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Erosive Lichen Planus an Epigone of Erythema Multiformae – A Case Report

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ABSTRACT: Oral lichen planus (OLP) is a chronic inflammatory disease with an unclear aetiology that affects the stratified squamous epithelium in the oral cavity. OLP affects 0.1% to 4% of the general population, with perimenopausal women being more likely to develop it. The oral cavity is impacted by several immune-mediated inflammatory mucocutaneous disorders; the majority of these diseases manifest with varied degrees of erosions, ulcers, and pain. Early and accurate diagnosis of such diseases is essential for patient treatment because they may have severe implications if not treated. This case report attempts to describe an ELP that mirrored erythema multiforme on a 54-year-old woman. On examination, features include flexor scarring, crustations over the lips, palmar crease erosion and plantar keratosis, dry flaky white lesions of the eyelids, and an annular pattern of papules in the groin and axillary region. On the buccal mucosae and dorsoventral surface of the tongue, there were many erosions with surface sloughing. Erosive lesions with bloody remnants were visible on the labial mucosae. Systemic steroids were administered to the patient, since there was no progression, systemic and oral topical immunosuppressants was given. The patient was under regular follow up for 3 months with no new complaints. The rate of healing and ability to ward off malignancy will both depend on an accurate diagnosis of the lesion.

Keywords: Erosive lichen planus, Oral lichen planus, erythema multiforme, Mimicking, mucocutaneous diseases.

INTRODUCTION

Lichen planus (LP) is a chronic mucocutaneous disorder of the stratified squamous epithelium that affects oral and genital mucous membranes, skin, nails, and scalp. The mucosal equivalent of cutaneous LP is called oral lichen planus (OLP). The Indian subcontinent has a particularly high incidence of disease. LP is estimated to affect 0.5% to 2.0% of the general population (Gupta & Jawanda 2015). It presents frequently in the fourth decade of life and affects women more than men in a ratio of 1.4:1 (Lavanya *et al*., 2011). The distribution of oral lesions - buccal mucosa 80%, tongue 65%, lips 20%, labial mucosa, floor of the mouth, gingiva and palatal occurrence are 10% (Omal *et al*., 2012). The six clinical patterns of OLP are reticular, plaque-like, erythematous, erosive/ulcerative, papular, and bullous. Erosive lichen planus is the second commonest type of LP.ELP is a variant of lichen planus which involves chronic and painful ulceration of the skin and mucosal surfaces (Krupaa *et al*., 2015). It might be present with other forms of lichen planus or by drug exposure. The oral lesions will have an irregular shaped central erosive lesion covered with pseudomembranous plaque and peripheral white radiating reticular striae. ELP has greater potential to undergo malignant changes. Erosive lichen planus is often confused with autoimmune diseases like pemphigus vulgaris and Mucous membrane pemphigoid which will have similar erythema and ulcerations. It can be clinically differentiated by the presence of concomitant reticular areas. This case report describes the occurrence of erosive lichen planus in a 54-year-old female, mimicking clinical features of Erythema multiforme.

CASE REPORT

A 54-year-old female reported to the Department of Oral Medicine and Radiology with the complaint of soreness with difficulty in chewing and swallowing for the past 6 months. Patient revealed a history of abdominal pain before 7 months with sudden onset of oral ulcers following medical management of the same. Patient also had severe burning sensation in her oral cavity while chewing and swallowing food. Patient visited nearby hospital for the oral ulcers and was treated with steroids for 3 months, but showed no improvement. Patient also developed flaking of skin, eyelids, neck, and lips on continuing the medication.

Then she developed cracks and fissures over her palms and soles. The lesions continued to progress with no episodes of regression with steroids, meanwhile she got admitted in the hospital for the complaint of back pain which was prevailing for past 1 month, where she was diagnosed with pyelonephritis and was treated for the same. Past medical history revealed history of hypertension for 1 year and was under regular medication. Extra oral features revealed scarring with dried flaky white lesions of eyelids, crustations over the lips, palmar crease erosions and plantar keratosis, flexor scarring, annular pattern of papules was noted in groin and axillary region (Fig. 1).

