Case Report

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Oral Lichen Planus, 2 Varied Clinical Presentations and Its Therapeutic Intervention

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Abstract

Lichen planus is a relatively common mucocutaneous disorder affecting the oral mucosa and causes discomfort thereby compromising the quality of life. Among various types of oral lichen planus, papular type is relatively rare. Here we are reporting two cases of papular lichen planus of the oral mucosa, one of which is associated with multiple areas of hyperpigmentation and other case of angina bullosa hemorrhagica coexisting with papular lichen planus.

Keywords: Angina Hemorrhagica Bullosa; Hyperpigmentation; Oral Lichen Planus.

Introduction

Lichen planus is an autoimmune mucocutaneous disorder which involves the oral mucosa, skin, scalp, genital mucosa and nails [7]. It was first described by Wilson in 1869 [3]. Oral Lichen planus(OLP) is prevalent in 1-2% of the general population and skin lesions are usually present in almost 15% of the patients with OLP [7,11]. OLP is mainly affecting adults more than 40 years old, with female to male ratio of 1.4:1 [3]. Andreasen classified OLP in six clinical presentations as reticular, plaque-like, papular, erosive, bullous and atrophic [11]. The lesion mainly involves the buccal mucosa, gingiva and tongue while occurrence of the palatal region is rare [3]. The reticular type of OLP is the most common type, and is characterised by plaque and papules with interlacing white lines called Wickham's striae, named after Louis Frederic Wickham [3,11]. Occurrence of papular type is rare, and it clinically presents as small white papules and fine striae at its periphery. The oral lesions appear few weeks or months before the cutaneous lesions [5]. The diagnosis of OLP should be based on clinical and histopathological examinations. Though various treatment modalities are available, Corticosteroids are the drug of choice for management of the lesions [7].

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Case Report 1

A 32 year old male patient reported to our department with chief complaint of burning sensation and difficulty in eating since 1 month. Patient gave history of burning sensation which aggravated on eating spicy food. Patient gave no significant medical history. On examination there was no associated skin lesions. Intraoral examination revealed multiple diffuse white raised papules with multiple well demarcated dark brownish central pigmentation, bilaterally on buccal mucosa [Figure 1a, 1b]. Similar papules were also evident on the upper and lower labial mucosa, vestibule and soft palate. Lesion was clinically compatible with OLP associated with hyperpigmentation. An incisional biopsy on his left buccal mucosa was performed and the histopathological evaluation showed features suggestive of OLP [Figure 5a]. As the lesion was involving multiple oral sites, orally Tab Prednisolone 30 mg per day was started for 1 week followed by tapering of the dose for 2 weeks. Tab. Levamisole (immunomodulator) 150mg once daily for 3 days and systemic antioxidant were also prescribed. A complete

resolution of burning sensation, papular lesions and pigmentation was seen in 10 days [Figure 2a, 2b]. No recurrence of oral lesions was seen on follow up period of 1 month.

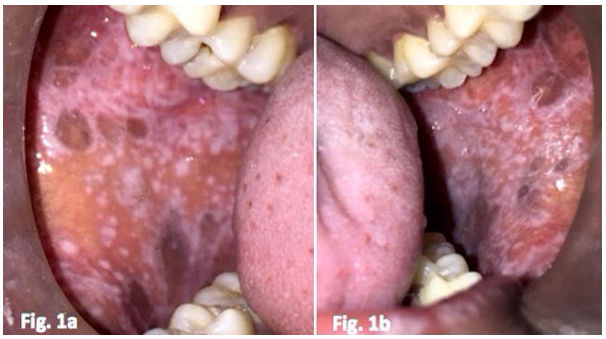

Fig. 1a, 1b: Showing OLP with well demarcated areas of hyperpigmentation on right and left buccal mucosa

