

Symptomatic benign migratory glossitis: report of two cases and literature review

CASE REPORTS

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Abstract

Benign migratory glossitis (geographic tongue) is a common clinical finding in routine pediatric dentistry. The condition usually is discovered on routine clinical examination, appearing as an asymptomatic, ulcer-like region on the dorsum of the tongue. The lesion may recur at different sites on the tongue, creating a migratory appearance, and in many cases, will resolve completely. The presentation of symptomatic geographic tongue in children is rare. This article presents two cases of symptomatic geographic tongue. Both children presented with a chief complaint of significant oral pain which was affecting daily activity, eating, and sleeping. Both patients presented with a classical clinical presentation of ulcer-like regions on the dorsum of the tongue in which the filiform papillae were denuded. Successful management was achieved with topical and systemic antihistamine. The clinician should be aware that this condition may be symptomatic in children. (Pediatr Dent 14:392-96, 1992)

Introduction

Benign migratory glossitis (BMG) is a condition referred to in the literature by a variety of names, such as: geographic tongue, erythema migrans, annulus migrans, and wandering rash of the tongue.¹⁻⁴ This inflammatory condition first was reported by Rayer¹ in 1831.

It is a benign, inflammatory disorder occurring most commonly on the dorsum of the tongue, possibly extending onto the lateral borders. The characteristic appearance includes 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. The white border is composed of regenerating filiform papillae and a mixture of keratin and neutrophils. The surface is nonulcerated, but appears ulcerated due to the loss of the surface papillae and keratin. These well-defined, elliptical lesions vary in size from a few millimeters to several centimeters. The location and pattern undergo change over time, thereby accounting for the name "migratory." This apparent migration is due to a concurrent epithelial desquamation at one location and proliferation at another site.⁵⁻⁸

The prevalence of BMG in the general population is between 1.0 and 2.5%.^{2-4, 9} Various age groups can be affected with no apparent racial predilection; however, the condition appears to be more common in females with reported female-to-male ratios of 5:3 and 2:1.^{1, 2, 10}

Redman¹¹ observed a 1% prevalence of BMG in schoolchildren, with an equal distribution between males and females. A similar finding was noted in an investigation of university students by Meskin.⁴

Very high rates of occurrence of BMG in children in Japan (8%) and Israel (14%), with a peak age of 2-3 years, were reported in studies conducted on hospitalized pediatric patients.^{12, 13} This sampling bias could

account for the increased prevalence observed, since BMG may be seen more often in children with associated major illnesses. Both articles also suggested that the increased incidence may reflect the different racial/ethnic backgrounds of the samples examined.^{12, 13}

The etiology of geographic tongue is still unknown. Some consider the condition to be a congenital anomaly and others believe it to represent an acute inflammatory reaction. Attempts have been made to demonstrate an association between various systemic and/or psychological conditions and BMG. These conditions include psoriasis,^{7, 14} Reiter's syndrome,¹⁴ anemia, gastrointestinal disturbances, nutritional disturbances, candidiasis, lichen planus, hormonal imbalance,¹⁰ psychological upsets,¹⁵ and allergies.¹⁶ A definitive causal relationship has not been established.

Heredity may play a role in the etiology of BMG. Redman¹⁷ postulated a polygenic mode of inheritance for geographic tongue. Eidelman et al.¹⁸ determined that the prevalence of BMG in parents and sibling combinations was significantly higher than that observed in the general population. They concluded that geographic tongue was a familial condition in which heredity plays a significant role.

Marks and Tait¹⁹ provided additional support for a genetic basis of BMG by demonstrating an increased incidence of tissue type HLA-B15 in atopic patients with geographic tongue. Wysocki and Daley⁵ investigated the prevalence of BMG in patients with juvenile diabetes, because it is known that HLA-B15 occurs more frequently in insulin-dependent diabetic patients.²⁰ They discovered a prevalence of 8% for BMG in diabetic patients and concluded that BMG may be a clinical marker for insulin-dependent diabetes mellitus.⁵

Marks and Simons²¹ discovered a significantly increased frequency of atopy among patients with geographic tongue, as compared to the normal population. In a study of atopic patients with a history of asthma and/or rhinitis, Marks and Czarny¹⁶ found a 50% prevalence of BMG. They also observed that the frequency of geographic tongue increased significantly in the control group with no clinical history of atopy, but who had a positive skinprick test to common inhalant allergens. They concluded that a positive association between geographic tongue and atopy exists, and further postulated that geographic tongue and asthma/rhinitis may have a similar pathogenesis. Both conditions are recurrent, inflammatory, and can be initiated by contact with external environmental irritants. Geographic tongue is probably a sign common to those individuals who have a tendency to develop a recurrent acute inflammatory reaction on surfaces which are in contact with the external environment.

