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Case Report

Eosinophilic Ulcer of Tongue: A Case Report

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Abstract

Eosinophilic ulcer is a rare self-limiting chronic benign lesion of the oral mucosa with pathogenesis still unclear. It may resemble malignancies, traumatic ulcerations and some infections such as deep fungal infections, tuberculosis and primary syphilis. However, trauma has been found to be a contributing factor in a majority of the cases. As the condition is perhaps unknown to medical personal and due to the confusion in the literature with eosinophilic granuloma, we think the following case report deserves presentation. This is a case report of ulcerative granuloma in the lateral border of tongue with history of recurrent trauma from sharp tooth. The ulcer rapidly healed after incisional biopsy and definite diagnosis was achieved only combining histologic finding and clinical follow up.

Keywords: Eosinophilic Ulcer, Tongue

Introduction

Eosinophilic ulcer (EU) is a rare self-limiting chronic benign condition. This entity was first described in 1881 by the Italian physician, Antonio Riga, and as additional cases were subsequently published by F. Fede in 1890, it has been known as Riga-Fede disease.[1] Clinically, the ulceration has been most frequently found in tongue and it is characterized by the presence of mildly indurated borders which may resemble malignancies, traumatic ulcerations and some infections such as deep fungal infections, tuberculosis and primary syphilis.[2] EU also has been referred as traumatic ulcerative granuloma with stromal eosinophilia, traumatic eosinophilic granuloma, traumatic granuloma and ulcerative eosinophilic granuloma. In infants, usually on the ventral surface of the anterior tongue secondary to trauma from newly erupted primary teeth, EU is referred as Riga-Fede disease.[2] Trauma has been found to be a major cause. Microscopically, it is characterized by a diffuse polymorphic cell infiltrate composed predominantly of eosinophils, B and T lymphocytes, macrophages, and large atypical cells involving the superficial mucosa and extending deep into the submucosa causing degeneration of the underlying muscle.

Case Report

A 45 years old female presented with a painful ulcer on the right side of tongue since one month. She gave history of pain being moderate, continuous. There was no relief after medication. On clinical examination an ulcer was found on right lateral border of tongue opposite right first molar tooth. The ulcer was 01 x 0.5 cm in size, painful, containing borders bringing two aspects, central ulceration with white-yellow fibrous base. Patient's oral hygiene was poor and dental caries was found with sharp corner in right first molar tooth. There was no cervical lymphadenopathy. The ulcer was excised and sent for histopathology. Histopathology report revealed adjoining hyperplastic mucosa and infiltration of polymorphs rich in eosinophils in lamina propria. The patient was sent in dental department for correction of dental problem. After fifty days tongue was found completely healed.

