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CASE REPORT

A recalcitrant case of erosive Oral Lichen Planus

Renita Lorina Castelino, Sajad Ahmad Buch, Devika S Pillai, Chethana Dinakar

Lichen Planus is an inflammatory condition of the skin and mucous membrane that can also affect oral mucosa in a variety of forms. The mucosal form has six types and at least two of them carry a risk for malignant transformation and thus warrant a follow-up. The cutaneous form is pruritic and sometimes can be self-limiting but oral lichen planus (OLP), is a chronic inflammatory disease with relapses and remissions. A case of erosive lichen planus, treated with topical clobetasol propionate 0.05% and systemic corticosteroids is reported. The lesion healed after 3 weeks of treatment, following which the patient was referred for needful restorative treatment. The patient had a recurrence during the follow-up and the second course of treatment was administered. There is no well-defined treatment for OLP yet steroids have a vital role in symptomatic relief. Although topical steroids are the mainstay, recurrent, multiple, and large lesions are supplemented with systemic corticosteroids.

Dr Renita Lorina Castelino
BDS, MDS
NITTE,
Department of Oral Medicine and Radiology,
AB Shetty Memorial Institute of Dental Sciences (ABSMIDS),
Mangalore, India
ORCID ID : 0000-0002-8696-549X

***Dr Sajad Ahmad Buch**
BDS, MDS
Department of Clinical Oral Health Sciences,
School of Dentistry
IMU University,
Kuala Lumpur, Malaysia
ORCID ID : 0000-0002-1241-4679

Dr Devika S Pillai
BDS, MDS
Department Oral Medicine and Radiology & Special Care Dentistry,
Saveetha Dental College and Hospitals
Chennai,
Tamil Nadu, India
ORCID ID : 0000-0001-7549-6395

Dr Chethana Dinakar
NITTE,
Department of Oral Pathology,
AB Shetty Memorial Institute of Dental Sciences (ABSMIDS),
Mangalore, India
ORCID ID : 0000-0002-5024-4906

**Corresponding author*
buchh.sajad@gmail.com

Lichen planus is a mucocutaneous disease of inflammatory origin affecting around 1-2% of the general population. It can occur in single or multiple cutaneous and non-cutaneous (mucosal) areas within the body. Consequently the disease can affect either skin alone, the oral cavity alone, or occur in both the areas simultaneously; it may also affect other sites namely nails, scalp, oesophagus, and the genital areas.[1,2]Although the aetiology is yet elusive, the various factors attributed include genetics, immunologic factors, medications, and hepatitis C infection. The literature shows six distinct types of OLP, namely reticular, atrophic, erosive, plaque-type, popular, and bullous type;[3]one or more forms can coexist in a single patient. The atrophic and erosive types are reported to carry an increased risk for malignant transformation. OLP is usually found in a bilateral fashion but need not be symmetrical. The common sites for OLP are buccal mucosa, dorsal surface of the tongue, and gingiva, at times occurring on other rare sites as well.[4] The reticular OLP is the most common form which is asymptomatic and is usually diagnosed on routine oral examination. The erosive OLP is the most noteworthy of all the types having characteristic symptoms and consisting of the lesions in the form of a network formed by radiating keratinized striations;[5] the other types with symptoms and difficult to treat include the atrophic and the bullous types. The erosive OLP becomes multifocal with time and patients complain of varying amounts of pain and interference with the normal mastication process. Topical or systemic corticosteroids remain the mainstay of treatment for OLP; however, it is advised to use further adjuvants to enhance the process of healing and assist in modulation of the immune process in the OLP microenvironment. We report a case of erosive OLP effectively treated with topical clobetasol propionate and systemic prednisone.

