

Oral bullous lichen planus: Case report and review of management

ARCHANA PATIL, SHIVA PRASAD, L. ASHOK, G. P. SUJATHA

Abstract

A 34-year-old female patient with the chief complaint of burning sensation in the oral cavity associated with generalized pruritis, scalp and skin lesions diagnosed as Bullous lichen planus and treated with systemic prednisolone, levamisole, benzydamine oral rinse. Patient is in follow up since 1 year and free of lesions. Here we report the case and review current modalities in the management of oral lichen planus.

Keywords: Bullous lichen planus, pruritis, burning sensation

Introduction

Oral lichen planus (OLP) is a common disorder that affects stratified squamous epithelium virtually exclusively. It is seen worldwide, mostly in the fifth to sixth decades of life, and is twice as common in women than in men. Here we report case of bullous lichen planus.

Case Report

A 34-year-old Indian female patient reported to the Department of Oral Medicine and Radiology with the chief complaint of burning sensation of oral cavity since 4 days. Burning sensation was insidious in onset, preceded by eruption of fluid filled vesicles that used to burst within few seconds, continuous, severe intensity aggravated on having spicy food substances and no relieving factors, episodes of such occurrence of blisters 10–15 times per day. She was also associated with generalized body itching and mild fever. History of recurrent episodes of burning sensation since 8 years but this time associated with severity. Her medical history revealed occurrence of dermal and scalp lesions, loss of hair, generalized pruritis for which she was taking treatment by a dermatologist since 10 years and presently was

on the following medications: pimecrolimus (pacroma cream 0.01%), halobresol propionate cream (halox cream 0.05%), and ketoconazole shampoo (Keto Az shampoo). History of anal hemorrhoids treated 4 years back. Her past dental history revealed that extraction of mobile teeth and gingival flap surgery was done 8 years back.

No significant family history. She was married since 13 years blessed with two children (girls) and stays in a joint family, often her life was associated with stress and tension as reported by the patient. Vegetarian by diet. No history of any deleterious habits. She brushed once daily in horizontal manner using brush and paste. General physical examination revealed that she is moderately built and nourished, appeared depressed and sad. Depression Anxiety and Stress Scale [DASS] score indicated moderate depression, mild anxiety, and moderate stress. Scalp had isolated central area of hair loss, solitary ulcerative lesion measuring about 3 × 4 cm, margins were irregular, floor covered with yellow slough, tender on palpation. Nails present with mild longitudinal ridging. Black healed pigmented lesions of varying sizes were evident on legs, hands, and back [Figure 1].

On intra-oral examination solitary mixed red and white lesions, irregular in shape, measuring about 2 × 3 cm i.r.t right posterior buccal mucosa extending anteriorly from mesial of 45 posteriorly distal to 48, superiorly from occlusal plane of 14 inferiorly up to buccal vestibule i.r.t 45 46 47 48 regions, surface presented with bullae filled with clear fluid measuring about 1 × 0.8 cm, margins are irregular with pseudo membrane slough in the posterior buccal mucosal region, surrounding buccal mucosa was normal [Figure 2].

Left buccal mucosa presented with solitary red and white lesion measuring about 3 × 3.5 cm irregular in shape, extending along the line of occlusion extending supero-inferiorly about 1 cm on either side, has central area of erythema surrounded surface by white striae [Figure 2].

Department of Oral Medicine and Radiology, Bapuji Dental College and Hospital, Davangere, Karnataka, India

Correspondence: Dr. Archana Patil, Department of Oral Medicine and Radiology, Bapuji Dental College and Hospital, Davangere- 577 004, Karnataka, India.
E-mail: archanapatil2803@gmail.com

Access this article online	
Quick Response Code: 	Website: www.contemplindent.org
	DOI: 10.4103/0976-237X.103634

Hard palate showed solitary white lesion measuring about 1.5 × 1 cm irregular in shape, extending anteriorly from mid-palatal raphe posteriorly to line joining distal surface of 13 23, white papules, interspersed with striae, surrounding mucosa was normal.

On palpation inspeactory findings were confirmed as tender, smooth, and nonscrappable.

