ACTA SCIENTIFIC DENTAL SCIENCES

Volume 9 Issue 2 February 2025

Case Report

Traumatic Ulcerative Granuloma with Stromal Eosinophilia (TUGSE)-A Rare Case Report of Diagnostic Dilemma

Bhuvan Nagpal[1], Rujuta Patil[2], Aparna Pathak[3], Geetpriya Kaur[4]* and Shivani Ramesh Mungala[5]

[1]*Director and Sr. Oral Pathologist, Tohana Manglam Diagnostics, Tohana and Centre for Oral Pathology and Maxillofacial Diagnostics (COPMD)™, Hisar, Haryana, India*

[2]*Periodontist, Former Post Graduate Student, Department of Periodontology, The Oxford Dental College, Bangalore, Karnataka, India*

[3]*Consultant, Paradise Diagnostics, New Delhi, India*

[4]*Director, Paradise Diagnostics, New Delhi, India*

[5]*SVS Institute of Dental Science, Mahabubnagar, Telangana, India*

***Corresponding Author:** Geetpriya Kaur, Director, Paradise Diagnostics, New Delhi, India.

DOI: 10.31080/ASDS.2025.09.1969

Received: December 23, 2024

Published: January 04, 2025

Abstract

Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is a rare, benign, and self-limiting oral mucosal lesion characterized by a persistent, painful, and indolent ulcer. The exact etiology remains unclear, but it is thought to be associated with local trauma or irritation. In this case report, we present the clinical, histopathological, and management aspects of a patient diagnosed with TUGSE. A 70-year-old female patient presented with a chief complaint of a painful ulcer on the left side of the anterolateral aspect of the tongue for the past 10 days. She gave a history of pain being moderate, continuous, and radiating to the cheek. There was no relief post-medication. The histological analysis displayed hyperkeratosis and areas of ulceration with dense inflammatory cells like eosinophils and lymphocytes. There was also evidence of these cells infiltrating tongue musculature. Based on the clinicopathologic features, a diagnosis of Traumatic Ulcerative Granuloma with Stromal Eosinophilia (TUGSE) was rendered. Complete healing was noticed after 1 month. No treatment was required other than regular observation and routine check-ups. The close resemblance of TUGSE to Oral Malignancies makes it challenging to diagnose. Thorough clinical examination and histopathological analysis serve as essential components of the diagnosis of TUGSE. These reactive lesions are self-limiting and respond well to conservative treatment with desired healing.

Keywords: Eosinophilia; Reactive Benign Lesion; Ulcerated Epithelium; Tongue

Introduction

Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is a rare, benign, reactive, and self-limiting lesion usually associated with trauma. A few non-trauma-related TUGSE cases are also reported [1]. Clinically, TUGSE presents as a rapidly developing solitary ulcer in oral soft tissue, majorly on the tongue. Other common sites are buccal mucosa, gingiva, floor of the mouth, retromolar area, lip, and palate [2,3].

The lesion generally persists from one week to one year [4]. It is characterized by rolled borders mimicking squamous cell carcinoma [5]. Mild to severe pain is commonly associated with the ulcer. TUGSE usually regresses on its own after excision of the oral lesion. The ulcer size varies between 0.5 cm to 6.5 cm. Both genders are typically affected, with no specific gender predilection. TUGSE can be prevalent in all ages, but mainly with two peak incidences. It can be noted in babies below two years of age, mostly associated with teething, and fourth to seventh decades of adult life [1].

Histopathological examination generally reveals surface ulceration and dense and mixed inflammatory cell infiltration in the connective tissue. Few eosinophils are typically found in the submucosa, but they can sometimes penetrate between the muscle fibers and minor salivary glands [6].

Case Presentation

A 70-year-old female presented with a chief complaint of a painful ulcer on the left side of the anterolateral aspect of the tongue for the past 10 days. She gave a history of pain being moderate, continuous, and radiating to the cheek. Intraoral clinical examination revealed a single erythematous ulcer measuring about 1× 1 cm with well-defined margins and induration. On palpation, the ulcer was smooth, tender, and firm in consistency. There were no signs of any cervical lymphadenopathy. Her medical history was unremarkable. A conservative excisional biopsy was performed under local anesthesia and the specimen was sent for histopathological examination.

Gross examination showed a single soft-tissue specimen measuring 0.7 cm x 0.8 cm. Histologically, hematoxylin and eosin (H and E) stained soft-tissue section revealed an ulcerated, hyperplastic stratified squamous surface epithelium displaying hyperkeratosis with associated fibrovascular connective tissue. The areas of ulceration were infiltrated with dense mixed inflammatory cells chiefly composed of eosinophils and lymphocytes (Figures 1A, B). These inflammatory cells extend deep into the musculature of the tongue with evidence of infiltrating muscle fibers. The infiltrated tissue was well vascularized. No atypical cells were seen (Figures 2A, B).

Based on the clinicopathologic Eosinophilia features, a diagnosis of Traumatic Ulcerative Granuloma with Stromal Eosinophilia (TUGSE) was rendered. Complete healing was noticed after 1 month. No treatment was required other than regular observation and routine check-ups.