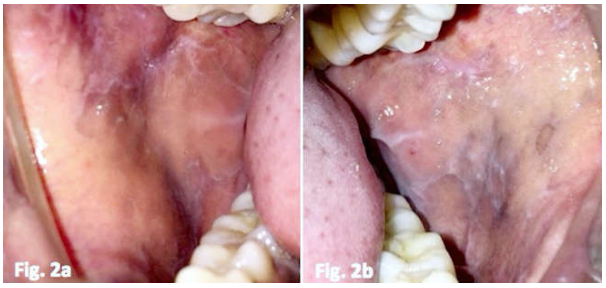

Fig. 2a, 2b: Showing complete resolution of papular lesions and pigmentation on right and left buccal mucosa after 10 days following treatment

Case Report 2

A 60 year old female patient reported with complaint of burning sensation in mouth since 3 months. Patient gave no contributory medical and dental history. General examination revealed no associated skin lesions. On intraoral examination, patient was completely edentulous with evidence of multiple diffuse white raised papules bilaterally on buccal mucosa and a solitary bluish purple blood blister of 1 cm diameter was evident on left buccal mucosa [Figure 3a, 3b]. The lesion was diagnosed as papular lichen planus with angina bullosa hemorrhagica (ABH). Intraoral finding on subsequent visit of next day revealed ruptured blood blister [Figure 3c]. A biopsy on left buccal mucosa was performed and the histopathological evaluation showed features suggestive of OLP [Figure 5b]. After counselling and reassurance, topical corticosteroids therapy with 0.1% triamcinolone acetonide was initiated along with systemic antioxidant. Complete resolution of oral lesions and symptoms were observed in 3 weeks with no recurrence for a period of 3 months [Figure 4a,4b].

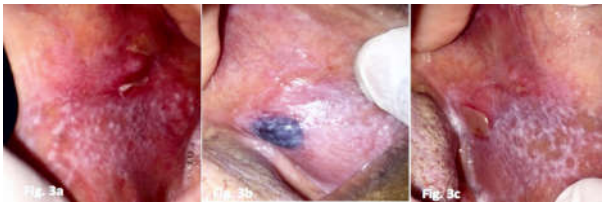

Fig. 3a: Showing papular lichen planus on right buccal mucosa.
Fig. 3b: Showing papular lichen planus with angina bullosa hemorrhagica on left buccal mucosa.
Fig. 3c: Showing ruptured blood blister on day 2

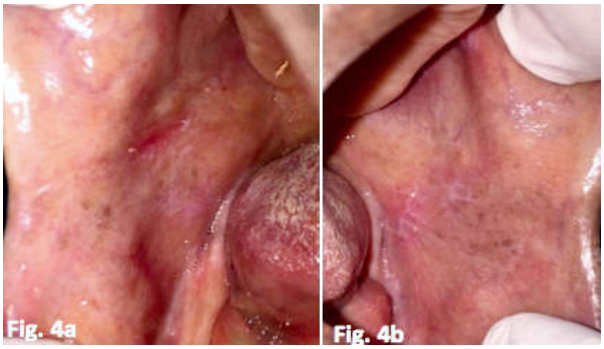

Fig. 4a, 4b: Showing post treatment images with complete resolution of oral lesions after 3 weeks

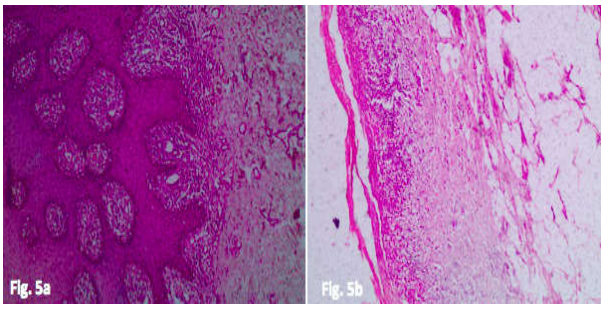

Fig. 5a, 5b: Showing histopathological pictures of first and second case respectively with dense band of chronic inflammatory infiltrate at sub epithelium along with basal cell degeneration