CASE REPORT

A 60-year-old male patient with no underlying systemic anamnesis approached our department of oral medicine, for a therapeutic opinion for a painful ulcer of 6 months duration on the left buccal mucosa. The patient had no history of smoking or tobacco chewing. The ulcer on the left buccal mucosa had increased in size in the last 6 months. In addition, the patient complained of pain and tenderness during phonetics and while having food. The patient was using Tess buccal paste (Triamcinolone acetonide 0.1%); advised by a local Dentist he had visited around 15 days back, but without any significant relief. On examination patient had an irregular shallow ulcer around 3cm x 2cm on the posterior

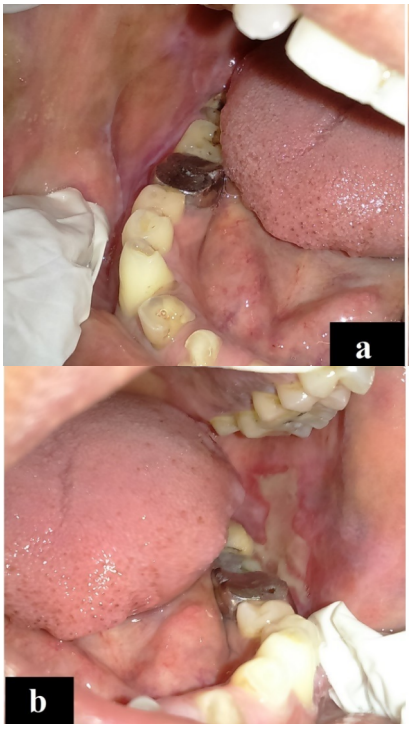

Figure 1 *A) Reticular lichen planus on the right buccal mucosa. B) Erosive lichen planus on the left*

region of the left buccal mucosa; the right buccal mucosa had white striations which the patient was not aware, possibly due to lack of any symptoms. (Figure 1a & 1b) The hard tissue examination revealed metal crowns on teeth numbers 36 and 46, and amalgam restoration with respect to tooth number 37. The approximation of amalgam restorations and the metal crowns prompted us for a provisional diagnosis of oral lichenoid reaction (OLL), and a differential diagnosis of OLP Incisional biopsy of the lesion on the left buccal mucosa was carried out and showed features of erosive lichen planus without any dysplastic features. Stratified squamous parakeratinised epithelium was noticed with saw tooth rete ridges in a few areas. (Figure 2a) Areas of ulceration surrounded by hyalinization and a thick band of inflammatory infiltrate can be appreciated below the dense mixed inflammatory infiltrate; chiefly neutrophils. (Figure 2b) Most of the areas

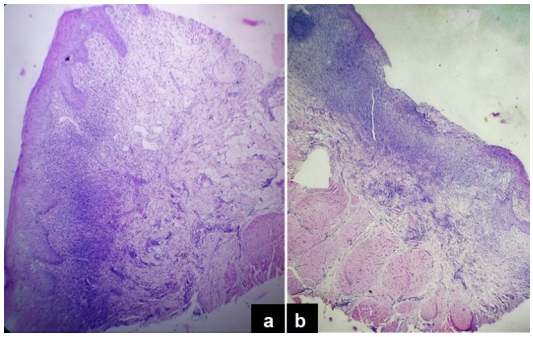

Figure 2 *A) Stratifed squamous parakeratised epithelium with saw tooth rete ridges and dense subepithelial lymphocytic infiltration. B)Ulceration surrounded by hyalinization and thick band of inflammatory infiltrateposterior buccal mucosa.*

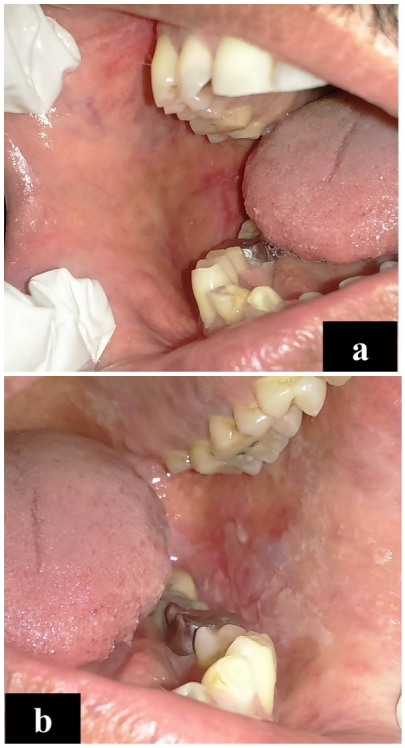

Figure 3 *A) Resolution of lesion on the right buccal mucosa at 7 days of treatment B) Marked improvement on the left buccal mucosa after 7 days of treatment*

