Case Report

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A RARE CASE REPORT OF EROSIVE ORAL LICHEN PLANUS

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ABSTRACT:

Lichen planus is a chronic autoimmune, mucocutaneous disease, which can affect the oral mucosa, skin, genital mucosa, scalp and nails, and commonly seen in adults. Childhood lichen planus is a rare entity, which is characterized by skin lesions with the oral involvement being extremely uncommon. There are only a few reports on this subject in the literature. Early and correct diagnosis with adequate management is very important to avoid further complications as it is a pre-malignant condition. Herewith, we report a case of 12 year old boy having erosive oral lichen planus. This is to add another case of lichen planus in a child and to emphasize its consideration in the differential diagnosis of oral mucosal red and white lesions in children.

Keywords: Erosive lichen planus, childhood lichen planus, paediatric lichen planus

INTRODUCTION

Lichen planus is a common chronic inflammatory disease of skin and mucous membranes. It was first described and named by British physician *Erasmus Wilson* in 1869[1]. Oral lichen planus in childhood (OLP) is rare and it was first reported in the 1920s. Oral mucosal involvement in adults itself accounts for 0.5% to 19%, while in children, it is very uncommon, the youngest case documented in a child aged 3 months. Paucity of reported cases of juvenile OLP may be due to lack of patient and parent awareness of lesions, lack of recognition by practitioners, low incidence of autoimmune diseases and precipitating factors such as stress[2]. Familial LP differs from the classical form clinically, with earlier age at onset, more generalized involvement, and more common mucosal involvement[3].

Herewith, we report a case of 12 year-old boy having erosive oral lichen planus without cutaneous involvement, which responded well to the treatment. This is to add another case of lichen planus in a child and emphasize its consideration in the differential diagnosis of oral red and white lesions, particularly, in children.

CASE DETAIL:

A 12 year old boy reported to the Department of oral medicine and radiology with a complaint of burning sensation in the mouth since 2-3 months. History revealed burning present on both right and left buccal mucosa, particularly, while consuming hot and spicy food. Patient's father had history of tuberculosis

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6 years back of which he has taken complete treatment. Patient was not having any habit history. There was an asymptomatic cutaneous lesion of size 1.5 x 1.5 cm present on the nape of the neck which was present since patient was 5 years old.

On examination atrophic and ulcerative areas of size approximately 1 x 0.5 cm along with grayish white elevated striae were present on left buccal mucosa extending from distal of 85 upto pterygomandibular region, and similar lesion with lesser ulcerations present on right buccal mucosa extending from distal of 75 upto pterygomandibular region of that side.

The patient's oral hygiene was poor without any dental restorations. Dental hard tissue examination revealed dental caries with 75 and 85. Based on clinical findings and history of the patient, a provisional diagnosis of erosive oral lichen planus with clinical differential diagnosis of lichenoid reaction and Discoid Lupus erythematosus was made. Investigations advised were complete blood count and ESR, reports of which came out to be within the normal range.

The incisional biopsy was done, and histopathologic features were significant with atrophic parakeratinized stratified squamous epithelium, saw teeth rete pegs and well-defined bands of chronic inflammatory cells subepithelially. Few degenerating keratinocytes interfaced between epithelium and connective tissue forming colloid or civette bodies. These features were suggestive of an atrophic oral lichen planus.

Patient was advised to undergo all required dental treatments like oral prophylaxis and restorations. General measures for management of the OLP included meticulous oral hygiene and avoidance of any form of physical injury to oral mucosa. Patient was instructed to have diet rich in fresh vegetables and fruits. Specific treatment for OLP was with topical 0.1 % w/v betamethasone drops. Chewable mebendazole tablet was advised once OD along with multivitamins as dietery supplements and patient was kept on follow ups. First review of the patient after one week showed a significant reduction of about 30-40% in both symptoms and signs of the oral lesions. At second follow-up after 15 days, there was almost 60% improvement in the condition. Erosive oral lesions had completely healed by one month of treatment, leaving asymptomatic reticular areas.

