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Original Research Paper

Dental Science

EOSINOPHILIC ULCER OF THE TONGUE: A DISTINCT ENTITY

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ABSTRACT Oral mucosa is the common site for ulcerations to occur and traumatic ulcers are frequent amongst them. Traumatic ulcerations occur in sites that may be injured by teeth, like tongue, lips and buccal mucosa. Traumatic ulcerative granuloma with stromal eosinophilia is a benign, self-limiting lesion with unknown etiology and presents as a round, elevated ulcers with indurated borders which is often confused with malignancy. Trauma to the oral mucosa results in surface ulcerations which usually heals within days but may remain for extended periods of time in certain instances. Herewith we report a case of eosinophilic ulcer with review of clinical, pathological and differential diagnosis.

KEYWORDS : Eosinophilic Ulcer, Traumatic Ulcerative Granuloma With Stromal Eosinophilia, Traumatic Ulcerations, Oral Ulcer

Introduction

Eosinophilic ulcer, also known as traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is considered to be benign, reactive, self-limiting lesion of the oral mucosa with unknown pathogenesis. It is known under a variety of names including traumatic granuloma of the tongue, eosinophilic ulcer of the mucosa, ulcerated granuloma eosinophilicum of the tongue etc. [1,2] TUGSE was described in children by Riga in 1881 and its histological feature was described by Fede in 1890. [1,3] In 1956, Popoff was first to describe TUGSE confined to oral mucosa in adult.[3]

Clinically it mimicks squamous cell carcinoma and due to its aggressive clinical course, it warrants a biopsy.[1] The diagnosis of TUGSE is difficult because different process may share similar clinical and histopathogical appearances.

Case Report

A 63 year old female patient reported to the hospital with the chief complaint of an ulcer in the left lateral border of the tongue since one month. History revealed that the patient had rheumatoid arthritis for the past 10 years and was under medication for the same.

On Intra-oral examination, irregular ulcer of size 1 x 1 cm was seen on the left lateral border of the tongue with irregular margins, floor of the ulcer was covered with yellowish slough (Figure 1).The surrounding mucosa was erythematous and margins were inverted. There was a mild swelling on the left side of the tongue. The ulcer was tender on palpation and there was no induration. Sharp tooth in relation to 35, 36, and 37 were evident. Single left submandibular lymph node was palpable, mobile and non-tender.

Due to the chronicity of the ulcer, and presence of sharp tooth, a provisional diagnosis of traumatic ulcer was made with a clinical differential diagnosis of non-healing ulcer of the tongue was given. Since the lesion was small about 1x1 cm, an excisional biopsy was performed under local anesthesia and hemostasis was achieved. Histopathological examination of H & E stained soft tissue section revealed hyperkeratotic stratified squamous surface epithelium with focal areas of discontuinity suggesting ulceration (Figure 2) . No evidence of dysplasia is seen. The underlying stroma shows mixed inflammatory infiltrate consisting of lymphocytes, plasma cells, neutrophils, few macrophages and blood vessels (Figure 3 & 4). Numerous binucleated cells with darkly stained eosinophilic cytoplasm suggestive of eosinophils are scattered throughout the connective tissue.

Based on the clinical and histopathological examination, a final diagnosis of Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) was given.

Discussion:

Oral mucosa is the common site for ulcerations to occur and traumatic ulcers are frequent amongst them. [4] Traumatic ulcerations occur in sites that may be injured by teeth, like tongue, lips and buccal mucosa.[5]

TUGSE is a chronic, slow growing, distinct clinical entity presenting as an ulcer with elevated margins. [6] The exact etiology of TUGSE is unknown. The role of trauma is considered to have a major role in the etiology of TUGSE although obvious trauma could be demonstrated in only 50% of cases.[3] Trauma may be due to malposed tooth, partial dentures, erupting teeth during nursing (Riga-Fede disease) ,overzealous tooth brushing, sharp tooth margins and very rarely in conditions related to self mutilation.[4] Velez at al considered trauma to be the main initiative factor for the development of TUGSE by enabling the entry of toxic agents and pathogens evoking a granulomatory inflammatory response. [4] In our case, the patient had a sharp tooth in relation to 36 which had induced the trauma and hence the lesion.

TUGSE is reported with slight female predominance in most of the cases with peak incidence between sixth and seventh decades of life. [9] Clinically, TUGSE manifests as a rapidly developing solitary ulcer, from few millimetres to several centimetres in diameter, with elevated and indurated borders. In our case, irregular ulcer of size 1 x 1 cm was seen on the left lateral border of the tongue with irregular margins, floor of the ulcer was covered with yellowish slough. The surrounding mucosa was erythematous and margins were inverted. The above said clinical features were suggestive of malignancy.[1]

Tongue is the most common location for the eosinophilic ulcer. The increased incidence of this lesion in the tongue which is more exposed to mastication explains the course of the lesion. [10] There had been instances where the ulcer may be related to enlarged regional lymph node as reported by El Mofty et al. [9]

The clinical differential diagnoses includes major apthous ulcers, several infectious disorders such as primary syphilis, oral tuberculosis, necrotizing bacterial lesions, granulomatous diseases like sarcoidosis, autoimmune disorders such as discoid lupus erythematosus ,malignancies such as squamous cell carcinoma, leukemia ,lymphoma and histiocytosis X .[3,11]

Based on the age, sex predilection, location and number of the ulcer and its clinical presentation, a provisional diagnosis of traumatic ulcer was made.

