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RESEARCH ARTICLE

TRAUMATIC ULCERATIVE GRANULOMA WITH STROMAL EOSINOPHILIA ASSOCIATED WITH ORAL SUBMUCOUS FIBROSIS- A RARE CASE

***,[1]Dr. Priya Lokanath, [2]Dr. Satheesha Reddy, B.H. and [3]Dr. Shilpa Patil**

[1]Dept. of Oral Medicine and Radiology, KVG Dental College and Hospital, Sullia (D.K.), India
[2]Dept. Of Oral Medicine and Radiology, AECS Maaruti College of Dental Sciences and Research Centre, Bangalore, India
[3]Shilpa Dental Care and Speciality Clinic, Bangalore, India

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ABSTRACT

Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is a benign lesion of the oral mucosa with an unclear pathogenesis. Clinically, this disease is characterized by the presence of chronic ulcerative lesion with elevated and indurated borders in the oral mucosa. It usually develops rapidly and may persist for several weeks or months. It presents mainly on the tongue but other areas of the oral mucosa such as gingiva, cheek and vestibular mucosa may also be involved. We describe here a rare case of Traumatic ulcerative granuloma with stromal eosinophilia of buccal mucosa associated with Oral submucous fibrosis.

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INTRODUCTION

Traumatic injuries of the oral mucosa are frequently observed in the oral cavity which can result in surface ulcerations. They may be acute or chronic in nature (Sivapathasundharam and Lavanya, 2005) and most of them usually heal within days. A histopathologically unique type of chronic traumatic ulceration of the oral mucosa is TUGSE which exhibits pseudoinvasive inflammatory reaction and is typically slow to resolve (Eleni *et al*., 2011). Rapid expansion with a clinical appearance of ulceration often provokes fear of malignancy despite its benign nature (Jason and Butler, 2017).

It is most often found on the tongue but can also occur on gingiva, buccal and vestibular mucosa (Azizi Taghi and Mohammad Hosein Kalantar Motamedi, 2009). TUGSE, clinically mimic squamous cell carcinoma (Bashar and Abdullah, 2011), but, histologically TUGSE shows a diffuse polymorphic inflammatory infiltrate, rich in eosinophils, involving the superficial mucosa and the deeper muscle layer (Budnick and Shiyong Li, 2009). The aim of this paper is to report a rare case of Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE), a reactive lesion associated with Oral submucous fibrosis.

***Corresponding author: Dr. Priya Lokanath**
Dept. of Oral Medicine and Radiology, KVG Dental College and Hospital, Sullia (D.K.), India.

CASE REPORT

A 29 year old male patient reported to the Department of Oral Medicine and Radiology complaining of a growth on the left cheek mucosa since 7 months. The patient noticed the growth since the commencement of orthodontic treatment (i.e. 7 months). It was initially a size of small bead and gradually progressed to the present size. Patient occasionally complained of pain in the growth while chewing food. He also had difficulty in opening the mouth since 1 year. The patient gave a history of chewing betel leaf and betel nut since 4 years. He also smoked around 4 cigarettes per day. On clinical examination, blanching of buccal mucosa was present bilaterally and a solitary sessile growth was present on the left buccal mucosa opposite to the second molar measuring about 1X1 cm in diameter. The surface appeared ulcerated and the ulcer was irregular in shape measuring about 0.5X0.5 cm in size with raised, rolled-up margins. The borders and floor of the ulcer were pale pink in colour. On palpation, the lesion was tender and the borders, base of the ulcer and the growth was firm in consistency with slight induration at the borders. The right buccal mucosa showed a white, keratotic area about 0.5X0.5 cm in diameter opposite to upper first molar which was non-tender on palpation. Indentation marks of the orthodontic arch wires and buccal tubes were present bilaterally on the buccal mucosa. Vertical palpable fibrotic bands were found bilaterally on the buccal mucosa and the

mouth opening was recorded to be about 35mm. Based on the chief complaint and clinical findings, provisional diagnosis of Traumatic fibroma of the left buccal mucosa and Frictional keratosis of the right buccal mucosa and Oral submucous fibrosis were made. A differential diagnosis of pyogenic granuloma and chronic exophytic ulcerative growth was given. Patient was advised for a complete blood count, bleeding and clotting time and all were under normal range. An excisional biopsy was performed, after an informed consent from the patient, under local anaesthesia and the tissue was subjected to histopathological examination. Histopathological examination of the section showed ulcerated epithelium in one area with loose and oedematous connective tissue and focal collection of chronic inflammatory cells with abundant stromal eosinophilia. An incisional biopsy was also performed on the same side cheek mucosa where palpable vertical fibrotic bands were present and the histopathological examination of the tissue revealed atrophic epithelium and muscle component in close proximity to the epithelium. The final diagnosis with the above histological and clinical findings was made as traumatic ulcerative granuloma with stromal eosinophilia associated with OSMF. Patient was advised to quit smoking and betel quid chewing. The post-operative healing period was uneventful.