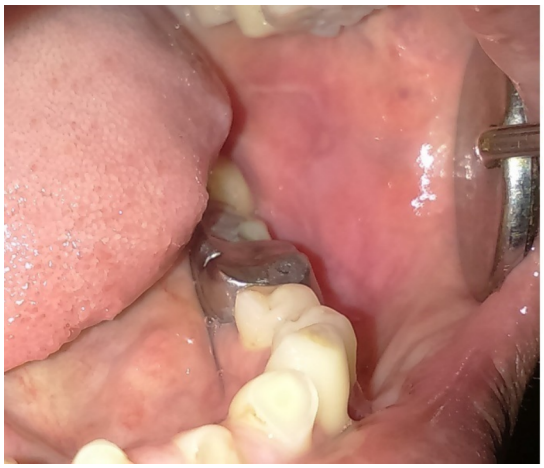

Figure 4 *Complete resolution of erosive lichen planus after initial course of corticosteroids, on the left buccal mucosa after 3 weeks of treatment.*

have subepithelial epithelium and the deep connective tissue has muscle fibers, adipose tissue, and extravasated RBCs. The patient was advised topical clobetasol propionate 0.05% and systemic prednisone in the form of 20 mg/day oral tablets for 1week. The patient was evaluated after 1 week with marked improvement noticed on both the right and the more severe left side. (**Figure 3a & 3b**) The patient was advised to continue topical clobetasol propionate 0.05% and systemic prednisone was tapered for another week. The patient showed remission in lesions bilaterally in 3rd week of treatment, and the erosive lesion on the left buccal mucosa was completely resolved. (**Figure 4**) The amalgam restorations and the metal crowns were replaced with ceramic restorations and the patient was kept on follow-up. The patient reported back with relapse even after the replacement of the restorations, (**Figures 5a & 5b**) supporting our clinicopathological diagnosis of OLP The patient was retreated with local and systemic corticosteroids and showed signs of healing during the second course of treatment. (**Figure 5 c & 5d**) The patient did not report for further evaluation.

DISCUSSION

The etiology of lichen planus is still not clear. Much of the data yielded through years of research, attributes immunologic mechanisms as the mainstay for the pathogenesis of the lichen planus.[6] The role of Langerhans cells, the mast cells, and their interactivity with the abundant T-cells assembled in the underlying connective tissue has been primarily focused on in the literature. The evidence gathered over the years suggests lichen planus is an autoimmune disorder mediated by T-cells in which apoptosis of basal keratinocytes occurs as a result of cluster of differentiation (CD8) + T cells. OLP is a non-infectious chronic inflammatory disorder of stratified squamous epithelium of oral mucosa and the underlying lamina propria with or without its

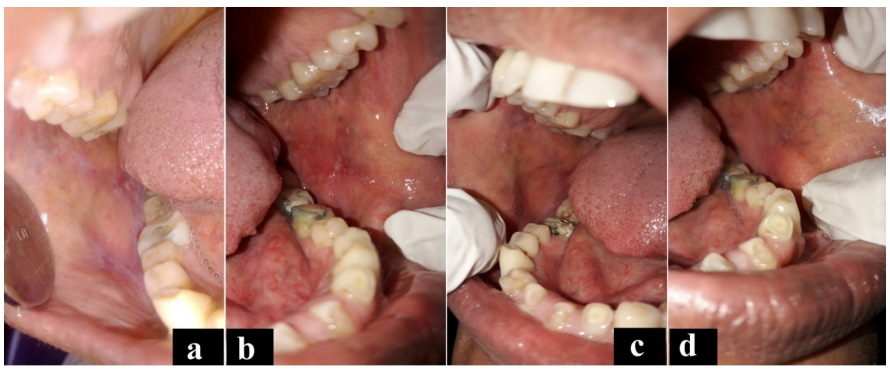

Figure 5 *A,B) Recurrence after replacement of metallic restorations. C,D) Improvement during second course of treatment.*

