

Lichenoid Granulomatous Stomatitis

- an oral medicine case book

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CASE REPORT

A 72-year-old male patient presented with a painful area on the labial mucosa of the upper lip adjacent to tooth 12. He complained of a non-healing “bruise” on the inside of his lip that had been present for about 4-weeks. The patient was a non-smoker and reported taking anti-hypertensive medication (Lisinopril, hydrochlorothiazide) for 10-years. A prior colonoscopy 4 years ago revealed adenomas.

On examination, a swelling was noted with the overlying mucosa appearing erosive and erythematous with peripheral white striae, clinically similar to lichen planus. The lesion also involved the facial gingiva associated with teeth 12 & 22 (Figure 1). This red-white lesion was excised as well as minor salivary glands that extruded during the biopsy procedure. No additional pharmacotherapeutic agents were administered.

The specimen submitted from the lip lesion consisted of a mucosa-covered tissue fragment measuring 12x5x4mm. Histological evaluation confirmed the presence of a tissue fragment surfaced by stratified squamous epithelium with areas of hyperparakeratosis, as well as vacuolar degeneration of the basal cell layer with associated apoptotic bodies. A band-like lymphohistiocytic infiltrate was seen in the underlying superficial lamina propria. Secondly, varying degrees of granulomatous inflammation within the superficial lichenoid inflammatory infiltrate was noted. These poorly formed granulomas were composed of epithelioid macrophages, however no giant cells or central necrosis could be appreciated. Additionally, lymphoid follicles were seen, with a striking

perineural and perivascular distribution (Figures 2 & 3). No foreign material was noted under polarised light. Periodic acid–Schiff (PAS) and Ziehl–Neelsen histochemical stains failed to highlight any fungal elements or acid-fast bacilli respectively.

In conclusion, a final diagnosis of lichenoid granulomatous stomatitis was made.

The patient was followed-up one month after total excision of the lesion to reassess for further treatment. He reported that healing was uneventful, and all symptoms had disappeared after the excision biopsy. Intra-oral examination showed an absence of any clinical signs in the original area (Figure 4).

The patient will be followed-up for routine examination every few months and was instructed to immediately report back should symptoms reappear.

DISCUSSION

The presence of lichenoid inflammation with concomitant granulomatous inflammation is an uncommon observation within the oral cavity. Many diseases are typified by either lichenoid or granulomatous inflammation.¹ However when both patterns occur simultaneously, problems arise in determining which pattern represents the primary disease process, or whether the coexistence of both patterns represents a distinctive disease entity.

The term lichenoid granulomatous stomatitis (LGS) was first described in literature by Robinson et al. in 2006.¹ Lichenoid inflammation may render the oral mucosa susceptible to the ingress of foreign material, resulting in granuloma formation. LGS has been reported in cases of foreign body gingivitis. In a series of 61 foreign body gingivitis cases, investigators reported the presence of both patterns of inflammation in 26% of biopsies studied.² In the present case, no foreign material could be identified under polarised light.

To date the largest review by Hakeem et al.³ in 2019 identified 47 patients with LGS. In this study, patient demographics showed a female predilection of 1.9:1 with a mean age of 59 years. Seventy-nine percent of patients were older than 55 years. Patients commonly presented with a solitary lesion, with most cases occurring on the attached gingiva followed by the buccal mucosa and vestibule. With regards to clinical description, 38% were described as erythroleukoplakia, 36% as leukoplakia, and 26% as purely erythematous lesions.

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2. André W van Zyl: Clinical case, treatment and follow-up - 25%
3. Willie FP van Heerden: Diagnosis, histological images and advisor - 25%

There was an equal incidence of presentation amongst patients regarding painful or non-painful lesions. The clinical impressions for all cases in this study (for which multiple were listed in some instances) included lichen planus (17 cases), dysplasia/carcinoma *in situ*/squamous cell carcinoma (11 cases), vesicubullous lesions (9 cases), trauma-associated (5 cases), leukoplakia (5 cases), allergy (2 cases) and other differentials (4 cases). No clinical diagnosis was reported in 7 cases.³

Histologically, LGS consists of three distinctive components. First, is the presence of lichenoid inflammation, characterised by hyperkeratosis, basal cell degeneration with associated apoptotic bodies and a band-like lymphohistiocytic inflammatory cell infiltrate. Secondly, variable degrees of granulomatous inflammation can be seen throughout the corium. Importantly, all granulomas consist of epithelioid macrophages without giant cells or areas of necrosis. Thirdly, lymphoid follicles are present in the corium showing a prominent perineural distribution.¹

Additional studies ruling out infective agents and foreign material should be performed in suspected cases. A study of six cases of LGS by Robinson et al.¹ found that

presence of fungal hyphae was not associated with a lichenoid inflammatory reaction.¹ Secondly, granulomatous inflammation is typical of deep mycoses and not superficial candidosis.

