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Case Report

Eosinophilic Ulcer of the Tongue in Smokeless Tobacco "Shamma User": Case Series Report

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Abstract: Oral eosinophilic ulcers (OEU) are rare, benign, reactive, and often self-limiting lesions. As the lesion is self-limiting, it develops rapidly, usually disappearing after several weeks. Although the exact pathogenetic mechanisms are unknown, the possibility that trauma may play a role has been postulated. The lesion frequently misleads to the suspicion of squamous cell carcinoma because of its prolonged duration. Despite trauma being suggested as the main culprit of oral eosinophil ulcers, the exact pathogenesis mechanism of this disease remains controversial. However, it may misdiagnose as malignancy, particularly in smokeless tobacco "Shamma users." The study emphasizes the clinical and histopathological aspects of diagnosing and treating this pathology. The study aims to demonstrate the importance of knowledge and its course. The novelty of our study can be described in terms of thorough early clinical diagnosis with confirmatory histopathological findings that can aid towards perfect diagnosis, as part of treatment - removal of etiological factors (trauma) saw the Oral eosinophilic ulcers (OEU) completely healed, which was earlier can be misdiagnosed as malignancy based on the use of Shamma smokeless tobacco by patients for a longer period. We present the two male cases of Saudi residents with a long history of painful ulcers on the lateral borders of the tongue. Both had a long history of being "Shamma users" of smokeless tobacco. The ulcers were ulceration with indurated and elevated borders and no evidence of regional lymph node involvement. Histopathologically, the lesion showed an infiltrate with several eosinophils, was ulcerated, and no cellular atypia was observed. After the treatment, the ulcer rapidly healed with an incisional biopsy and the cause removal. A definite diagnosis was achieved by combining histologic findings and clinical follow-up. A biopsy is not only the best approach for a proper diagnosis but also for its therapeutic management. Understanding this disease is necessary since its lesion can mimic an oral squamous cell carcinoma occurring as an indurated ulceration coupled with its frequent occurrence on the tongue.

Keywords: Diagnosis, Eosinophilic ulcer, Oral mucosa, Oral ulcerations

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1. INTRODUCTION

Oral eosinophilic ulcer (OEU) is an inflammatory disease of the oral mucosa featuring painful ulceration with a tendency to heal spontaneously. This lesion is also known as traumatic ulcerative granuloma with stromal eosinophilia [1-3].
Traumatic injury of oral mucosa can occur by the

- Mechanical
- Chemical
- Thermal
- Factitious
- Radiation
- Eosinophilic ulcer (Traumatic Granuloma)

OEU occurs predominantly in individuals between their 5th to 6th decades, but there are also cases reported in young individuals. Both males and females are affected with equal frequency. [4]. The cause and pathogenesis mechanism of this disease is obscure. Oral Eosinophilic Ulcer OEU) usually affects the tongue but may involve any other site of the oral mucosa[5]. Clinically, it presents as a chronic ulcer with indurated borders that tend to persist for several weeks only to progress into remission, usually after a surgical procedure, such as a biopsy. The biopsy procedure accelerates the healing process and is usually performed due to the resemblance of this lesion to a carcinomatous ulcer. [4]. OEU is considered a reactive lesion with a benign clinical course and has been known by several terms, including eosinophilic ulcer, eosinophilic granuloma of the tongue, traumatic granuloma, atypical histiocytic granuloma and traumatic ulcerative granuloma with stromal eosinophilia (TUGSE)[6]. Traumatic granuloma (TG) was originally described clinically by Riga[7] and histologically by Fede[8]. The term 'Traumatic ulcerative granuloma with stromal eosinophilia' (TUGSE) was coined by Elzay in 1983. However, trauma is considered an etiological factor, and a viral or toxic agent has been implicated in its pathogenesis. However, there is no definitive experimental proof. Oral mucosal ulceration that does not resolve within 2 weeks is troublesome, and the presence of induration leads to suspicion as it could mimic oral squamous cell carcinoma. The main clinical feature of eosinophilic ulcers is a lesion surrounded by uncertainty regarding its nature, etiology, and pathogenesis[9]. Despite trauma being suggested as the main culprit of oral eosinophil ulcers, the exact pathogenesis mechanism of this disease remains controversial. It occurs most commonly on the tongue and presents a clinically chronic, well-demarcated ulcer, which may mimic a squamous cell carcinoma. However, it may misdiagnose as malignancy, particularly in smokeless tobacco "Shamma users." The study emphasizes the clinical and histopathological aspects of diagnosing and treating this pathology. The objective of the study is to demonstrate the importance of the disease's knowledge and its course.