Gingiva was observed with generalized gingival inflammation, erythematous, bleeding on probing, tender, lower right side



Figure 1: Lesions on scalp and skin

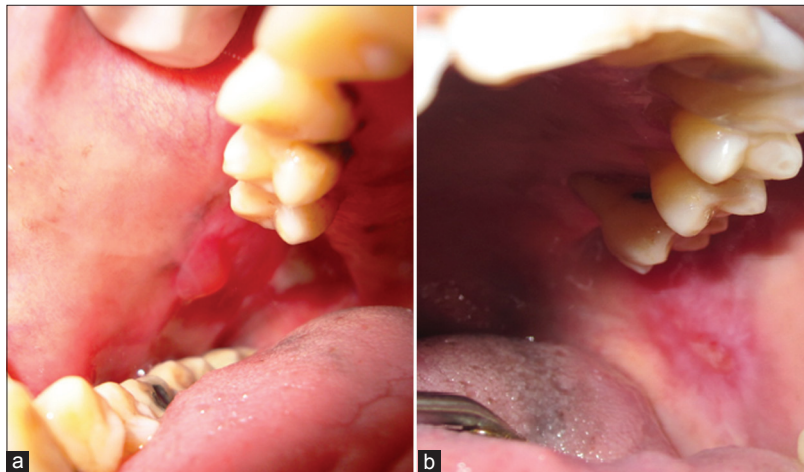


Figure 2: Lesions on right and left buccal mucosa



Figure 3: Posttreatment-healed lesions of scalp and buccal mucosa

of posterior gingiva had typical white striae. Generalized pockets were present. Hard tissue examination reveals teeth 15 14 13 12 11 21 22 23 3 24 25 27 48 47 46 45 44 43 42 41 31 32 33 34 35 38. 38 – Grade III mobility. Considering history and clinical features Bullous lichen planus (LP) was considered as provisional diagnosis with the differential diagnosis pemphigus vulgaris, Bullous pemphigoid, mucus membrane pemphigoid, Lichenoid contact stomatitis.

Patient was subjected to investigations like orthopantomogram (OPG), complete hemogram, blood sugar, cytosmear, and incisional biopsy from right buccal mucosa. All laboratory findings were within normal limits, and biopsy confirmed as LP, which showed typical histopathological features.

She was treated with Tab levamisole (Vermisol) 150 mg once daily for 3 days, Tab prednisolone (Wysolone) 10 mg 4 times a day for 10 days, benzydamine oral rinse 0.15% (Tantum oral rinse), ranitidine 150 mg (Histac EVT) once daily for 10 days. On second visit after 10 days, patient reported with 50% reduction in burning sensation and also episodes of occurrence of bullae was reduced from 10–15 times/day to 7–8 times/day. The dose of wysolone was tapered. On third visit after 15 days patient reported with complete reduction of burning sensation and was enjoying the taste of food. Episodes of bullae occurrence was 4 times in 15 days. On fourth visit after 15 days lesions on right and left buccal mucosa had healed for remaining white lesions, triamcinolone (caziq) was prescribed for topical application. On fifth visit her lesions on right and left buccal mucosa had healed, her scalp lesions too were healed and erythematous gingiva was much reduced [Figure 3]. Patient was referred to Department of Periodontics for oral hygiene prophylaxis. Patient is still under follow up since a year and free of lesions.

Discussion

Oral Lichen planus (OLP) is a common chronic immunological inflammatory mucocutaneous disorder that varies in appearance from keratotic (reticular or plaque like) to erythematous and ulcerative.^[1] The history of LP dates back to 1869, when Erasmus Wilson^[2] first delineated and named the disease. In 1895, Thieberg identified the oral lesion. About 1-2% of world population suffer from LP. 1.5%-of Indians suffer from this disorder, age range of occurrence is 30–70 years, with female predilection Male:Female-1:1.4.^[3]

The different etiological factors considered for LP are genetic background, dental materials, drugs, infectious agent, autoimmunity, immunodeficiency, food allergy, stress, habits, trauma, diabetes, hypertension, malignant neoplasm, and bowel diseases.^[4]

