CASE REPORT

Traumatic ulcerative granuloma with stromal eosinophilia: A diagnostic dilemma?
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Abstract
Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is a slow growing reactive lesion that commonly affects the tongue. The pathogenesis of this lesion is obscure. Chronic irritation from traumatic agents is considered to be one of the contributing factors in most of the cases. It usually presents as an ulcer or an indurated submucosal mass. Histopathologically, it is characterized by eosinophilic inflammatory infiltrate penetrating into the submucosal layers degenerating the underlying muscle. It is important to recognize this mucosal entity as it has a close resemblance with malignancy of the oral cavity. The pathognomonic feature of TUGSE is that it resolves on its own after excisional biopsy. A 25-year-old Sudanese boy had reported to us with an ulceroproliferative growth in the left side of the mouth.

Keywords
Biopsy, eosinophils, trauma, traumatic ulcerative granuloma with stromal eosinophilia

Introduction
Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is a unique and relatively recently delineated entity also called as traumatic granuloma and eosinophilic granuloma.[1] It is a benign lesion, possibly a lymphoproliferative disorder, which typically follows trauma.[1,2] The rapid growth, large ulceration and indurated borders resemble early squamous cell carcinoma; hence biopsy is performed to rule out that possibility.[1,2] Interestingly, many of these traumatic granulomas undergo resolution after incisional biopsy.[2]

Here, we report a case of 25-year-old Sudanese boy who presented to our department with a swelling on the left side of the lower 1/3rd of the face.

Case Report
A 25-year-old Sudanese boy had come to the hospital with the chief complaint of pain in the lower left back teeth region and asymmetry on one side of the face since 1½ months. In the history of presenting illness, pain was insidious in onset, severe in intensity since 1 week, intermittent in nature, aggravated on chewing food on the affected side and relieved on taking medications.

Swelling was noticed by the patient in the lower aspect of the jaw 1½ months back which was the size of a peanut and has gradually increased to its present size, associated with severe pain since 1 week. The medical and allergic history was not significant.

Patient revealed the habit of smoking 3-4 cigarettes/day since 6 years and has stopped the habit since 1 month. Occasionally, a patient would take to hookah smoking.

Extraoral examination revealed presence of a solitary swelling on the lower left side of the face roughly oval in shape measuring about 3 cm × 3 cm in size. It extended anteriorly 1 cm from the angle of mouth, posteriorly from the posterior border of the ramus of the mandible, superiorly 0.5 cm below the infraorbital margin and inferiorly till lower border of the mandible. It had well-defined margins, color over the swelling was normal, and no effect on surrounding structures [Figure 1].

On palpation, inspectory findings were confirmed. Swelling was tender, firm to hard in consistency. A solitary left submandibular lymph node was palpable, tender, 1 cm in size roughly, soft in consistency and freely movable. On biting, tenderness was elicited over the area of the left temporomandibular joint and mouth opening was restricted.

Intraoral soft tissue examination revealed presence of an ulceroproliferative growth on the left side of the oral cavity on
the buccal mucosa of size about 3 cm × 3 cm, roughly oval in shape, raised well-defined margins. On palpation, inspectory findings were confirmed. Tenderness on palpation was elicited, and the lesion had everted margins and indurated base [Figure 2].

On hard tissue examination, presence of partially erupted 38 adjacent to the growth [Figure 2]. Based on the clinical history and all the findings a provisional diagnosis of carcinoma of lower left buccal vestibular sulcus was given. An intraoral periapical radiograph (IOPAR) of 38 and an orthopantogram (OPG) was advised.

The IOPAR radiograph [Figure 3] revealed vertical impaction and OPG [Figure 4] revealed generalized horizontal bone loss and no involvement of bone with respect to the left mandible. A complete hemogram was advised followed by incisional biopsy of the lesion the next day. Patient was recalled after 1 week for the report. After the biopsy, there was a reduction in the size of the growth to 1 cm × 2 cm, hence, we suggested for an excisional biopsy to see the healing of the lesion. Along with the excisional biopsy, extraction of 38 was also done.

After 1 week follow-up, there was almost complete disappearance of the growth [Figure 5]. The histopathology report [Figures 6 and 7] revealed the presence of parakeratotic

Figure 1: Extraoral swelling on the left lower third of the face

Figure 2: Ulceroproliferative growth adjacent to an impacted 38 diagnosed as carcinoma of left mandibular buccal vestibular sulcus

Figure 3: Intraoral periapical radiograph revealed vertical impaction with 38

Figure 4: Orthopantomogram reveals generalized horizontal bone loss and intact left lower border of the mandible

Figure 5: Photograph taken after biopsy before the extraction was done
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epithelium with ulceration in some areas. Connective tissue showed numerous mixed inflammatory infiltrate predominantly composed of histiocytes and eosinophils. Lymphocytes and plasma cells were also seen infiltrating the muscles with muscle degeneration hence a final diagnosis of traumatic ulcer with stromal eosinophilia. No recurrence of lesions was noticed during the 3 months follow-up.

