

Paediatric Geographic Tongue: A Case Report, Review and Recent Updates

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ABSTRACT

Geographic tongue is a benign recurrent condition of uncertain aetiology affecting the tongue characterized by loss of epithelium especially filiform papillae giving a characteristic appearance. The clinical presentation may vary from asymptomatic to painful and burning ulceration. The condition is commonly seen in adults but few cases are reported in children. A case of asymptomatic geographic tongue in three-year-old male child and literature review with new insight in aetiology is presented here. Management depends on the clinical condition and underlying aetiology.

Keywords: Benign migratory glossitis, Children, Erythematous patches, Filiform papillae, Tongue, Wandering rash of tongue

INTRODUCTION

Geographic tongue (GT) is a benign harmless transient oral condition. It is also referred by various terminologies such as benign migratory glossitis, erythema migrans, annulus migrans and wandering rash of the tongue [1].

GT was first described as wandering rash of the tongue by Rayer in 1831 [2]. The characteristic pattern of the lesions gives the dorsal surface of the tongue the appearance of a map. The lesion persists for a period of time of several days to weeks depending on the individual and disappears only to reappear at a different location giving a different pattern [1]. Symptomatic GT is rare in children and may produce anxiety [3].

CASE REPORT

In an annual dental screening camp, geographic tongue was noticed in three year male child. Clinically, lesions characteristic of geographic tongue were found on the anterior two thirds of the dorsal surface as well as lateral surface of the tongue [Table/Fig-1]. Oblate pattern was observed in the present case. He was unaware of condition and had no pain or discomfort. His parents were informed about the condition. No contributing history was elicited. However, oral examination revealed dental caries in relation to 51, 61, 74, 84 teeth. Patient was asymptomatic and since GT is a self limiting condition reassurance, plenty of fluid intake, restoration of carious teeth, and oral hygiene maintenance was advised. Patient's parents were informed and a recall was advised after three months but patient didn't turn up.

DISCUSSION

Epidemiology

Overall prevalence of geographic tongue is approximately 1-2.5% of the population [4]. The prevalence of GT in the paediatric population ranges from 0.37% to 14.3% with unknown aetiology [5,6].

In India, prevalence is 0.89% [7]. The highest incidence of about 39.4% occurs in the age group of 20-29 years [8]. GT is more frequently reported in adults than in children [9] while others believe it to be common in younger individuals [10]. Females are more commonly affected than males with the ratio of 1.5:1 [8]. Some studies have reported this condition to affect males more frequently [10].

Google and Medline literature search revealed the prevalence of geographic tongue in paediatric age group worldwide as shown in [Table/Fig-2] [4,8,9,11-39].



[Table/Fig-1]: Red atrophic areas surrounded by white circinate borders seen on lateral and dorsal surfaces of tongue.

Aetiopathogenesis

The aetiopathogenesis of the geographic tongue remains unknown till date. Some investigators have classified it as congenital anomaly while others said it to be a hereditary disorder [40]. Few authors believed it to be a chronic inflammatory condition [40]. Several aetiological factors have been suggested.

Hereditary factors: Siblings of one parent affected by geographic tongue revealed significantly higher prevalence of geographic tongue than those siblings of unaffected parents suggesting the role of familial and hereditary factors [41].

Assessment of histocompatibility antigens among GT patients revealed significantly increased DR5 and DRW6 antigens in serum of GT patients when compared to controls while DR2 was reduced comparatively [42]. Recently, GT in five-year-old monozygotic twins with mild pain and discomfort on consumption of spicy food was reported supporting the genetic aetiology [43].

Association with other systemic diseases: GT is commonly associated with other systemic diseases such as atopy, allergy, stress, anaemia, psoriasis, gastrointestinal disturbances and hormonal variations [1,40]. It may be associated with syndromes like Reiter's syndrome, Down syndrome, Aarskog syndrome, Fetal hydantoin syndrome and Robinow's syndrome [1,44]. But none have shown a definite causal relationship.