DISCUSSION:

Oral lichen planus in childhood is exceptionally rare (<2%–3% of the total), and only a few reports are available in the literature. Most of these cases reported in the tropics, mainly from India, with some from the United Kingdom, Italy, Mexico, African America, and Kuwait. This could be due to specific genetic predisposition and environmental factors in the pathogenesis of lichen planus.[1]

In a UK study, four out of six OLP cases reported were from Asian origin. In another UK study, 21 of 26 LP cases were

from the Indian subcontinent. In a study from the Netherlands, two of the three patients with OLP were from Asian origin, in a region where approximately 1% of the population is Asian.[4]

Woo *et al*. 2007 did the literature review on childhood OLP from 1990 to 2005 and found the slight male predilection and common age of occurrence were 11 and 15 years with no ethnic predilection. Buccal mucosa was the commonly affected site, and most patients were with reticular pattern, like present case age of the patient was 12 years with both buccal mucosa involved however type was erosive lichen planus.[5]

Familial lichen planus has been reported as being uncommon. Milligan reported a family history present in 1-2% of cases. Childhood familial lichen planus is said to occur at an early age and with greater severity.[6]

Alam and Hamburger described 6 OLP patients, all males ranging from 6 to 14 years old. Based on their series, these authors proposed a possible male predilection for juvenile OLP.Eisen evaluated 723 OLP patients and only 5 (<1%) were children younger than 15 years old.7 All 5 had atrophic and erosive OLP, and all developed dermal LP within 2 years of oral onset. Of note, Eisen reported that each patient was initially misdiagnosed and treated incorrectly for other oral conditions, such as herpes simplex, candidiasis, and recurrent aphthous stomatitis[7].

With more extensive disease there are painful, persistent erosions on the gingiva, and ulcers on the buccal mucosa, tongue, and labial mucosa. Difficulty in eating results in weight loss and nutritional deficiencies such as iron deficiency. Painful erosions lead to suboptimal dental hygiene and increased tooth decay[8].

Nail involvement is rare in children while it occurs in 1-10% of adults. In different studies, nail involvement has been found in 0-8.7% of patients[3].

The differential diagnosis of childhood OLPc may be quite extensive and, of course, depends on the age of the patient, the clinical variant, and the severity and the persistence of the lesions and includes candidosis, morsicatio buccarum, leucoplakia, stomatitis migrans, auto-immune bullous diseases, lupus erythematosus, several viral infections (herpes simplex, Epstein-Barr, Coxsackie, HIV), recurrent aphthous stomatitis (caustic) traumata, erythema multiforme (major), allergic gingivostomatitis, gluten sensitivity enteropathy, and less commonly Crohn and Behçet diseases, oral lesions in immunodeficiencies, dyskeratosis follicularis, pachyonychia congenita, dyskeratosis congenita, white sponge nevus; allergy to flavorings may be relevant in- stead in the diagnosis of allergic contact stomatitis[4] .

The histopathological findings in our case were in accordance with essential features suggestive of OLP.

The aims of current OLP therapy are to eradicate mucosal erythema and

ulceration, alleviate symptoms, and reduce the risk for oral cancer. Corticosteroids administered topically, intra-lesionally or systemically, depending on the age and condition of the patient, site, and severity of the lesions is the mainstay of the therapy. Topical corticosteroids remain the treatment of choice in most patients with localized lesions[1,9,10]. The present case was managed successfully with topical steroids and multivitamin supplements.

Clinicians must be aware that OLP children also may have simultaneous or future involvement of skin and other mucosal sites; if lesions are reported elsewhere, appropriate referrals are necessary. The asymptomatic reticular variant of OLP appears to predominate in children. Therefore, pharmacologic treatment is often not necessary. In symptomatic patients, good oral hygiene should be encouraged as a means of reducing irritating factors such as plaque and calculus. The prognosis of juvenile OLP is unclear at this time, as long-term studies have not been published. Although Laeijendecker et al reported no OLP-related malignancies to date in the pediatric population, it appears that careful follow-up of all OLP patients is warranted[7].

The prognosis and the effect of treatment in OLP in children seem to be more favorable than in OLP in adults, which usually persists for many years in spite of intensive treatment and thorough investigation of associated factors.

Malignant transformation of ulcerative OLP in adults is 0.07% to 5%; however, malignant transformation of OLP in children is not documented in the literature till now[11].

