t primary maxillary right central incisor was arrived at. Pulpectomy w.r.t primary maxillary right central incisor and primary mandibular right second molar were done and semi-permanent restorations were given.

the change in the area, size and shape of presentation of the lesion during successive dental treatment a working diagnosis of benign migratory glossitis was arrived at. The parent was enquired about the similar condition in the family, which revealed a positive family history of the lesion in father (Figure 2) as well as elder sibling (Figure 3). The lesion in the father and the sibling were in the remission stage. The patient was followed up periodically over a period of six months (Figure 4).

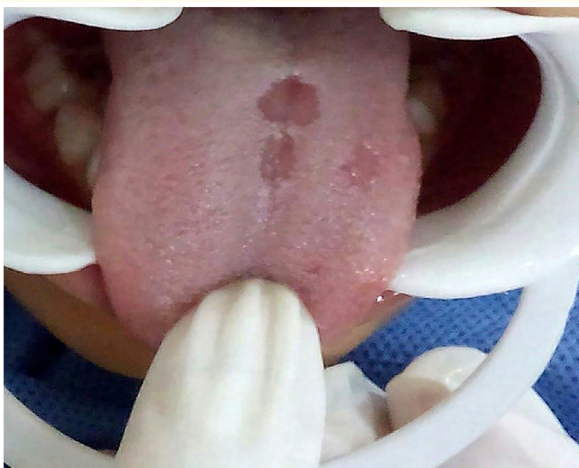


Figure 1: Clinical presentation of tongue of child at the first visit.

Follow up at 1 week showed, resolution of tongue lesion. However there was proliferation and presence of desquamation at another site. The lateral aspects of anterior one third of tongue showed an area of mild erythema and depapillation. Exfoliative cytology was performed to know the nature of the lesion. It showed absence of candida on Periodic Acid Schiff staining. Thus, due to

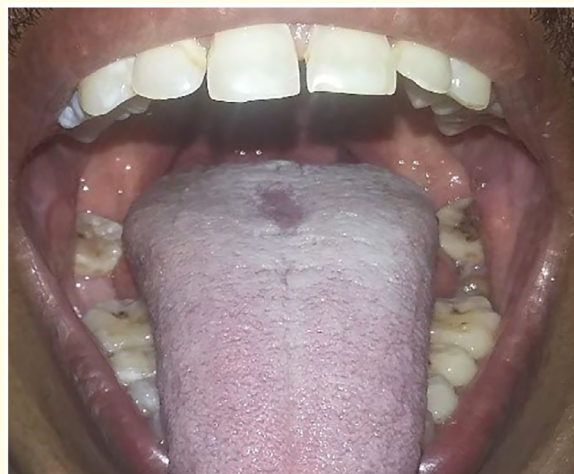


Figure 2: Lesion on the tongue of Father.

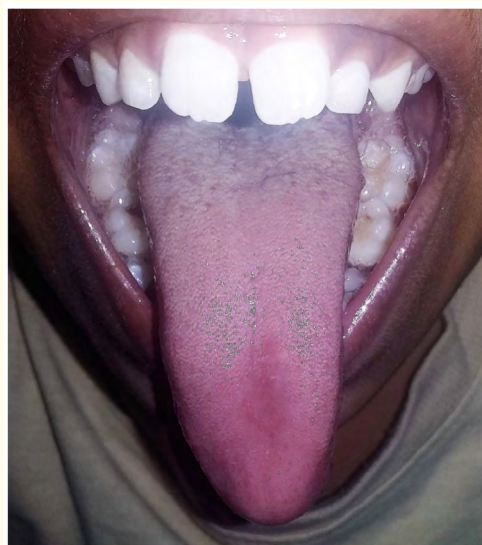


Figure 3: Lesion on the tongue of the Sibling.