Psoriasis, a cutaneous dermatological condition, appears to be related to an accelerated rate of epithelial turnover, resulting in epithelial hyperplasia and is seen clinically as erythematous papules and/or white scaly plaques.⁷ BMG has been suggested as an oral manifestation of psoriasis.^{7, 22, 23}

The association between fissured tongue and BMG supports a genetic basis for the development of the condition.¹⁸ The fissures may act as stagnation areas on the tongue surface in which glossitis may begin.⁸ Geographic tongue appears to be associated with geographic stomatitis^{6, 8} which has a clinical and histological appearance similar to BMG but occurs on an extraglossal intraoral site.²⁴

The diagnosis of BMG usually is based solely on the history and clinical presentation which would include characteristic migratory pattern and chronic nature. The vast majority are asymptomatic and noticed during the course of a routine oral examination, or self-examination.^{6, 8, 10, 25} Only Cooke⁸ reported that a significant number of patients complained of some oral sensitivity or discomfort, most often described as a "burning sensation." No oral sensitivity associated with cases of BMG in pediatric populations has been reported.

If a patient presents with suspected BMG, a differential diagnosis based on adult studies should include atrophic candidiasis, psoriasis, Reiter's syndrome, atrophic lichen planus, systemic lupus erythematosus, leukoplakia, and drug reaction.^{6, 25}

The histological features of BMG are those of a localized acute glossitis. The central erythematous portions represent an area of epithelial degeneration and the absence of the stratum corneum, with very little alteration in the basal layer of epithelium. Beneath the epithelium is a dense infiltration of inflammatory cells

with migration of polymorphonuclear leukocytes and lymphocytes toward the zone of epithelial degeneration. Munro abscesses also may be seen. The advancing margin is outlined clearly by a dense, polymorphonuclear infiltration of the acanthotic epithelium and corium. The border may demonstrate a zone of hyperkeratinization (parakeratosis). Tissues which were affected previously show a chronic inflammatory reaction.^{8, 10, 26, 27} Geographic tongue is characterized by periods of exacerbation and remission. During remission, lesions resolve without residual scar formation. When lesions recur, they tend to occur in a new location, thus producing the migration effect.

Since BMG is asymptomatic in most cases, no treatment is required other than patient reassurance of the benign and self-limiting nature of the disorder. If symptoms are present, the patient should be instructed to avoid any known irritants, such as hot, spicy, or acidic foods. If treatment is warranted, it should be palliative/symptomatic care using topical anesthetic rinses or gels, antihistamines, or steroids.^{6, 8, 10, 25, 28} A psychological component may contribute to the development of BMG, and tranquilizers may be considered in patient management.²⁵

A review of the literature on BMG failed to produce a reported case of symptomatic BMG in children. This article presents two cases of therapeutic management of children with symptomatic BMG.

Case Reports

Case One

A 4-year-old Caucasian boy was seen with a chief complaint of oral discomfort and increased salivation. Review of his medical history revealed an innocent heart murmur, sleep apnea until the age of 9 months, and recurrent otitis media which required placement of myringotomy tubes at the age of 1 year. At the initial visit there was no history of allergy to medications or environmental factors.

The patient's mother reported that approximately one month earlier, her son began to experience oral discomfort, evidenced by crying, placing objects in his mouth, and a marked increase in salivation and expectoration. The child reported that his mouth had an "awful taste."

He was placed on nystatin (Mycostatin[®], Bristol-Myers Squibb Canada, Montreal, Quebec, Canada), 100,000 units, three times a day, by his family physician without success. His general dentist suspected that the early eruption of his first permanent molars could be the source of the discomfort and referred him to our clinic for consultation. The mother also reported that no other family members had a similar condition.

On examination, the child appeared normal, healthy, and well-developed. There were no abnormal extraoral clinical findings. Intraoral examination revealed a complete, caries-free primary dentition, good oral hygiene, and no evidence of gingival inflammation. The dorsal surface of the tongue demonstrated a pattern consistent with a resolving geographic tongue, with irregular circumscribed areas devoid of filiform papillae (Figure). The surface was not erythematous and there was no evidence of leukoplakia or white curd-like pseudomembranes, as seen in fungal infections. The first permanent molars had not erupted and radiographic examination demonstrated that they were present at a normal stage of development with no evidence of communication between the crypt of the molar and the oral cavity.



Figure. Dorsal surface of the tongue in Case 1 demonstrating irregular areas devoid of filiform papillae consistent with the pattern seen in geographic tongue.

The anti-fungal agent was stopped and the mother was instructed to contact our clinic if an acute exacerbation occurred. Nine months later, the mother reported that one month earlier, her son had his DPT booster and for seven days had a fever of 101–102°F. At that time, the tongue lesions reappeared with oral pain, increased salivation with expectoration and a generalized agitation. He was placed on nystatin and a lidocaine (Xylocaine®, Astra, Mississauga, Ontario, Canada) gel for topical pain relief by his family physician, but this provided only temporary relief. Review of his medical history at this time revealed that he was developing allergies to environmental and food factors.