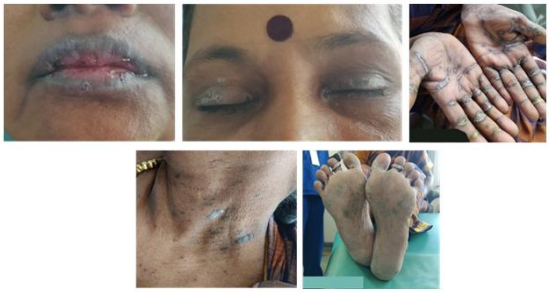

Fig. 1. Extraoral crustations of the lips, cicatrix of the eyelids, palmar crease erosion with plantar keratosis and post inflammatory hyperpigmentation.

Right and Left Submandibular lymph nodes were palpable and was roughly oval, measuring 0.3×0.5cm approximately, tender, mobile, with soft consistency. On intraoral examination, multiple erosions with surface sloughing were noted on both the buccal mucosae, dorsoventral surface of the tongue. Labial mucosae exhibited erosive lesion with bloody residues (Fig. 2 and 3).

Based on the patient's chief complaint, history of presenting illness, past medical history, and clinical examination a provisional diagnosis of Persistent erythema multiforme was given. Oral erosive lichen planus, paraneoplastic pemphigus, lichen planus pemphigoides were included in differential diagnosis. Complete blood profile revealed no significant changes. Cutaneous punch biopsy was taken from the annular papules of Vastus intermedius, oral perilesional and lesion biopsies were taken from the left buccal mucosa, to proceed with histopathological and direct immunofluorescent study (Fig. 4).

The histopathological studies showed features of lichen planus (hyper parakeratosis, acanthosis, saw tooth rete pegs) and immunofluorescence results were negative of IgG, IgM, C3 (Fig. 5).

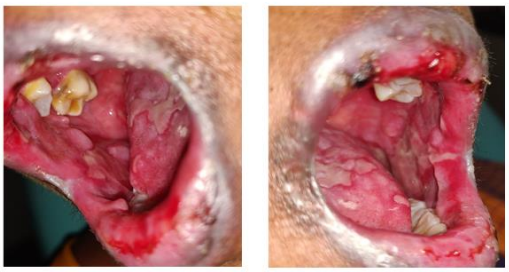

Fig. 2. Multiple erosions and sloughing of the buccal mucosae.

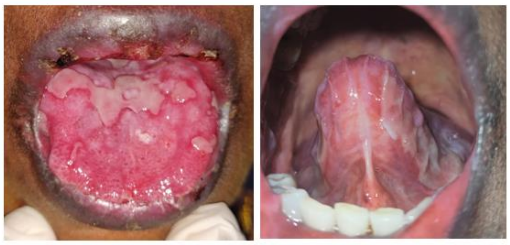

Fig. 3. Diffuse erosions and sloughing of the dorsoventral surface of the tongue.

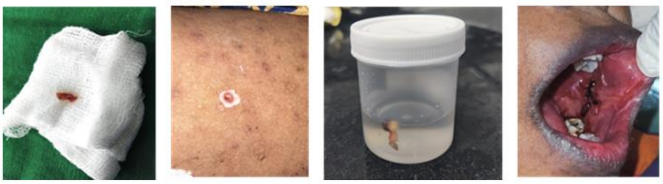

Fig. 4. Oral and cutaneous biopsy specimen.

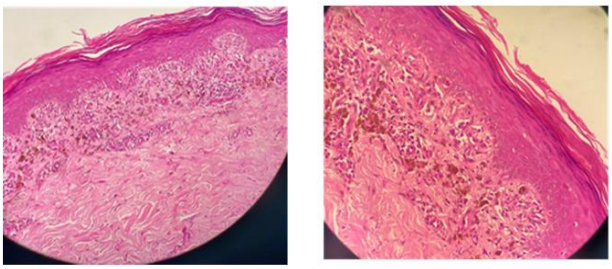

Fig. 5. Photomicrograph of H&E-stained histopathological image of 20X and 40X respectively.

Oral biopsy was performed at the left buccal mucosa, but the results were inconclusive. Correlating with histopathological and immunofluorescent studies, a final diagnosis of Erosive lichen planus was given.

