

Unusual Presentation Of Oral Lichen Planus Without Any Cutaneous Manifestation In A 5 Years Old Child –A Case Report

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Introduction

Lichen planus is Latin term (Lichen- Algae or fungi, planus- flat). The name has been suggested by British physician Erasmus Wilson, who first described the condition in year 1869 and reported a prevalence of 6% in general population. Wickham (1895) described the characteristic appearance of white striae that develops over flat surface or papules.

Oral lichen planus is a chronic mucocutaneous disease that is relatively common. It is predominantly a disease of the middle aged and elderly population with some female predilection having 2:1 female to male ratio.¹ The condition is rare in childhood and only few case reports have been reported.² The exact etiology remain obscure as condition is complex and multifactorial. In majority of cases the condition may be idiopathic while in others condition is exacerbated or even provoked by variety of drugs and dental materials. Viruses, genetic factor and lifestyle also plays a significant etiological role.³ The pathogenesis of lichen planus is not completely understood but T-lymphocyte infiltrate suggests cell-mediated immunological damage to epithelium. Both CD4 (Helper) and CD8 (cytotoxic) T-cell are involved in pathogenesis but increasing numbers and activation of CD8 cells is thought to contribute to the characteristic damage to basal epithelium.⁴

Currently employed treatment modalities for management of lichen planus include administration of corticosteroids topical, intralesional or systemic. Alternative therapies include topical and systemic retinoids, griseofulvin, Cyclosporine, and surgery. Other medical treatments and experimental modalities, including mouth PUVA have also been reported to be effective.

Case Report

A 5 year male child reported to the unit of pedodontics and preventive dentistry, department of oral health sciences center, PGIMER Chandigarh with chief complaint of pain and severe burning sensation in the mouth. Patient's parents also complain of black discoloration of left and right buccal mucosa and tongue and bleeding from the gums which they noticed since 10 months of age. Medical history was non-significant except the history of pneumonia at the age of 3 months. There was no history of any systemic illness or long term medication. History did not reveal any allergic or hypersensitivity reaction related to use of any oral chemical product neither there was any history of dental restorations in the past. There was a history of visit to a local dental practitioner for ulceration and black discoloration of oral mucosa who advised the biopsy of buccal mucosa and a diagnosis of oral lichen planus was confirmed after histopathological examination of biopsy. As there was no improvement in clinical signs and symptoms so the parent reported to the unit of pedodontics and preventive dentistry, department of oral health sciences center, PGIMER Chandigarh for further treatment. As there was no intraoral cause responsible for the condition, a complete systemic examination was carried out to rule out any systemic condition contributory to the oral involvement. The results of systemic investigation did not reveal any positive findings that could be suggestive of an association with oral lichen planus.

General physical and extraoral examination did not reveal involvement of skin or mucosa of any other part of the body.

Intraoral examination revealed black discoloration in right and left buccal mucosa and tongue. A reddish ulcer of approximately 1x1 cm dimension with rough surface and ill-defined margins was present in left

buccal mucosa at the level of occlusal plane opposite 74, 75. (Fig.1)

Intraoral examination also revealed bleeding and ulceration of gingiva in relation to 63, 64 region due to gingival erosion. (Fig.2)

The above clinical presentations along with histopathological findings (Fig.3) were confirmatory of childhood oral mucosal lichen planus.

Oral manifestations of lichen planus to such a severity is very less common in children. Considering the severity of oral involvement, consultation was sought from pediatric dermatology unit PGIMER Chandigarh for the management. Combination of systemic and topical corticosteroid was started at the onset of therapy which included Tab.Prednisolone 5mg twice daily orally in tapered dose along with topical Clobetasol ointment (0.05%) applied to painful areas 3-4 times daily for 15 days for remission of acute symptoms. 5% Amelazerox oral paste, 4 times a day was prescribed for relief of pain. Instructions were given not to eat spicy and acidic foods and to avoid alcohol containing mouthwashes. A significant relief in oral symptoms was noticed after 15 days of combination therapy. The patient had been under topical corticosteroid therapy (Clobetasol ointment 0.05% twice daily) for last 6 months and there has been a significant improvement in clinical signs and symptoms including the ulceration and burning sensation of oral mucosa. However, black discoloration of oral mucosa was still persisting although there was some improvement in its appearance.

