




Traumatic ulcerative granuloma with stromal eosinophilia of the tongue: case report in a pediatric patient

Yurgen Torrez Velásquez¹ , Mariela Jiménez Chacón¹ , Yadira Boza Oreamuno¹ .

Abstract: Introduction: Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE) is a rare, benign lesion of the oral mucosa, characterized by a prolonged clinical course and histopathological findings that can mimic malignant processes. Its presentation in the pediatric population is uncommon, which can hinder its recognition and timely management. We report a pediatric case of TUGSE to facilitate its clinical and histopathological identification and highlight its importance in the differential diagnosis of chronic oral ulcers. **Case presentation:** An 8-year-old boy presented with a three-month history of a progressive lingual ulcer that was refractory to topical treatment. Examination revealed an oval ulcer with indurated borders, and initial laboratory tests showed only mild inflammation. Given the clinical changes, differential diagnoses of chronic traumatic ulcer, lymphoproliferative disorder, and neoplastic epithelial lesion were considered. A biopsy was performed, and histopathological analysis showed an ulcer with fibrin, fibrosis, and scant exudate, along with negative CD30 and EBER results and positive CD68, consistent with TUGSE in the cicatricial phase. The outcome was favorable, with complete healing and no recurrence in two years. **Conclusions:** This case highlights the importance of recognizing TUGSE as a benign entity that can mimic neoplastic processes, especially in children, and emphasizes the value of histopathological examination to establish an accurate diagnosis and guide appropriate management.

Key words: Child, Eosinophilia, Granuloma, Pathology, Tongue Diseases.

Granuloma ulcerativo traumático con eosinofilia estromal en lengua: reporte de caso en un paciente pediátrico

Resumen: Introducción: El granuloma ulcerativo traumático con eosinofilia estromal (TUGSE) es una lesión benigna e infrecuente de la mucosa oral, caracterizada por un curso clínico prolongado y hallazgos histopatológicos que pueden simular procesos malignos. Su presentación en población pediátrica es poco común, lo que puede dificultar el reconocimiento y manejo oportuno. Se reporta un caso pediátrico de TUGSE para facilitar su identificación clínica e histopatológica y resaltar su importancia en el diagnóstico diferencial de úlceras orales crónicas. **Presentación de caso:** Niño de 8 años presentó una úlcera lingual de tres meses, progresiva y refractaria a tratamiento tópico. La exploración mostró una úlcera ovalada con bordes indurados, y los exámenes de laboratorio iniciales solo evidenciaron inflamación leve. Ante cambios clínicos, se establecieron diagnósticos diferenciales de úlcera traumática crónica, proceso linfoproliferativo y lesión epitelial neoplásica. Se realizó biopsia, cuyo análisis histopatológico mostró una úlcera con fibrina, fibrosis y exudado escaso, junto con CD30 y EBER negativos y CD68 positivo, compatibles con TUGSE en fase cicatricial. La evolución fue favorable, con cicatrización completa y sin recidiva en dos años. **Conclusiones:** Este caso resalta la importancia de reconocer la TUGSE como una entidad benigna que puede simular procesos neoplásicos, especialmente en niños, y enfatiza el valor del examen histopatológico para establecer un diagnóstico preciso y orientar un manejo adecuado.

Palabras clave: Niño, Eosinofilia, Granuloma, Patología, Enfermedades de la lengua.

¹Universidad de Costa Rica, San José, Costa Rica

Granuloma ulcerativo traumático com eosinofilia estromal da língua: relato de um caso num doente pediátrico

Resumo: Introdução: O granuloma ulcerativo traumático com eosinofilia estromal (TUGSE) é uma lesão benigna rara da mucosa oral, caracterizada por um curso clínico prolongado e achados histopatológicos que podem mimetizar processos malignos. A sua apresentação na população pediátrica é pouco frequente, o que pode dificultar o seu reconhecimento e tratamento atempado. Relatamos um caso pediátrico de TUGSE para facilitar a sua identificação clínica e histopatológica e realçar a sua importância no diagnóstico diferencial das úlceras orais crônicas. **Apresentação do caso:** Um rapaz de 8 anos apresentou-se com um historial de três meses de úlcera lingual progressiva, refratária ao tratamento tópico. O exame revelou uma úlcera oval com bordos endurecidos, e os exames laboratoriais iniciais mostraram apenas uma inflamação ligeira. Face às alterações clínicas, foram considerados os diagnósticos diferenciais de úlcera traumática crónica, distúrbio linfoproliferativo e lesão epitelial neoplásica. Foi realizada uma biópsia, tendo a análise histopatológica revelado uma úlcera com fibrina, fibrose e escasso exsudato, para além de resultados negativos para CD30 e EBER e positivos para CD68, compatíveis com TUGSE na fase cicatricial. A evolução foi favorável, com cicatrização completa e sem recidiva em dois anos. **Conclusões:** Este caso realça a importância de reconhecer a TUGSE como uma entidade benigna que pode mimetizar os processos neoplásicos, especialmente em crianças, e enfatiza o valor do exame histopatológico para estabelecer um diagnóstico preciso e orientar o tratamento adequado.

