CASE REPORT

Eosinophilic ulcer of the tongue in an 80-year-old Iranian woman after a psychologically stressful event

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SUMMARY

Eosinophilic ulcer of the oral mucosa is a benign self-limiting, chronic lesion more frequently seen in the oral cavity or tongue that can mimic squamous cell carcinoma. The pathogenesis of this problem is unknown. In this paper, we present a case of an 80-year-old woman with eosinophilic ulcer of the tongue after a psychologically stressful event.

BACKGROUND

Eosinophilic ulcer of the oral mucosa (EUOM) is an uncommon self-limiting oral condition most commonly presenting as an ulcer or sometimes as a raised and indurated submucosal mass.^{1–10} It has been referred to as various entities in medical literature such as: ulcerated granuloma eosinophilicum diutinum, traumatic granuloma of the tongue, eosinophilic granuloma of the tongue, eosinophilic ulcer of the tongue, traumatic eosinophilic granuloma, atypical histiocytic granuloma (AHG) and ulcerative eosinophilic granuloma of the tongue.^{1–3 6–10} It also occurs in infants under 2 years of age at the anterior labial mucosa or at the ventral surface of the tongue, which is considered as a result of friction between the tongue or lip and teeth and is termed Riga-Fede disease.^{1–3 6–10} Although some specialists propose that the trauma caused by accidental bites or by repeated thrusting against sharp, misplaced or fractured teeth has a major role in this disease, however; its exact pathogenesis is obscure.^{2–10} Stress-induced psychoneuroimmunological factors may also have a role in pathogenesis.⁵ In this paper, we present a case with this disease occurring subsequent to a psychologically stressful event.

CASE PRESENTATION

The patient was an 80-year-old woman referred to a private ENT office with a painful white lesion in the lateral aspect of her tongue. The lesion suddenly presented since 2 weeks previously, after her husband's death, and grew rapidly. She recalled no traumatic event. The patient complained of odinophagia; however, she could not explain the dysphagia. She was not a smoker and did not drink alcohol. The patient had taken atenolol for hypertension. On physical examination, she was found to be in good general condition. Head and neck examination revealed a 3 cm tender and indurated ulcer with inflamed borders in the lateral aspect of her tongue that was covered with a fibroid membrane. There was no lymphadenopathy. Since the lesion mimicked a squamous cell carcinoma

(SCC) of the tongue, an excisional biopsy was performed of the specimen and sent to the pathologist.

INVESTIGATIONS

Pathological examination revealed polymorphic inflammatory cells including neutrophils, macrophages and numerous eosinophil infiltration that extended to the submucosa and muscles (figure 1).

DIFFERENTIAL DIAGNOSIS

Owing to the rapid growth, ulcerative nature and indurated borders of the usually single lesion, this condition can mimic SCC.^{1 2 6} In our patient, the clinical feature was so similar to SCC that the ENT specialist talked to the patient and her family about the necessity of hemiglossectomy and ipsilateral lymph node dissection after confirming the diagnosis by pathology. Other tumoral lesions such as lymphoma, lymphangioma, minor salivary gland tumours and metastatic tumours may be included in the differential diagnosis.¹ Some infectious diseases such as primary syphilis, oral tuberculosis and fungal infections are found in the clinical differential diagnosis list.² Sarcoidosis, discoid lupus erythematosus, Wegener's granulomatosis and eosinophilic granuloma are also included in the list of differential diagnosis.² Microscopic examination, however; easily diagnosis EUOM from other clinical conditions, such as that of our patient.

The disease is microscopically similar to AHG, angiolymphoid hyperplasia with eosinophilia and Kimura disease.²

TREATMENT

Chlorhexidyn mouth wash, penicillin V tablets 500 mg four times a day and ibuprofen tablets 400 mg three times a day were prescribed.

OUTCOME AND FOLLOW-UP

Two weeks later, the lesion recovered completely. Now, 14 months later, the patient is well and her disease has not recurred.

DISCUSSION

The aetiology and pathophysiology of EUOM remain poorly understood. Some authors propose a relation with trauma. The evidences that reinforced this theory include: the case of some patients remembering accidental biting of their tongue before the disease; the disease having two peaks of age incidence, the first one in children at the primary teeth eruption, and the second in old age on the occasion of missing and more commonly

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Figure 1 Polymorphic inflammatory cells especially eosinophils infiltration that is extending to the submucosa and muscles.

malposed teeth, dental appliances and dentures.² The disease is more common in the ventral and lateral aspects of the tongue that are most exposed to trauma. In experimental studies, repeated injuries to the tongue in rats demonstrate similar lesions.⁴ Others have suggested that trauma is only a contributing factor in the development of EUOM and could facilitate the entry of unidentified aetiological factors such as microorganisms, toxins or foreign proteins into the underlying tissue to cause an inflammatory response.^{1 2 4} On the other hand, trauma is identified in less than 50% of cases.¹ Histologically, EUOM shows a diffuse polymorphic inflammatory infiltrate, rich in eosinophils, involving the superficial mucosa and a deeper muscle layer. A typically large mononuclear cell scattered within the inflammatory infiltrate has been described and its origins have been a matter of debate in some patients;^{2 4 8} however, they were not seen in our patient. According to a suggestion about the immunohistochemical characteristics of these cells by Regezi *et al*, they are the combinations of macrophages and the oral counterparts to dermal dendrocytes. They also found an abundance of smaller CD3-positive T-lymphocytes.¹¹ El-Mofty *et al*¹² suggested that they may be myofibroblasts. Ficarra *et al*¹³ and Segura *et al*⁴ described the proliferation of CD30 atypical large mononuclear cells then as leading to the conclusion that EUOM could be included within the spectrum of CD30 lymphoproliferative disorders. Alobeid *et al*¹⁴ described three patients with oral mucosal lesions and EUOM features in one of whom the oral lesion was followed by skin nodules. All oral lesions contained atypical large lymphoid cells, which expressed T-cell markers and CD30. PCR analysis showed a monoclonal rearrangement of the TCR γ chain gene in all lesions and, in one patient, the same rearrangement in the oral and cutaneous specimens. These studies led to the conclusion that EUOM could be included within the spectrum of CD30 lymphoproliferative disorders. On the other hand, Elzay¹⁵ reported the increased numbers of mast cells in this disease and suggested that the interactions between mast cells and eosinophils might play a pathogenetic role. Atopic dermatitis (AD) is a chronic and relapsing inflammatory skin disease characterised by eczematous skin lesions, xerosis, lichenification and severe pruritus.¹⁶ This disease has some similarities to EUOM. Both of them have an obscure aetiology and similar inflammatory cells such as