CONCLUSION

Oral lichen planus in childhood is rare, especially erosive form; diagnosis should be based on children presenting with ulcerative white lesion in oral cavity. The schedule of follow up of OLP in children should be 7 days, 15 days, and 30 days after diagnosis to assess healing. Patient should be reviewed twice a year for regular follow ups after complete progress of the present condition. However, generally, the prognosis of oral lichen planus in childhood seems to be more favorable compared to adults.

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FIGURES:

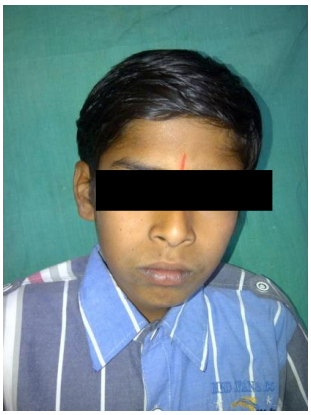

Figure 1 showing extraoral picture of 12 year old boy

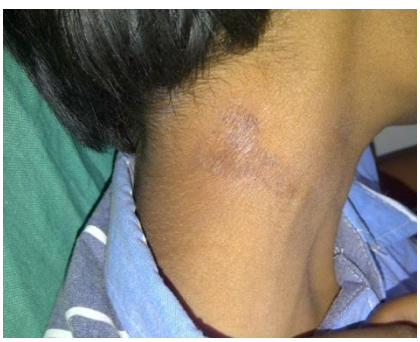

Figure 2 showing hyperpigmented macular lesion present on the neck.

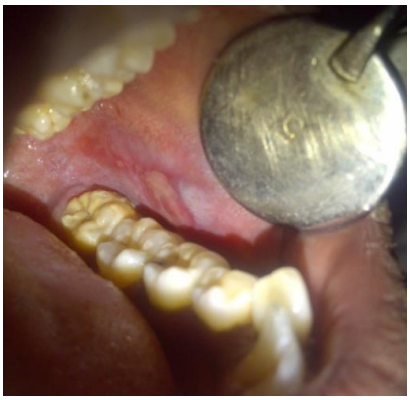

Figure 3 showing atrophic ulcerative lesion with fine grayish white striations present on left buccal mucosa

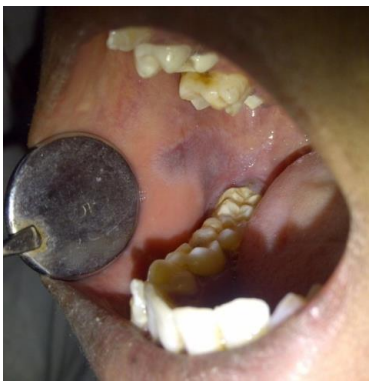

Figure 4 showing greyish white fine radiating striated lesion present on left buccal mucosa

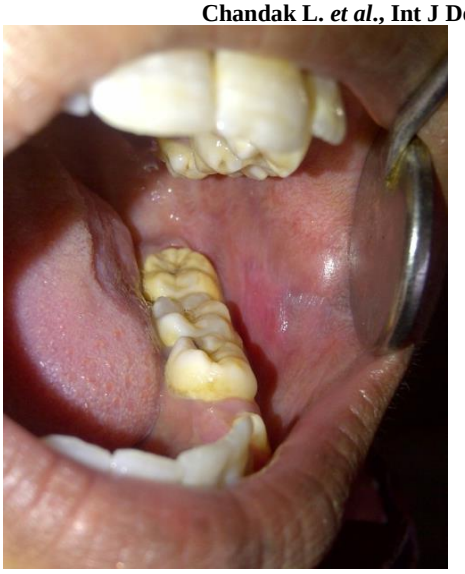

Figure 5 showing improvement in the lesion on left side after duration of 1 month

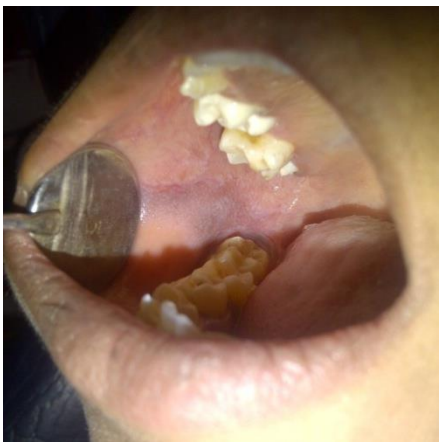

Figure 6 showing improvement in the lesion onright side after duration of 1 month