CASE REPORT
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Lichenoid and Granulomatous Stomatitis Diagnosis and Treatment: Report of Three Cases

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Abstract:

Objective(s): Lichenoid and granulomatous stomatitis (LGS) is a rare inflammatory condition of the oral mucosa that clinically mimics oral lichen planus, posing diagnostic and therapeutic challenges. **Case:** This report described three cases of LGS in adults aged 55–63, each presenting with persistent burning sensations and chronic ulcers of the upper labial mucosa. Histopathologic evaluation in all cases revealed features consistent with lichenoid and granulomatous inflammation. Despite initial misdiagnosis and ineffective corticosteroid treatment, all patients showed marked improvement with chlorhexidine mouthwash. **Conclusion:** Accurate diagnosis of LGS is essential due to its resistance to standard corticosteroid therapies commonly used for similar lesions. Chlorhexidine may be an effective alternative, and early histopathological confirmation is recommended for proper management.

Keywords: Lichenoid Granulomatous Stomatitis; Lichen Planus; Oral Chronic Ulcers

Introduction

Lichenoid and granulomatous reactions may involve skin and oral mucosa, known as lichenoid granulomatous dermatitis (LGD), and lichenoid granulomatous stomatitis (LGS), respectively.[1] LGS is an inflammatory disease affecting the oral mucosa. When the oral cavity is involved, it presents a lichen planus-like appearance.[1] The exact etiology of this lesion remains unknown; however, in some cases, adjacent dental restorations, microbial agents (such as local plaque or calculus), medications, or systemic factors have been reported as potential triggers.[2]

It is more commonly observed in women over the age of 50. The main complaint of most patients is a burning sensation and the presence of oral ulcers. The most common site is the upper labial mucosa, although the lower labial mucosa or other areas of the mouth may also be affected. Clinically, it appears as erythematous mucosa, sometimes accompanied by small papules, white striae, or erosions.

Treatment may include antibiotics such as clarithromycin, with or without corticosteroids, although the most effective treatment is chlorhexidine.[3]

The aim of this study was to describe three cases of LGS with a newly identified etiology.

Cases

Case 1:

A 55-year-old man with multiple oral ulcers and a sensation of swelling in his upper lip for the past six months was referred to the Oral Medicine Department of the Dental School, Shahid Beheshti University of Medical Sciences, Tehran, Iran. An incisional biopsy was performed by a dentist, but the diagnosis was unclear, with lichen planus considered a differential diagnosis. The dentist prescribed diphenhydramine, dexamethasone, and nystatin, but these were not effective. The patient had no history of smoking or alcohol consumption. He had type 2 diabetes and was taking metformin twice daily as prescribed by his physician. He worked in a laboratory and reported previous contact with formalin at work, that he believed triggered the initial lesion on his lip. Intraoral examination revealed multiple chronic, map-like ulcers covered with a white to yellow pseudomembrane and erythematous (erosive) margins on both the upper and lower labial mucosa (Figures 1,2). Involvement of marginal and attached gingiva of maxillary anterior teeth was observed, presenting as desquamative gingivitis (Figure 3).

Figure 1: Map-like Ulcers covered with white to yellow pseudomembrane and erythematous (erosive) margins on upper labial mucosa

Figure 2: Map-like Ulcers covered with white to yellow pseudomembrane and erythematous (erosive) margins on lower labial mucosa

Figure 3: Desquamative gingivitis

Oral hygiene was fair. Extraoral examination revealed only mild swelling of the upper lip. Based on the clinical presentation, the most probable diagnosis was mucous membrane pemphigoid. An incisional biopsy was performed from the upper labial mucosa (affected area) and buccal mucosa (normal tissue). The samples were submitted for histopathological and direct immunofluorescence (DIF) examination. Histopathological sections revealed mucosal tissue covered by non- to parakeratinized stratified squamous epithelium with exocytosis and basal cell degeneration. The underlying connective tissue showed intense lymphocytic infiltration immediately adjacent to the surface epithelium. Deeper nodular aggregates of lymphocytes were observed with perivascular and perineural distribution. The epithelium was partially ulcerated and replaced with a fibrinopurulent membrane. Based on the clinical and histopathological findings, the diagnosis was consistent with LGS. DIF testing was negative for IgM, IgG, IgA, and C3. Treatment with chlorhexidine was initiated for two weeks. Fortunately, the patient's lesions improved significantly with this treatment. On follow-up visits, no new ulcers were observed, and the oral mucosa appeared completely normal. The patient was advised to inform the dentist in case of recurrence.