The pathogenesis of LP is thought of from four mechanisms Antigen specific cell mediated immune response (heat shock proteins, CD4+ T helper cells, CD8+ cytotoxic T cells) Nonspecific mechanism (epithelial basement membrane, mast cells, chemokines, matrixmetaloproteinases) autoimmune response, humoral immunity (circulating autoantibodies to desmoglein 1 and 3).^[4]

Extra-oral manifestations

Patients with OLP frequently have concomitant disease in one or more extra-oral sites. Approximately 15% of patients develop cutaneous lesions.^[5] The classic appearance of skin lesions described by the six p's: planar, plaque, pruritic, purple, polygonal, and papular. Typically cutaneous lesions develop after the appearance of oral lesions and severity of oral lesions does not correlate with cutaneous lesions. Undoubtedly the most frequent extra-oral site in 20% of female patients with OLP is genital mucosa. Association of LP of vulva, vagina, and gingiva is recognized as vulvovaginal–gingival syndrome. When LP affects genital mucosa the erosive form of disease is predominant type. The penogingival syndrome represents male equivalent of vulvovaginal syndrome.^[6]

Lichen planopilaris represents LP involvement of scalp and hair follicles causing a scarring alopecia. Typically occurs in three forms classic Lichen Planopilaris (LPP), frontal fibrosing alopecia, Graham-Little syndrome. The combination of follicular LP with scarring alopecia of scalp and non-scarring alopecia of axilla and pubis or other areas is known as Graham-Little syndrome.^[7]

LP can also involve nails producing longitudinal ridging of nail plate, onycholysis, subungual hyperker-atosis, trachyonychia, pterygium formation. Other sites of involvement in LP are esophagus, ocular, bladder, nasal, laryngeal, otic, gastric, and anal.^[8]

Oral manifestations

The red and white components of lesion can be part of following textures.

Reticular

White fine striae/network can show annular/circular patterns. The striae often present with peripheral erythemous zone, which reflects sub epithelial inflammation. Most frequently observed bilaterally in buccal mucosa and rarely on mucosal side of lips.^[9]

Papular

Present in initial phase of disease, clinically characterized by small white dots which in most times intermingle with reticular form.^[9]

Plaque like

Homogenous well demarcated white plaque often but not always surrounded by striae.^[9]

Erythematous/atrophic

Characterized by homogenous red area, striae are frequently seen at periphery. Some may exclusively display erythematous OLP of attached gingiva, which represents desquamative gingivitis.^[9]

Erosive: Ulcerative, Bullous

Ulcerative and bullous are most disabling. Clinically present with fibrin coated ulcers surrounded by erythematous zone frequently displaying radiating white striae. Bullous lesions vary from 4 mm to 2 cm and rupture leaving erythematous zone.^[9]

Diagnosis

The characteristic clinical aspects are sufficient to make a correct diagnosis if classic lesions are present. An oral biopsy with histopathologic study is recommended to confirm clinical diagnosis and also to exclude dysplasia and malignancy.^[10] The value of direct immunofluorescence for confirmation of disease is well accepted, especially with nondiagnostic histopathologic features and for desquamative gingivitis.^[7]

Management

To date no cure for OLP or dermal counterpart. The treatment goal is 2-fold, that is, alleviation of symptoms, monitoring of dysplastic changes.^[11]

Corticosteroids have shown to be predictable and effective medications for controlling signs and symptoms. The following topical medications have been tried in the treatment of OLP: -Fluocinonide 0.05% in an adhesive base improved OLP with no adverse effects;^[12] Betamethasone showed effectiveness in symptomatic OLP in another study;^[13] Hydrocortisone hemisuccinate aqueous solutions with little benefit in treating OLP;^[14] Fluticasone propionate spray and betamethasone sodium phosphate mouth rinse have been used effectively in short-term management of symptomatic OLP;^[15] Mometasone furoate microemulsion resulted in significant reduction in pain in erosive ulcerative OLP and significantly reduced surface area of erythema and ulceration;^[16] Clobetasol propionate 0.05% in various forms like orabase, ointment or aqueous solution shown to be effective for OLP in many studied subjects;^[17,18] Application of clobetasol 17 propionate orabase paste 0.05% plus 100,000 IU/ml of nystatin by means of tray appeared to be efficacious treatment for severe erosive gingival lesions and showed complete response in 33 cases over 48-week period.^[19] Clobetasol propionate 0.05% was found to be as useful as tacrolimus 0.1% in treatment of OLP in another study.^[20]

Triamcinolone acetonide 0.1% in orabase showed better results than cyclosporin,^[21] Pimecrolimus 1% cream,^[22] Betamethasone oral minipulse therapy, Fluocinolone acetonide 0.1% orabase,^[23] which have also been used for treatment of OLP.