The initiation of treatment was with systemic corticosteroids Tab. Methylprednisolone 16mg/day for 2 weeks and was tapered to 12mg/day for 2 weeks and 8mg/day for 2weeks subsequently since patient showed no progression, steroids were replaced with

immunosuppressants, Tab. Cyclosporine 250 mg/day for 4 weeks and Tab. Dapsone 100mg for 4 weeks along with topical liquid paraffin for the skin lesions. For oral lesions topical Triamcinolone acetonide I.P 0.01% was given to apply over the oral lesions 4 times/day and Cap. Doxycycline 100mg was diluted in 10 ml of water and insisted to be used as a mouth rinse for 4 times/day for 4 weeks. Following less response with topical steroids for oral lesions, Topical tacrolimus 0.03% thrice daily for 4 weeks and Cap. Lycopene, Zinc Sulphate Monohydrate, Selenium Dioxide Monohydrate, Mixed Carotene, and Refined Wheat Germ Oil once daily for 1 month were prescribed. The patient was under regular followed up for 3 months with no new complaints(Fig. 6) and (Fig. 7).

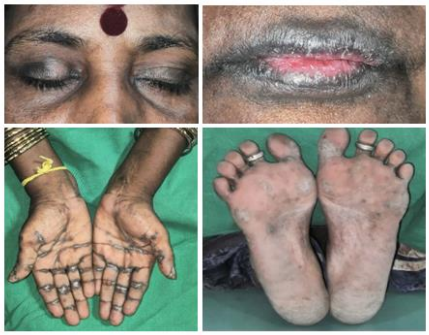

Fig. 6. Healing of extraoral lesions.

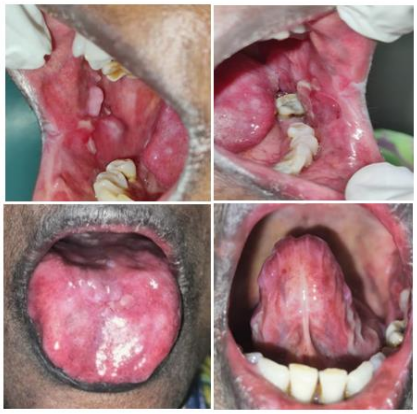

Fig. 7. Healing of intraoral lesions with systemic and oral corticosteroids.

DISCUSSION

Lichen planus is a chronic inflammatory, mucocutaneous disease involving skin, oral and genital mucosa. It is chronic in nature, with periods of exacerbations and remission. The Latin term "planus" indicates flat, while the Greek word "leichen" signifies tree moss. Lichen planus was first described by British Physician Wilson Erasmus in 1869. In 1895, Wickham noted the characteristic reticulate white lines on the surface of LP papules, today recognized as Wickham striae. The first formal description of the LP-related histopathological alterations is attributed to Darier (Gupta & Jawanda 2015). Lichen planus' aetiology is yet unknown. There may be a hereditary tendency for LP if there are familial examples of the disease (Gupta & Jawanda 2015). LP has been linked to gene polymorphisms for various HLA indicators as well as inflammatory cytokines and chemokines. Stress and anxiety, the hepatitis C virus (HCV), autoimmune illnesses, internal cancers, dyslipidaemia, and viral infections are a few of the associated causes and disease states identified in LP. In LP patients, anxiety is a recognised risk factor or coexisting condition (Anjum *et al.,* 2012). Even though the varicella zoster virus is uncommon in OLP patients, a recent study found that the virus plays a substantial role in zosteriform LP but not in linear LP. OLP appears to have a smaller impact on patients' quality of life compared to oral bullous illnesses, but a larger impact compared to recurrent aphthous stomatitis. There are six clinical patterns of OLP - reticular, plaque-like, erythematous, erosive/ulcerative, papular, and bullous. Erosive lichen planus (ELP) is a type of lichen planus that causes painful, chronic skin and mucosal ulcers. ELP is believed to be caused by basal cell layer destruction from an autoimmune process that is mediated by activated CD8 T cells. The oral mucosa or genitalia are most frequently affected by erosive lichen planus (ELP). However, it is extremely rare for it to affect the external auditory canal, bladder, anus, oesophagus, larynx, or eyelids. Patients experience erythematous erosions and ulcerations that can last for several months or longer and are painful. Lesions on the tongue, buccal mucosa, labial mucosa, or vermillion border might develop as a result of oral erosive LP. The lesions are frequently symmetrical and bilateral. Another typical occurrence is the Koebner phenomenon, which is the emergence of new erosions in areas of minimal damage. Desquamative gingivitis and gingival ulceration are most frequently caused by LP, while blisters are seldom produced (Scully *et al*., 1997). The vulvovaginal-gingival (VVG) syndrome, a severe variation of ELP in females affecting both the genital and oral mucosa, was first identified by Pelisse *et al*., in 1983. This condition is a triad of gingivitis, vaginitis, and (erosive or desquamative) vulvitis. The Peno-Gingival (PG) Syndrome, which was first identified by Cribier *et al.,* in 1993, is the name of the similar disorder in men. Given that ELP can affect several bodily sites, handling affected people involves several healthcare professionals, including doctors of oral medicine, dermatology, gynaecologists, and, if the oesophagus is involved, gastroenterologists (Cheng *et al*., 2012). Nutritional deficits, such as an iron deficiency, and weight loss are consequences of eating difficulties (Eisen, 1999). Painful erosions cause poor oral hygiene and more tooth decay. The presence of erythroplakic lesions (reddened patches with a velvety surface found in the mouth) and sites involving the tongue, gingival, or buccal mucosa are high-risk factors for malignant transformation in oral LP (Scully & Carrozzo 2008). Other risk factors include smoking and excessive alcohol consumption. Whether early ELP treatment lowers the risk of cancer is unknown. Depending on the affected area, the differential diagnosis for erosive lichen planus might vary. Clinical symptoms of erythema multiforme, Behcet's syndrome, Stevens-Johnson syndrome, paraneoplastic pemphigus, lichen planus pemphigoides can all resemble those of