Clinical examination again indicated a resolving pattern on the dorsum on the tongue, and a significant amount of drooling with a complaint of foul taste. Exfoliative cytology was performed and was negative

for *Candida*. The nystatin was stopped immediately and the mother was informed to call if signs and symptoms recurred.

Four weeks later he experienced similar symptoms. Examination revealed irregular, circumscribed erythematous areas on the dorsum of the tongue. These areas were devoid of filiform papillae. The lesions had margins with a raised white appearance that could not be scraped off. The regions were not tender to touch. Exfoliative cytology was repeated and did not show any evidence of candidiasis. A complete blood count with differential was within normal limits.

The boy was instructed to rinse with 5 cc of a diphenhydramine HCl suspension (Benadryl®, Parke-Davis, Morris Plains, NJ, 12.5 mg/5 cc) up to four times per day, holding it over his tongue and then swallowing it. All the symptoms were relieved within 48 hr of initiation of antihistamine therapy.

In the subsequent six months, the condition recurred twice and immediate treatment with the diphenhydramine suspension provided symptomatic relief within 24 hr.

Case Two

A 3-year-old Caucasian girl gave a chief complaint of oral pain that prevented eating and drinking. She had multiple environmental allergies and was suspected to have asthma. The remainder of her medical history was unremarkable. No other family members had a similar oral condition. Her father reported that her mouth became extremely painful every month for two to three days and this pain would resolve spontaneously without treatment.

Examination revealed a well-circumscribed, irregular pattern on the dorsum of the tongue devoid of filiform papillae, consistent with BMG. The areas were asymptomatic and appeared to be resolving, so no treatment was performed. The parent was informed about the nature of the condition and instructed to return the child to our clinic if the lesions and symptoms recurred.

The child returned six months after the initial examination with a similar clinical presentation but during a period of acute pain. The diagnosis of symptomatic BMG was made and the child was instructed to rinse with 5 cc of diphenhydramine HCl suspension (Benadryl, 12.5 mg/5 cc) up to four times per day, holding it over her tongue for 1–2 min and then swallowing it. She was relieved of all her symptoms within 24 hr of initiation of antihistamine therapy.

Discussion

Geographic tongue or BMG is a common finding during routine examination of children. In previous investigations, the condition was asymptomatic. Only

Cooke,⁸ in 1962, reported that a significant number of adults with BMG had varying degrees of oral sensitivity associated with the condition. To our knowledge, these are the first two cases of symptomatic geographic tongue reported in pediatric patients. In both cases, symptoms were severe enough to interfere with sleeping and eating.

BMG is capable of producing symptoms in children that are significant enough to require management. The differential diagnosis of BMG in children should include atrophic candidiasis, drug-induced reactions, local trauma and a severe neutropenia. Psoriasis, Reiter's syndrome, atrophic lichen planus, malignancy, and systemic lupus erythematosus can produce similar lesions, but are rare in children.

The child with symptomatic tongue ulcerations should have a complete medical and dental history taken, followed by a comprehensive extra- and intraoral examination. If a diagnosis of BMG cannot be made based on the history and examination due to an atypical, symptomatic presentation, then a complete blood count with differential should be obtained to rule out neutropenia and to assess the general state of health. In addition, exfoliative cytology of the area should be performed to rule out candidiasis. If a definitive diagnosis still cannot be made, a biopsy of a representative region of the lesion would be warranted.

Most patients require no definitive treatment other than observation and reassurance of the benign nature of the condition. For painful BMG, recommended supportive and symptomatic management would include a bland diet, plenty of fluids, acetaminophen for systemic pain relief, and a topical anesthetic agent such as viscous lidocaine or benzydamine (Tantum™, Riker/3M, London, Ontario, Canada) rinse for local pain relief. If available, benzydamine may be preferred because of a reported combined analgesic and anti-inflammatory effect that lasts for up to 3 hr.²⁹

If the lesions should recur, or their severity is such that the child does not have adequate relief from the symptomatic therapy, then an antihistamine, such as diphenhydramine HCl (Benadryl) should be used. The child should rinse with 12.5–25 mg (1 to 2 teaspoons) depending on age and weight, holding it over the tongue for a few minutes, and then swallowing, three to four times per day for up to seven days.²⁹

If the lesions do not respond to antihistamine therapy, then a corticosteroid, such as betamethasone as a 500-µg tablet dissolved in water, can be used as a rinse for a few minutes and swallowed, twice daily for seven to 14 days.²⁹ The steroid only should be used in patients who do not respond to either supportive/symptomatic or antihistamine therapy. The child's physician should be consulted when using steroids.

The etiology of BMG is unknown, but the condition may be linked to allergies.²¹ It is interesting to note that both of these children presented with various environmental allergies.

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