1.1. Ethical Statement

Permission was obtained from the institution to conduct and publish the case study. Participants were explained the study's purpose, and written informed consent was obtained from both participants before conducting the study. The study was conducted according to the guidelines of the Declaration of Helsinki for biomedical research involving human subjects.

2. CASE REPORT 1

A 75 years old Saudi male presented and was referred to our oral medicine clinic for evaluation of a painful lesion on her tongue for more than three months (Figs. 1). The lesion had raised and indurated edges and was large ulceration. The latest episode began as an erythematous plaque with minor pain and evolved into a severe deep ulcer over a few days. From then on, it continued to be painful, preventing regular feeding. Intraoral examination revealed a solitary ulcer was about (1.5X2 cm). The Patient has been a smoker and Shamma user for over twenty years. Therefore, we suspected squamous cell carcinoma (SCC). The medical history was positive for diabetic mellitus (DM) and hypertension (HTN). No extra oral abnormality was detected. A clinical differential diagnosis of the traumatic and malignant ulcer was made, and a provisional diagnosis was made. An incisional biopsy was done, and the results of the histopathological analysis revealed an ulcerated stratified squamous epithelium overlying a dense mixed inflammatory cell infiltrate that was primarily lymphocytes followed by eosinophils, as well as features of epithelial hyperplasia and mild epithelial dysplasia that reached deep into the submucosa. (Fig 2 A, B) The sharp teeth were rounded off. The lesion entirely healed without event over the six weeks while we followed the Patient. A review of the histological section revealed that the lesion was an eosinophilic ulcer. (EU). (Fig 3 A, B)

Fig 1: The ulcer in the right lateral border of the tongue was about (1.5x2cm) related to sharp molar teeth

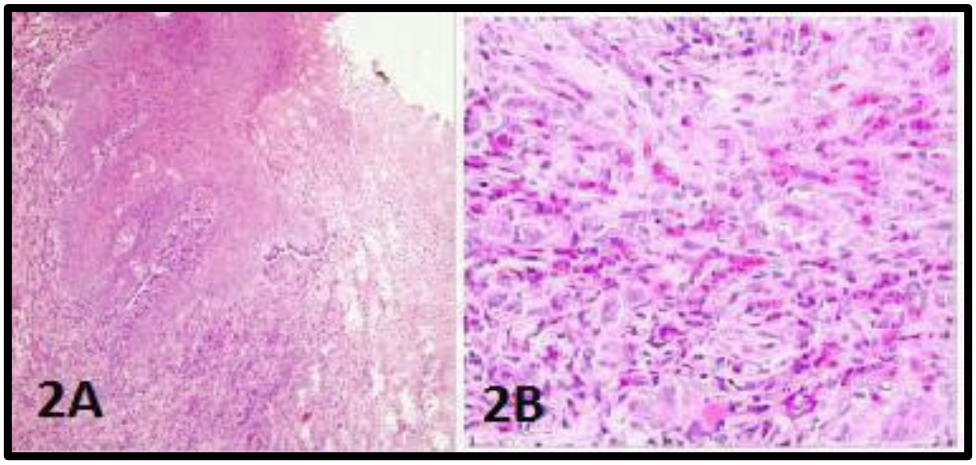

Fig 2A: Histopathological analysis revealed an ulcerated stratified squamous epithelium overlying dense mixed inflammatory cell extending deep into the submucosa
Fig 2B: Dense, mixed inflammatory cell infiltrate predominantly consisting of eosinophils, followed by lymphocytes

Fig 3A: Three weeks after the biopsy and removal of the cause (sharp teeth). Healing in the border.
Fig3 B: Six weeks after biopsy and removal of the cause (sharp teeth) and the lesion completely healed without event

3. CASE REPORT 2

A 53 years old Saudi male medically fit presented to our clinic complaining of a painful and indurated ulcer at the right lateral border of the tongue for more than two months. No extraoral abnormality was detected. Intraoral examination revealed a solitary ulcer (1X1.5cm) with elevated and whitish borders related to sharp posterior teeth (Fig4). The Patient has been a Gat and Shamma user for over ten years. Smoothening of the sharp teeth was done, we followed the Patient for six weeks, and the lesion completely healed without event upon review of the histological section; the lesion proved to be an eosinophilic ulcer (EU). (Fig 5 A, B)

Fig 4: The ulcer in the right lateral border of the tongue was about (1x1.5cm) related to sharp molar teeth

Fig 5A: Three weeks after biopsy and removal of the cause (sharp teeth). Healing in the border.
Fig5 B: Six weeks after biopsy and removal of the cause (sharp teeth) and the lesion completely healed without event

4. DIFFERENTIAL DIAGNOSIS

Based on the history and clinical examination, a provisional diagnosis of traumatic ulcer with secondary induration was made, with a differential diagnosis of an eosinophilic, malignant, and major aphthous ulcer.