Case 2:

A 63-year-old man was referred to the Oral Medicine Department with a chief complaint of burning sensation and ulcers on his upper lip. The patient had first noticed the lesions approximately one year ago and had consulted several doctors, but unfortunately, the medications prescribed including corticosteroids had no effect. The patient had no significant medical history and was not taking any medications. Intraoral examination revealed map-like ulcers and erosions covered by a pseudomembrane, each measuring less than 0.2 × 0.2 cm, located on the upper lip. Additionally, generalized erythema of the anterior maxillary gingiva was noted, consistent with desquamative gingivitis (Figures 4, 5).

Figure 4: Map-like ulcers and erosion with pseudomembrane and various sizes lower than 0.2*0.2 cm on upper lip

Figure 5: Desquamative gingivitis

Figure 6: Follow up after one year of treatment

Extraoral examination was within normal limits. To establish a diagnosis, an incisional biopsy was taken from the upper lip. The specimen was submitted to the university's pathology department for histopathological evaluation. Microscopic examination revealed a section of epithelial tissue partially covered by erosive, parakeratinized stratified squamous epithelium, with areas of hydropic degeneration. The underlying fibrovascular connective tissue showed lymphocytic infiltration beneath the neurovascular bundles. Based on the clinical and histopathological findings, the diagnosis was consistent with LGS.

The patient was treated with chlorhexidine mouthwash, and at the two-week follow-up, the lesions had completely resolved. The one-year follow up is shown in Figure 6. The patient was advised to contact the oral medicine specialist in case of any recurrence.

Case 3:

A 59-year-old woman was referred to the Oral Medicine Department with a chief complaint of swelling and burning sensation in the upper lip. The patient reported using antibiotics such as amoxicillin intermittently, which led to partial improvement of the lesions. She was otherwise healthy and had no history of systemic disease, medication use, alcohol consumption, or smoking. Extraoral examination was unremarkable. Intraoral examination revealed generalized erosion of upper labial mucosa associated with a burning sensation (Figure 7). For initial treatment, 0.1% triamcinolone oral paste was prescribed, and a follow-up visit was scheduled for two weeks later.

At the follow-up, the patient's condition had worsened. Intraoral examination revealed map-like ulcers of various sizes on the upper labial mucosa, covered by a pseudomembrane and surrounded by inflamed margins, without any tissue tags (Figure 8). With a provisional diagnosis of bullous lichen planus, an incisional biopsy was performed on the upper lip lesions. The specimen was sent to the university pathology department for histopathological evaluation.

Figure 7: Generalized erosion in upper labial mucosa with burning sensation

Figure 8: Map-like ulcers (in various size) with pseudo membrane and inflamed margin and without tissue tag in upper labial mucosa

Microscopic examination showed fibrous connective tissue covered by hyperplastic, orthokeratinized stratified squamous epithelium. Edema was noted in the superficial lamina propria, along with dilation of lymphatic vessels and diffuse lymphocytic infiltration. Lymphocytes and epithelioid cells were observed around blood vessels and nerve bundles (Figure 9). Mild sialadenitis was also evident. The final diagnosis was LGS. Initially, topical corticosteroids were prescribed, but the response was unsatisfactory. Therefore, treatment was switched to chlorhexidine. At subsequent follow-ups, the patient's symptoms showed significant improvement. The patient was advised to contact her oral medicine specialist in case of recurrence.