Palavras-chave: Criança, Eosinofilia, Granuloma, Patologia, Doenças da língua.

Introduction

Traumatic ulcerative granuloma with stromal eosinophilia (TUGSE), also known as eosinophilic ulcer or traumatic eosinophilic ulcer, is an uncommon reactive lesion of the oral mucosa, characterized by a chronic ulcer associated with an inflammatory infiltrate rich in eosinophils and macrophages¹.

Although TUGSE can affect individuals of any age, studies show that its incidence is higher in adults between 40 and 60 years of age². However, cases have been reported in the pediatric population, particularly in infants or very young children, under the term Riga-Fede disease, which is associated with repeated trauma from primary teeth during the forward and backward movements of the tongue³.

Clinically, TUGSE may mimic more serious conditions such as squamous cell carcinoma, persistent infections,

or granulomatous diseases due to its ulcerated appearance with indurated borders, prolonged duration, and sometimes tissue necrosis⁴. Therefore, the differential diagnosis is broad and may require histopathology and, in some cases, immunohistochemistry to rule out malignancy or systemic infectious processes^{5,6}.

Management of TUGSE depends on eliminating triggering traumatic factors, performing a biopsy to confirm the diagnosis, and, in many cases, conservative treatment^{7,8}. In general, the lesion has a benign course and tends to resolve spontaneously following biopsy or removal of the irritative source⁹.

The aim of this report is to present a case of TUGSE affecting the lingual mucosa of a pediatric patient, with the goal of contributing to the clinical and pathological understanding of this rare benign entity and highlighting

the importance of considering it in the differential diagnosis of chronic ulcerative lesions of the oral cavity.

Case report

An 8-year-old Costa Rican boy presented with a lingual ulcer of three months' duration. According to the mother, the lesion was initially very painful but gradually became asymptomatic, although it still caused discomfort while eating. During this period, the lesion progressively increased in size and did not respond to topical healing agents, such as Oddent® hyaluronic acid gel (0.2% hyaluronic acid). In the medical history, the mother denied relevant systemic conditions, chronic diseases, or previous similar episodes. There was no report of fever, weight loss, night sweats, or exposure to toxic substances, including tobacco or alcohol.

On clinical examination, an oval ulcer approximately 10 × 5 mm was observed on the left lateral border of the tongue, with slightly indurated whitish borders and a yellowish base (Figure 1), tender on palpation. The remaining oral structures were within normal clinical limits, although the lingual cusps of the left mandibular first molar were markedly prominent. No cervical or facial lymphadenopathy was palpable, and there were no alterations in facial contour or associated skin lesions.

Considering the clinical characteristics of the lesion and the patient's age, the differential clinical diagnosis included a chronic traumatic ulcer, given its subacute course and the presence of prominent



Figure 1. Initial clinical examination of the ulcer on the left lateral border of the tongue.

dental cusps in contact with the affected site. Additionally, the possibility of a lymphoproliferative process (lymphoma) or an epithelial neoplastic lesion was considered.

Laboratory tests were requested to evaluate the patient's general systemic status: complete blood count (to rule out anemia, leukocytosis, or hematologic disorders), erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) (to rule out inflammatory or infectious processes), fasting blood glucose and glycated hemoglobin (HbA1c) (to exclude diabetes mellitus), total immunoglobulin E (IgE) (to rule out local allergic reaction), as well as serum levels of vitamin B12, iron, iron-binding capacity, and folic acid (to identify possible nutritional deficiencies). Coagulation times were also determined as part of the preoperative evaluation for the biopsy.