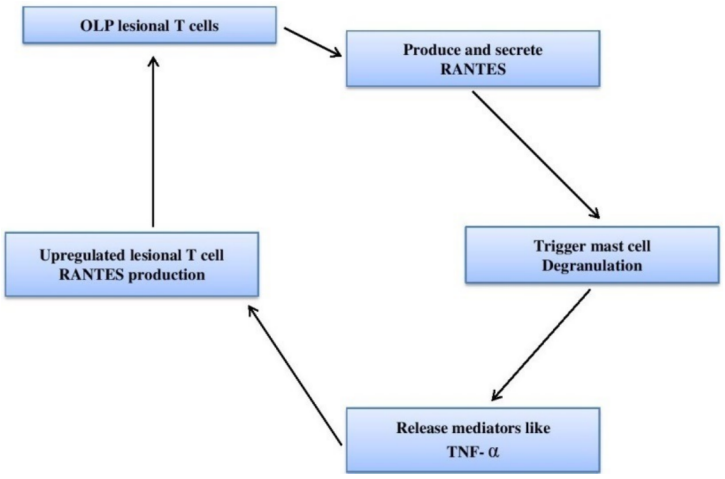

Figure 6 *The disease chronicity of oral lichen planus promoted by cyclic mechanism*

cutaneous form. It is estimated that 30% to 50% of cases with OLP have cutaneous manifestations, and such skin lesions can aid in the diagnosis of this disorder. Lichen planus occurs on cutaneous sites only, in 35% of cases, and 25% of cases have only a mucosal presentation. OLP usually occurs in the middle-aged and elderly with a female to male ratio of 1.5: 1. OLP has significantly higher chronicity than cutaneous lichen planus. Although this inconsistency in chronicity between OLP and its cutaneous counterpart is not clear, the role of T-cell RANTES (regulated upon activation, normal T-cell expressed and secreted) and mast cell degranulation, that releases tumor necrosis factor α and a cyclical process, where interleukin 4 and interferon may forecast the chronic nature of the disease in certain cases. (Figure 6)[1]

Clinical characteristics of OLP are dictated by the severity of the disease and present in the range of clinical appearances. The four types of OLP lesions most often described and discussed are reticular, erosive, plaque-like, and bullous. (Table 1) Topical and systemic `corticosteroids are the most widely used agents for OLP with the aim to modulate a patient's immune response. Topical corticosteroids constitute the mainstay and depending upon the degree of severity, the agents commonly used in increasing order of their potency are triamcinolone acetonide, fluocinonide, or fluocinolone acetonide (fluorinated steroids), betamethasone phosphate, and halogenated clobetasol propionate.[7] Topical corticosteroid resistant and recalcitrant OLP can alternatively be managed by other topical agents, like tacrolimus or cyclosporine, both calcineurin inhibitors, and sometimes by retinoids.[8] Systemic corticosteroids have a decent role in recalcitrant, erosive, or when topical agents are ineffective. A number of studies have established systemic corticosteroids as the most effective modality of treatment for OLP[9] As corticosteroids dosage has a wide range, and further due to variable patient responses to systemic corticosteroids, a number of dosing options exist. The oral dose of prednisone ranges from 10–20 mg/day for moderate OLP to as high as 35 mg/day for extreme cases.[7] It is challenging to treat and diagnose symptomatic OLP in a definite manner due to the lack of a uniform method and also due to variable individual factors.[10]

