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Benign Migratory Glossitis in Toddlers: Report of Two Cases and Review

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Abstract

Benign migratory glossitis is an often asymptomatic, characteristically migratory, recurrent, inflammatory condition involving, mostly dorsum of the tongue or any other part, with a typical "map-like appearance". It is a condition frequently encountered in adults, with a controversial etiology.

Despite being benign and self-resolving, owing to its relative uncommon occurrence in children, it can be a matter of significant concern for parents. Here we report two cases of asymptomatic geographic tongue in children below 2 years of age.

Key words: Child, Preschool; Glossitis, Benign Migratory; Tongue

Introduction

Benign migratory glossitis(BMG) or glossitis areata migrans is an idiopathic inflammatory condition that usually presents as asymptomatic erythematous patches with diagnostic circinate or serpiginous borders, resembling a map (hence commonly referred to as Geographic tongue).[1] Initially described by Rayer in 1831 as wandering rash of the tongue, it is also referred in various literature as annulus migrans, erythema migrans, benign wandering glossitis, exfoliatioareata linguae, or transitory benign plaque of the tongue.[2]

Localized loss of filiform papillae and a thinning of the epithelium leading to smooth ulcer-like lesions that change the size, color and location over a few days to weeks are characteristic. Variable presentation from asymptomatic to painful and burning ulceration may be present.[2,3]

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Case report

Case 1

A 15 months old female child was brought to Dermatology Out Patient Department with a complaint of asymptomatic lesions over the lateral aspect of tongue noticed since two months' duration. The mother noted the red and whitish lesions while feeding the child. On local examination, irregular, circinate, erythematous patch sized 1 x 2 cm^2 with central normal mucosa was present on the right dorsolateral surface of the tongue (Figure 1). Within the patch, loss of filiform papillae was noted. Mother gave a history of similar lesion over the dorsal aspect of tongue 15 days back, which had subsided on its own. No history suggestive of atopy or other skin conditions were present. The child achieved normal developmental milestones as per her age. Medical history, family history and general physical examination were not significant. She was diagnosed clinically as BMG; parents were counseled about the condition and reassured.

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Figure 1. Circinate patch on the lateral aspect of the tongue

Case 2

A two-year-old female child reported to our OPD with complaints of reddish lesions over the tongue, for 4 months' duration. The child had no discomfort associated with the lesions. Her parents reported concerns as the lesion seem to subside on its own and reappear almost every other week. Examination revealed multiple erythematous patches with circinate white elevated borders over the entire dorsal anterior half of the tongue (Figure 2). There was no history of drug intake, atopy or any skin diseases in family or child. A clinical diagnosis of BMG was made and since the lesion was asymptomatic, reassurance was done and oral hygiene maintenance recommended.

Figure 2. Erythematous patch with elevated white border and loss of lingual papillae

Discussion

The occurrence of benign migratory glossitis is quite common, ranging from 1-2.5% of the population. However, the prevalence of BMG in the pediatric population itself is rather low, ranging from 0.37% to 14.3%.[1] Studies across the world suggest a highly variable prevalence of BMG, including The United States reporting 1.8%[4], India 0.84 -16.4%[5,6], Libya 17.2%[7] and Iran 27%.[8] A study from central-southern Nepal concluded a prevalence of BMG to be 0.98% overall with women (1.2%) and men (0.96%).[9] Data on true prevalence amongst the pediatric group is difficult to ascertain mainly due to the asymptomatic and inconsistent nature of the condition. In India, geographical tongue was reported in children in 0.89%.[10] A prevalence of 1.41% among school children with equal distribution among males and females, in a US study[11] and a prevalence of 14.29% amongst infants less than 2 years of age in Israel were reported.[12]

Aetiopathogenesis

The exact aetiopathogenesis of BMG remains unclear. It has been classified as a congenital anomaly or even hereditary disorder by some literature while others believe it to be a chronic inflammatory condition.[2] Hereditary factors are suggested by higher prevalence in parents and siblings of affected individuals, suggesting a polygenic mode of inheritance and multiple reports of BMG affecting monozygotic twins.[13,14] HLA studies have suggested significantly increased HLA DR5 and DRW6 antigens in serum and reduced DR2 in comparison to control.[15]

