

CASE REPORT

Eosinophilic Ulcer Of Tongue - A Rare And Confusing Clinical Entity

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ABSTRACT

Introduction: Eosinophilic ulcer is a rare, chronic, benign and often self limiting lesion of the oral mucosa. The ulcer most frequently occurs on the tongue and is characterized by the presence of indurated borders resembling malignancy. It is considered to be a reactive lesion with a benign clinical course and has been known by a number of terms, including eosinophilic ulcer, eosinophilic granuloma of tongue, traumatic granuloma, atypical histiocytic granuloma and traumatic ulcerative granuloma with stromal eosinophilia (TUGSE). Although trauma is considered as an etiologic factor, less than 50% of the patients recall the history of trauma. Similar lesions are seen in infants on the ventral surface of the tongue due to trauma from newly erupted primary incisors. This entity was first described in 1881 by the Italian physician, Antonio Riga, and subsequently published by F. Fede in 1890, hence it is known as *Riga-Fede disease*. Oral mucosal ulceration which does not resolve within two weeks is troublesome and presence of induration leads to suspicion, as it could mimic oral squamous cell carcinoma. This is the main clinical feature of Eosinophilic ulcer, a lesion surrounded with uncertainty regarding its nature, etiology and pathogenesis.

Case Report: This is a case report of a 65 year old female patient with eosinophilic ulcer on the lateral border of the tongue. The ulcer healed rapidly after an incisional biopsy and topical steroid application. The final diagnosis was achieved following clinical and histopathological examination.

Conclusion: In conclusion, it can be pointed out that though TUGSE is a self limiting lesion, its accurate diagnosis is important as it mimics oral squamous cell carcinoma clinically.

KEYWORDS: Eosinophilic ulcer, TUGSE, Infiltration

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INTRODUCTION

TUGSE is a reactive, benign, asymptomatic, self-limiting lesion of the oral mucosa first described by Antonio Riga, that clinically may mimic Squamous cell carcinoma.^{1,2} Though the etiology is quite obscure, trauma may be one of the factor causing TUGSE. Trauma may be due to malposed teeth or a partial denture. It affects any age group and has a slight male predilection. The common site of occurrence is anteroventral and dorsal surfaces of the tongue and may also be seen in gingiva, palate and mucobuccal fold.^{3,4} The lesion is covered by a removable yellow fibropurulent membrane with erythematous borders. Because of its close resemblance to SCC, histopathologic evaluation of the lesion is important. In this paper we have presented an interesting case of TUGSE occurring on the lateral border of the tongue.

CASE PRESENTATION

A 65-year-old female patient presented with a chief complaint of an ulcer on left side of her tongue since 20 days (figure-1). The ulcer was insidious in onset, small in size and gradually increasing in size to reach the present size. There was no history of trauma or noticing of any white or red patch or fluid filled blisters prior to the onset of ulcer. There was no history of bleeding or pus discharge. The ulcer was associated with pain, which was mild, intermittent and localized and no aggravating or relieving factors were present. There was no history of restricted tongue or jaw movements since the onset of ulcer and no history of difficulty in chewing and swallowing food. There was no history of recurrent ulcers in the oral cavity or elsewhere in the body. There was no other relevant history. Patient's medical history was remarkable, as she was a diabetic and was on oral hypoglycemic drugs for the same. Extra-oral examination revealed a palpable solitary, left submandibular lymph node, with no local rise of temperature, measuring about 1x1cm in size, oval in shape, soft in consistency, non-tender and not fixed to the underlying structures. The skin over the node appeared normal. No ipsilateral or contralateral distant lymph nodes were palpable. On intraoral examination, left maxillary second molar was supraerupted with grade III mobility and was impinging on the ulcer. On examination of the tongue, a solitary, round ulcer was evident on the left posterior lateral border of the tongue, measuring about 1.0 x 1.0 cm with raised edges and the floor was covered with yellow fibrinous exudate. The ulcer was surrounded by hyperkeratotic mucosa (figure 2). The ulcer was non-tender on palpation, edges and base of the ulcer were indurated. Tongue movements were not restricted. On routine investigation, patient's complete blood picture and blood sugar level were within the normal range. An incisional biopsy followed by histopathological examination was performed.

