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Traumatic Ulcerative Granuloma with Stromal Eosinophilia (TUGSE): A Case Report

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Abstract

Eosinophilic ulcer of the oral mucosa is considered to be a benign, reactive, and self-limiting lesion, with unclear pathogenesis. However, trauma has been found to be a contributing factor in a majority of the cases. Clinically, it often presents as an isolated ulcer or an indurated submucosal mass. 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. TUGSE is rare and may be easily mistaken for a cancer or microbial infection, but it is self-limiting and tends to resolve spontaneously. Thus, awareness of this entity is important to emphasize the correct diagnosis of indurated ulcerated lesions and deliver appropriate and effective treatment.

Keywords: CD30, Eosinophilic ulcer, Riga-fede disease, Traumatic ulcer.

Introduction

Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is a chronic, benign, self-limiting lesion of the oral mucosa, manifesting as an ulcer with elevated margins. The most common location is the tongue, although other locations in the oral mucosa are possible. Various terms have been used to describe TUGSE, including eosinophilic ulcer, eosinophilic

granuloma of soft tissue, ulcerative eosinophilic granuloma, and traumatic ulcerative granuloma with stromal eosinophilia. In infants, TUGSE is called Riga-Fede disease. Histologically, TUGSEs show a diffuse polymorphic inflammatory infiltrate, rich in eosinophils, involving the superficial mucosa and the deeper muscle layer. Atypical large mononuclear cells scattered within the inflammatory infiltrate have been described in some cases, thus the term, atypical histiocytic granuloma. The pathogenesis of TUGSE is controversial. Although trauma was considered to have a major role in its pathogenesis, obvious trauma could not be demonstrated in most cases. We present a patient with eosinophilic ulcer affecting the right buccal mucosa.

Case description and results:

A 19-year-old male presented with a chief complaint of painless ulcer on right side of buccal mucosa since 5 months. Initially it was small in size with a gradual increase in size upto present size. He had a history of trauma on right side of buccal mucosa from the fixed orthodontic brackets placed since four months. He was apparently fit and well and not on any medication. Clinical examination revealed a large ulcer in the right buccal mucosa near to corner of mouth. The ulcer was measuring about 2×2 cm^2 with a yellow fibrinous base. No bleeding or pus discharge. On palpation the ulcer was smooth, non-tender, with well-defined margins. Borders were raised and thickened without induration. There was no fixation to the deeper structures. Palpable bilateral Level 1b lymph nodes, tender and mobile. Provisional diagnosis of a chronic traumatic ulcer was made. An excisional biopsy was performed under local anaesthesia for histopathological examination. Fixed orthodontic appliance was removed after consultion with the orthodontist. Microscopic examination showed superficial hyperplastic epithelium displaying hyperkeratosis with a central area of ulceration. Superficial epithelial cells exhibited mucopolysaccharide keratin dystrophy. The area of ulceration was infiltrated with dense mixed inflammatory cells chiefly composed of eosinophils and macrophages. These cells extend deep into the musculature with evidence of infiltrating the muscle fibers. The infiltrated tissue was well vascularized. No atypical cells were seen. Based on the clinico-pathologic features, a diagnosis of Traumatic Ulcerative Granuloma with Stromal Eosinophilia (TUGSE) was arrived. Complete healing was noticed after 1 month. No treatment was required other than regular observation and routine check-up. Figure 1showing raised and thickened ulcer on the right buccal mucosa and figure 2 showing one-month postoperative photograph after excisional biopsy.

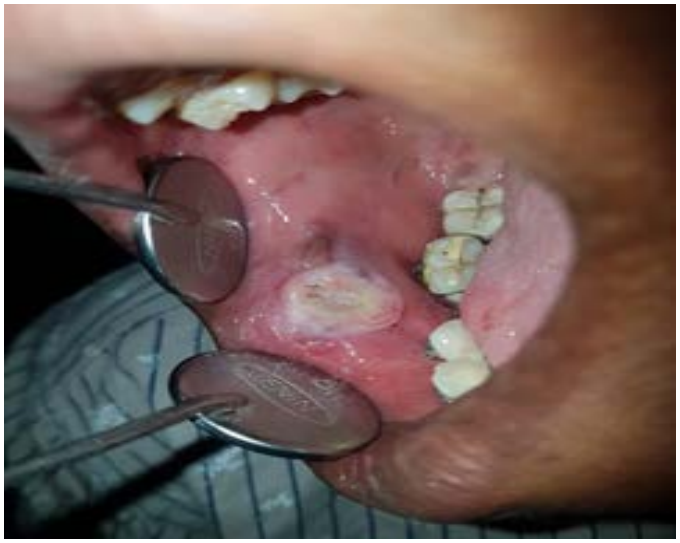


Figure 1: Clinical photograph showing raised and thickened ulcer on the Figure right buccal mucosa.

Figure 2: One-month postoperative photograph after excisional biopsy.

Discussion

TUGSE is a rare and unique lesion with uncertain nature, aetiology and pathogenesis. It is considered to be a benign, reactive and chronic but self-limiting reactive ulcer of the oral mucosa. TUGSE was originally described clinically in 1881 by Riga and histologically in 1890 by Fede. The term TUGSE was coined by Elzay in 1983[1]. in adult by Popoff in 1956, it was identified Described as a distinct entity by Shapiro and Juhlin in 1970[2]. This lesion has been known by different names, Riga-Fede disease in infants and neonates, sublingual granuloma, traumatic granuloma, eosinophilic granuloma, eosinophilic ulcer, and ulcerative eosinophilic granuloma [1,3].

