

Traumatic ulcerative granuloma with stromal eosinophilia: A puzzle

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Abstract

Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is an ulcerative condition of the oral mucosa amid a chronic course and delayed healing. It is a reactive lesion that usually affects the tongue. TUGSE is a rare mucosal lesion to aid as an ulcer or an indurated submucosal mass and might cause diagnostic problems as it mimics a traumatic or neoplastic ulcer. However, chronic irritation from traumatic agents is well thought out to be a major initiating aspect. Histopathologically, it is categorized by an eosinophilic inflammatory infiltrate penetrating into the underlying muscle. It is significant to make out this mucosal lesion as it mimics malignant ulcers; the condition tends to resolve on its own. Thus, attentiveness of this entity is imperative to emphasize the accurate diagnosis of indurated ulcerated lesions and convey suitable and effective treatment. The present case highlights the clinical aspects, etiopathogenesis and histopathology of this unusual lesion.

Keywords: Anatomic pathology, eosinophilic ulcer, hematology, head and neck, traumatic ulcer

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INTRODUCTION

Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) was first described in an adult patient by Popoff in 1956, and the first case was reported in 1960. In 1970, Shapiro and Juhlin proposed this lesion as a distinct entity.^[1] Other terms have been used to describe the lesion: traumatic granuloma of the tongue and eosinophilic granuloma of tissue. The comparable entity in infants and neonates is called Riga–Fede disease as it was first clinically described in 1881 by Riga and then histologically in 1890 by Fede. TUGSE is an unusual self-limiting condition of the oral mucosa.^[2] The lesion manifests as an isolated ulcer that can be either asymptomatic or associated with mild-to-severe pain, and in most cases, it affects the tongue.

TUGSE lesions may mimic malignancy such as squamous cell carcinoma, CD30-positive lymphoproliferative disorder or infectious diseases such as primary syphilis, tuberculosis or Epstein–Barr virus mucocutaneous ulcer.^[3]

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 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

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of indurated ulcerated lesions and deliver appropriate and valuable treatment. The present case highlights the clinical aspects, etiopathogenesis and histopathology of this scarce lesion.^[4]

Histologically, owing to the presence of large histiocyte-like cells, the lesion is confused for atypical histiocytic granuloma, angiolymphoid hyperplasia with eosinophilia and Langerhans cell histiocytosis. Most cases heal without any intervention. Since a rift of these cases, particularly the recurrent ones, shows monoclonality for T0cell marker or CD 30, all the recurrent cases necessitate immunohistochemical assay for CD30 marker clonality and enduring follow-up.^[5]

Here, we intend to report a case of TUGSE sited on the lower right lateral border of the tongue, to review clinical and histopathological features and to point out some differential diagnoses and clinical workup of this entity.

CASE REPORT

A 65-year-old male presented with a chief complaint of ulcer on the right lateral aspect of the tongue for 1 year [Figure 1]. History of present illness revealed a small-sized ulcer in the initial phase of peanut shape which is gradually reached to the present size. The pain was moderate, continuous and radiating to the cheek and neck region which did not relieve after medication. The patient did give a history of local trauma by adjacent teeth. On clinical examination, an ulcer was seen on the right lateral border of the tongue, opposite teeth 45 and 46. Close inspection revealed an erythematous ulcer of 2 cm diameter with a whitish surrounding halo. On palpation, the ulcer was smooth, tender and firm in consistency, with well-defined margins among everted rolled and

induration. There was no fixation to the deeper structures. Submandibular lymph nodes were tender and palpable, while deeper cervical lymph nodes were nonpalpable. Provisional diagnosis of traumatic ulcer was made. An excisional biopsy was performed under local anesthesia for histopathological examination.

The possibility of malignancy was ruled out by ultrasonography. Microscopic examination revealed under scanner view/low-power view, two pieces of tissue showing irregular ulcerated epithelium overlying intensely inflamed connective tissue stroma. The deeper stroma shows a group of muscles fiber bundles with some areas showing inflammatory cell infiltration [Figure 2]. Under high-power view, it reveals that epithelium is stratified squamous parakeratinized type. The connective tissue stroma depicts loose to dense bundles of collagen fibers with plump to spindle-shaped fibroblasts. Intense chronic inflammatory infiltrate predominantly comprising lymphocytes, eosinophils and macrophages/histiocytes is evident. The inflammatory components appear to infiltrate deep into the underlying muscle fiber bundles. In some areas, mild degenerative changes are also absorbed in muscle fibers [Figures 3 and 4]. The clinical differential diagnosis may include traumatic neuroma, oral lymphangioma atypical histiocytic granuloma. A traumatic neuroma is a type of neuroma which results from trauma to a nerve, usually during a surgical procedure. The most common oral locations are on the tongue and near the mental foramen of the mouth. The lesion clinically visualized as TUGSE.