5. HISTOPATHOLOGICAL FINDINGS

The tissue had been covered in an ulcerated, hyperplastic stratified squamous epithelium in one area of the sections, and the connective tissue beneath had a severe inflammatory infiltrate that penetrated deep into the muscle layer. These features are suggestive of TUGSE. Thus a final diagnosis of eosinophilic ulcer of the tongue was made. (Fig2A, B).

6. MANAGEMENT

After smoothing out the sharp teeth, we followed the Patient for six weeks, and the lesion completely healed. The Patient was advised to apply a 0.1% triamcinolone acetonide paste topically three times daily for one week.

7. 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[10]. Since then, names such as eosinophilic ulcer, traumatic eosinophilic granuloma of the tongue, traumatic granuloma, atypical histiocytic granuloma, and TUGSE have been used to describe the lesion. Our cases also represent the etiology of eosinophilic ulcers as trauma. Trauma is considered to play a major role in the etiology of the ulcer, considered to be a benign, reactive, and self-limiting lesion of the oral mucosa. Although mechanical trauma (accidental bites, repeated thrusting against sharp or misplaced teeth) coexists in more than half of the reported cases, see a similar study of Marszalek A[11] and Ada S[12]. Eosinophilic ulcers usually occur in the fifth and seventh decades of life with equal distribution between males and females; however, a slight male predilection was noted by Fonseca et al. 1, which is similar to our cases. The tongue is the most commonly involved site in about 60% of the cases. Other sites that can be involved include the buccal mucosa and labial mucosa, the floor of the mouth, and the vestibule, where the underlying skeletal muscle is found.[13]The lesion typically appears as an ulcer with 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. These features and rapid development can clinically mimic squamous cell carcinoma, making biopsy necessary to rule out malignancy. Lymphadenopathy can be observed in extremely rare cases.[14] In the present case, a solitary ulcer was present on the left posterolateral surface of the tongue, measuring about 1.5×2 cm in the first case and 1.0 x1.5cm in diameter in the second case, surrounded by keratosis. Its margins and base were indurated and covered with yellow fibrinous material, as in similar cases presented in the case study by Nagarajan NP[15] Histopathologically, the ulcer exhibits a serious neuroinvasive inflammatory reaction and is typically slow to resolve. However, complete resolution of the lesion after the incisional biopsy has also been noted.[16] The literature has described a variety of therapeutic modalities for treating eosinophilic ulcers, including waiting it out, using antibiotics, topical, intralesional, and systemic corticosteroids, curettage, cryosurgery, and surgical excision. However, the most frequently performed therapy is simple surgical incision/ excision.[17] Our case showed no recurrence, as normally reported in OEU[18].

8. CONCLUSION

Knowledge of this disease is necessary since its lesion can mimic an oral squamous carcinoma occurring as an indurated ulceration coupled with its frequent occurrence on the tongue and targeting predominantly individuals in their 50s and 60s. The biopsy is not only the best approach for a proper diagnosis but also for its therapeutic management. The cause of this lesion and the reason it often disappears after a biopsy intervention remains an enigma.

1. The pathogenesis of the eosinophilic ulcer remains uncertain, and a combination makes the diagnosis of clinical and histopathological features.
2. Eosinophilic ulcers of the oral mucosa are characterized by rapidly growing ulcers with indurated borders that include a wide clinical differential diagnosis.
3. The lesion is characteristically self-limiting with a benign course.
4. More research is needed with more cases to evaluate the clinical diagnosis outcomes to be measured precisely.

9. AUTHORS CONTRIBUTION STATEMENT

Dr. Shreefah Faris conceptualized and gathered the data about this case report. Ali M. Makrami gave the necessary inputs and managed the literary searches. Dr. Afnan A Essa provided valuable input on the manuscript. Fareedi Mukram Ali curated the data and provided valuable inputs toward the design of the manuscript. Finally, all authors discussed and contributed to the final version of the manuscript.

10. CONFLICT OF INTEREST

Conflict of interest declared none.

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