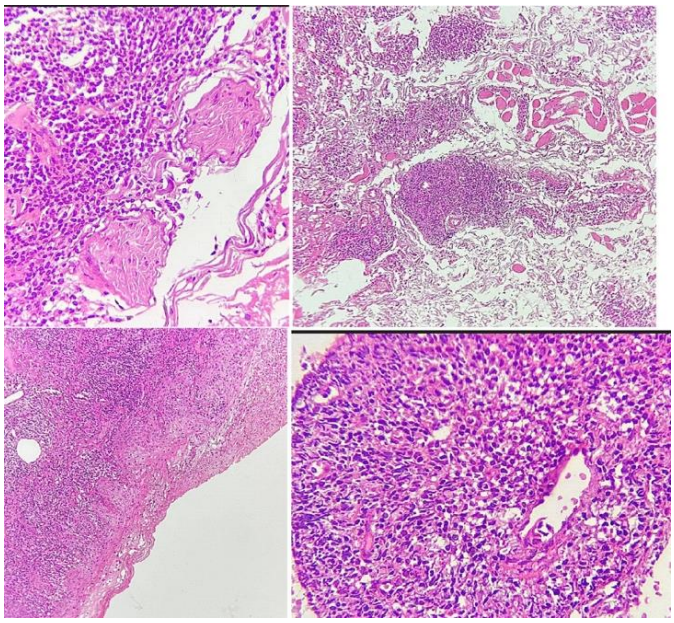

Figure 9: Histopathological examination of the lesion

Discussion

LGS typically involves the skin and oral mucosa, with oral manifestations often resembling lichen planus in clinical appearance.[4] In the cases presented in this study, all three patients only exhibited oral lesions, which appeared clinically similar to lichen planus. The prevalence of these lesions is higher in women, with an average reported age of 59 years. In this study, two patients were male, but their ages were close to the average reported in previous studies.[5] The etiology of LGS is not yet fully understood. However, possible contributing factors include adjacent dental restorations, microbial agents (such as local plaque or calculus), medications, or other systemic agents.[3] Historically, these lesions were referred to as "lichenoid reactions to composite restorations." However, due to their association with lip swelling and resistance to corticosteroid therapy which is typically effective in managing lichenoid reactions—a new term, lichenoid and granulomatous stomatitis, has been adopted.[3] In one of the present cases, the patient reported that the onset of lip lesions coincided with exposure to formalin, whereas the other patients did not report any clear etiological factors. Most reported cases involve the upper lip and upper labial mucosa, although involvement of the lower lip has occasionally been documented.[6] In the current study, all cases involved the upper lip and labial mucosa, with only one case showing brief involvement of the lower lip. The clinical appearance was similar across all cases.

While extraoral examination is usually normal, some patients may report a sensation of lip swelling or enlargement. Histopathologically, LGS closely resembles lichen planus but is characterized by perineural inflammation and discomfort for the patient.[7] The current cases showed parakeratinized stratified squamous epithelium with exocytosis and basal cell degeneration. The underlying connective tissue exhibited dense lymphocytic infiltration, with deeper nodular aggregates of lymphocytes in a perivascular and perineural distribution. DIF is not always necessary, but when performed, the findings are typically negative.

Treatment of LGS remains controversial, with various approaches reported in the literature. Identifying and eliminating the etiological factor, when possible, is the key. In one of the present cases, the suspected etiological factor was an anterior composite restoration; thus, replacing the restoration may be beneficial. Other reported treatments include antibiotics (e.g., clarithromycin) and antiseptic mouthwashes (e.g., chlorhexidine). The effectiveness of corticosteroids is inconsistent. In the present cases, corticosteroid therapy was ineffective, and chlorhexidine mouthwash was prescribed for two weeks. After treatment, all patients showed significant improvement, and the mucosa appeared normal. No recurrences were observed during follow-up visits.[8]

Conclusion

Accurate diagnosis of LGS is crucial due to its clinical and histopathological resemblance to other conditions, particularly lichen planus. Correct identification of LGS facilitates more effective management, as these lesions typically do not respond well to corticosteroid therapy, which is commonly used for similar conditions**.**

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All cases included in the study were reported with the participants' consent and without revealing their identities.

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Using AI: No artificial intelligence (AI) tools were used in the design, analysis, or writing of this study.

Conflict of Interest: "The authors declare no conflict of interest.".

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