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TUGSE- CASE SERIES

Dental Science

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ABSTRACT

Background: Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is an uncommon benign, reactive lesion of oral mucosa that usually occurs on tongue. Etiopathogenesis of this lesion is uncertain, though trauma is considered to be possible predisposing factor in majority of the cases. Clinically it presents as an ulcer or submucosal indurated mass. Microscopically it shows mononuclear inflammatory cell infiltrate predominantly eosinophils. Here we report five cases of TUGSE emphasizing on its benign nature. This lesion is rare and may be easily mistaken for a cancer or microbial infection. The clinician's awareness of this entity is paramount in delivering an effective treatment.

Materials and methods: A total of five cases were reported to our department from 2012 to 2016. Data was retrieved from archives of the department.

Results: All the five cases were seen in the lateral border of the tongue, with sharp tooth as predisposing factor. For all the patient's incisional biopsy was performed after which the lesion regressed spontaneously and there was no recurrence.

KEYWORDS

Eosinophils, Incisional biopsy, Mast cell degranulation, Riga Fede disease.

INTRODUCTION:

TUGSE is a rare, benign, chronic self-limiting reactive lesion of the oral mucosa commonly manifesting as an ulcer of unknown etiology. The word TUGSE was first coined by Elzay in 1983. Various terms have been proposed in the literature to designate this lesion such as ulcerated granuloma eosinophilicum diutinum, traumatic granuloma of tongue, eosinophilic granuloma of tongue, eosinophilic ulcer of tongue, traumatic eosinophilic granuloma, and ulcerative eosinophilic granuloma of the tongue [1]. In infants, it is known as sublingual ulcer/sublingual granuloma/reparative lesion of tongue/Riga-Fede disease as it was first described by Riga clinically and Fede histologically in 1881 and 1890 respectively [2]. Etiopathogenesis of TUGSE is obscure but traumatic injury caused by accidental biting, chronic irritation by sharp teeth, misplaced/malposed teeth, fractured restorations and ill-fitting dentures have been implicated [3]. Some authors suggest that trauma could only be a contributing factor causing ulceration and letting the viral/toxic agents enter the underlying connective tissue triggering an inflammatory response. Usually the lesion presents as an irregular ulcer with rolled, elevated borders and the base of the ulcer may have yellowish exudate and can project outwards akin to a tumour. Tongue is the most commonly affected site and the lesion can be asymptomatic/occasionally associated with pain [4]. Microscopically, it is characterized by inflamed granulation tissue that extends deep in to the submucosa showing varying degrees of degeneration.

Here we report five cases of TUGSE emphasizing on its benign nature. This lesion is rare and may be easily mistaken for a cancer or microbial infection. The clinician's awareness of this entity is paramount in delivering an effective treatment.

CASE SERIES: A total of five cases had been reported to our institution from 2012-2016.

Case-1: 75 year old male patient reported with a chief complaint of pain in the left side of the tongue since ten days. Patient has a habit of smoking since 35 years. Examination revealed a solitary, firm, ulcer approximately size of 1.5x2 cm with indurated margins on left lateral border of the tongue. Histopathologically it shows hyperplastic ulcerated epithelium with spongiosis (Figure 1). Inflammatory infiltrate consists of lymphocytes, plasma cells, macrophages and few eosinophils, neutrophils. Inflammatory cell infiltrate extended deep into the connective tissue and caused muscle degeneration (Figure 2)

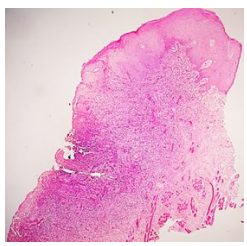


Figure 1: H&E stained sections in 4x view shows hyperplastic ulcerated epithelium with inflammatory infiltrate in the submucosa

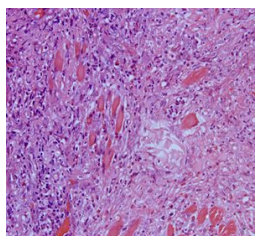


Figure 2: H&E stained sections in 40X view shows dense inflammatory infiltrate in the deep connective tissue and in between the muscle fibers and causing muscle degeneration.

