CASE REPORT

Eosinophilic ulcer of the tongue: a rare and confusing clinical entity

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Accepted 18 March 2015

SUMMARY

Eosinophilic ulcers are rare, benign, reactive and often self-limiting lesions of the oral cavity. Although the aetiology is not clear, trauma is believed to play a role in their development. Clinically, the lesion manifests as an isolated ulcer, with raised, indurated borders and a yellow fibrinous floor; because of its long duration it often leads to the suspicion of squamous cell carcinoma. Although the ulcer is benign in nature, a biopsy is necessary to rule out malignancy. Histopathologically, the ulcer is characterised by the presence of dense inflammatory infiltrate extending into the deeper muscle layers with sheets of lymphocytes intermixed with eosinophils. This is a case report of a 65-year-old woman with an 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.

BACKGROUND

An 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 characterised by the presence of indurated borders resembling malignancy.¹ 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 the tongue, traumatic granuloma, atypical histiocytic granuloma and traumatic ulcerative granuloma with stromal eosinophilia (TUGSE).² Although trauma is considered as an etiological factor, less than 50% of patients recall the history of trauma.³ A viral or toxic agent implication has been hypothesised in its pathogenesis; however, there is no definitive experimental proof.⁴ Similar lesions are seen on the ventral surface of the tongue in infants, and are 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.⁵ Oral mucosal ulceration that does not resolve within 2 weeks is troublesome and 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 with uncertainty regarding its nature, aetiology and pathogenesis.⁴

CASE PRESENTATION

A 65-year-old woman presented with an ulcer on the left side of her tongue for 20 days. The ulcer

was insidious in onset and initially small in size, gradually increasing to its present size. There was no history of trauma, white or red patches, or fluid filled blisters prior to the onset of the ulcer. There was no history of bleeding or pus discharge. The ulcer was associated with pain, which was mild, intermittent and localised; no aggravating or relieving factors were noted. There was no history of restricted tongue or jaw movements since the onset of the ulcer and no history of difficulty in chewing and swallowing food. There was no history of recurrent ulcers in the oral cavity or of ulcers elsewhere in the body. There was no history of other associated symptoms such as fever, nausea, or loss of weight or appetite.

The patient's medical history was remarkable, as she was diabetic and on oral hypoglycaemic drugs. Local extraoral examination revealed a palpable solitary, left submandibular lymph node, with no local rise of temperature, measuring about 1×1 cm in size, oval in shape, soft in consistency, non-tender, mobile 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, the 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 1×1 cm with raised edges, and the floor was covered with yellow fibrinous exudate. The ulcer was surrounded by white hyperkeratotic mucosa (figure 1). The ulcer was non-tender on palpation, and the edges and base were indurated; no discharge was elicited. Mobility of the tongue was not restricted.

INVESTIGATIONS

On routine investigation, the 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

On the basis of the history and clinical examination, a provisional diagnosis of traumatic ulcer with secondary induration was made with a differential diagnosis of eosinophilic ulcer, malignant ulcer and major aphthous ulcer. The sections showed tissue covered by hyperplastic stratified squamous epithelium that was ulcerated in one area and the underlying connective tissue with intense

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To cite: Lingaraju N, Besagarahally Gaddelingiah Y, Shivalingu MM, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2015-210107

Figure 1 Ulcer with raised edges and covered with yellow fibrinous exudate.

inflammatory infiltrate consisting predominantly of lymphocytes and eosinophils extending deep into the muscle layer (figures 2 and 3). These features are suggestive of TUGSE. Thus a final diagnosis of eosinophilic ulcer of the tongue was made.

TREATMENT

Extraction of the left maxillary second molar was carried out and the patient was advised topical application of 0.1% triamcinolone acetonide paste three times per day for 1 week.

OUTCOME AND FOLLOW-UP

The patient returned for follow-up on the eighth day after incisional biopsy. Resolution of the ulcer was observed (figure 4).

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.^{6 7} Since then, names such as

Figure 2 H&E stain showing abundant eosinophils.

Figure 3 H&E stain showing inflammatory infiltrate extending deep into the muscle layer.

eosinophilic ulcer, traumatic eosinophilic granuloma of the tongue, traumatic granuloma, atypical histiocytic granuloma and TUGSE have been used to describe the lesion.

An eosinophilic ulcer is considered to be a benign, reactive and self-limiting lesion of the oral mucosa. Trauma is considered to play a major role in the aetiology of the ulcer, however, trauma could be observed in only 50% of cases.⁸ According to some authors, trauma is only a contributing factor, and could lead to viral and toxic agents entering the underlying tissue, causing inflammatory response. Most 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.

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

Figure 4 Resolution of the ulcer after treatment.

Fonseca *et al.*⁷ 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, floor of the mouth and the vestibule, where underlying skeletal muscle is found.³ 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 along with rather quick development can clinically mimic squamous cell carcinoma and this 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 posterolateral surface of the tongue, measuring about 1×1 cm in diameter, surrounded by keratosis. Its margins and base were indurated and covered with yellow fibrinous material associated with left submandibular lymphadenopathy.

Histopathologically, the ulcer exhibits a deep pseudoinvasive inflammatory reaction and is typically slow to resolve. However, complete resolution of the lesion 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 ulcers have been reported in the literature, including a 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

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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Learning points

- The pathogenesis of the eosinophilic ulcer still remains uncertain and the diagnosis is made by a combination of clinical and histopathological features.
- Eosinophilic ulcers of the oral mucosa are characterised by rapidly growing ulcers with indurated borders that include a wide clinical differential diagnosis.
- The lesion is characteristically self-limiting with a benign course.

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