Discussion

OLP is insidious in onset and usually patient complaints of roughness, pain, red and white patches, ulceration of the oral mucosa, sensitivity to hot and spicy food while some patients present with lesions on the skin, nails, scalp and genital mucosa [7]. Although its etiology remains unclear, it has been found to be associated with autoimmunity, genetic predisposition, infectious agents such as viral and bacterial infections, dental restorative materials, medications, stress, diabetes and hypertension [2,5]. Cell mediated immunity plays an important role in the pathogenesis of OLP. CD8+ cytotoxic T-cells trigger the apoptosis of keratinocytes of oral epithelium, through the binding of antigen linked with major histocompatibility complex-1 (MHC) on

keratinocyte [7]. OLP is characterized by whitish grey, velvety appearance, thread like papules in retiform, linear and annular arrangement, forming lacy, reticular patches, streaks and rings. A tiny elevated white dot is seen at the white line intersection called as striae of Wickham. The lesions are bilateral and symmetrical in presentation [5].

The papular type is a rare presentation and characterized as raised papules of small size (0.5 to 1.0 mm) [7]. Hyperpigmentation is another clinical sign that is frequently seen in OLP [3]. The term "pigmented pattern" was used by Cooke in 1954 for classifying this subtype of OLP [10]. The reason for hyperpigmentation is not clearly known, but it could be due to inflammatory changes associated with OLP [3]. The presence of hyperpigmentation along with papular lichen planus in the first case report may be due to the long standing inflammation. Histopathological description of OLP was first given by Dubreuill in 1906, and Shklar later put forward three histologic features which are lymphocytic infiltrate within the connective tissue, liquefaction degeneration of basal layer and overlying keratinization [7]. Other histological features include variable degree of para or orthokeratosis, hypergranulosis, acanthosis and vacuolar degeneration of basal layer keratinocytes [9]. Direct immunofluorescence helps in differentiating OLP from other vesiculo-bullous conditions such as benign mucous membrane pemphigoid, pemphigus vulgaris and linear IgA bullous dermatosis. Various biomarkers have been proposed for predicting the onset and severity in individuals with OLP such as CD275, urinary prokallikrein, PLUNK, serum antibodies to desmogleins 1 and 3 [7].

Chronic inflammatory mucosal disorders such as lichen planus, pemphigoid or pemphigus can cause pigmentation of the mucosa because of melanin deposition within the connective tissue. Usually, resolution of the inflammation allows healing of oral pigmentation [4]. The first case showed response to the treatment causing subsidence of hyperpigmentation, along with the oral lesions. The second case is unique because of papular lichen planus coexisting with ABH. Badham coined the term angina bullosa hemorrhagica in 1967 for blood blister in oral mucosa that occurs without evidence of vesiculobullous disorder or blood dyscrasias. Diabetes and steroid inhalers are the possible aetiological factors. The pathogenesis of the ABH may be due to Minor mucosal insults. Clinically, the lesion appears as solitary, painless, dark red, tense and blood filled blister mainly involving the soft palate, although infrequently affecting lateral border of the tongue and buccal mucosa. This blood-filled blister rapidly expand and rupture followed by spontaneous healing within 7- 10 days, [1,6] as evident in our second case.

The patient should be counselled about course, nature of the disease and various treatment modalities. Treatment options of OLP include use of topical corticosteroids in mild to moderate lesions. Systemic corticosteroids should be used for erosive OLP or with mucocutaneous involvement. Immunosuppressant such as topical tacrolimus have shown better results for OLP as compared to topical corticosteroids [8].

Conclusion

OLP has varied clinical presentations. Here we highlighted two cases of papular lichen planus, which can coexist with hyperpigmentation or entirely distinct lesion of angina bullosa hemorrhagica. Proper diagnosis by clinical evaluation and histopathological examination is essential for the treatment of these lesions. Because of the high recurrence rate, long-term follow-up of the patients is required. Management of patients with oral and cutaneous lesions needs interdisciplinary approach of dermatologists and oral medicine specialists to improve the quality of life.

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