Case-2: 55 year old male patient reported with a two months history of ulcer on the left side of the tongue. Patient has a habit of cigarette smoking since 10 years. Examination revealed solitary firm ulcer approximately 2x1 cm in size on left lateral border of the tongue. Microscopic examination showed hyperkeratotic parakeratinised stratified squamous epithelium with surface ulcerations (Figure 3). Inflammatory cell infiltrate consists of eosinophils, lymphocytes and plasma cells that extend deep into the connective tissue and invade the muscle causing degeneration (Figure 4).

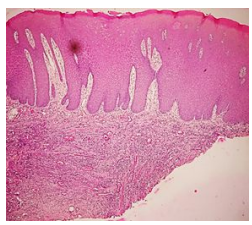


Figure 3: H&E stained sections in 4X view shows ulcerated hyperplastic epithelium with diffuse inflammatory infiltrate.

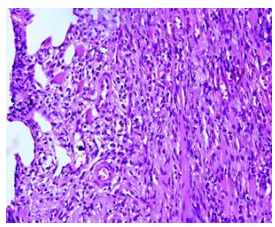


Figure 4: 40X view of H&E stained sections shows dense inflammatory cell infiltrate invading muscle fasciculi causing degeneration.

Case-3: 55 year old female patient reported with a chief complaint of ulcer on the right side of the tongue since 20 days. Examination revealed a solitary, firm ulcer approximately 0.5x0.5 cm in size on right lateral border of the tongue with elevated, sloping margins and floor covered with white slough. Microscopic examination revealed hyperplastic epithelium with spongiosis (Figure 5) and inflammatory cell infiltrate which extend into the submucosa and invade the muscle fasciculi, causing degenerative changes in the muscle (Figure 6).

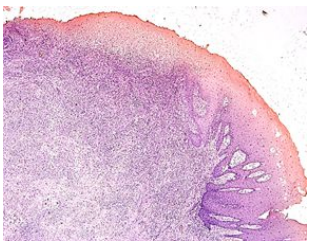


Figure 5: 4X view of H&E stained sections shows hyperplastic stratified squamous epithelium with underlying dense inflammatory cell infiltrate.

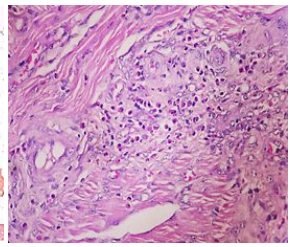


Figure 6: 40X view of H&Estained sections shows dense inflammatory cell infiltrate contains lymphocytes, plasma cells and eosinophils.

Case-4: 50 year old female patient with a chief complaint of pain in the left side of the tongue since 3 months. Examination revealed a solitary ulcer approximately of size 0.5x0.4 cm on the left lateral border of the tongue. Microscopically, specimen showed parakeratinised ulcerated epithelium (Figure 7) with mixed inflammatory cell infiltrate extending deep into the connective tissue causing muscle degeneration (Figure 8).

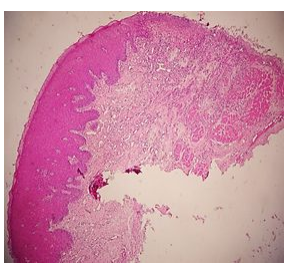


Figure 7: 4X view of H&E stained sections shows hyperplastic epithelium with surface ulcerations and underlying dense inflammatory infiltrate.

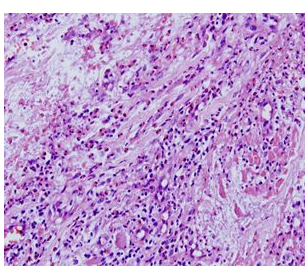


Figure 8: 40Xview of H&E stained sections shows inflammatory cell infiltrate in deep connective tissue invading muscle causing degeneration.

Case-5: 38 year old male patient reported with a chief complaint of inability to eat and swallow food due to pain on right side of the tongue since 15 days. Examination revealed a solitary ulcer approximately of size 1x1.5 cm on right lateral border of the tongue. Microscopic examination showed, ulcerated surface epithelium (Figure 9) with chronic inflammatory cell infiltrate extending into submucosa and between the muscle fasciculi thereby causing its degeneration (Figure 10).

