CASE REPORT

Oral lichen planus bullous variety: A case report

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Abstract
Lichen planus (LP) is a common immune-mediated chronic mucocutaneous disorder with a worldwide distribution. It mainly affects the skin, nails, hair, and the genital area. Initial presentation of these lesions may occur in the oral mucosa without any other cutaneous manifestations. Hence, it is necessary for the dental professionals to be both familiar with its varied clinical presentations, course of this condition, and their management. Here, we report a case of bullous LP in a 23-year-old male patient, discussing clinical features, etiopathogenesis, and management.

Introduction
Oral lichen planus (OLP) is a well-recognized potentially malignant disorder which affects epithelium of skin, oral mucosa, and genitalia. Clinically, the lesion looks like lace-like pattern produced by symbolic algae and fungal colonies on the surface of rocks (lichens) and “planus” stands for flat. It is most commonly seen in females in the age group of the third to sixth decades of life. Malignant transformation is reported to be 0.4-5% in a duration of 0.5-20 years.

Although LP is considered as an autoimmune disease, its exact cause is not clear. Few researchers have suggested that LP is a chronic, cell-mediated immune disorder involving activated lymphocytes and upregulated cytokine production. OLP has varied clinical presentations such as reticular, erosive, atrophic, papular, plaque-like, and bullous subtypes of which bullous is considered to be the rarest. The signs and symptoms can vary according to the clinical subtypes. Reticular OLP usually presents as asymptomatic while the erosive and atrophic forms will be associated with pain or burning sensation.

Bullous lichen planus (BLP) is rare and may be confused with other sub-epidermal bullous dermatoses. Here, we present a case report of bullous OLP in a young male patient with initial involvement of oral mucosa alone.

Case Report
A 23-year-old male patient (Figure 1) presented with a history of burning sensation and recurrent fluid-filled blisters in the buccal mucosa since 2 weeks. The boils would subsequently burst. The patient also noticed white colored patches on the buccal mucosa bilaterally. Medical history was negative and he was not under any medications.

Cutaneous examination did not show any abnormality. On intraoral examination, diffuse white interlacing lines with reticular pattern were noticed on the buccal mucosa bilaterally. Linear white colored striae were noticed in the mandibular buccal vestibule bilaterally, extending anteriorly from mesial of 45 posteriorly distal to 48 superiorly from the occlusal plane of 14 inferiorly up to buccal vestibule i.r.t 45, 46, 47, and 48 regions. A solitary vesicle of was evident on the right buccal mucosa adjacent to the white colored striae at the level of the first molar. Surface presented with bullae filled with clear fluid measuring about 1 cm × 0.8 cm, margins are irregular with pseudomembrane slough in the posterior buccal mucosal region, surrounding mucosa was normal. There were no adjacent dental restorations. Soft and hard palate, tongue, and floor of the mouth did not show any abnormalities. On palpation the lesion was tender, smooth, and nonscrappable.

Clinical features were consistent with BLP. For further confirmation of the lesion, an incisional biopsy of the...
A vesicular lesion was performed and subjected to direct immunofluorescence. Anti-hepatitis C virus (HCV) test was performed and the result was negative.

Histopathological examination revealed hyperkeratotic cornified epithelial layer, focal hypergranulosis, and irregular epithelial hyperplasia. Papillary dermis shows civatte bodies with dense lichenoid, lymphohistiocytic infiltrate.

Direct immunofluorescence revealed linear deposition of IgA and C3 in the epidermal layer (Figure 4).

Based on the clinical correlation to the presence of recurring oral lesions a final diagnosis of BLP was given. The patient was initially treated with clobetasol mouth rinse for 1 week. The patient reported back with bullae. Considering it, the patient was treated topically with 0.1% tacrolimus for 15 days. After 15 days, on review, there was complete remission of vesicle and reduction in burning sensation. The patient is under follow-up since a year and is free of lesions to date (Figure 5).

Discussion

The term LP was identified earliest in the year 1866, by Erasmus Wilson.

The prevalence of OLP in the general population varies between 0.5% and 2.6%. The disease is more common in females of middle age group. The patient, in this case, was a male of 23 years of age, which does not fall into the age range in which LP is most commonly reported.

Autoimmune diseases are known to have multifactorial etiology. LP can be triggered by various potential etiological agents such as, for example, viral or bacterial antigens, metal ions, drugs or physical factors, stress, genetics, and systemic diseases such as diabetes and hypertension, dental restorative materials, drugs, infectious agent, immunodeficiency, food allergy, trauma, malignant neoplasm, chronic liver disease and HCV, tobacco chewing, graft versus host disease, and bowel diseases.

LP usually manifests as multiple lesions in a bilateral, symmetrical pattern with episodes of quiescence and exacerbation. Intraorally, LP is commonly seen on the buccal mucosa, tongue, gingiva, and lips. Oral lesions are characterized by minute white or gray velvety thread-like papules in a linear, annular, or reticular arrangement. A characteristic feature is the presence of fine lacy white lines covering the lesions called as Wickham’s striae. Of the various clinical subtypes, the reticular form is the most common while bullous is the rarest. Bullous OLP is seen as small vesicles or bullae that range in size from few millimeters to several centimeters in diameter. It is commonly seen on the buccal mucosa followed by lateral margins of the tongue.

The presence of cutaneous lesions may be serve as a diagnostic clue LP. About 30-70% of patients with skin lesions have oral involvement, while 15% present with only oral involvement. In the present case, only oral involvement could be appreciated. Thus, histopathological examination was undertaken to confirm the diagnosis of BLP, to differentiate it from other oral vesiculobullous diseases and also to exclude dysplasia and malignancy.

Keratinocyte apoptosis as a result of activation of cell-mediated immune response is the primary step in the pathogenesis of LP. Laboratory diagnosis is based on the evidence of focal ortho or
parakeratosis and irregular acanthosis, a saw-toothed appearance to the rete pegs, lymphocytic infiltrate in the subepithelial region in band-like patterns, liquefactive degeneration of the basal cell layer without epithelial dysplasia, the presence of civatte bodies, which are eosinophilic colloid bodies in the spinous and basal cell layers and lamina propria.\(^7\)

Immunofluorescent studies of biopsy specimens from lesions of LP aid in distinguishing them from other vesiculobullous disorders such as pemphigus, oral mucus membrane pemphigoid and lupus erythematosus. Direct immunofluorescence patterns of OLP include a shaggy band of fibrinogen in the basement membrane zone alone or with positive IgM, IgA, or C3 deposition at colloid bodies.

Topical and oral corticosteroids form the mainstay of therapy for controlling signs and symptoms in OLP. The time and dosage regimens are determined by patient’s medical status, severity of disease, and previous treatment responses. Topical application of retinoids, calcineurin inhibitors (cyclosporine and tacrolimus), have been suggested as a second-line therapy for OLP.\(^9\) In the present case, we initiated the management with topical steroid, no improvement was seen. Following it, the patient was treated with tacrolimus 0.1% topically. Lesion resolved after which no remission occurred till date.

References
