

# Mucocutaneous Enigma – A Case Report (Multidisciplinary Management for Successful Treatment Outcomes)

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## ABSTRACT

**Introduction:** Lichen planus is a complex, mucocutaneous, chronic inflammatory disease of unknown etiology. The association of lichen planus with autoimmune disease has been found with equatorial racial and cosmopolitan distribution with increased frequency of HLA-A3, A28, B5, B7 and DRW9.

**Case report:** The paper highlights a case report of a female patient of 19 years reporting to department of periodontics with chief complaint of burning sensation in mouth and oral lesions. A detailed case history and clinical features were suggestive of mucocutaneous lesions. The paper stresses on the importance of case history recording with special emphasis on medical history. An integrated approach involving the dermatologist in the diagnosis of the lesions was done. The paper also outlines on various diagnostic tests undertaken to arrive at diagnosis and further multidisciplinary management of oral and systemic lesions associated with lichen planus.

**Conclusion:** Lichen planus have been reported to be a multifactorial disease. Co-existence of multiple symptoms of an oro-systemic disease is of particular interest of dentist. Interdisciplinary management is advocated in these cases.

**Keywords:** Autoimmune, Oral Lichen Planus, Hydroquinone, Local Drug Delivery, Hepatitis.

## INTRODUCTION

Lichen planus is a chronic, immunological, mucocutaneous disease with a wide range of clinical manifestations is affecting 2 percent of population.<sup>1</sup> Although the disorder may occur in all age groups, women over the age 50 years are most commonly affected. The disease can affect the skin and any lining mucosa (stratified squamous epithelia). It can involve the oral, esophageal, vaginal mucosa as well as the skin. Often, it is found only in the oral cavity.<sup>2</sup> The article elaborates a case of oral lichen planus with systemic involvement.

## CASE REPORT

A 19 years old female patient reported to the department of periodontics and oral implantology, Jaipur Dental College and Hospital with the chief complaint of burning sensation in mouth with itching and oral lesions since last 4 months with no relevant medical, family history and no deleterious habits and good oral hygiene.

Upon extraoral examination flat violaceous papules with a fine scaling on the surface were seen on the skin. Similar kind of lesions has been noted on chest, axillary and external genital areas by physician.

Upon intraoral examination pigmentation on the dorsum of tongue and desquamation of marginal gingival and isolated pockets in first molars were observed.

On clinical examination white reticular striations were present bilaterally on buccal mucosa and retromolar region with pigmentation in between.

Based on a detailed history and clinical examination our differential diagnosis were of oral erosive lichen planus, mucous membrane pemphigoid, pemphigus vulgaris, reticular lichen planus.

A hemogram, biochemical investigations, biopsy and swab for fungal growth was advised.

Haematological and biochemical investigations (Table 1):-

## Histopathology

Punch biopsy from perilesional tissue from right buccal mucosa was stained and showed irregularly hyperplastic epithelium and is infiltrated in its lower 1/3<sup>rd</sup> by lymphocytes without much spongiosis.<sup>3</sup> Occasional colloid bodies can be seen within the lower epithelium. The granular layer is irregularly thickened and the stratum corneum shows laminated orthokeratosis with scanty parakeratosis.

Oral swab upon culturing showed presence of fungal hyphae suggestive of Candidial infection.

## Management

Observing this we decided to opt for interdisciplinary approach i.e. Medical and dental. The medical team further consisted of dermatologist, gastroenterologist and ophthalmologist.

## Treatment

For lesions dermatologist prescribed hydroxychloroquine (hcq) along with multivitamins tablets, topical steroids.<sup>4</sup> For raised SGOT, SGPT gastroenterologist was consulted, since HbsAg, HCV were negative, no related gastrointestinal disorder was found.

For side effects following use of HCQ an ophthalmological opinion was taken and needful treatment was taken care.

Since no syndromic association was ascertained the dermatologist confirmed it as a case of erosive lichen planus. Patient was advised to take medications regularly and come for check up at one month interval. The dental treatment part included scaling and root planning and topical antifungals was advised for 15 days twice daily.

At 1 month interval patient was recalled. Fungal swab was taken again and periodontal examination was done. The persistent pockets were subjected to local drug delivery following

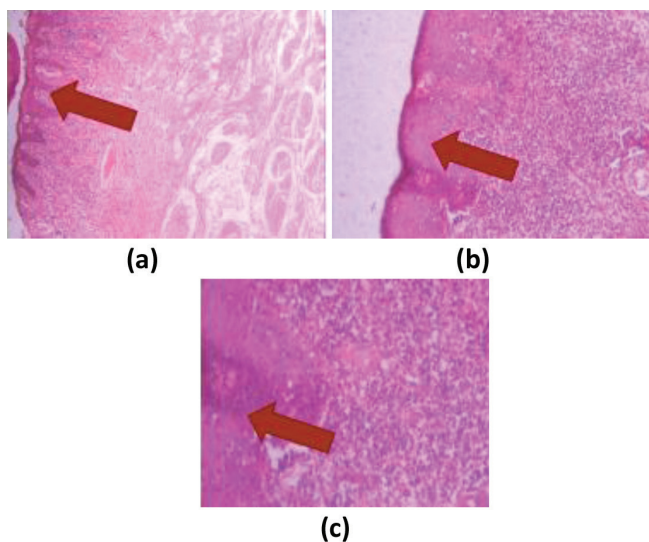
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**Figure-1:** Shows skin lesions; **Figure-2:** Represents intra-oral lesion.