erosive LP. Erythema multiforme (EM) is a mucocutaneous inflammatory disease that manifests on the skin, most frequently on the oral mucosa. It is acute, recurring, and self-limited. It typically happens after the administration of medication or an illness. The most frequent risk factor for the development of EM mild is HSV infection. It has been demonstrated that EM may precipitate from HSV types 1 or 2. An EM (small or large) lesion often starts 10–14 days after the clinical signs of an HSV infection. In instances of HAEM, the lip is the location of prior HSV infection most frequently (Kamala *et al*., 2011). In this case report initially the clinical features were suggestive of erythema multiforme since patient had diffuse ulcerations, bloody crustations of the lips and palmar crease fissures, erosions with plantar keratosis but persistence of EM for 5 months even after the withdrawal of the drug excluded drug induced erythema multiformae and negative HSV titre excluded HIEM. Lichen sclerosus and erosive LP might be mistaken when they affect the female genitalia. Oropharyngeal candidiasis, leukoplakia, squamous cell carcinoma, leukoedema, and allergic contact mucositis should be included in the differential diagnosis for erosive LP. Erosive lichen planus (ELP) is normally diagnosed through a clinical examination and review of medical history. However, a biopsy aids in validating the diagnosis and excluding the possibility of cancer. The basal layer liquefaction, Civatte bodies (eosinophilic masses signifying apoptotic keratinocytes), saw-toothed rete ridges, hyperkeratosis without parakeratosis, hyperkeratosis without parakeratosis, and a band-like lymphohistiocytic infiltrate close to the dermal-epidermal border are characteristic histopathological findings (Gall & Navarro-Fernandez 2023). Use of topical corticosteroids for symptomatic relief is the primary line of treatment for oral lichen planus. In cases of erosive or erythematous lichen planus, systemic corticosteroids might be used if topical corticosteroids are ineffective (Piñas *et al*., 2017).

Immunosuppressives, retinoids, and immunomodulators are employed as additional topical forms. As a preventive approach, maintaining good oral hygiene and removing local triggers are options. Tetracycline mouthwashes are beneficial against erosive lichen planus and desquamative gingivitis because plaque components or bacteria may be implicated in the development of gingival desquamation. Immunosuppressants that are effective against erosive lichen planus, such as Topical Tacrolimus 0.1%, may be administered in some circumstances. Micronutrients like antioxidants have been shown in several studies to increase immunological function. Patients with erosive or ulcerative lichen planus should receive regular follow-up due to the condition's potential for malignant change.

CONCLUSIONS

Oral lichen planus is a multifactorial disease and one of the most prevalent oral dermatological conditions that dentists commonly encounter in their practice. In this case report, it is noticed that erosive lichen planus occasionally mimics the characteristics of erythema multiforme. So, it is important to diagnose the lesion and administer proper medications at the earliest along with educating the patient and lifestyle modification to ensure a positive outcome.

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Conflict of Interest. None.

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