Discussion

TUGSE was first described by Popoff in 1956. In 1970, this lesion was proposed as a distinct entity by Shapiro and Juhlin. Since then different names have been used to define it.

The first case of TUGSE was reported in 1960 and it was included in the spectrum of granuloma faciale and some authors also proposed the term ulcerated granuloma eosinophilicum diutinum of the tongue.

It is a chronic, slow growing, spontaneously healing mucosal entity, presenting as an ulcer with elevated margins. The most common location is the tongue, although other locations in the oral mucosa are possible.

It presents as a spontaneously healing single ulcer which stays on for weeks to months after the trauma. Many times the source of constant irritation cannot be found. The lesion characteristically has firm margins along with fast occurrence and slow healing; it can be confused with oral malignancy such as squamous cell carcinoma.

In the literature, the pathogenesis of TUGSE was mainly trauma leading to ulceration, hence ingress of viral/toxic agents/foreign bodies induces an exaggerated inflammatory reaction leading to release of mast cells and eosinophils.

The degranulation of mast cells leads to release of mediators, which causes inflammation and also attracts eosinophils by release of eosinophil chemotactic factor of anaphylaxis. The aryl sulfates and histamines are released by the eosinophils, which inhibits slow reacting substances of anaphylaxis and histamines. This leads to suppression of basophils and mast cell degranulation and inhibit the release of other mediators of inflammation by the mast cells. Eosinophils also produce major basic protein, which causes tissue destruction.

It is most frequently diagnosed in patients aged between 30 and 50 years. It can also occur in infants as well as in elderly people. Male to female ratio is 1:1 or slightly more elevated in females. Tongue is the most common location for eosinophilic ulcer, but it can occur in other areas of the mouth (lips, buccal mucosa, palate, gingiva and floor of the mouth). Clinically, the lesion presents as an ulcer with raised, indurated margins and yellow fibrinous base.

The most common location for TUGSE is the tongue, but in our case report, a 25-year-old Sudanese boy had an ulceroproliferative growth with everted and indurated margins adjacent to a partially erupted 38 in the posterior mucobuccal sulcus of 1 ½ months duration hence adding to its rarity.

Ficarra et al. reported a case with multiple recurrent episodes of ulcers occurring mainly on the buccal mucosa and gingiva, lasted for 3-4 weeks and slowly underwent spontaneous healing after repeated incisional biopsies. This was similar to our case where the ulceroproliferative growth healed after multiple biopsies and extraction of partially erupted 38 with no recurrence.

Sometimes, TUGSE shows delayed self-healing. Some authors explained the reason for the delayed self-healing as the lack of synthesis of transforming growth factor by eosinophils in the inflammatory infiltrate. These eosinophils in the inflammatory infiltrate release other cytokines, such as tumor necrosis factor, which enhances tissue damage and keeps the inflammatory response unfinished.

The histopathology report in various cases showed ulceration of the oral mucosa with lymphocytes and multiple eosinophils extending deep into the soft tissues. It also revealed large, atypical, mononuclear cells with irregular nuclear margins and small nuclei on higher magnification.

A similar histopathology report was present in our case. The immunohistochemistry shows large, atypical, mononuclear cells. In the literature, researchers said the origin of these atypical cells...
were from macrophages (CD68-positive), dendritic cells (factor XIII-positive) and myofibroblasts (vimentin-positive).\(^{(11)}\)

Many different therapeutic approaches have been tried for TUGSE the most common being simple surgical excision and its recurrence is rare.\(^{(12,13)}\)

Topical mouthwashes and steroids can also be prescribed. The other treatment modalities are intralesional or oral corticosteroids, topical antibiotics, curettage and cryotherapy.\(^{(13,14)}\)

The removal of traumatic agents was considered as the mainstay of the treatment. The lesion shows regression after the incisional or excisional biopsy. Prognosis of TUGSE found to be good.\(^{(15)}\) In our case the only treatment given was incisional biopsy and extraction of 38 and showed regression of the lesion.

**Conclusion**

Diagnosis of this eosinophilic ulcer is on the basis of history, clinical presentation and microscopic findings that include the presence of an ulcer or a submucosal mass with a predominant eosinophilic inflammatory infiltrate with lymphocytes. The infiltrate extends into deeper layers, along with giant atypical histiocyte-like cells with large prominent nuclei and nucleoli and presence of mitotic figures.

As oral diagnosticians, TUGSE should be kept in mind as one of the differential diagnosis, when we see a solitary ulcer with everted and indurated margins that is a mimicker of squamous cell carcinoma.

**References**