| Author(s) | Country | Year | Prevalence of Geographic Tongue (%) |
|---------------------------|----------------|------|-------------------------------------|
| Rahamimoff [11] | Israel | 1957 | 14 |
| Togo et al., [12] | Japan | 1961 | 8 |
| Luigi [13] | Italy | 1963 | 28.7 |
| Redman [14] | USA | 1970 | 1.41 |
| Ghose et al., [15] | Iraq | 1982 | 43 |
| Kullaa et al., [16] | Finland | 1982 | 2.5 |
| Sawyer et al., [17] | Nigeria | 1984 | 3 |
| Sedano et al., [18] | Mexico | 1989 | 1.9 |
| Kleinman et al., [19] | Denmark | 1994 | 0.6 |
| Arendorf et al., [20] | South Africa | 1996 | 1.6 |
| Bezerra et al., [21] | Brazil | 2000 | 21 |
| Voros-Balog et al., [22] | Hungary | 2003 | 5.7 |
| Rabiei [23] | Iran | 2003 | 13.4 |
| Avcu & Kanli [24] | Turkey | 2003 | 1.2 |
| Sanei [25] | Iran | 2003 | 6.2 |
| Sadolah et al., [26] | Iran | 2003 | 3 |
| Garcia-Pola et al., [27] | Spain | 2002 | 4.48 |
| Bessa et al., [28] | Brazil | 2004 | 9.8 |
| Shulman et al., [4] | USA | 2005 | 1.05 |
| Jainkittivong et al., [8] | Thailand | 2005 | 39.4% (both C+A) |
| Ugar-Cankal et al., [29] | Turkey | 2005 | 1.8 |
| Khozeimeh et al., [30] | Iran | 2006 | 4.9 |
| Mozarrad & Viziri [31] | Iran | 2008 | 27 |
| Mathew et al., [32] | Southern India | 2008 | 0.84 |
| Byahatti et al., [33] | India | 2010 | 17.2 (both C+A) |
| Darwazeh et al., [9] | Jordan | 2011 | 4.8 |
| Ambika et al., [34] | India | 2011 | 1.4 |
| Yilmaz et al., [35] | Turkey | 2011 | 2.68 |
| Jahanbani et al., [36] | Iran | 2012 | 0.8 |
| Madera et al., [37] | Colombia | 2013 | 0.8 |
| Patil et al., [38] | India | 2013 | 16.4 (both C+A) |
| Rezai et al., [39] | Iran | 2015 | 7.86% (both C+A) |

[Table/Fig-2]: Prevalence of geographic tongue in Paediatric population worldwide [4, 8, 9, 11-39].
C-Children, A-Adults.

Asthma/Atopy: Marks and Tait demonstrated an increased incidence of tissue type HLA- B15 in atopic patients with geographic tongue which provided an additional support for genetic basis.

They observed a positive association between GT and atopy/asthma which led the authors to postulate a similar pathogenesis for both the disease entities [45]. GT was more commonly found in patients with allergies to drugs, food or others in a study conducted by Jainkittivong et al., [8].

Association with psoriasis: Due to similarities in clinical, histopathologic and immunohistochemical findings, few researchers consider geographic tongue as an oral manifestation of psoriasis. Histopathologic similarity may support the hypothesis that GT may be considered as an expression of oral psoriasis.

Few studies have concluded that GT is an oral manifestation of psoriasis [46,47] while others argue that it is not [48]. Highly significant association with Human Leukocyte Antigen (HLA)-Cw6 and weakly significant association with B13 in both GT and psoriasis are reported [49].

Recently Tarakji et al., in their review have mentioned that GT is more common in psoriatic patients however a strong evidence is still lacking [50].

Few authors concluded that GT and psoriasis do not share a common aetiology and rather occur co-incidentally [51,52].

A case-control study in India examined 600 adult patients with psoriasis and revealed a strong association between psoriasis and GT. Authors concluded that GT more frequently affects males and those patients with severe psoriasis [10].

Significant occurrence of GT in early onset psoriasis and fissured tongue in late-onset psoriasis was observed in a recent study. Authors believed that GT could be used as a marker of the severity of psoriasis [53].

Association with fissured tongue: Significant co-existence of GT and fissured tongue was noticed in 60.1% patients in a study conducted by Jainkittivong et al., [8]. Ghose et al., suggested a genetic linkage between these two conditions [15].

GT and fissured tongue may share a common genetic aetiology. Deep fissures on the dorsum of the tongue ensure stagnation and subsequent development of glossitis [8].

GT and fissured tongue have been reported in association with chronic granulomatous disease [54].

Psychosomatic factors and stress: Psychosomatic factors appear to have significant role in the aetiology of GT. Redman et al., demonstrated higher prevalence of geographic tongue in mentally ill patients [14].

Ebrahimi et al., demonstrated the association between the stress and geographic tongue. They also found that decreasing stress in geographic tongue patients can be helpful in healing the lesions [55].