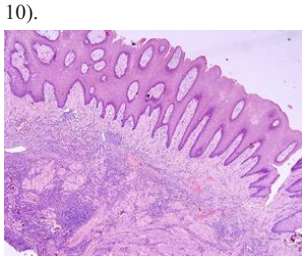


Figure 9: 4X view of H&E stained sections Hyperplastic stratified squamous epithelium with surface ulcerations and dense inflammatory cell infiltrate in the deep connective tissue.

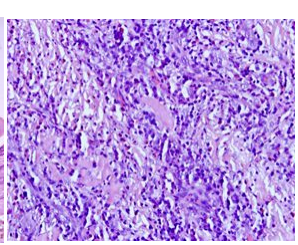


Figure 10: 40X view of H&E stained sections shows dense inflammatory cell infiltrate contains lymphocytes, plasma cells in the deep connective tissue and muscle degeneration

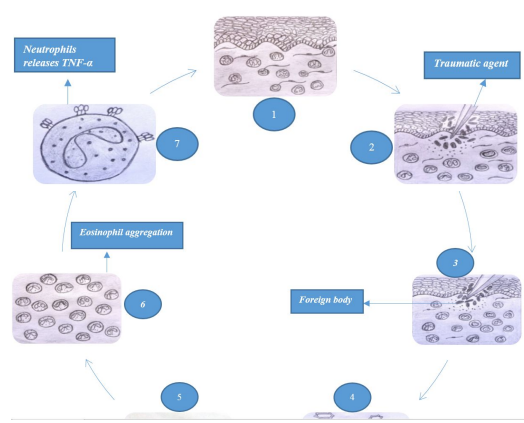
In all cases the site effected was the lateral border of the tongue, which was associated with a sharp tooth that act as predisposing factor causing irritation and ulcer, with no relevant medical history and no associated lymphadenopathy. Out of five cases provisional diagnosis of three cases were given as traumatic ulcer and remaining two cases as carcinoma of tongue because of the clinical appearance resembling the carcinoma. (The details of the cases are enlisted in table 1).

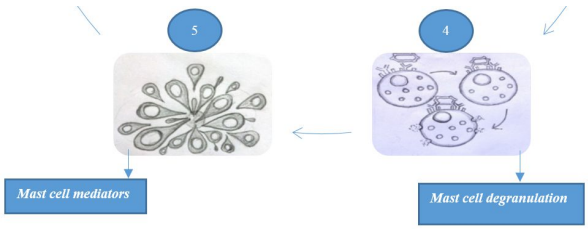
Table 1 showing the details of cases reported to our department

Case No	Age in years	Gender	Site	Duration	Etiological Agent	Lesion Description	Provisional diagnosis
1	75	Male	Left lateral border of tongue	Since 10 days	Sharp tooth irt 37	1.5x2cm Tender Ulcer with indurated margins	Traumatic ulcer
2	55	Male	Left lateral border of tongue	Since 2 months	Sharp tooth irt 38	2x1cm firm, tender ulcer	Carcinoma of tongue
3	55	Female	Right lateral border of tongue	Since 20 days	Sharp tooth irt 47	0.5x0.5cm firm, tender ulcer with elevated sloping margins, floor is covered with white slough.	Traumatic ulcer
4	50	Female	Left lateral border of tongue	Since 3 Months	Missing left lower teeth, upper supra erupted teeth impinges on tongue	0.5x0.4cm tender ulcer	Traumatic ulcer
5	38	Male	Right lateral border of tongue	Since 15 days	Sharp tooth irt 48	1x1.5 Cm sessile, tender ulcer with indurated raised margins	Carcinoma of tongue

PATHOGENESIS:

