

Atypical median rhomboid glossitis: A case report

Priyanka Nath¹, Saroj Kumar Rath²

¹Department of Oral and Maxillofacial Surgery, ²Periodontics and Implantology Dental Hospital, Army Dental Corps, Dharamshala, Himachal Pradesh, India

Correspondence to: Dr. Priyanka Nath, P7/4 lower SNS, Yol Cantt Dharamshala 176052, Himachal Pradesh, India.
E-mail: nath.priyanka5@gmail.com

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ABSTRACT

Median rhomboid glossitis or glossal central papillary atrophy is characterized by an area of redness and loss of filiform papillae, situated on the dorsum of the tongue in front of the circumvallate papillae. Smoking, denture wearing, use of corticosteroid sprays or inhalers, and human immunodeficiency virus are few of the predisposing factors. Previously, this condition was believed to be a developmental defect of the tongue, caused by a failure of the tuberculum impar to be covered by the lateral processes of the tongue; however, now, it is thought to be due to chronic candidiasis. This paper presents an atypical case of rhomboid glossitis in a 4-year-old girl, discussing the clinical presentation, etiological factors, and differential diagnosis.

Keywords: *Candidiasis, Diabetes, Kissing lesion, Median rhomboid glossitis, Tuberculum impar*

Median rhomboid glossitis (MRG) was first described by Brocq in 1914 and occurs in <1% of the general population. Men are affected more often than women (3:1) and adults more than children [1]. MRG, also known as central papillary atrophy of the tongue, is clinically characterized by an erythematous lesion in the center of the posterior part of the dorsum of the tongue and anterior to the circumvallate papillae. This area of erythema results from atrophy of the filiform papillae. The lesion may appear flat, maculated, and usually raised by 2–5 mm. Some lesions are depressed or eroded; others display a coarse granular texture [2]. While some cases are asymptomatic, other patients complain of persistent pain, irritation, or pruritus [2]. When MRG is found in association with palatal inflammation corresponding to contact with the involved area on the tongue, it is called kissing lesion; immunosuppression should be suspected, and it has been considered a marker of AIDS [3].

There are various schools of thought regarding the etiology of MRG; it has been proposed that MRG may be attributed to chronic candidiasis or that it may be of embryological, inflammatory, or immunological origin. It is considered to be a dysembryoplastic process, with the lesion provoked by the mechanical actions of mastication and swallowing, in which other complications and secondary infections such as chronic candidiasis or pigmented nevus may occur. The prevalence of this condition is higher in immunosuppressed patients, diabetics, and patients on broad-spectrum antibiotics [4]. This paper reports an atypical case of rhomboid glossitis in a 4-year-old girl with a discussion on its clinical presentation, etiological factors, and differential diagnosis.

CASE REPORT

A 4-year-old female visited the outpatient department of our dental unit with a chief complaint of burning sensation in the posterior dorsum of the tongue for 1 week. A history of present illness revealed that the patient had been suffering from erythematous raised lesions on the posterior part of the dorsum of the tongue since birth (Fig. 1). This was accompanied by mild burning and itching sensation for 1 year. This sensation gradually increased in severity leading to discomfort for 1 week, prompting the parents to seek medical advice. The burning sensation aggravated on consumption of sour and spicy food. Past medical history was non-contributory. There was no history about the administration of antibiotics or steroids in the past. At birth, the parents had sought medical advice regarding the erythematous tongue lesions and a biopsy was advised. The biopsy had been carried out when the patient was 2 years old at a different medical centers; however, the biopsy report was inconclusive due to an insufficient specimen.

Clinical examination of the mouth revealed multiple areas of depapillation 2 mm × 2 mm on the dorsal surface of the tongue just anterior to circumvallate papillae and medial portion of the tongue. The surface was erythematous smooth and raised; the palatal mucosa was normal. No associated symptoms such as fever lymphadenopathy were detected.