Discussion

Popoff initially reported Traumatic Ulcerative Granuloma with Stromal Eosinophilia (TUGSE) in 1956, and the first case was reported in 1960. However, research by Shapiro and Juhlin in 1970 led to the recognition of TUGSE as a separate entity [7]. TUGSE is an uncommon but benign ulcerative lesion of the oral mucosa. While oral TUGSE lesions mostly affect the tongue, they can also affect other oral mucosal sites like the buccal mucosa, retromolar region, floor of the mouth, vestibular mucosa, gingiva, and palatal mucosa [8]. While several etiological factors, including sharp tooth margins and ill-fitting dentures, have been suggested, trauma has been determined to be the primary cause of TUGSE [7]. Studies have indicated that males are more likely to present with this rare phenomenon than females [8]. However, Elzay et al., after examining 41 clients, concluded there was no gender predilection and reported that females were as equally affected as males [9].

As a pathognomonic feature of tongue lymphangioma, oral lesions often present as irregular nodularity with a translucent hue and pebbly appearance due to its superficial location; the lesions of atypical histiocytic granuloma are clinically similar to squamous cell carcinomas or specific granulomatous ulcerations. The lesions can also be gray, pink, or yellowish in color [10]. Microorganisms and frequent toxins infiltrate the injured location after severe ulceration. These substances induce an excessive inflammatory response, which attracts eosinophils and mast cells to the area. Furthermore, the production of eosinophilic chemotactic components of anaphylaxis by mast cell degranulation draws in more eosinophils. Histamines and aryl sulfates secreted by these eosinophils prevent mast cell degranulation, inhibit basophils, and limit the production of further inflammatory mediators [7].

The H and E-stained sections in our case showed an ulcerated, hyperplastic stratified squamous surface epithelium with hyperkeratosis and associated fibrovascular connective tissue. Deeply into the tongue musculature with evidence of muscle fiber infiltration, these inflammatory cells were infiltrated with dense mixed inflammatory cells, primarily eosinophils and lymphocytes (Figures 1A, B). Our results aligned with those of Lakkam., *et al*., who reported that inflammatory cells like lymphocytes, eosinophils, and macrophages/histiocytes penetrated deeply into the underlying muscle fiber bundles [10]. The infiltrated tissue was extremely vascularized and contained no atypical cells (Figures 2A, B). Furthermore, Shen et al. found that eosinophils were uniformly distributed throughout the muscle fibers in these types of injuries; our results were consistent with their findings [11].

The case was diagnosed as TUGSE, based on clinicopathologic characteristics. Complete healing was noticed after one month. Other than routine check-ups and monitoring, no treatment was necessary.

Although uncommon, TUGSE is benign and readily confused with SCC, CD30-positive LPD, or infectious illnesses like EBV-associated mucocutaneous ulcer or primary syphilis [12-15]. The appearance of eosinophils can help differentiate TUGSE from other conditions like histiocytosis X, granuloma faciale, allergic reactions, angiolymphoid hyperplasia with eosinophilic lymphoid granuloma, eosinophilic fascitis, and insects or parasites [16,17].

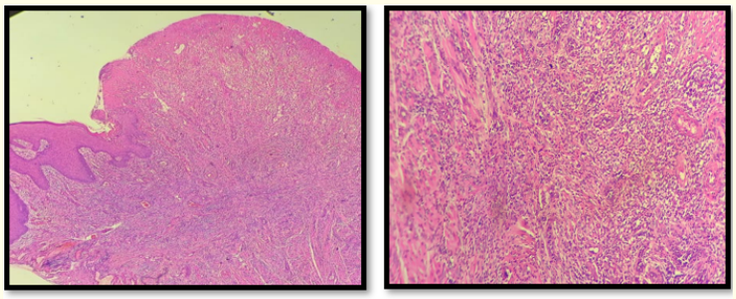

Figure 1A Figure 1B

Figure 1: (A) The histopathological photomicrograph exhibited an ulcerated, hyperplastic stratified squamous surface epithelium with hyperkeratosis (H and E, 4x), (B) Dense mixed inflammatory cells, mostly eosinophils, and lymphocytes, were deeply infiltrated into the tongue musculature, exhibiting signs of muscle fiber infiltration (H and E, 10x).

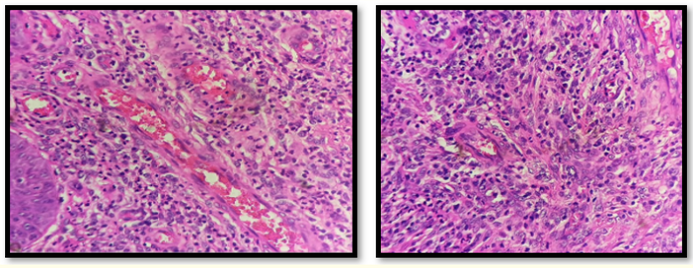

Figure 2A, B: The invaded tissue, which was highly vascularized, did not contain atypical cells (H and E, 40x).