Diagrammatic representation of pathogenesis of TUGSE





TUGSE mainly a traumatic lesion caused by sharp teeth, fractured restorations, ill-fitting dentures (2) leading to ulceration, as a result of which there is ingress of viral/toxic agents/ foreign bodies (3). The foreign body acts as an antigen induces an inflammatory reaction leading to release of mast cells and eosinophils. Mast cell activation occurs when antigen binds with IgE antibody usually present on its surface eventually degranulation of mast cells (4) leads to release of mediators, like histamines, leukotrienes, eosinophil chemotactic factor (5), which causes inflammation and also attracts eosinophils (6) with the help of eosinophil chemotactic factor of anaphylaxis. Histamines and leukotriene's helps in contraction of the smooth muscle there by helps in vasodilation. Eosinophils release aryl sulfates and histamines, which inhibits slow reacting substances of anaphylaxis. This leads to suppression of basophils and mast cell degranulation and inhibit the release of other mediators of inflammation by the mast cells. Eosinophils also produce major basic protein i.e., tumour necrosis factor (TNF-α) (7), which causes tissue destruction.

DISCUSSION:

TUGSE is a unique entity of oral mucosa which is a lesion of unusual clinical behaviour, obscure etiology and pathogenesis. Synonyms to describe this lesion are eosinophilic granuloma of the soft tissue, traumatic granuloma of the tongue, ulcerative eosiniphilic granuloma, eosinophilic ulcer, eosinophilic ulcer of the oral mucosa, ulcerated granuloma with eosinophilicum of the tongue, ulcerated granuloma eosinophilicum diutinum of tongue [1]. In infants and neonates it is also called as cardarelli apthae [6] or Riga Fede disease as it was described by Riga clinically and Fede histologically in 1881 and1890 respectively [2]. It was first described by Popoff (1956) in adults [7]. Shapiro and Juhlin in 1970 considered it as distinct entity [8]. Most authors suggested that this lesion is traumatic in origin caused by accident bites, chronic irritation by sharp tooth, misplaced or malposed teeth, fractured restorations, ill-fitting dentures [6]. Velez et al suggested that trauma is only a contributing factor cause's ulceration and could lead to viral or toxic agents entering the underlying connective tissue and cause inflammatory response [9]. One third to one half reported cases showed the history of traumatic injury. Tongue is the most common site as it is exposed to trauma through its continuous movement [8]. This lesion is seen in all age groups from pediatric to geriatric patients. There is a slight female predilection. There is no geographical restriction or no racial preponderance [10]. Lymphadenopathy observed in extremely rare cases. In present study all cases showed sharp tooth as the etiological factor. In our cases mean age of 54 years with male: female ratio of 3:2 is observed. In all cases site of the lesion is lateral border of the tongue. There is no associated lymphadenopathy. Clinically lesions manifest as an ulcer with indurated and elevated margins covered with yellow fibrinopurulent slough. Lesion can range from few millimetres to few centimetres. Clinically our cases show solitary, firm ulcer of size ranging from 4mm to 2cm with indurated borders. In infants ventral surface of anterior tongue is the most common site due to trauma from newly erupted primary teeth [8]. According to Velez et al [9] and alobeid etal lesion is symptomatic in 17-100% of cases. Histopathologically lesions show inflammatory cell infiltrate predominantly of small lymphocytes, T and B cells, large atypical mononuclear cells and eosinophils. Studies by Elmofty et al in 38 cases showed large atypical cells was found to be myofibroblasts as they show positive for vimentin [11].

Differential diagnosis includes squamous cell carcinoma, atypical histiocytic granuloma, Langerhans cell histiocytosis, lymphocyte rich CD30+ lymphoproliferative disorder, angiolymphoid hyperplasia with eosinophilia [2].

This lesion is usually treated by conservative incisional or excisional biopsy, results in spontaneous resolution of the lesion with no further recurrence. Other treatment approaches includes topical steroids or mouth washes, intralesional or oral corticosteroids, topical antibiotics, curettage and cryosurgery. Incase of Riga Fede disease surgical removal of the anterior primary teeth is not recommended although it may resolve the lesion. NSAIDS or topical anaesthetics may be used to get temporary relief [11]

CONCLUSION:

TUGSE is a benign lesion of the oral mucosa with trauma as the predisposing factor. As it mimics oral cancer, clinico-histopathological data is essential to procure appropriate diagnosis and is important for conservative treatment. In most of the cases, the lesions heal spontaneously so there is no need for more radical surgery.

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No conflicts of interest related to this study.

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