



CASE STUDY

TRAUMATIC ULCERATIVE GRANULOMA WITH STROMAL EOSINOPHILIA (TUGSE)

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ABSTRACT

Acute or chronic trauma to the oral mucosa may result in surface ulcerations. Eosinophilic ulcer or traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is a chronic benign, reactive and self-limiting lesion. Its etiopathogenesis is still uncertain but trauma seems to play a fundamental role. Clinically the lesion manifests as an isolated ulcer, showing a raised and indurated border in addition to a white or yellowish bed. Histopathologically, it is characterized by eosinophilic inflammatory infiltrate penetrating into the submucosal layers degenerating the underlying muscle. Hereby, reporting a case of a 62-year-old female patient who presented with a chief complaint of non-healing painful ulcer on the right ventral aspect of tongue. The lesion was excised and sent for histopathologic examination. Microscopic examination showed an ulcerated epithelium and the underlying connective tissue showed dense inflammatory infiltrate predominantly PMNs, lymphocytes, plasma cells and significant number of eosinophils. Based on microscopic findings, the lesion was diagnosed as "TUGSE".

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INTRODUCTION

Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is considered to be a benign, reactive and self-limiting lesion. It is known under a variety of names including traumatic granuloma of the tongue, eosinophilic ulcer of the oral mucosa and ulcerated granuloma eosinophilicum of the tongue (Kirammayi *et al.*, 2014). It exhibits a deep pseudoinvasive inflammatory reaction and is typically slow to resolve. Many traumatic granulomas undergo resolution after incisional biopsy. Clinically, the condition is characterized as a chronic, benign self limiting lesion of the oral mucosa, manifested as an ulcer with elevated margins with the tongue being the most common location. The rapid growth and indurated borders makes it mimic squamous cell carcinoma and that is where the condition derives its importance. Clinical features suggest that trauma plays a role in the development of the condition; especially that it is often observed on the tongue where traumas are frequent. (Bashar *et al.*, 2011) It can be diagnosed at any age, but most commonly it is found as a rapidly developing lesion in 4th - 5th decade of life. It can also occur in infants as well as in elderly people. Male to Female ratio is 1:1 or slightly more elevated in females. Most frequent location is on the tongue but can occur in other areas like lips, palate. Buccal mucosa and floor of mouth. Clinically, the

lesion presents as an ulceration with mild indurated borders and yellow fibrinous base. Microscopic examination revealed an ulcerated mucosa with underlying granulation tissue containing sheets of inflammatory cells, including eosinophils, lymphocytes, macrophages, plasma cells, neutrophils, and large atypical mononuclear cells, extending deep into the underlying soft tissue and between muscle fibers. Here, we report a case of a 62-year old female who had an ulcer in the right ventral aspect of the tongue since 25 days which turned out to be TUGSE after histopathologic confirmation.

Case report

A 62 year old female reported with pain in relation to the right ventral surface of tongue since 25 days. Patient has a history of panchewing since 5 years 5 times per day. On intraoral examination an ulcer was revealed on the right ventral aspect of the tongue extending from 45 to 46 region measuring 1.5 x 2 cm in size. The lesion was oval in shape reddish in colour that had indurated margins and was firm in consistency. No regional lymphadenopathy was noted. Sharp cusps were noted in relation to 46. Based on the clinical features a provisional diagnosis of traumatic ulcer was made. The histopathological section stained with H & E shows ulcerated epithelium. The remaining epithelium has hyperplastic stratified squamous epithelium of varying thickness (Fig.1).

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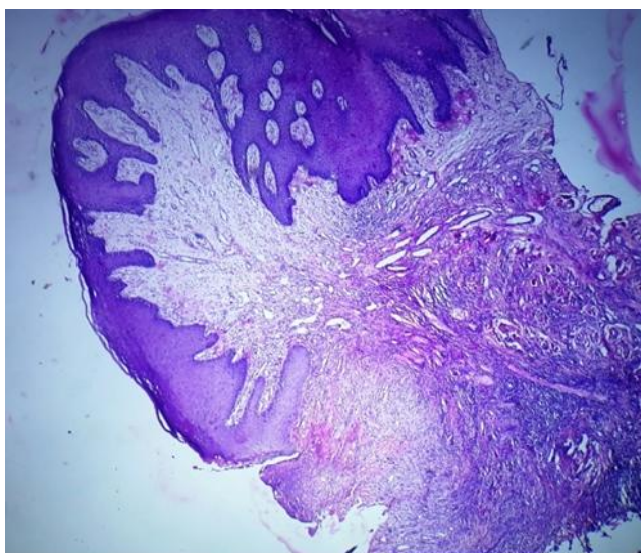


Fig. 1. Photomicrograph showing hyperplastic stratified squamous epithelium with ulcerated areas and lesional tissue (H&E, 4X)