A complete blood workup was advised to rule out an immunosuppression. No abnormality was detected. A working clinical diagnosis of “MRG” was made. Immune booster syrup, probiotics, and non-steroidal oral rinse with local anaesthetic and analgesic properties (Tantum Oral Rinse) were prescribed. The patient was asked to come for a follow-up after 1 month. The



Figure 1: Erythematous raised lesions on the posterior part of the dorsum of the tongue

patient showed improvement in terms of burning sensation and irritation; however, no regression of the lesion was seen.

DISCUSSION

Considerable proportions of oral mucosal lesions involve the tongue and hence are of health concern to both the oral health-care providers and the public. Examination of the tongue and oral mucosa is an essential part of general physical examination; thus, dental clinicians need to recognize and know the spectrum of disorders that affect the tongue [5].

Martin and Howe [6] gave an explanation for the pathogenesis of this disease based on an embryologic fault in the development of the tongue. According to them, tuberculum impar “may persist in its most perfect, rhomboid form, but it can be anteriorly or laterally displaced and of any shape; it can be split and appear in either two or three segments and be dispersed anywhere in the tongue except posterior to the foramen caecum and the sulcus terminalis.” Gougerot and Dechaume reported a series of lingual plaques, some of which were not located in the midline of the tongue and was termed as an atypical form of MRG, as was seen in this case [7].

This lesion is now believed to be related to chronic infection by *Candida albicans*, and in many cases, candida hyphae are present in the parakeratinized surface layers of the tongue epithelium. The exact role of this fungus in the pathogenesis of the lesion is not clear [8]. Like candida, *Actinomyces* also has been shown to induce pseudoepitheliomatous hyperplasia of the tongue and inflammatory hyperplasia of the underlying connective tissue, resulting in the characteristic elevated lesion [9]. Tapper-Jones *et al.* [10] in their study showed a direct relationship between smoking and candidiasis in both diabetic and healthy subjects.

Willis *et al.* [11] found that patients with diabetes who also smoked had significantly higher load of candidiasis than diabetic patients who did not smoke. Furthermore, Guggenheimer *et al.* [12] depicted MRG to be one of the most observed oral candidial infections in insulin-dependent diabetes mellitus patients. Gumru *et al.* [13] observed a relationship between denture stomatitis and MRG. However, in contrast, Farman and

Nutt [14] revealed that neither the association between MRG and denture stomatitis nor the association between MRG and denture wearing was statistically significant.

According to Arendorf and Walker [15], 44% of the population harbor candidiasis fungus as part of their normal oral flora, and they also stated that the tongue is the primary oral reservoir for candida. In particular, the midline of the tongue is suitable for an intense overgrowth of candidiasis fungus. Since the tongue maintains close contact with the palatal mucosa during swallowing and at rest, the area of the tongue contacting the palate corresponds well to the area in which MRG develops. When MRG is found in association with palatal inflammation corresponding to contact with the involved area on the tongue, it is called the kissing lesion [3].

MRG is easily recognized by its unique location and clinical appearance. Although the diagnosis of MRG is basically clinical, sometimes histopathology is required to differentiate MRG from irritation fibroma, mucocele, granular cell tumor, tertiary syphilis, lingual thyroid (usually further posterior) and, in rare cases, squamous cell carcinoma. There have been three previous reports of malignant transformation of MRG [16]. However, Delamarre and Van der Wall [7] suggested that there is no clear relationship between MRG and cancer.

There is no definite treatment for this condition. In recent times, however, antifungal therapy either topical or systemic has been reported to be beneficial [2]. Numerous antifungal agents such as polyenes (nystatin) or azole group antimycotics (fluconazole and clotrimazole) are being prescribed. Inappropriate prophylactic and empirical therapies have led to an outbreak of widespread resistance, and hence, alternative strategies have to be adopted [17]. Moreover, potential adverse reactions such as hepatotoxicity, possible cardiac arrhythmias, and drug interactions should be discussed with the patient’s primary care physician before any particular agent is prescribed. If treatment does not resolve with antifungal therapy, a biopsy may be warranted.

CONCLUSION

The occurrence of MRG in young children is compatible with its supposed developmental etiology. MRG is a benign condition and requires reassurance and explanation, with little to no treatment. Further research is required to study the etiopathogenesis and risk factors of MRG.

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