Figure 6 *The disease chronicity of oral lichen planus promoted by cyclic mechanism*

OLP type	Clinical features and favoured location
Reticular	Asymptomatic and most common type and hence diagnosed often during routine examination; lacy whitish streaks, surrounded by an erythematous border. Reticular OLP may change into one of the severe subtypes, like erosive form. Most commonly occurs on buccal mucosa bilaterally.
Erosive	The most advanced subtype, can present clinically as atrophic or erythematous ulcerations and erosions of the mucosa with faint radiating white striae. Occasionally the ulcers are covered by a pseudomembrane. The atrophic and ulcerative form confined to gingiva is giving rise to a pattern, desquamative gingivitis. This pattern may also represent pemphigus vulgaris or mucous membrane pemphigoid and thus warrants histopathological examination. It causes mild discomfort to severe episodes of pain and might cause dysgeusia on dorsum of the tongue.
Plaquelike	Large homogenous white patches are typical of plaque-like OLP. As most focal leukoplakia lesions have similar clinical picture, therefore leukoplakia should be ruled out. Most commonly seen in tobacco smokers, it occurs commonly on tongue.
Bullous	Rarely OLP may show bullous lesions, with bullae size ranging up to 2 cm. The bullae rupture in the oral cavity similar to other vesiculobullous lesions, leaving ulcerations on an inflamed mucosa. Bullous type most commonly involves posterior aspect of buccal mucosa.

CONCLUSION

The present case with a history of 6 months duration and ineffective results with topical triamcinolone acetonide was managed with a combination of a super potent topical agent (clobetasol propionate) and a systemic (oral) corticosteroid. The patient had a relapse during the course of the treatment. There is a need for large group studies of clinical trials to unravel potential medications for the effective treatment of OLP

Declaration Of Patient Consent

Duly signed informed consent was taken from the patient for publication purposes. It was conveyed to the patient that his name and initials shall be concealed, and all efforts would be taken to hide his identity although anonymity cannot be guaranteed.

REFERENCES

1. De Rossi SS, Siarrocca K. Oral Lichen Planus and Lichenoid Mucositis. Dent Clin North Am. 2014; 58: 299-13.
2. Lucchese A, Dolci A, Minervini G et al. Vulvovaginal Gingival Lichen Planus: Report of two cases and review of literature. Oral Implantol (Rome). 2016; 9:54-60.
3. Canto AM, Müller H, Freitas RR et al. Oral Lichen Planus (OLP): Clinical and Complementary Diagnosis. An Bras Dermatol. 2010; 85:669-75.
4. Ismail SB, Kumar SK, Zain RB. Oral Lichen Planus and Lichenoid reactions; Etiopathogenesis diagnosis, management and malignant transformation. J Oral Sci. 2007; 49:89-106.
5. Sharma S, Saimbi CS, Koirala B. Erosive Oral Lichen Planus and its management: A case series. J Nepal Med Assoc. 2008; 47: 86-90.
6. Thornhill MH. Immune Mechanisms In Oral Lichen Planus. Acta Odontol Scand. 2001; 59:174-77.
7. Mester A, Lucaciu O, Ciobanu L et al. Clinical features and management of Oral Lichen Planus (OLP) with emphasis on the management of Hepatitis C Virus (Hcv)-Related OLP. Bosn J Basic Med Sci. 2018; 18:217-23.
8. Al-Hashimi I, Schifter M, Lockhart Pb et l. Oral Lichen Planus And Oral Lichenoid Lesions: Diagnostic And Therapeutic Considerations. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2007; 103:E1-12.
9. Alrashdan Ms, Cirillo N, Mccullough M. Oral Lichen Planus: A Literature Review and Update. Arch Dermatol Res. 2016; 308:539-51.
10. Thongprasom K. Oral Lichen Planus: Challenge And Management. Oral Dis. 2018; 24:172-3.