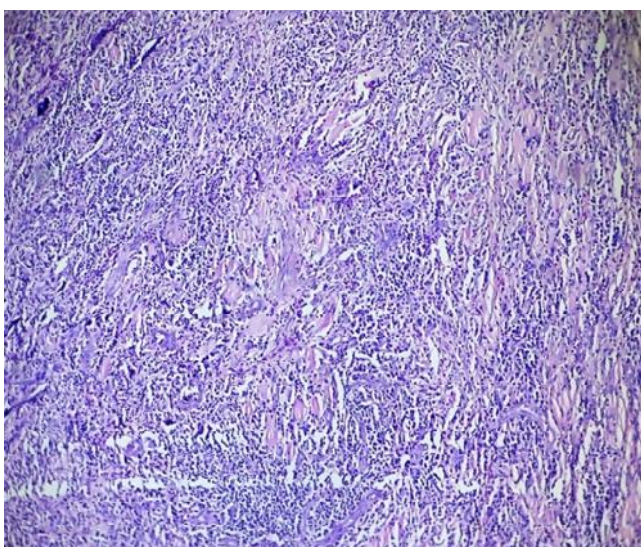


Fig.2. Photomicrograph showing connective tissue with inflammatory cells with PMN's, lymphocytes, plasma cells with significant number of eosinophils (H& E, 10X)

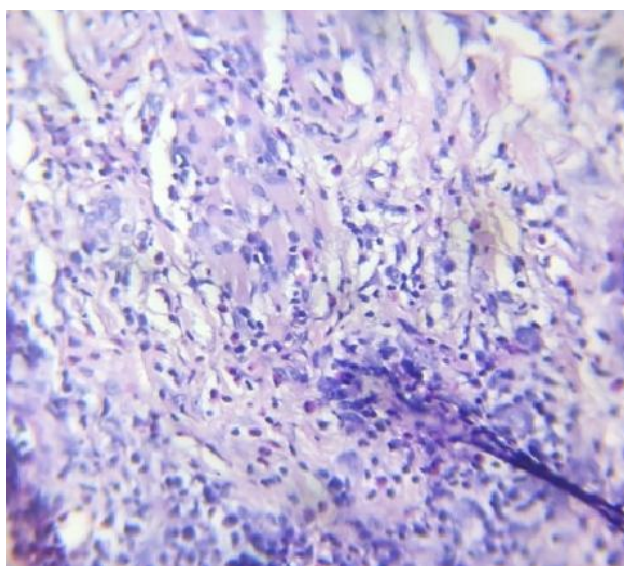


Fig. 3. Photomicrograph showing connective tissue with inflammatory cells with PMN's, lymphocytes, plasma cells with significant number of eosinophils (H& E, 40X)

The underlying connective tissue shows dense infiltration of chronic inflammatory cells with PMN's, lymphocytes, plasma cells with significant number of eosinophils (Fig. 2, 3). Degeneration of the muscle fibers are also evident. The section also exhibited numerous blood vessels.

DISCUSSION

Traumatic ulcerative granuloma with stromal eosinophilia, also known as eosinophilic ulcer of the oral mucosa, eosinophilic granuloma, atypical histiocytic granuloma is a disorder which has been delineated as a unique entity. The term was suggested by Elzay in 1993 to delineate the term eosinophilic ulcerations from more aggressive lesions such as the eosinophilic granuloma of histiocytosis X. TUGSE is a benign, asymptomatic, self limiting lesion of the oral mucosa. Clinically it may mimic squamous cell carcinoma. The etiology remains obscure and may be associated with trauma. Trauma may be due to malposed teeth, partial denture or sharp cusps. Cell mediated immunity plays an important role in its etiopathogenesis as it contains T lymphocytic infiltrate. (Chandra *et al.*, 2014) In infants the erupting teeth can cause sublingual ulcerations and is referred as Riga – Fede disease. It occurs usually in 1 week to 1 year of life. The lesions usually occurs in the anteroventral surface of the tongue caused with the erupting mandibular incisors. These associated teeth are usually natal or neonatal teeth. Atypical eosinophilic ulcerations is a rare lesion and exhibits sequential ulcerations, necrosis and self – regression. They are not associated with trauma and are believed to represent the oral counterpart of a T- cell cutaneous lymphoproliferative disorder (Alobeid *et al.*, 2004). Eosinophilic ulcerations can occur at any age with significant male predilection. Common sites are the anteroventral and dorsal surfaces of the tongue. Other sites are the gingiva, palate and mucobuccal fold. The ulcerations usually persist weeks to months and resemble traumatic ulcers. The center of the lesion is covered by a removable fibropurulent membrane with erythematous borders. 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. Ulceration resulting from trauma permits ingress of microorganisms, toxins and foreign proteins into the connective tissue which induces a severe inflammatory response resulting in an exaggerated mast cell eosinophilic reaction. Degranulation of the mast cell leads to release of eosinophilic chemotactic factor of anaphylaxis. 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. (Hirshberg *et al.*, 2006) Many different therapeutic approaches have been tried for TUGSE the most common being simple surgical excision and its recurrence is rare. The other treatment modalities are intralesional or oral corticosteroids, topical antibiotics, curettage and cryotherapy. The removal of traumatic agents was considered as the mainstay of the treatment. The lesion shows regression after the incisional or excisional biopsy. Prognosis of TUGSE found to be good. In our case the only treatment given was incisional biopsy and extraction of 46 and this showed regression of the lesion.

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