Psychological factors, such as stress, may represent a potentially modifiable risk factor that could influence the frequency of recurrence of GT in adults [56]. Stoopler et al., in their clinical observation suggested that these factors may be associated with GT even in the paediatric population similar to the adult population [57].

Hormonal factors: Wysocky and Daley found association between the geographic tongue and diabetes and found fourfold increase in the prevalence of geographic tongue in diabetic patients. They found a prevalence of 8% in type 1 diabetic patients [58]. On the contrary, Guggenheimer et al., reported no significant correlation between geographic tongue and insulin dependent diabetes mellitus [59].

Drug induced: Geographic tongue may develop as a result of hormonal fluctuation. Women on oral contraceptive pills have shown to develop GT lesions which got severe on the seventeenth day of the cycle [60].

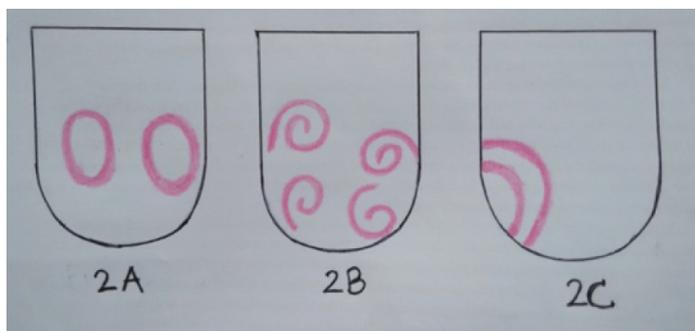
Drug induced GT due to angiogenesis inhibitors has also been reported. Authors believed that therapeutic inhibition of VEGF or its receptors in oral mucosa by the drugs could induce GT [61].

Tobacco smoking: A protective influence of smoking has been observed since smokers had less prevalence of geographic tongue than nonsmokers [4,24].

Vitamin deficiency: Deficiency of vitamin B6, B12, folic acid, iron and zinc have been proposed to be contributing factors as well [37,40].

Recent aetiopathogenesis according to dynamics based on physics: Recently in 2015, Seiden and Curland explained geographic tongue in terms of media dynamics based on physics. They observed different evolutionary phases between "a healed (rest) state, a highly inflamed (excited) state, and a healing (recovering state)". Similarities between dynamics of GT and other excitable media such as forest fires, cardiac arrhythmias, chemically driven reaction-diffusion systems and morphogenesis in multicellular organisms was noted by the authors [62]. There exist different patterns of geographic tongue – oblate pattern, wavy 1D pattern and a ring shaped concentric pattern. Seiden and Curland have explained dynamics of circular or oblate or closed pattern which are more typically observed and secondly spiral or open ended pattern [Table/Fig-3] [62].

In circular pattern, shape of lesions will remain same as they expand on tongue unless there is some obstacle or inhomogeneity in the



[Table/Fig-3]: Different clinical patterns of geographic tongue - Oblate or circular pattern (A), Spiral pattern (B), Wavy 1 D pattern (C)

epithelium. If circular pattern is exhibited in a patient it is more likely that tongue is gradually affected and subsequently healed [62].

While spiral patterns tend to be self-sustaining and will linger for a longer duration of time. These patterns result due to inhomogenities in the medium or due to external intervention in other excitable media. However, in GT more studies are further needed.

Apart from its unique composition, tongue is also exposed to external conditions like temperature variations with different foods, rubbing of tongue's edge against teeth or gums which may trigger the phenomenon in GT. They also observed GT in a one-year-old boy with teething supporting the environmental origin [62].

Clinical Presentation

Lateral margins and tip of the tongue are the most commonly involved sites followed by dorsal and ventral surfaces [8]. If the lesions occur in different sites other than tongue, then the term *ectopic geographic tongue* is employed as first coined by Crooke in 1955 [1]. The extra glossal sites include the labial mucosa, buccal mucosa, gingival floor of the mouth, soft palate and uvula. GT is also known by various names such as geographic stomatitis, wandering rash of tongue, erythema migrans, benign migratory glossitis and stomatitis areata migrans [2].

Clinically, lesion of the geographic tongue reveal erythematous atrophic areas with loss of filiform papillae surrounded by the white circinate borders [1].

Periods of remission and exacerbation of varying duration are usually evident. The lesion recurs in the new locations thereby producing the migrating pattern. Continuously changing patterns and migration of lesions on the tongue surface with unusual appearance of the tongue are the usual chief complaints of the patient [8,40].

