Traumatic Ulcerative Granuloma with Stromal Eosinophilia- A Mimic of Malignancy- Report of Two Cases with Review Of Literature.

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Abstract: Traumatic ulcerative granulomas are self limiting reactive lesions mimicking malignancy clinically and grossly, causing apprehension in the patient. The etiopathogenesis of these lesions is still uncertain though trauma is said to play a fundamental role often. CD 30 positivity of the large atypical cells seen in these lesions places them in the spectrum of CD 30 positive reactive lymphoprolifeative disorders.

Keywords: Ulcer, Oral mucosa, traumatic eosinophlic granuloma, CD 30 positive lymphoproliferative disorders.

I. Introduction

Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is one of the self healing reactive proliferations mimicking malignancy clinically and grossly[1]. They have recently been delineated a unique distinct entity [2]. Clinically they mimic malignancy raising anxiety in the patients, but are self healing lesions with low risk of recurrence. CD 30 positivity of these lesions places them in the spectrum of lymphoproliferative disorders [3]

II. Case Report

2.1 -Case 1

A 55yr old male presented with a complaint of a non healing oral ulcer present over the tongue for about two months duration. An edge biopsy of the lesion was done with a diagnosis of malignancy.

Histopathology revealed superficial fibrinopurulent exudate covering the ulcerated area with unhealthy granulation tissue infiltrated by dense collections of eosinophils, lymphocytes and plenty of large round to oval cells with eosinophilic cytoplasm and oval vesicular nuclei with prominent nucleoli. (Fig-1 and Fig-2)

The cells were seen infiltrating the underlying striated muscle bundles. Clinical history revealed that the patient was an epileptic who was taking treatment irregularly. He sustained the ulcer during one of the epileptic attacks and it persisted due to a repeated attack.

A diagnosis of Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) was made. An excision biopsy was done which showed the same picture. The wound healed completely and follow up for a period of 9months revealed no recurrence.

Immunohistochemistry was not performed as none of the markers are specific for the large atypical cells [5]

2.2 -Case 2

A 17 yr old male present with a growth over lower lip on the right side for the past one and half months. The lesion was excised and sent for HPE. Grossly we received a 1cm diameter gray brown soft tissue mass. Cut section was gray white and unremarkable.

Histopathology revealed an ulcerated lining epithelium with unhealthy granulation tissue infiltrated by eosinophils, lymphocytes and large cells with prominent nucleoli extending into the underlying muscle fibers.

A diagnosis of traumatic ulcerative granuloma with stromal eosinophilia was made despite the absence of a history of trauma.

III. Discussion

Traumatic ulcerative granuloma with stromal eosinophilia, also known as eosinophilic ulcer of the oral mucosa, esoinophilic granuloma, atypical histiocytic granuloma, Riga-Fede disease(in infants and children) is a disorder which has been delineated as a unique entity only recently ,though it was first described in 1956 in adults by Popoff and in children by Riga and Fede individually as early as 1881 and 1890.[4]

They are often single lesions varying in size from 0.3cms to 2.0cms in diameter. The common site of occurrence is the tongue with other sites being the lip, floor of the mouth, gingiva and palate. They are usually self limiting ulcers without any recurrence except when there is a known cause of repeated trauma. [3]

Histopathology reveals dense inflammatory cell infiltrate composed of eosinophils, lymphocytes and large atypical cells with vesicular nuclei and single nucleolus (histiocyte like cells) infiltrating the underlying muscle bundles.

Immunohistochemically these large cells show variable positivity for CD68, S-100, Factor XIII and vimentin. These cells also show variable positivity for CD 30, a marker originally described for RS cells and expressed commonly by activated B and T cells in certain lympho-proliferative disorders, suggesting that TUGSE is possibly a part of this spectrum. [3]

Unlike most traumatic oral ulcers which are devoid of eosinophils, TUGSE show prominent eosinophilic infiltrates which result due to release of eosnophilic chemotactic factors by mast cells. The cytokines and chemotactic factors released by these eosinophils contribute to the development of TUGSE. A lack of synthesis of Transforming growth factors by the eosinophils explains the delayed healing of these lesions. [1].

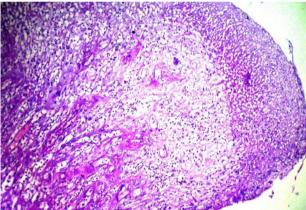
Clinically, diagnosis of this entity is difficult as many conditions have a similar appearance. (Table- 1) .Histopatholoically the closest mimics are Atypical histiocytic granulomas, Angiolymphoid hyperplasia with eosinophilia and Kimura disease . [1].

In Atypical histiocytic granulomas the infiltrate are more superficial and rarely extend into the underlying muscle.

In Angiolymphoid hyperplasia with eosinophilia, mucosal involvement is rare. The lesions show marked vascular proliferation with bizarrely shaped vessels and lymphocytic infiltrates forming follicles or aggregates.

Kimura disease is a chronic inflammatory process in which oral mucosal involvement is rare. They lack the large atypical cells seen in TUGSE despite having a prominent inflammatory infiltrate.

TUGSE are generally self limiting lesions. But various therapeutic modalities like wait and watch policy, intralesionsl steroids, Cryotherpay, Incision and excision biopsy have been advocated depending on the clinical setting.[6].



Fig—1 H&E, x 10x, showing superficial fibrinopurulent exudateand subepithelial granulation tissue with inflammatory cell infiltrate.

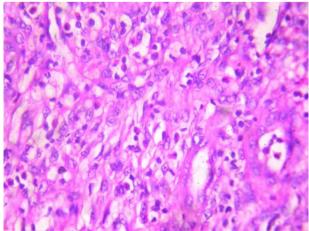


Fig-2. H&E stain, x 40x, showing eosinophils and large atypical cells.

Table-1 - Clinical differential diagnoses of Oral ulcers Infectious disease Tuberculosis Syphilis Histoplasmosis Autoimmune disorders Wegener's granulomatosis Discoid lupus eryhematosis Neoplastic Proliferations Squamous cell carcinoma Histicytosis X Lymphoma. Miscellanoeus TUGSE

IV. Conclusion

Traumatic ulcerative granulomas with stromal esoinophilia are benign lesions which closely mimic malignancy clinically, raising a lot of apprehension and anxiety in the patients. An awareness of this entity and histopathological examination of the oral ulcers helps confirm the diagnosis and alleviates patient's fear.

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