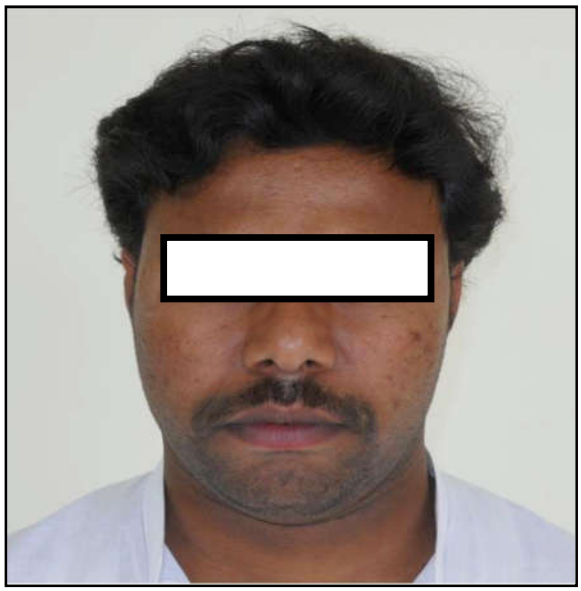

Fig. 1. Patient reported to the Department

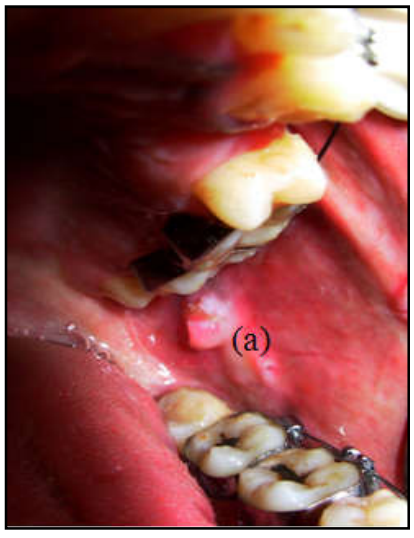

Fig. 2. The lesion on the left buccal mucosa presented clinically. Ulceration is present on the surface of the lesion at its apical part

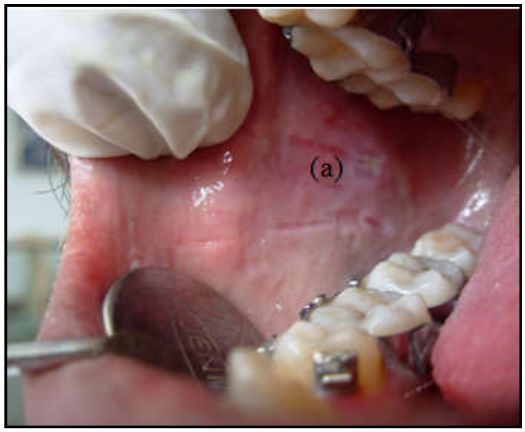

Fig. 3. White, keratotic area (a) opposite to upper first molar with indentations of Orthdontics wire

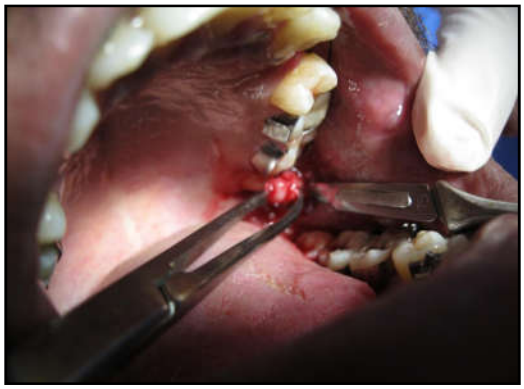

Fig. 4. Excision of the lesion for histopathological examination

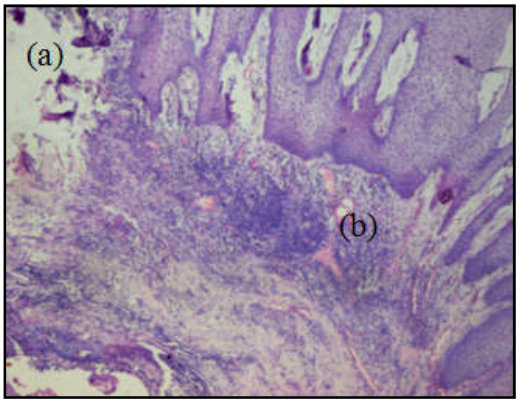

Fig. 5. Histopathological picture of the lesion (10X) showing break in the continuity of the epithelium (a); Blood vessels in the connective tissue stroma