Oral lesions frequently appear on the tongue showing irregular nodularity demonstrating pebbly appearance and translucent hue owing to its superficial location and having gray, pink or yellowish color, which is also a pathognomonic feature



Figure 1: The clinical picture of ulcer on the right side of the lateral aspect of the tongue

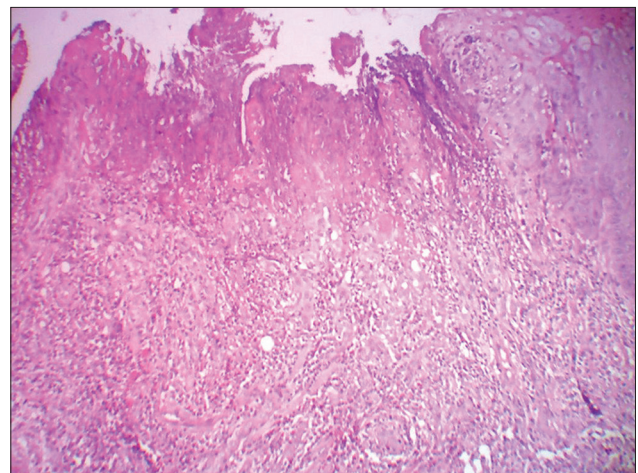


Figure 2: The histopathological photomicrograph showing discontinuous epithelium with underlying highly fibro cellular stroma. (H&E, ×4)

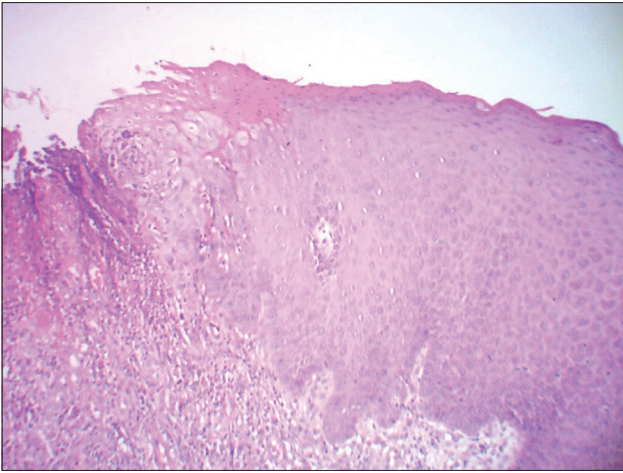


Figure 3: The histopathological photomicrograph showing epithelial proliferation at the border of the ulcerative lesion with underlying inflamed stroma. (H&E, x10)

of lymphangioma of the tongue. The lesions of atypical histiocytic granuloma are clinically similar to squamous cell carcinomas or specific granulomatous ulcerations. Several therapeutic approaches such as topical steroids, mouthwashes, topical antibiotics, curettage and cryotherapy have been reported.^[6] The most frequently performed therapy is simple surgical excision. No local recurrences are usually noted after excision.^[7,8] In our case, simple excision was performed and a rapid improvement of the ulcer was seen similar to what was described by other authors.^[9]

DISCUSSION

TUGSE acquaint like a chronic slow progressive lesion with fourth to sixth decades was most commonly reported.^[10] Clinically, most of them appear as a promptly budding ulcer with elevated or indurated margins.^[11] In addition, multifocal lesions, submucosal masses and recurrences enclose.^[12] It may be indicative associated with mild-to-severe pain in ensembles of cases, indicative of malignancy and infections such as deep fungal infections, tuberculosis and primary syphilis and this is where the situation becomes significant.^[13]

It can bear for several weeks or months and heals devoid of any treatment. This lesion occurs subsequently malposed teeth and partial denture. Chiefly, TUGSE appears to be more extensive in males than females (1.6:1). It appears like a red lesion in the oral mucosa with rolled, elevated and indurated margins by central areas of fibrinopurulent membrane mimicking squamous cell carcinoma. Even though the tongue is the chief common site, erstwhile regions such as the buccal mucosa, vestibular mucosa, gingiva, lip, mucobuccal fold, retromolar area and palate also may reveal TUGSE.^[14]