Histopathology shows a granulation tissue in the ulcer bed with a fibrinous exudates that supports mixed inflammatory cell infiltrate of lymphocytes, histiocytes, neutrophils and plasma cells intermixed with eosinophils often extending in to the deeper structures.[4] The granulation tissue usually shows increased number of capillaries with prominent endothelial cells.[12] Our case findings were also consistent with the above features.

Histological differential diagnosis includes atypical histiocytic granuloma (AHG) or pseudolymphoma and Angiolymphoid hyperplasia with eosinophilia (ALHE).

AHG are ulcerative lesions without a clear history of trauma and exhibits cellular pleomorphism, atypia and mitotic activity that often suggest an initial diagnosis of lymphoma. ALHE is associated with regional lymphadenopathy and blood eosinophilia and shows proliferation of hyperplastic vessels with characteristic vacuolated or plump endothelial cells. [4] However all the above lesions are excluded in this case by careful clinic-pathologic correlation.

Eosinophils are present in a variety of cutaneous and gastrointestinal conditions, often related to allergic, parasitic or drug related causes, or as a part of systemic eosinophilia.[13] However, most traumatic ulcers are devoid of eosinophils. It had been suggested that eosinophilic infiltrate is a tissue response to mucosal trauma due to introduction of unknown antigen in to the submucosa.[1] A possible direct pathogenic role of cytokine and chemotactic factors released by eosinophils has also been hypothesized. Interaction among mast cells, release of chemotactic factors and tissue eosinophilia has also been postulated.[9]

Sometimes, TUGSE also shows delayed self healing. This nature is explained by some authors as the lack of synthesis of tumor growth factor (TGF) by eosinophils in the inflammatory infiltrate. Along with this, eosinophils produce a wide spectrum of other cytokines such as Tumor Necrosis Factor (TNF) which enhances the tissue damage.[14]

The treatment of TUGSE includes oral antibiotics, topical, intra-lesional and/or systemic corticosteroids, cryosurgery and surgical excision.[15] The favourable antibiotic which can be used is that of penicillin. However, 0.1% triamcinalone acetonide can be effectively used as a topical steroid both in the form of oral paste and/or mouth wash.

Recurrence of TUGSE has been reported by Shen WR et al in two of his cases with CD 30+ cells and mononuclear arrangement of TCR-gamma chain gene. However, the recurrence was related to clinical failure of eliminating the possible causative factors. The recurrent lesions in his study demonstrated higher level of eosinophilic infiltrate than the initial lesion.[12] However, in our case, surgical excision was performed and there were no recurrences observed during the follow-up.

Conclusion

Our case presented here is of interest to the clinicians because it provides useful information on the clinical, pathological features, as well as suggesting the possible etiology of this uncommon lesion. As oral pathologists, eosinophilic ulcer should be kept in mind as one of the differential diagnosis, as it represents group of related disorders with overlapping clinical and histopathological features. Careful evaluation and diagnosis prevents aggressive surgical interventions. Further research in cases with recurrence is required as eosinophilic ulcer shows T cell clonality and shares common features with CD30+ lymphoproliferative disorders.

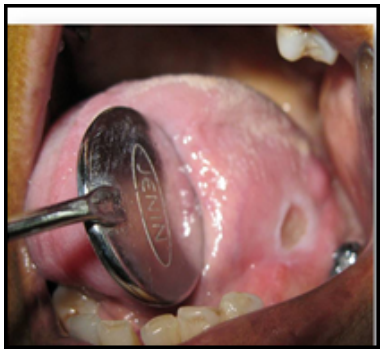

Figure 1: Clinical photograph showing an ulcer with raised margins on the lateral border of the tongue

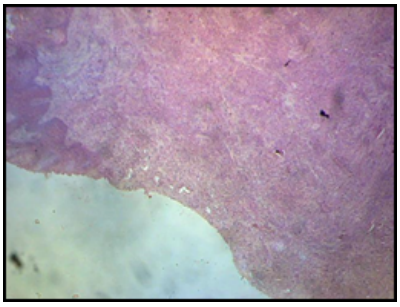

Figure 2: Histopathological picture showing stratified squamous surface epithelium with area of ulceration and the underlying connective tissue stroma.

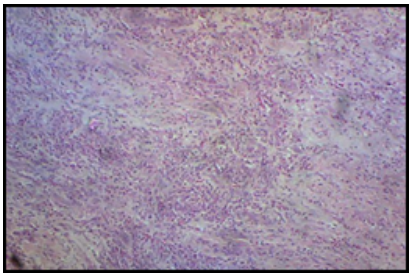

Figure 3: Histopathological picture (10X) showing granulation tissue with numerous lymphocytes, macrophages and eosinophils

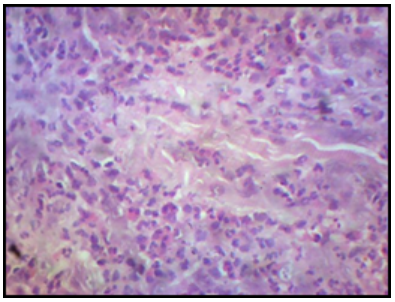

Figure 4: Histopathological picture (40X) shows mixed inflammatory cells with Eosinophils.

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