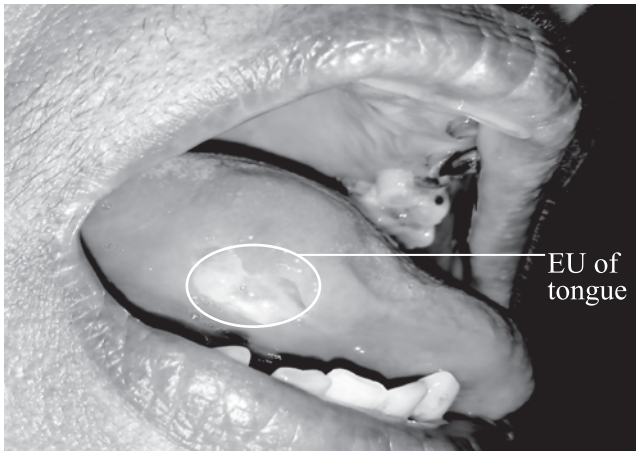

Fig-1: EU in right side of tongue

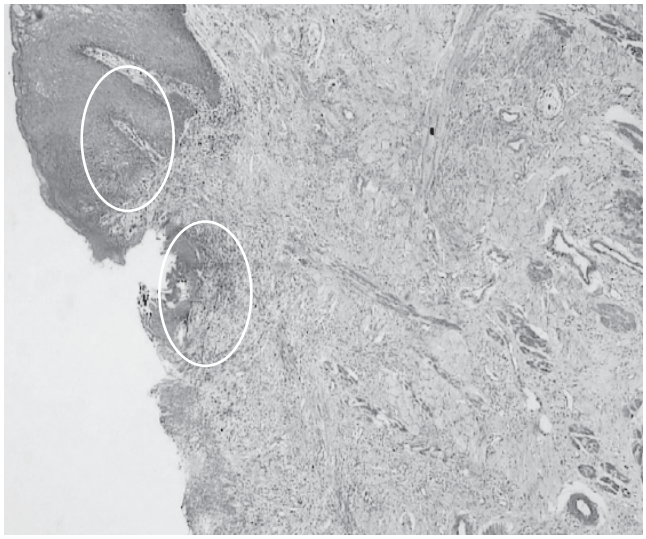

Fig-2: Polyinflammatory infiltrat

Discussion

Eosinophilic ulcer is an unique lesion with uncertain nature, aetiology and pathogenesis. Eosinophilic ulcer, traumatic eosinophilic granuloma of the tongue, traumatic granuloma, atypical histiocytic granuloma and TUGSE have been used to describe the lesion. An eosinophilic ulcer is considered to be a benign, reactive and self-limiting lesion of the oral mucosa. Trauma is considered to play a major role in the aetiology of the ulcer, however, trauma could be observed in only 50% of cases.[3] It is considered to be a benign, reactive and chronic but self-limiting reactive ulcer of the oral mucosa. Eosinophilic ulcers usually occur in the fifth and seventh decades of life with equal distribution between males and females; however, a slight male predilection was noted by Fonseca et al. [4] The entity brings several clinical differential diagnoses which include mainly malignancies, infectious diseases, autoimmune diseases or others. In this case histopathology shows the lining epithelium is partly ulcerated, the adjoining mucosa is hyperplastic and the lamina propria shows many polymorphic inflammatory infiltrate rich in eosinophil. In our case differential diagnoses included squamous cell carcinoma, non-Hodgkin's lymphoma and traumatic granuloma.

A rapid improvement of the ulcer occurred after the incisional biopsy similarly to what was described by other authors,[1, 2, 5] and the reason for that behavior is unknown. Accordingly to Eleni et al.[6] This behavior may be indicative that the healing response is reactivated to the surgical intervention. Different therapeutic approaches for eosinophilic ulcers have been reported in the literature, including a wait-and-see approach, antibiotics, topical, intralesional and/or systemic corticosteroids, curettage, cryosurgery and surgical excision. The most frequently performed therapy is simple surgical incision/ excision.[7] In this case after excision of ulcer and dental treatment topical steroid was given. Intralesional and systemic corticosteroids are not an option since deep fungal infection where considered as clinical hypothesis. Accordingly to el-Mofty et al.[8] in a review of 38 cases, suggest that cell-mediated immunity might play an important role in the pathogenesis of EU, since that topical, as well as systemic prednisone, is effective in the treatment of this disorder.

The latest survey reveals that these are CD30 positive cells originating from T lymphocytes. CD30 is a histological marker for lymphoproliferative disease, Hodgkin's lymphoma and RS cells. However, it also occurs in many non-neoplastic cutaneous disorders such as atopic dermatitis, adverse drug reaction, molluscum contagiosum and insects and spiders bites.[9] Segura and Pujol reviewed eosinophilic ulcer of the oral mucosa and considered it as a non-specific reactive pattern rather than a distinct entity.[10]

Surgical excision is the most commonly cited treatment procedure among the different therapies used. But surgical excision and topical steroid showed to be a reliable method with an excellent end result in this case report. Considering the benign and self-limiting behavior of EU, achieving a correct diagnosis and monitoring these patients are important to avoid possible overtreatment due to the difficulties that arise in the histopathological examination, since there is no specific hallmark of the disease and, therefore, the histopathological diagnosis of EU is by exclusion.[10] Monitoring is also important because low-grade lymphoma in routine microscopic exam, for example, may only be recognized retrospectively, when lesions recur several times, spread to other areas, or develop more pronounced malignant features microscopically.[11]

Conclusion

Diagnosis of Eosinophilic ulcer (EU) is made by the combination of clinical and histopathological features.

The pathogenesis of this condition remains uncertain and its histogenesis still remains controversial and this condition is characteristically self-limiting with a benign course. Different therapeutic approaches for eosinophilic ulcers have been reported in the literature, including a wait-and-see approach, antibiotics, topical, intralesional and/or systemic corticosteroids, curettage, cryosurgery and surgical excision. The most frequently performed therapy is simple surgical incision/excision.

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