Treatment options for TUGSE lesions include antibiotics like penicillin, removal of the traumatic agent, and excision if the lesion is still bothersome. Several therapeutic techniques have been reported, such as topical steroids, mouthwashes, topical antibiotics, curettage, and cryotherapy. The primary therapeutic intervention is surgical excision. Usually, no local recurrences have been observed post-excision [10]. In the event of Riga-Fede disease, appropriate management of the offending tooth is advised [16]. Liquid nitrogen, oral antibiotics, topical and systemic steroids, electrocoagulation, radiation, and 0.1% triamcinolone acetonide mouthwash are additional treatment options [17]. Most cases show rapid healing after surgery, but a few of them show a retarded healing phase due to the lesion's aggressiveness. Although TUGSE has a good prognosis, a lengthy follow-up of at least two years is required [18].

Conclusion

TUGSE is one of the rare presentations of ulcerative reactive oral lesions. Its resemblance to malignancies and infectious diseases makes it challenging to diagnose. Meticulous clinical examination combined with histopathological evaluation form essential components for the diagnosis of TUGSE. It should be included in the differential diagnosis of Ulcerative oral lesions resembling oral squamous cell carcinoma. TUGSE is self-limiting post-biopsy, usually responds well to surgical intervention with no adjuvant aids, and satisfactory healing can be achieved.

Bibliography

1. Mohamad AF., *et al*. "A Case Report of Traumatic Ulcerative Granuloma with Stromal Eosinophilia (TUGSE): Clinical and Histopathological Diagnostic Challenges". *Cureus* 15.11 (2023): e48481.

2. Shen WR., *et al*. "Oral traumatic ulcerative granuloma with stromal eosinophilia: a clinicopathological study of 34 cases". *Journal of the Formosan Medical Association* 114 (2015): 881-885.

3. el-Mofty SK., *et al*. "Eosinophilic ulcer of the oral mucosa. Report of 38 new cases with immunohistochemical observations". *Oral Surgery, Oral Medicine, Oral Pathology* 75 (1993): 716-722.

4. Farooq S., *et al*. "Traumatic ulcerative granuloma with stromal eosinophilia: a clinical study". *International Journal of Applied Dental Sciences* 4.2 (2018): 80-82.

5. Shen WR., *et al*. "Oral traumatic ulcerative granuloma with stromal eosinophilia: a clinicopathological study of 34 cases". *Journal of the Formosan Medical Association* 114 (2015): 881-885.

6. Pushpa TS and Balamurugan R. "Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE): a rare presentation and case report". *The Canadian Journal of Dental Hygiene* 56.1 (2022): 39-41.

7. Shapiro L and Juhlin EA. "Eosinophilic ulcer of the tongue report of two cases and review of the literature". *Dermatologica* 140 (1970): 242-250.

8. T-cell receptor gene rearrangement and CD30 immunoreactivity in traumatic ulcerative granuloma with stromal eosinophilia of the oral cavity". *American Journal of Clinical Pathology* 132 (2009): 722-727.

9. Elzay RP. "Traumatic ulcerative granuloma with stromal eosinophilia (Riga-Fede disease and traumatic eosinophilic granuloma)". *Oral Surgery, Oral Medicine, Oral Pathology* 55 (1983): 497-506.

10. Lakkam BD., *et al*. "Traumatic ulcerative granuloma with stromal eosinophilia: A puzzle". *Journal of Oral and Maxillofacial Pathology* 25 (2021): S42-45.

11. Shen WR., *et al*. "Oral traumatic ulcerative granuloma with stromal eosinophilia: a clinicopathological study of 34 cases". *Journal of the Formosan Medical Association* 114 (2015): 881-885.

12. Marszalek A and Neska-Dlugosz I. "Traumatic ulcerative granuloma with stromal eosinophilia. A case report and short literature review". *Polish Journal of Pathology* 62.3 (2011): 172-175.

13. Ficarra G., *et al*. "Traumatic eosinophilic granuloma of the oral mucosa: a CD30+ (Ki-1) lymphoproliferative disorder?" *Oral Oncology* 33.5 (1997): 375-379.

14. Dojcinov SD., *et al*. "EBV positive mucocutaneous ulcer--a study of 26 cases associated with various sources of immunosuppression". *The American Journal of Surgical Pathology* 34.3 (2010): 405-417.

15. Feller LL., *et al*. "Oral squamous cell carcinoma in relation to field precancerisation: pathobiology". *Cancer Cell International* 13.1 (2013): 31.

16. Sivapathasundharam B and Lavanya S. "Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE)". *Journal of Oral and Maxillofacial Pathology* 9 (2005): 30-33.

17. Kumar SK., *et al*. "Indurated tongue lesion". *The Journal of the American Dental Association* 139 (2008): 159-161.

18. Sarangarajan R., *et al*. "Traumatic ulcerative granuloma with stromal eosinophilia - Mystery of pathogenesis revisited". *Journal of Pharmacy and Bioallied Sciences* 7.2 (2015): S420-423.