Differential diagnosis

Based on the history and clinical examination differential diagnosis of - Traumatic ulcer, eosinophilic ulcer, malignant ulcer, major aphthous ulcer.

On H&E staining, the sections showed hyperplastic stratified squamous epithelium which was ulcerated in one area and there was intense inflammatory infiltrate in the underlying connective tissue consisting predominantly of lymphocytes and eosinophils, extending deep into the muscle layer (figure 3 and 4). These features are suggestive of traumatic ulcerative granuloma with stromal eosinophilia. Thus a final diagnosis of eosinophilic ulcer of tongue was made.

Treatment

Extraction of left maxillary second molar was carried out and the patient was advised topical application of 0.1% triamcinolone acetonide paste three times daily for one week

Outcome and follow-up

Patient came for follow-up on 8th day after incisional biopsy. Resolution of the ulcer was observed

DISCUSSION

Eosinophilic ulcer of the oral mucosa was first described in adults by Popoff in 1956 and in 1970, it was identified as a distinct entity by Shapiro and Juhlin.^{1,2} Eosinophilic ulcer is considered to be a benign, reactive and self-limiting lesion of the oral mucosa. It usually occurs in the 5th and 7th decades of life with equal distribution between males and females; however a slight male predilection was noted by Fonseca FP et al.² it is known by different names such as eosinophilic ulcer, traumatic eosinophilic granuloma of tongue, traumatic granuloma, atypical histiocytic granuloma and traumatic ulcerative granuloma with stromal eosinophilia (TUGSE).³ The tongue is the most commonly involved site in about 60% of the cases and induration is a constant finding⁴ The other sites that may be involved include the buccal mucosa and labial mucosa, floor of the mouth, and the vestibule, where underlying skeletal muscle is found.³ Similar ulcers occurring in infants, on the ventral surface of tongue due to trauma from erupted primary incisors is seen in Rega Fede disease^[6] Trauma, though considered to be a major etiologic factor,

could be observed only in 50% of the cases.⁷



Figure 1 Figure 2 Figure 3 Figure 4

Figure 1: Extraoral view of the patient; **Figure 2:** Ulcer with raised edges and covered with yellow fibrinous exudate; **Figure 3:** H and E section showing abundant eosinophils; **Figure 4:** H and E section showing inflammatory infiltrate extending deep into the muscle layer.

According to few authors, trauma leads to viral and toxic agents entering the underlying tissue causing inflammatory response.⁸ Most of the oral traumatic ulcers are devoid of eosinophils and contain polymorphous infiltrate, whereas prominent eosinophilic infiltrates are observed in these lesions with a possible role of cytokine and chemotactic factors released by eosinophils in its development.⁹ The exact mechanism of pathogenesis still remains obscure. The ulcer is characterized by raised or punched out borders, with surrounding erythema or keratosis. The floor of the ulcer is usually covered with yellow fibrinous exudate and the surrounding tissue is indurated. All these features along with rather quick development clinically mimic squamous cell carcinoma and makes biopsy necessary to rule out malignancy. Lymphadenopathy can be observed in extremely rare cases.¹⁰ In the present case, a solitary ulcer was present on the left postero-lateral surface of the tongue, measuring about 1x1 cm in diameter, surrounded by keratosis. Its margins and base were indurated covered with yellow fibrinous material associated with left submandibular lymphadenopathy. In the case presented here, the sections stained with H&E revealed a dense and deeply infiltrative lymphoproliferation, showing epitheliotropism and massive eosinophilia. Sections showed sheets of lymphocytes and histiocytes along with hyperplasia of the vascular connective tissue leading to elevation of the surface ulceration. However complete resolution of the lesions after incisional biopsy has also been noted.^[11] Recurrences are not common, although some believe surgical removal is necessary to achieve a complete resolution. Different therapeutic

approaches for eosinophilic ulcer have been reported in literature, these include wait-and-see approach, antibiotics, topical, intralesional and/or systemic corticosteroids, curettage, cryosurgery and surgical excision. The most frequently performed therapy is simple surgical incision/excision.⁹

CONCLUSION

In conclusion, it can be pointed out that though TUGSE is a self limiting lesion, its accurate diagnosis is important as it mimics oral squamous cell carcinoma clinically.

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