GT can occur either as isolated or multiple lesions, can be asymptomatic or present with symptoms like pain, burning sensation, discomfort, dysgeusia, sensitivity to hot, spicy and sour food pain in ears or ipsilateral submandibular lymphadenopathy [8,40]. Majority of the patients will be asymptomatic. Two symptomatic cases in paediatric age group with environmental allergies presenting with oral discomfort, increased salivation, altered taste, pain while eating and drinking were reported by Sigal et al., [40].

Regezi et al., suggested that presence of deep fissures infected with candida organisms could be responsible for symptoms [63]. However, this has to be further investigated.

Histopathology

Microscopic examination reveals a thick keratin layer infiltrated by mixed inflammatory cell infiltrate, thin elongated rete ridges and epithelial oedema. The inflammatory cells produce small micro abscesses called Monro's abscesses in the keratin and spinous layer. Superficial areas may reveal candidal hyphae and spores [63]. Areas corresponding to the red patches show increased oedema, acanthotic epithelium and complete absence of filiform papillae [7]. The areas from the centre of the atrophic areas show complete

absence of filiform papillae and parakeratotic layers. The elevated white borders include subepithelial infiltration of neutrophils, micro abscesses, leukocyte invasion into the epithelium, intraepithelial oedema, acantholysis, glycogen deposits in the epithelial cells and exfoliation of necrotic cells in the surface layer [64].

Under scanning electron microscopy, two types of abnormal mucosa have been demonstrated from the surface of geographic tongue: an atrophic area on which the extensions of filiform papillae are absent but the bodies appear typical and the white margin of desquamating cells. Micro fissures were observed between the atrophic and normal mucosa [65].

Differential Diagnosis

GT is usually diagnosed by its characteristic history and unique clinical features. Histologic confirmation is rarely needed. In case of atypical lesions, differential diagnosis of GT includes atrophic candidiasis, neutropenia, psoriasis, Reiter's syndrome, leukoplakia, lichen planus, systemic lupus erythematosus, herpes simplex and drug reaction [7,66]. Isolated/solitary geographic tongue lesions may sometimes show striking resemblance to leukoplakia which is a precursor to squamous cell carcinoma of tongue (at times causing cancerphobia in patients), in which case the risk factors that are associated with oral cancer (such as old age, alcohol consumption, and tobacco use) are not associated with GT [5,67].

If the clinician encounters a solitary ulcer on the oral mucosa, the first consideration should be conditions associated with ulcers of short duration (less than 3 weeks), including trauma, recurrent aphthous ulcer, infection, and vesiculobullous disease. If the ulcer persists beyond three weeks after removing any local irritants, consideration should be given to chronic trauma, chronic infection, major aphthous ulcer, gumma (from syphilis), and leukoplakia.

Investigations are required if diagnosis of GT cannot be made by history and examination due to atypical and symptomatic presentation. Blood investigations would rule out anaemia and neutropenia, while Periodic acid Schiff stained cytosmeears can rule out candidiasis. If a definite diagnosis is still not made then biopsy from most representative area is warranted to rule out cancerphobia in patients [5,7,40].

Treatment

Geographic tongue typically does not require any treatment if asymptomatic. Periodic follow up to confirm diagnosis is required in case of first visit and when history is unclear. Reassure the patient about the benign and self-limiting nature of the lesion. If there is excessive pain and discomfort, medications like analgesics such as acetoaminophen, anti-inflammatory drugs, antihistamines like diphenhydramine hydrochloride, mouth rinses with topical anaesthetics like lidocaine gel, topical corticosteroids like betamethosone, cyclosporine, vitamin A therapy like tretinoin, zinc and vitamin K₂ supplements can be advised [1,68,69]. Complete reversal of condition has been noted after a few weeks of supplements.

Topical Tacrolimus application has been effective in recalcitrant cases [70]. Satisfactory improvement was observed by systemic administration of cyclosporine microemulsion pre-concentrate, with initial dose of 3mg/kg/day and maintenance dose of 1.5mg/kg/day two months later [71].

Discomfort from the geographic tongue can be limited or avoided by avoiding alcohol, tobacco products, hot, spicy and sour foods, acidic fruits and beverages, toothpaste that contains tartar control additives, heavy flavouring or whitening agents, dried, salty nuts which may aggravate the condition [67]. However, none of these treatments have been effective.

CONCLUSION

Geographic tongue is a benign condition of uncertain aetiology. The clinical presentation may cause anxiety in young patients as well as parents. Reassurance and follow up of patients especially in Paediatric population is needed to know about effective treatment modalities.

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