eosinophils and mast cells. The clinical occurrence of AD is often associated with psychological stress. In response to stress, the upregulation of neuropeptide mediators in the brain, endocrine organs and peripheral nervous system directly affects immune and resident cells in the.¹⁶ The substance P (SP) becomes elevated in the brain in response to many different types of psychological stressors. Lymphocytes, leucocytes, macrophages and mast cells have receptors for SP and these cells can be stimulated by SP and through this route. Our patient's case was similar to that reported by Ribeiro *et al*.⁵ She had experienced a psychologically stressful event before her problem occurred. Stressors may play a role in developing EUOM.¹⁷

Learning points

- Eosinophilic ulcer of the oral mucosa (EUOM) can mimic squamous cell carcinoma and must be considered in differential diagnosis.
- The aetiology of EUOM is unknown. Psychological stressors may have a role.
- EUOM is usually self-limited and recovers after 2–4 weeks.

Competing interests None.

Patient consent Obtained.

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REFERENCES

- 1 Joseph BK, BairavaSundaram D. Oral traumatic granuloma: report of a case and review of literature. *Dent Traumatol* 2010;26:94–7.
- 2 Segura S, Pujol RM. Eosinophilic ulcer of the oral mucosa: a distinct entity or a non-specific reactive pattern? *Oral Dis* 2008;14:287–95.
- 3 Vélez A, Alamillos FJ, Dean A, *et al*. Eosinophilic ulcer of the oral mucosa: report of a recurrent case on the tongue. *Clin Exp Dermatol* 1997;22:154–6.
- 4 Segura S, Romero D, Mascaró JM Jr, *et al*. Eosinophilic ulcer of the oral mucosa: another histological simulator of CD30+ lymphoproliferative disorders. *Br J Dermatol* 2006;155:460–3.
- 5 Ribeiro AL, Mendes FR, Alves Sde M Jr, *et al*. Eosinophilic ulcer: the role of stress-induced psychoneuroimmunologic factors. *Oral Maxillofac Surg* 2011;15:179–82.
- 6 Abdullah BH. Traumatic ulcerative granuloma with stromal eosinophilia (a clinicopathological study of 18 cases). *J Bagh Coll Dent* 2011;23:59–64.
- 7 Bortoluzzi MC, Passador-Santos F, Capella DL, *et al*. Eosinophilic ulcer of oral mucosa: a case report. *Ann Stomatol (Roma)* 2012;3:11–13.
- 8 Hirshberg A, Amariglio N, Akirsh S, *et al*. Traumatic ulcerative granuloma with stromal eosinophilia a reactive lesion of the oral mucosa. *Am J Clin Pathol* 2006;126:522–9.
- 9 Horie N, Shimoyama T, Kato T, *et al*. Eosinophilic ulcer of tongue. A case report with immunohistochemical study. *Oral Med Pathol* 1994;4:25–9.
- 10 Vasconcelos MG, Souza LB, Silveira ED, *et al*. Eosinophilic ulcer of the lateral tongue: case report. *RSBO* 2011;8:459–63.
- 11 Regezi JA, Zarbo RJ, Daniels TE, *et al*. Oral traumatic granuloma. Characterization of the cellular infiltrate. *Oral Surg Oral Med Oral Pathol* 1993;75:723–7.
- 12 El-Mofty SK, Swanson PE, Wick MR, *et al*. Eosinophilic ulcer of the oral mucosa. Report of 38 new cases with immunohistochemical observations. *Oral Maxillofac Pathol* 1993;75:716–22.
- 13 Ficarra G, Prigmano F, Romagnoli P. Traumatic eosinophilic granuloma of the oral mucosa: a CD-30+ (Ki-1) lymphoproliferative disorder? *Oral Oncol* 1997;33:375–9.
- 14 Alobeid B, Pan LX, Milligan L, *et al*. Eosinophil-rich CD30+ lymphoproliferative disorder of the oral mucosa. A form of "traumatic eosinophilic granuloma". *Am J Clin Pathol* 2004;121:43–50.
- 15 Elzay RP. Traumatic ulcerative granuloma with stromal eosinophilia (Riga-Fede's disease and traumatic eosinophilic granuloma). *Oral Surg Oral Med Oral Pathol* 1983;55:497–506.
- 16 Suárez AL, Feramisco JD, Koo J, *et al*. Psychoneuroimmunology of psychological stress and atopic dermatitis: pathophysiologic and therapeutic updates. *Acta Derm Venereol* 2012;92:7–15.
- 17 Black PH. Stress and the inflammatory response: a review of neurogenic inflammation. *Brain Behav Immun* 2002;16:622–53.

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