Association with various systemic diseases such as atopy, allergy, stress, anemia, psoriasis, gastrointestinal disorders and syndromes like Reiter's syndrome, Down syndrome, Aarskog syndrome, Fetal hydantoin syndrome and Robinow's syndrome without a definite causal relationship has been described.[2,13] Psychosomatic factors and stress, Hormonal factors (prevalence of 8% in type 1 diabetic patients), Drug (oral contraceptives, angiogenesis inhibitors), Tobacco smoking (possible protective influence), Vitamin deficiency (vitamin B6, B12, folic acid, iron and zinc)[2] are also implicated.[2,16-18]

Seiden and Curland in 2015, went on to explain variation in clinical presentation in terms of media dynamics based on physics, suggesting a practical way of assessing the severity of the condition, based on the characteristic patterns observed. Three stages of evolution of inflammation were stressed on, including resting state (healed epithelium), excited-state (highly inflamed epithelium) and recovering state (healing epithelium). They suggested that in a circular pattern, the shape of lesions will remain the same as they expand unless there is some obstacle or inhomogeneity in the epithelium. If the circular pattern is exhibited in

a patient, it is more likely that the tongue is gradually affected and subsequently healed. However, spiral patterns, which result due to inhomogeneity in the medium or due to external intervention in other excitable media, tend to be continuous, self-sustaining and will linger for a longer duration of time.[19]

Clinical features

Solitary or multifocal, erythematous atrophic areas with loss of filiform papillae with slightly elevated white, yellow or greyish margins, which may be circinate or serpiginous are usual presentation. Migratory lesions with periods of remission and exacerbation of varying duration may be present. Lesions are usually asymptomatic but symptoms like pain, burning sensation, discomfort, dysgeusia, sensitivity to hot, spicy and sour food, pain in ears or ipsilateral submandibular lymphadenopathy are associated.[1,2,10,13] "Geographic", pertaining to a resemblance to an aerial view of an area of land masses and ocean on a map is thus used to describe the lesion. Three patterns: oblate, spiral and wavy 1D pattern of the geographical tongue along with the evolution have been described by Seiden and Curland.[19] Most common sites are lateral margins and the tip of the tongue followed by dorsal and ventral surfaces. However, extra lingual sites including labial mucosa, buccal mucosa, the gingival surfaces, floor of the mouth, soft palate and uvula have also been described.[20]

Histopathology

Loss of filiform papillae with the resultant flattened mucosal surface with irregular rete pegs is typically present. There is epithelial degeneration with an absence of stratum corneum. Inflammatory infiltrates beneath epithelium and migration of polymorphonuclear leukocytes and lymphocytes are seen.[10] The elevated white borders include subepithelial infiltration of neutrophils, microabscesses, leukocyte invasion into the epithelium, intraepithelial oedema, acantholysis, glycogen deposits in the epithelial cells and exfoliation of necrotic cells in the surface layer.[21]

Diagnosis and Differentials

Clinical diagnosis is often possible with a classical presentation, which is a relatively asymptomatic lesion of migratory nature and a "geographic appearance" with characteristic white serpiginous borders. Histopathological confirmation is rarely required. In instances, they need to be differentiated from candidiasis, psoriasis, leukoplakia, lichen planus, Reiter's syndrome, herpes simplex, systemic lupus erythematosus, drug rash and squamous cell carcinoma of the tongue.

Management

Asymptomatic cases require no treatment other than reassurance regarding the benign and self-resolving nature of the lesion. Even in symptomatic cases, there is no gold standard or scientifically proven treatment.[22] Avoidance of coarse food and irritant like alcohol, tobacco products, hot, spicy and sour foods, acidic fruits and beverages should be advised. For alleviation of excessive pain or discomfort, oral analgesics, anti-inflammatory, antihistamines or mouth rinses with topical anesthetics like lidocaine gel can be prescribed. Topical corticosteroids like betamethasone, topical tretinoin and nutritional supplements including vitamin A therapy, zinc and vitamin K_2 have been used.[2,13,22]

Successful treatment with cyclosporine and topical tacrolimus has also been reported in resistant cases.[23-25]

Conclusion

BMG is a benign condition of uncertain etiology. When present in young children they may cause anxiety in parents. Reassurance and routine follow up are advocated for the majority of the cases.

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