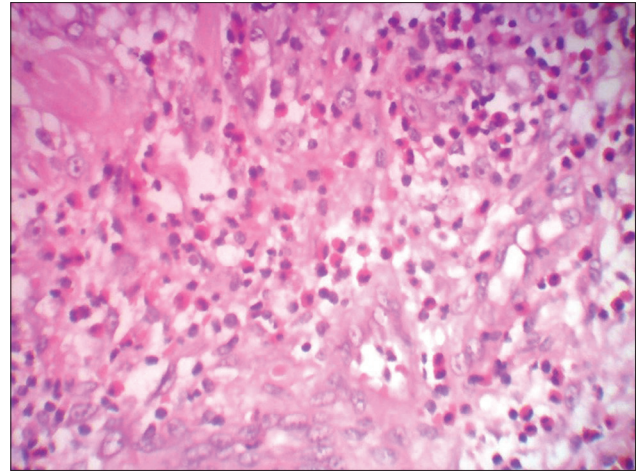


Figure 4: The histopathological photomicrograph showing chronic inflammatory cells like lymphocytes, eosinophils and histiocytes infiltrating around the muscle fibers. (H&E, x45)

Histopathologically, TUGSE reveals ulceration concerning diffuse polymorphic inflammatory infiltrate (exhibiting epitheliotropism) extends from the superficial epithelium into the deep connective tissue. This intense chronic inflammatory infiltrate includes small-round T-lymphocytes, large atypical mononuclear cells, B-lymphocytes massive eosinophils, granular macrophages, plasma cells and histiocytes within the superficial layer and deep muscle layers. Proliferating endothelial cells, blood vessels and focal areas of necrosis are seen drape all over the lesion.^[15] However, being there of enormous eosinophilia is not enforced in this lesion. A TUGSE lesion is often seen in infants.^[16] Others have recommended that persistent trauma leads to disparity of tissue antigens or introduces toxins, microorganisms, endogenous ruin products or foreign proteins into the tissues; moreover, event could provoke a local immune reaction.^[17] Some authors have even reported a probable interface among mast cells, release of eosinophil chemotactic factor and tissue eosinophilia.^[8] However, an fixed linkage among trauma and the development of such lesions was not established in the entire studies.^[18]

Due to an assortment of cells originate in inflammatory infiltrate, TUGSE is thought to be a benign, reactive lesion. El-Mofty *et al.* showed that cell-mediated immunity might play an important role in the pathogenesis.^[17]

Ficarra *et al.*^[19] were the earliest to report a case in which CD30+ cells were established in an ulcerated lesion histologically resembling TUGSE. The patient had multiple episodes of recurrent, self-healing eosinophilic ulcers in the oral mucosa. It was suggested that TUGSE of the oral mucosa may represent the oral counterpart of cutaneous CD30+ lymphoproliferative disorder. Alobeid *et al.*^[10] recently described three patients with oral TUGSE

containing CD30+ atypical cells in which T-cell clonality was demonstrated by using polymerase chain reaction analysis of the TCR γ chain gene. In one case, the lesion healed completely following incisional biopsy; in the other two cases, additional self-healing oral and extraoral lesions developed. Contrary to the findings of Ficarra *et al.*,^[19] and Alobeid *et al.*,^[10] believed that the lesions they reported represented a subset of cases of TUGSE that share common features with primary cutaneous CD30+ lymphoproliferative disorders.^[7]

In our case, the TUGSE was present on the right lateral border of the tongue measuring about 2 cm in diameter, with a glossy red surrounding area and pale yellow color of the floor. Its margins were indurated and tender on palpation. It occurred in a male patient, in the seventh decade of life, and presented without lymphadenopathy. The clinical differential diagnosis may include traumatic ulcer, traumatic neuroma, salivary gland tumors, primary syphilis, metastasis, atypical histiocytic granuloma and proliferative myositis. Histopathologically, TUGSE shows irregular ulcerated epithelium overlying intensely inflamed connective tissue stroma. The deeper stroma shows group of muscle fiber bundles with some areas showing inflammatory cell infiltration, in higher magnification epithelium showing squamous parakeratinized type, connective tissue stroma depict loose to dense bundles of collagen fibers with plum to spindle shaped fibroblasts. Intense chronic inflammatory infiltrate predominantly comprising lymphocytes, eosinophils and macrophages/histiocytes is evident. The inflammatory components appear to infiltrate deep into the underlying muscle fiber bundles. In some areas, mild degenerative changes are also absorbed in muscle fibers. In our case, simple excision was performed and a rapid improvement of the ulcer was seen similar to what was described by other authors.^[6,9]

CONCLUSION

It is significant to make out this mucosal lesion as it mimics malignant ulcers; the condition tends to resolve on its own. Thus, attentiveness of this entity is imperative to emphasize the accurate diagnosis of indurated ulcerated lesions and convey suitable and effective treatment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity

cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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