**Figure-3:** Histological picture from punch biopsy of perilesional tissue from right buccal mucosa. (a) Saw tooth rete pegs (b) Basal cell degeneration (c) Chronic inflammatory cells

TLC	6200/mm
DLC- Neutrophils	64%
Lymphocytes	33%
Eosinophils	02%
Monocytes	01%
Basophils	00%
Haematocrit	43.9%
Haemoglobin	13.4%
Total RBC	4.98ml/mm <sup>3</sup>
MCHC	32.8%
MCV	88.3ft
MCH	28.9pg
Platelets	2,93000/mm <sup>3</sup>
Bleeding time	2 min 40 sec
Clotting time	4 min 45 sec
Anti-HCV serum	Not detected
Raised A/G ratio	1.2:1
G6PD quantitative	11.8u/ghb
Raised SGOT and SGPT	45.6 and 47.8 respectively
GGTP	93
ALP	166

**Table-1:** Haematological and biochemical investigations

a thorough curettage. Application of topical antioxidants, antifungals (clotrimox), topical steroids (fluocinonide) for one month period was advised.

## DISCUSSION

Erasmus Wilson in 1869 described lichen planus for the first

time. Aetiology of it is considered as idiopathic, autoimmune.<sup>5</sup> It is stated that T-cell mediates autoimmune course of disease where the cytotoxic CD8+ T cells trigger apoptosis of the basal cells of the oral epithelium.<sup>6</sup> Increased expression of the vascular adhesion molecules (VAM), CD62E, CD54, and CD106, by the endothelial cells of the sub-epithelial vascular plexus have been found in OLP.<sup>7</sup> The history, typical oral lesions, and skin involvement are sufficient to diagnose oral lichen planus (OLP), in the subject. Furthermore laboratory test and biopsy in consultation with the medical specialist confirmed the diagnosis.

Upto 70% of patients with cutaneous lichen planus have demonstrable oral lesions but only about 10% of patients presenting with oral lichen planus have skin involvement. Erosive lichen planus has been found to be associated with chronic liver disease.

In the present case fungal growth was recorded in the oral swab obtained as also noted in study by Masoumeh Mehdipour et al<sup>8</sup> who found an increased incidence of *candida albicans* infection in patients with oral lichen planus. Periodic acid-schiff (pas) staining of biopsy specimens and candidal cultures or smears may be performed. The fungal infection was treated by antifungals.

In the biochemical investigation SGOT and SGPT found to be elevated. Following this HbsAg and HCV were also investigated but found to be negative revealing no association with chronic liver disease.

As in the literature the association between chronic liver disease was first proposed by Mokni et al. in 1991.<sup>9</sup> This association of Hepatitis C virus and oral lichen planus is most prevalent in the mediterranean regions and japan.

Hematological investigations were found to be normal otherwise. Biopsy was suggestive of erosive type of lichen planus.

Syndromic association of lichen planus has also been found as in Greenspan syndrome which involves lichen planus, hypertension, diabetes mellitus; Good syndrome it involves vulvo-vaginal-gingival lichen planus with thymoma and immunodeficiency (The triad of lichen planus, thymoma and liver cirrhosis/hepatoma).<sup>10</sup> In the present case assessed there was no syndromic findings as confirmed by the medical specialists.

For long term use and to prevent remission of disease immunomodulatory and anti-inflammatory drugs like calcineurin inhibitors, retinoids, dapsone, mycophenolate mofetil and enoxaparin in topical preparation can be used. For cosmetic corrections of lesions photodynamic therapy and laser can be considered.

## CONCLUSION

Exact etiology of lichen planus is not known but prime role of genetics and immunity is considered. Research explains it as body's reaction towards an antigen within the skin surface or mucosa. Others quote it as an autoimmune disorder where skin cells lining the mouth are attacked by the white blood cells, still further research needs to be carry out. Some categorises lichen planus as a cell-mediated immune response and think that it is premature to classify the disorder as autoimmune because a particular antigen has not been recognized.

Co-existence of multiple symptoms of an oro-systemic disease

along with periods of remission and exacerbation present the challenge for the dental profession raising the possibility whether the lesion is benign or malignant. Further researches on immunological aspects and histopathological investigations are required for appropriate treatment.

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