In lesions recalcitrant to topical therapy intralesional corticosteroids can be effective, often triamcinolone acetonide 5 mg/ml combined with local anesthetic to inject 0.1 ml/cm³. Systemic steroids are indicated for brief treatment of severe exacerbations of OLP. Prednisone 30–60 mg depending on severity of lesion is usually administered.^[24]

Retinoids are useful and are frequently used in combination with topical steroids as adjuvant therapy. Topical immunosuppressive drugs have been very much effective in treating recalcitrant cases of OLP. Cyclosporin mouth rinse (containing 100 mg of cyclosporine per milliliter) three times daily. Despite of encouraging results in double-blind trials, its use is limited to due to hydrophobicity, high cost, and poor taste.^[25] In addition to concern over its role in viral reproduction and malignant change have restricted its use. 0.1% topical tacrolimus/pimecrolimus ointment used effectively in treatment of erosive LP.^[24]

Apart from these other treatment modalities are Dapsone 100 mg once daily for 3 months, PUVA therapy, Azathioprine: 150 mg/day, Levamisole: 150 mg/day for 3 consecutive days in 1 week, Thalidomide: 200 mg/day or topical 1% paste, giesofulvin have reported to be effective in treatment of OLP in various case reports in literature,^[25] unfortunately randomized control studies are lacking in this aspect.

Lichen planus: A premalignant lesion

The possible premalignant character of LP is subject of controversy and ongoing debate in literature, but the range of malignant transformation is reported to be between 0.4% and 5% over period of observation from 0.5 to 20 years.

Conclusion

The term OLP is a heterogeneous group of patients afflicted with mucosal disease, identifying and eliminating multifactorial agents associated with the disease is essential. Relief can be achieved in most of patients with topical steroids alone or in combination with other immunomodulatory topical agents. Infrequently patients require prolonged use of systemic medications. Patient should also be kept under long-term follow up due to malignant tendency of LP. All treatments are nonspecific and directed at eliminating inflammation and therefore are partially successful.

Acknowledgment

Dr. Pramod B M, PG student Department of Orthopedics, JJMC, Davangere.

References

- Greenberg MS, Glick M, Ship JA. Red and white lesions of oral mucosa. *Burkets Oral Medicine*. 11th ed. BC Decker Inc