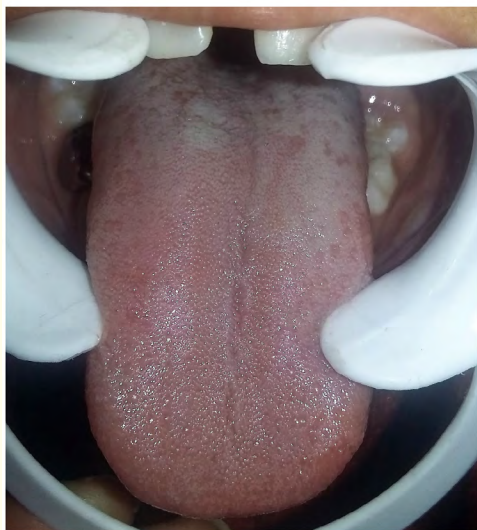


Figure 4: Lesion on the tongue of child at 6 months recall.

Discussion

Benign migratory glossitis or geographic tongue is a common disorder of unknown etiology. The lesion is known to occur on the tip, lateral borders, dorsum of the tongue, and sometimes extends to the ventral portion of tongue [3,5]. When the lesions are seen on other oral mucosal sites such as buccal mucosa, soft palate and floor of mouth the condition is known as ectopic geographic tongue.

The prevalence of BMG varies and generally is low [3]. Prevalence of 14% has been reported in Israeli children between ages 0 to 2 years [2]. A study on Indian population reported a higher prevalence of 16.4% [8]. There is no specific sex predilection for the condition. According to certain authors the condition is most prevalent among boys [8], while others believe it to be more common in girls [3,9].

BMG is thought to be genetically determined and may have a history of familial occurrence [6,7]. The analysis of pedigrees initially suggested a polygenic mode of inheritance. With this model, it would be expected that a higher proportion of affected siblings are present in families of probands where at least one parent is affected. The occurrence of this condition in parent and sibling pairs is significantly higher than that of the general population [6]. In our case, similar lesion was observed in father and an elder male

sibling of the child and was asymptomatic. Further, BMG has been reported in eight month old monozygotic twins.

The classical clinical presentation is of multifocal, circinate, irregular erythematous patches bounded by a slightly elevated, keratotic band or line. The erythematous patches represent loss of filiform papillae and a thinning of the epithelium. This white margin is usually 1-2 mm wide, which surrounds an erythematous atrophic area devoid of filiform papillae. The size of the individual lesions may vary from 0.5 cm to larger in diameter [2,3,5]. In the present case multiple areas of depapillation of varying size with slight inflammation was noted. These areas were irregular in shape. The lesion was also found to be asymptomatic and not associated with burning sensation.

Its clinical presentation may vary from asymptomatic to painful and burning ulceration with altered taste sensation. It is characterized by periods of exacerbation and remission. During remission, lesions resolve without residual scar formation but a differential diagnosis with red and white lesions should be made even at an early age. A differential diagnosis of drug-induced reactions, atrophic candidiasis, local trauma, chemical burns and in some cases atrophic lichen planus can be given [5]. However; these causes were non-contributory in the present case.

BMG and Hereditary BMG are not two different entities. Although BMG is not uncommon in clinical practice among adults, cases of hereditary BMG have not been reported much in literature, more so in pediatric population. Further cases of hereditary BMG warrants recognition and requires examining family members for proper diagnosis to avoid unnecessary medication and diagnostic tests when confused with other similar mucosal conditions. The few published cases of hereditary/familial BMG have been reported [7]. Thus the present case report would add to the paucity of such cases in literature in pediatric population. Further, researchers and clinicians can focus on the genetic basis of causation of Hereditary BMG.

Since BMG is asymptomatic in most cases and as reported in our case, no treatment was required. Parent was reassured of the self-limiting and benign nature of the condition.

Conclusion and Clinical Significance

Hereditary BMG is an uncommon condition in children and is generally asymptomatic. Hereditary BMG can mimic other oral mucosal lesions and when not diagnosed can lead to unnecessary in-

vestigations and interventions. Reassuring the patient and the parent about the benign nature of the condition followed by periodic recall is generally recommended.

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