Discussion :

The prevalence of lichen planus is less common in children though there are some case reports and case series reported in the literature.^{5,6,7} Samman et al.⁸ in a series of 200 patients with cutaneous lichen planus found childhood prevalence to be only 1% whereas studies from India show a wide range from 1.16% to 11.2%.^{5,6} Majority of these cases showed mucocutaneous involvement with few having oral manifestation. Kumar et al.⁵ (1993) in series of 25 cases in children with cutaneous involvement found oral mucosal lesion in only one patient. Similarly, Kanwar et al.⁶ (1991) in their case series found lip involvement in only one child out of 17 children with

mucosal lichen planus. On the contrary, in the present case severe involvement of oral mucosa was evident without any cutaneous manifestations which is an unusual presentation of lichen planus. The condition may involve any part of oral cavity with frequent involvement buccal mucosa, tongue and gingiva. Also, the clinical appearance of the condition is variable ranging from the classical pattern where bilaterally symmetrical reticular network found on the buccal mucosa to widespread debilitating ulcerative lesions found in the present case.³ The condition may be asymptomatic in majority of cases but in some may be associated with severe burning sensation. The clinical presentation and site predilection in the present case are consistent with the classical presentation of oral mucosal lichen planus.

This condition has very complex and multifactorial etiology. An association has been suggested between oral mucosal lichen planus and glucose 6-phosphate dehydrogenase deficiency and glucose intolerance.⁹ Genetic factor and life style also plays a significant etiological role. Familial nature of the condition has also been suggested by many. Some contact allergen and dental materials had also been found to be responsible for condition. However, in the present case there was no history of any dental restorations neither there is any associated systemic condition nor any familial history that can be contributory.

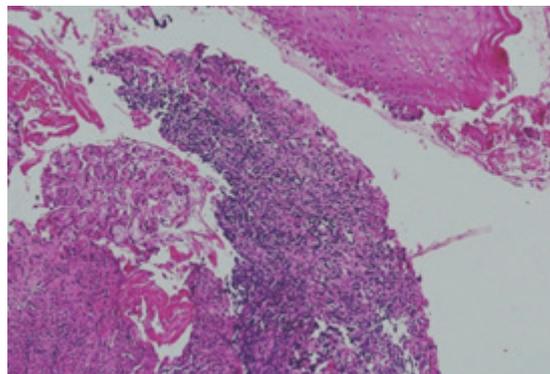
There is no complete cure for oral lichen planus and only symptomatic treatment is provided which varies according to severity of involvement. The possibility of Lichenoid drug reactions should be ruled out before beginning of treatment. Topical corticosteroids remain the mainstay in the general treatment of oral lichen planus.³ However, when there is an acute exacerbations of oral symptoms systemic steroids are recommended in doses of 1-2 mg/kg/day. Adverse effect of long term use of corticosteroids should be weighed against the benefits.¹⁰ In the present due to severity of oral symptoms, a combination therapy consisting of oral and topical corticosteroid had been used for first 15 days. Prednisolone was systemic corticosteroid of choice used (10 mg/day) along with topical Clobetasol ointment (0.05%) which is most potent of all topical steroids and had leads to improvement in clinical signs and symptoms. After 15 days systemic steroid was withdrawn and patient was advised to continue topical



Figure1 Severe blackish discoloration in right buccal mucosa (a) A reddish ulcer of approximately 1x 1 cm in dimension present in left buccal mucosa at occlusal plane, surface appears rough, edges ill defined (b)



Figure 2 Bleeding is evident from gingival margin in relation to 63,64 region



3 Histopathological section of left buccal mucosa showing atrophic epithelial ridges with hyperkeratosis. Degeneration of basal layer with dense subepithelial lymphocytic infiltrate is also seen

Clobetasol ointment (0.05%) twice daily for 6 months. There was a significant improvement in oral symptoms including burning sensation and inflammation of oral mucosa with little improvement in it's appearance. The present case is unique in the aspect that it represents a severe variety of oral mucosal lichen planus without any cutaneous involvement which is quite unusual in pediatric population.

Summary :

A 5 years old male child reported with severe burning sensation, gingival erosion and blackish discoloration of oral mucosa without any cutaneous manifestations with no other systemic illness or local factor associated. Combination of systemic and topical steroid had been tried as mainstay of treatment with remission of clinical

signs and symptoms.

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