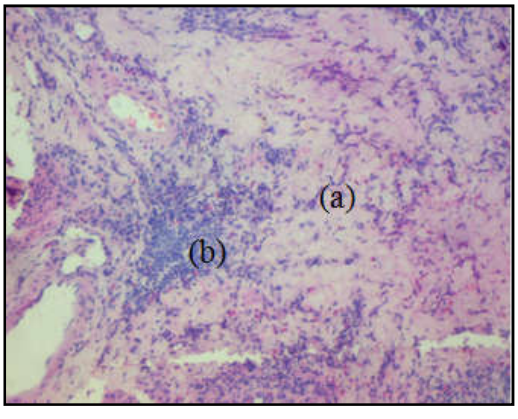

Fig. 6. Histopathological picture of the lesion (40X) showing dense connective tissue stroma(a) with abundant inflammatory cells(b)

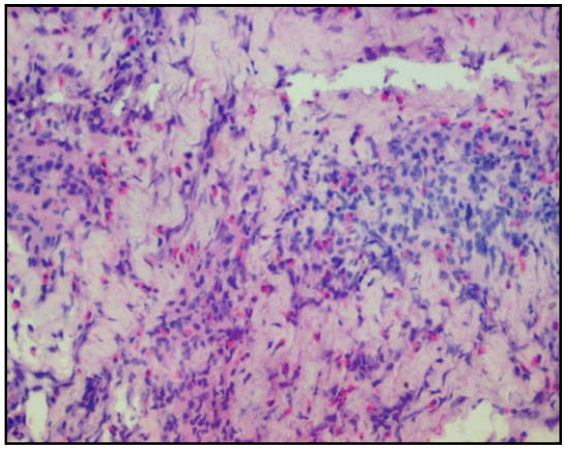

Fig. 7. Histopathological picture (40X) shows abundant Eosinophilia in loose connective tissue stroma

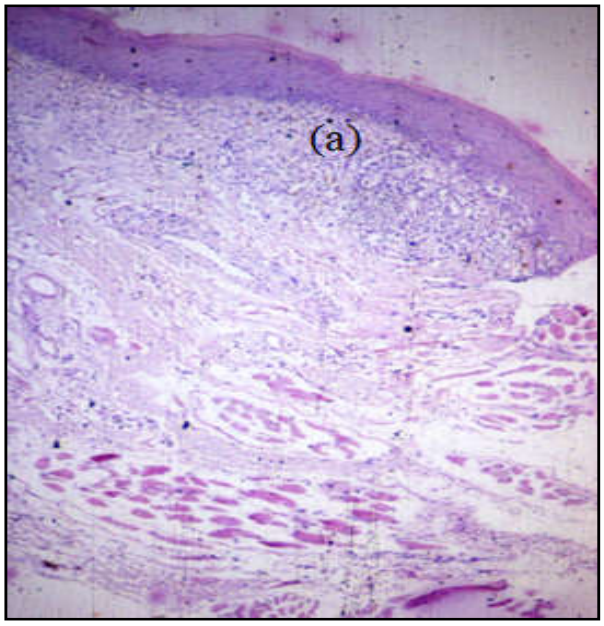

Fig. 8. Shows histopathological picture (10X) of the site where vertical fibrotic bands were palpable. Atrophied epithelium is seen with loss of rete ridges (a)

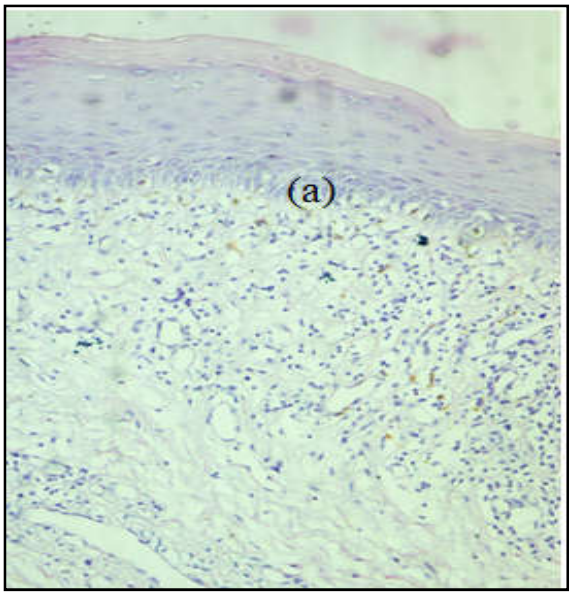

Fig. 9. Shows histopathological picture (40X) of the site where vertical fibrotic bands were palpable. Atrophied epithelium is seen with loss of rete ridges (a)