Three days later, the patient returned with the laboratory results, which were within normal limits except for a moderately elevated ESR (22 mm/h; reference value ≤ 13 mm/h) and a slightly increased CRP (6 mg/L; reference value < 3 mg/L).

During the follow-up clinical examination, significant changes in the lesion were observed: it had acquired an irregular shape and increased in size (15 × 8 mm), with well-defined, slightly raised borders and an erythematous base partially covered by a yellowish pseudomembrane (Figure 2A). On palpation, the lesion was tender, but tongue mobility was not limited. No signs of active bleeding or purulent discharge were observed.

An excisional biopsy was performed under left mandibular block anesthesia using 2% lidocaine, with an oval incision around the ulcer; the tongue musculature was not affected. Hemostasis was achieved, and the defect was closed with a simple 5/0 silk suture (Figure 2B). The specimen, marked with silk at its mesial end, was sent for histopathological examination in 10% formalin. Recommendations were given for maintaining proper oral hygiene and avoiding carbonated beverages, citrus fruits, and irritating, hard, or toasted foods. The patient was prescribed Cataflam® (Diclofenac) 5 ml every 8 hours for 3 days, Prelone® Syrup 15 mg/5 ml (Prednisolone) 2.5 ml every 12 hours for 3 days, and Oddent® Periodontal Gel (0.20% chlorhexidine digluconate) applied to the surface three times daily for 5 days.

At the eight-day follow-up, healing by secondary intention was observed, with most sutures having disappeared; only one suture remained, though loose (Figure 2C).

Histopathological examination revealed an ulcer covered by fibrin, with bacterial colonies and fibrosis in the base fragmenting skeletal muscle fibers. The inflammatory exudate was sparse, composed of histiocytes, neutrophils, and eosinophils. No neoplastic lesion, vasculitis, or lymphocytic infiltrate suggestive of lymphoma was observed (Figure 3A). Immunohistochemistry showed negativity for CD30 and EBER, ruling out a lymphoproliferative lesion or Epstein-Barr virus (EBV)-associated lesion, while CD68 was positive in histiocytes without a proliferative pattern (Figure 3B). These findings were consistent with a traumatic eosinophilic ulcer or a healing-phase ulcer, with no evidence of malignancy.

At the one-month follow-up, the healing process was evident (Figure 4A). At the two-year follow-up, findings were within normal limits, with no recurrence and preserved oral function and comfort (Figures 4B-C).

The patient's father provided written informed consent for the publication of this case.

Discussion



Figure 2. A. Asymmetric ulcer with raised borders on the left lateral border of the tongue. B. Simple suture following excisional biopsy. C. Healing by secondary intention at eight days, with one loose suture remaining.

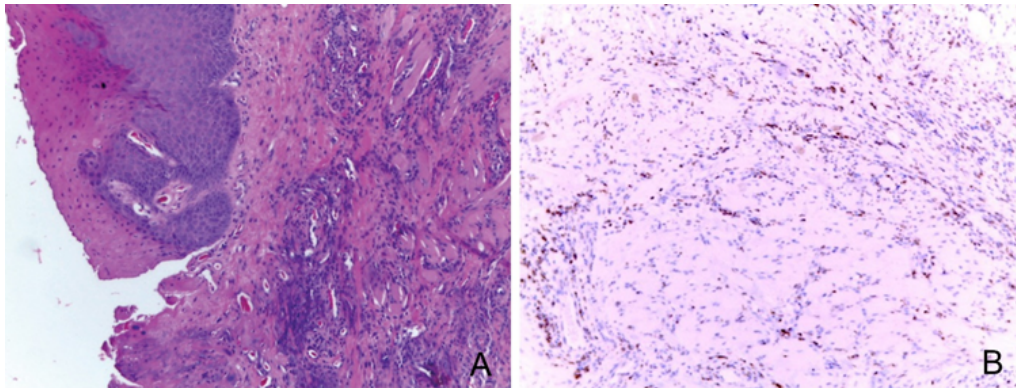


Figure 3. A. Histological section stained with hematoxylin and eosin showing an area of ulceration covered by fibrin, fibrosis at the base, and an inflammatory infiltrate composed of histiocytes, neutrophils, and eosinophils (40×). B. Immunohistochemical staining for CD68 highlighting the histiocytic cells present in the inflammatory tissue without a neoplastic proliferative pattern (20×).



Figure 4. Post-surgical evaluation: A. One-month follow-up. B. Three-month follow-up. C. Two-year follow-up.