Patients taking certain medications may develop LGS, which may ultimately resolve with discontinuation of the medication.³ Additionally, the case review by Robinson et al., reported two patients known to be on medications that have an association with lichenoid eruptions, namely Naproxen (Non-steroidal anti-inflammatory drug), Atenolol (β -adrenoceptor blocker), and Ramipril (Angiotensin-converting enzyme inhibitor).⁴ Furthermore, these groups of drugs have also been implicated in both lichenoid and granulomatous dermatitis.

Equally rare is the presence of both patterns of inflammation in dermatological conditions. Lichenoid granulomatous dermatitis (LGD) was first described by Gonzalez in 1986.⁵ A study by Magro and Crowson⁶ reported a series of 40 patients with skin lesions showing lichenoid dermatitis with a granulomatous component. The majority of these cases had confounding medical problems associated with the disease, however one-fifth of the cases were considered idiopathic. Furthermore, in 12 cases an infective cause was implicated. The agent was either a



Figure 1. Initial clinical presentation.

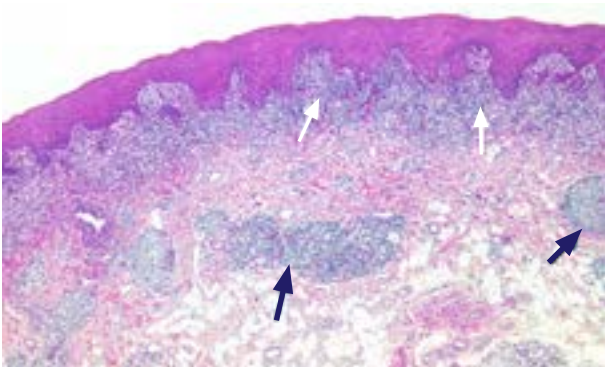


Figure 2. H&E-stained section showing the band-like inflammatory cell infiltrate (white arrows) and lymphoid aggregates in a perineural and perivascular distribution (black arrows) (original magnification x 40).

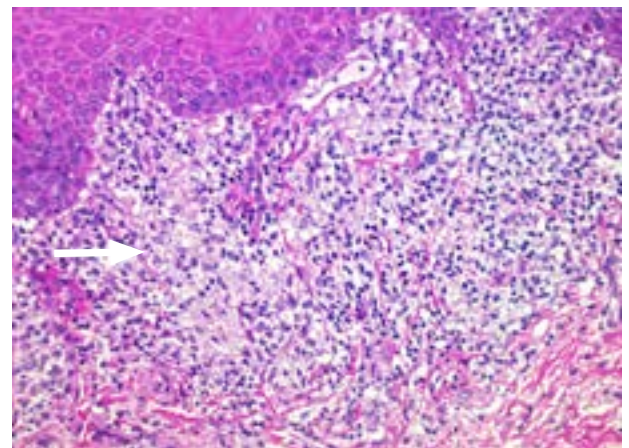


Figure 3. H&E-stained section showing a poorly formed granuloma (white arrow) within the superficial inflammatory cell infiltrate (original magnification x 200).



Figure 4. Clinical presentation one month post initial biopsy.

or bacterial infection and not fungal in origin.⁶ Both inflammatory patterns have also been reported to coexist in a rare skin condition, lupus erythematosus profundus.⁷

In total approximately 57 cases have been previously reported as LGD. The gender ratio reported in the prior cases showed a slight female predilection of 1.3:1 with mean age of 48 years. The trunk, arms, and legs were the most common location. Dermatologic lesions mostly presented as erythematous or as maculopapular entities.^{6,8-9}

Although many similarities were found when comparing histological features of LGS and LGD, some important differences were noted. Cases from the oral mucosa did not show an interstitial array between collagen fibers surrounded by palisaded histiocytes, granuloma annulare-like appearance, focal Langhans giant cells or granulomatous vasculitis. Additionally, a prominent perivascular inflammatory infiltrate, as seen in LGS cases, was not emphasised in descriptions of lesions involving the skin.³

Literature is sparse regarding the treatment of LGS, however, it appears to respond well to similar regimens used in treating conventional lichen planus.³

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