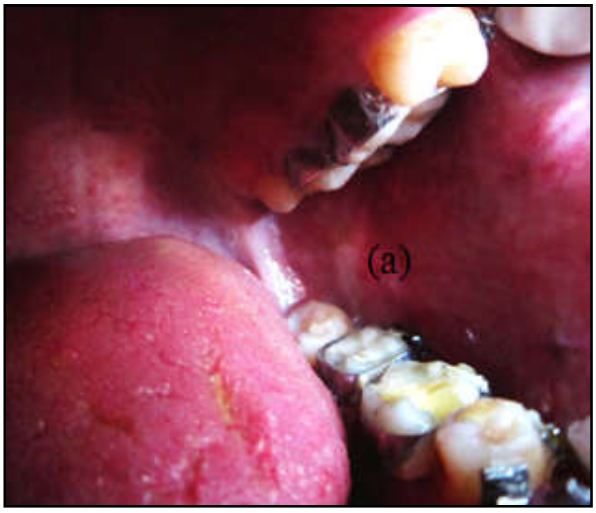

Fig. 10. Post operative healing (a) after 2 months

DISCUSSION

Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is a typically self-limited lesion of the oral mucosa with unclear pathogenesis (Eleni *et al*., 2011; Budnick and Shiyong, 2009). A variety of other terms have been used to describe this lesion, including eosinophilic ulcer, eosinophilic granuloma of soft tissue, traumatic eosinophilic granuloma, ulcerative eosinophilic granuloma, and Riga-Fede disease. It commonly manifests as a rapidly developing ulcer with elevated and indurated borders that persists for several weeks or months. It is most often found on the tongue but can occur in other areas, including the gingiva, buccal mucosa, and vestibular mucosa (Kühl Sebastian *et al*., 2008; Alobeid *et al*., 2004). Clinically, TUGSE may mimic squamous cell carcinoma, needing a biopsy or excision. Rapid healing after a biopsy or excision is typical. Histologically, these lesions are characterized by a dense, mixed, inflammatory infiltrate with prominent eosinophilia and a large, atypical, mononuclear cell infiltrate that extends deeply into the underlying muscle and soft tissues (Budnick and Shiyong Li, 2009).

The presence of eosinophils is not completely understood because most traumatic oral ulcers are devoid of eosinophils. It has been suggested that eosinophils represent a tissue reaction to some unknown antigen introduced via mucosal breakdown following trauma (Budnick and Shiyong Li, 2009; Sateesh S. Chavan and Purushotham Reddy, 2013). In this typical case we present the lesion as one of the cause being the tissue reaction to the antigens from the orthodontic wires (Ni-Ti) which would have entered the connective tissue through an ulcer caused due to trauma. Past studies examining the immunohistochemical profile of the large atypical cells have suggested a myofibroblastic or histiocytic origin (Budnick and Shiyong Li, 2009). Recent studies have demonstrated expression of CD30 and T cell–lineage antigens, suggesting the atypical mononuclear cells are of T-cell origin. Moreover, a few case reports have demonstrated molecular evidence of T-cell clonality within TUGSE, raising that possibility that a subset of these lesions could be classified as low-grade CD30+ T-cell lymphoproliferative disorders (Segura *et al*., 2006). The implication by recent case reports in the literature that TUGSE may represent the oral counterpart of primary cutaneous CD30+ T-cell lymphoproliferative disorders warrants further

understanding of the findings of T-cell clonality and CD30 positivity in TUGSE (Shen *et al*., 2013).

Conclusion

The occurrence of TUGSE along with OSMF has not been reported in the literature till date. The association itself is a rare one as the occurrence of granuloma, which contains proliferating fibroblasts, sprouting blood vessels and elongated rete ridges, while OSMF presents with a histological picture of loss of rete ridges and epithelial atrophy which are contradictory to each other. The association of these two lesions simultaneously has been a hallmark in the present case and further studies about the development of this lesion have to be done to know the exact aetiology of the occurrence.

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