TUGSE is a rare, benign, and self-limiting entity of the oral mucosa, with limited reports in the global literature^{10,11}. Its incidence shows two peaks: one during the first two years of life, mainly related to neonatal dentition, and another between the fifth and seventh decades of life, with a slight female predominance⁵. Our case differs, as it presented in an 8-year-old child. This aspect is particularly relevant because, although TUGSE can occur in the pediatric population, it is usually described in infants under the term Riga-

Fede disease³, and most recent reports focus on adults.

Although the etiopathogenesis is not fully understood, it has been associated with microtrauma or chronic irritation of the oral mucosa. Chronic oral ulcerations have been reported in the pediatric population, including repetitive local irritation of the tongue by the anterior primary teeth during forward and backward tongue movements³, as well as constant friction from rotated teeth or prominent cusps

^{2,12}. In our case, the lesion showed a possible, though not fully determinative, association with chronic microtrauma caused by the prominent lingual cusps of the left mandibular first molar, reflecting a mechanism similar to that described in the literature.

Regarding location, the tongue—especially the lateral border—is the most common site of presentation ^{7,12}, consistent with this case. The clinical presentation, characterized by a painful ulcer, initially oval and later irregular, with indurated borders and a yellowish pseudomembrane, aligns with literature reports, in which lesions may increase in size and change morphology during the first weeks, potentially mimicking neoplastic processes ^{8,11,13}.

The differential diagnosis of oral ulcers in children includes a wide range of entities, such as aphthous ulcers, viral infections, neoplastic lesions, and autoimmune diseases¹⁴. The differential diagnosis of a chronic ulcer on the lateral border of the tongue requires correlation of clinical findings with laboratory tests and histopathology¹¹. In our case, blood tests showed moderately elevated ESR and slightly increased CRP, suggesting a low-grade chronic inflammatory process but no systemic infection or hematologic disease. Lack of ulcer healing and induration of the borders should alert clinicians to perform a biopsy to avoid diagnostic delay.

Histopathologically, TUGSE is characterized by an ulcer covered with fibrin, with a deep, mixed inflammatory infiltrate predominantly composed

of eosinophils, lymphocytes, plasma cells, and histiocytes, often extending into the underlying muscle tissue^{2,9}. Immunohistochemically, it features a dominant polyclonal T-lymphocyte (CD3⁺) infiltrate, eosinophils, and macrophages, with occasional large mononuclear cells positive for CD30, no ALK (anaplastic lymphoma kinase) expression, and a moderate proliferation index by Ki-67, findings that support a reactive rather than neoplastic nature^{9,15}. CD68 positivity in histiocytes supports the inflammatory component, while EBER negativity rules out a lymphoproliferative or EBV-associated lesion, helping to differentiate this entity from malignancies such as carcinoma or lymphoma, as well as chronic infections ^{5,9}. In this sense, the use of immunohistochemistry in our report was consistent with literature recommendations for ruling out these entities.

TUGSE has a variable clinical course, with resolution times ranging from a few weeks to several months, mainly depending on the persistence of the local irritative factor^{16,17}. In most cases, lesions tend to resolve spontaneously or after biopsy, surgical excision, or removal of the traumatic stimulus ⁵. Although a history of trauma is suggested in most studies, less than 50% of cases present a clearly identifiable traumatic event ¹⁸. Prognosis is generally favorable, but long-term follow-up of at least two years is recommended¹⁹. Complete functional recovery and absence of recurrence in our case are consistent with the typical course of TUGSE.

This case highlights the importance

of a broad differential diagnosis to guide appropriate management and prevent complications. Correlation of clinical findings, laboratory tests, and histopathology allowed confirmation of TUGSE. Although it is a single case, its documentation provides valuable regional evidence and emphasizes the relevance of individualized treatment to optimize clinical outcomes and prevent recurrence.

limited response to conventional treatment. Its clinical course may mimic neoplastic processes, especially in pediatric patients. Histopathological and immunohistochemical studies remain essential to establish an accurate diagnosis and avoid unnecessary interventions. The favorable clinical course and absence of recurrence confirm the self-limiting nature of this entity when correctly identified and managed.

Conclusions

This case underscores the importance of considering TUGSE in the differential diagnosis of chronic oral ulcers with

Conflict of interest and funding

None declared.

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Correspondencia: Yadira Boza Oreamuno, correo: yadira.boza@ucr.ac.cr