- publication, Hamilton; 2008.
2. Wilson E. On lichen planus. *J Cutan Med Dis Skin* 1869;3:117-32.
 3. Rajendran R. Oral lichen planus. *J Oral Maxillofac Pathol* 2005;9:3-5.
 4. Roopashree MR, Gondhalekar RV, Shashikanth MC, George J, Thippeswamy SH, Shukla A. Pathogenesis of oral lichen planus—a review. *J Oral Pathol Med* 2010;39:729-34.
 5. Eisen D. The evaluation of cutaneous, genital, scalp, nail, esophageal, and ocular involvement in patients with oral lichen planus. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999;88:431-6.
 6. Rogers RS 3rd, Eisen D. Erosive oral lichen planus with genital lesions: The vulvovaginal-gingival syndrome and the peno-gingival syndrome. *Dermatol Clin* 2003;21:91-8, vi-vii.
 7. Eisen D, Carrozzo M, Bagan Sebastian JV, Thongprasom K. Number V Oral lichen planus: Clinical features and management. *Oral Dis* 2005;11:338-49.
 8. Souto P, Sofia C, Cabral JP, Castanheira A, Saraiva S, Tellechea O, *et al.* Oesophageal lichen planus. *Eur J Gastroenterol Hepatol* 1997;9:725-7.
 9. DeRossi SS, Ciarrocca KN. Lichen planus, lichenoid drug reactions, and lichenoid mucositis. *Dent Clin North Am* 2005;49:77-89, viii.
 10. van der Meij EH, Reibel J, Slootweg PJ, van der Wal JE, de Jong WF, van der Waal I. Interobserver and intraobserver variability in the histologic assessment of oral lichen planus. *J Oral Pathol Med* 1999;28:274-7.
 11. McCreary CE, McCartan BE. Clinical management of oral lichen planus. *Br J Oral Maxillofac Surg* 1999;37:338-43.
 12. Voûte AB, Schulten EA, Langendijk PN, Nieboer C, van der Waal I. Cyclosporin A in an adhesive base for treatment of recalcitrant oral lichen planus. An open trial. *Oral Surg Oral Med Oral Pathol* 1994;78:437-41.
 13. McGrath C, Hegarty AM, Hodgson TA, Porter SR. Patient-centred outcome measures for oral mucosal disease are sensitive to treatment. *Int J Oral Maxillofac Surg* 2003;32:334-6.
 14. Holbrook WP, Kristmundsdóttir T, Loftsson T. Aqueous hydrocortisone mouthwash solution: Clinical evaluation. *Acta Odontol Scand* 1998;56:157-60.
 15. Hegarty AM, Hodgson TA, Lewsey JD, Porter SR. Fluticasone propionate spray and betamethasone sodium phosphate mouthrinse: A randomized crossover study for the treatment of symptomatic oral lichen planus. *J Am Acad Dermatol* 2002;47:271-9.
 16. Aguirre JM, Bagán JV, Rodríguez C, Jimenez Y, Martínez-Conde R, Díaz de Rojas F, *et al.* Efficacy of mometasone furoate microemulsion in the treatment of erosive-ulcerative oral lichen planus: Pilot study. *J Oral Pathol Med* 2004;33:381-5.
 17. Sardella A, Demarosi F, Oltolina A, Rimondini L, Carrassi A. Efficacy of topical mesalazine compared with clobetasol propionate in treatment of symptomatic oral lichen planus. *Oral Dis* 1998;4:255-9.
 18. Carbone M, Conrotto D, Carrozzo M, Brocchetto R, Gandolfo S, Scully C. Topical corticosteroids in association with miconazole and chlorhexidine in the long-term management of atrophic-erosive oral lichen planus: A placebo-controlled and comparative study between clobetasol and fluocinonide. *Oral Dis* 1999;5:44-9.
 19. Gonzalez-Moles MA, Ruiz-Avila I, Rodriguez-Archilla A, Morales-Garcia P, Mesa-Aguado F, Bascones-Martinez A, *et al.* Treatment of severe erosive gingival lesions by topical application of clobetasol propionate in custom trays. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2003;95:688-92.
 20. Radfar L, Wild RC, Suresh L. A comparative treatment study of topical tacrolimus and clobetasol in oral lichen planus. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008;105:187-93.
 21. Yoke PC, Tin GB, Kim MJ, Rajaseharan A, Ahmed S, Thongprasom K, *et al.* A randomized controlled trial to compare steroid with cyclosporine for the topical treatment of oral lichen planus. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2006;102:47-55.
 22. Gorouhi F, Solhpour A, Beitollahi JM, Afshar S, Davari P, Hashemi P, *et al.* Randomized trial of pimecrolimus cream versus triamcinolone acetonide paste in the treatment of oral lichen planus. *J Am Acad Dermatol* 2007;57:806-13.
 23. Buajeeb W, Poburksa C, Kraivaphan P. Efficacy of fluocinonide acetonide gel in the treatment of oral lichen planus. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000;89:42-5.
 24. Thongprasom K, Dhanuthai K. Steroids in the treatment of lichen planus: A review. *J Oral Sci* 2008;50:377-85.
 25. McCreary CE, McCartan BE. Clinical management of oral lichen planus. *Br J Oral Maxillofac Surg* 1999;37:338-43.

How to cite this article: Patil A, Prasad S, Ashok L, Sujatha GP. Oral bullous lichen planus: Case report and review of management. *Contemp Clin Dent* 2012;3:344-8.

Source of Support: Nil. **Conflict of Interest:** None declared.