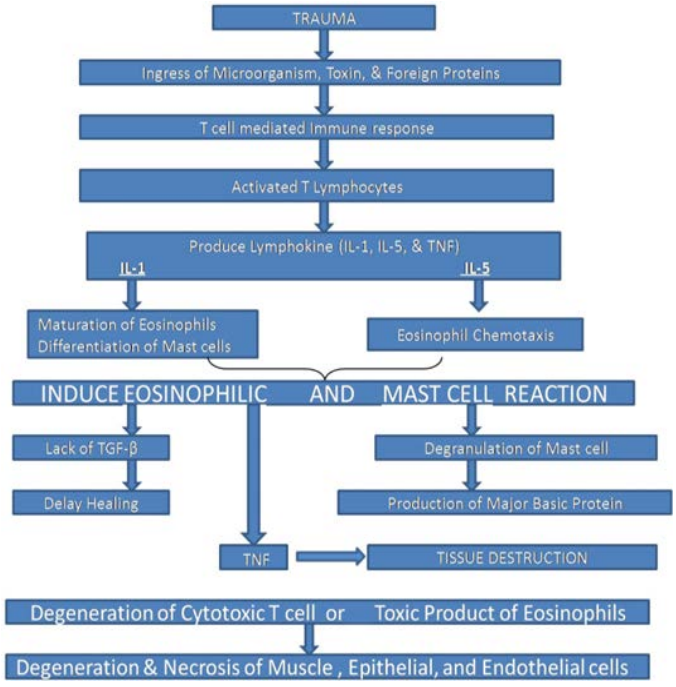
A bimodal age distribution, with the first peak occurring at early childhood and the second during the 6th and 7th decade of life, has been reported. No significant gender predilection has been highlighted, although some authors have evidenced a slightly higher female prevalence. The etiology of TUGSE is mostly unknown, although mechanical trauma has been considered as a typical trigger. Some authors suggested that trauma could be a contributing factor in the development of ulcers, by leading to microbial infections followed by intense inflammatory response. Others authors stated that cell-mediated immunity, CD30 lymphoproliferative disorders, mast cell- eosinophil interaction and stress may play a primary role in the pathogenesis of the lesion. Delayed healing of TUGSE lesions has been reported to be associated with the lack of secretion of transforming growth factor-α (TGF-α) and TGF-β by eosinophils infiltrating the lesions [4].

Clinically, the lesion appears as a singular entity. The lesion usually presents with raised, indurated borders and a yellow fibrinous floor [4,5]. It usually appears as a sharp, punched-out ulcer, often with a surrounding pale indurated rim [6] that is, clinically suspicious for malignancy. There may be exuberant polypoidal pyogenic granuloma-like granulation tissue. It may be present for weeks and may or may not be painful. Multiple ulcers may occur, either simultaneously or recurrently [7,8]. A case of multiple lesions has been reported in a young man affected by Riley-Day syndrome [4]. These ulcers are often self-healing but many may persist for weeks or longer [2.9]. Lymphadenopathy can occur but it is extremely rare [1]. Because of its clinical appearance as well as the frequent long-standing duration, a suspicion of malignancy is usually included in the differential diagnosis. Differential diagnosis of TUGSE may include mucosal burns (chemical or thermal), traumatic ulcer, malignancies (e.g., SCC or B-cell lymphoma), major aphthae (Sutton's disease), Bechet's syndrome, primary syphilis or the recently documented Epstein-Barr virus positive muco-cutaneous ulcer (EBV MCU), granulomatous disorders (tuberculosis, histoplasmosis, Wegener's granulomatosis and sarcoidosis), necrotizing bacterial infection, discoid lupus erythematosus and Langerhans cell histiocytosis [4].

Microscopically there is usually (as in this case) a deep ulcer with associated dense acute and chronic inflammation extending into deep underlying tissue, sometimes involving voluntary muscle. The infiltrate includes numerous lymphocytes and eosinophils, together with some normal histiocytes. Large, atypical, CD30-positive mononuclear cells may be present in some patients, raising the possibility that these lesions may be part of an evolving CD30 + lymphoproliferative disorder. Further investigations may then be indicated to exclude this possibility, depending on clinical evaluation, and any relevant history [9,10].

The inflammatory infiltrate in TUGSE exhibits sheets of atypical mononuclear cells (predominantly T-cell lymphocyte,population),eosinophils,macrophages/histiocytes, and few plasma cells. In earlier studies, immunohistochemistry revealed these atypical mononuclear cells to be belonging to the macrophage and myofibroblast lineage. More recent studies point toward the T lymphocytic function of these cells immunohistochemically, with positivity against all T-cell specific antigens. Of late, TUGSE is agreed upon as a reactive lesion dominated by predominant clonal T-cell population. However, this lesion requires more attention, owing to its tendency to mimic malignancy. The exact role of these inflammatory cells to produce such an exaggerated inflammatory response mimicking a malignancy is still unknown. Correlating various studies, this appears similar to a hypersensitivity type reaction (predominant T-cell infiltration exhibiting a delayed type of cell-mediated immunity. The ulcers often spontaneously heal after the biopsy. However, recurrence is common, particularly if there is persistent trauma. Where there is associated atypical epithelial hyperplasia, clinical follow up is important to exclude the possibility of developing malignancy.

Depicting pathogenesis of TUGSE [1].



Conclusion

The presence of TUGSE is rare in the oral cavity. This condition likely represents a group of related disorders with overlapping clinical and histopathologic features. diagnosis of TUGSE 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-healing with a benign course. We, as clinicians, must have adequate knowledge regarding TUGSE pathogenesis, and perform biopsy for a histological analysis along with a background immunohistochemistry technique, to rule off other neoplastic lesions at a very early stage.

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