Abstract
Lichen planus is a mucocutaneous disorder that predominantly affects older patients and less frequently the paediatric population. Oral lichen planus (OLP) can cause severe morbidity if undiagnosed and it carries low risk of malignant transformation potential too. Although OLP in children is rare, it should be included in the differential diagnosis of oral mucosal ulcerative disease because it is important to prevent the serious morbidity that may result from disease. This paper describes a case of bullous OLP in a 9 year old child that was diagnosed and successfully managed.

Introduction
Lichen planus (LP) is a relatively common condition, which was first described by Eramus Wilson in 18691 and it is also a chronic inflammatory disease that affects the skin and the mucous membrane. Oral lichen planus (OLP), the mucosal counterpart of cutaneous lichen planus, presents frequently in the fourth decade of life and affects women more than men in a ratio of 1.4:1. The disease affects 1–2% of the population2, 3. It is estimated that 50% to 70% of adult LP patients have both skin and oral lesions 4 and approximately 25% of patients present with oral lesions alone5. Cutaneous LP is characterized by purple, pruritic, polygonal papules with overlying reticular striations that tend to localize on the extremities and lower back6. In contrast to skin lichen planus OLP demonstrates clinical variability. It is seen clinically as reticular, papular, plaque-like, erosive/ulcerative, atrophic /erythematous and bullous types as classified by Andreason7. Intraorally, the buccal mucosa, tongue and the gingiva are commonly involved although other sites may be rarely affected8.

The disease process is characterised by quiescence and exacerbations9. OLP is a T-cell mediated autoimmune disease in which the auto-cytotoxic CD8+ T cells trigger apoptosis of the basal cells of the oral epithelium10.

Reticular OLP is usually asymptomatic while atrophic, erosive or bullous types cause pain and burning sensation11. The histopathology of OLP shows variable hyperkeratosis, basal cell degeneration, and a band-like predominantly lymphocytic infiltrate in close proximity to surface epithelium. While OLP is widely recognized in adults, its occurrence in children is uncommon. The exact incidence of paediatric LP is unknown, as percentages greatly vary from practice to practice. However, several retrospective reviews, have estimated that only 1% -16% of LP patients are less than 15 years old5, 12. Moreover, juvenile OLP, which is defined as OLP in patients younger than 20 years old,
has rarely been documented in medical/dental literature. Proposed factors responsible for this paucity of reports include lack of patient and parent awareness of lesions, and misdiagnosis or lack of recognition by practitioners. In this paper, we report a case of bullous type of oral lichen planus in a 9 year old child.

Case report

A 9 year old boy was presented to the Oral Medicine Clinic of the University Dental Hospital Peradeniya, Sri Lanka with oral mucosal ulcers of more than 1 month duration. During this period, he had experienced difficulty in eating and swallowing. Previous incisional biopsy done at a different hospital was not conclusive. There was no family history of oral ulcers of a similar nature. The patient was allergic to Cloxacilline. He had no history of erosions or ulcers on the skin or other mucosal surfaces.

Clinically, both upper and lower lips were dry and there were white reticular lesions with black pigmentation on both lips (Figure 1). Intra orally there were atrophic, erosive areas on both buccal mucosa and white striated appearance on dorsal tongue. Full blood count revealed elevated white blood cell count (WBC= 13.10) and elevated levels of differential WBC counts of monocytes, eosinophills, basophills. Also reduced levels of mean corpuscular volume (MCV=73.8 fl) and mean corpuscular haemoglobin (MCH=24.8). Provisional diagnosis is of vesiculobullous disorder was made. As the patient was symptomatic an antiseptic mouth wash (0.2% chlorhexidine) was prescribed.

On the second visit, an oral mucosal incisional biopsy was performed under local anaesthesia for routine histopathology and direct immunofluorescence. Microscopically there was a dense band of lymphocytes admixed with scattered plasma cells which was associated with basal cell destruction and apoptosis. Subepithelial cleft formation was also evident (Figure 2). Direct immunofluorescence was negative for IgG, IgM, IgA and C3. Considering above features, bullous OLP was made as the definitive histopathological diagnosis.

The patient was managed with topical Betamethasone. After six weeks the condition subsided leaving a mild burning sensation of the oral mucosae and plaquelike lesions on the dorsalsurface of the tongue suggestive of candida. The smear was positive and one tablet of nystatin was prescribed daily for a period of one month along with topical triamcinolone acetonide three times a day. Currently the patient is under regular follow-up.

Discussion

Many illnesses and conditions are associated with LP occur in older patients. Thus, explaining the occurrence of OLP in the younger population is a challenge. Important factors in the development of juvenile OLP include: (1) previous hepatitis B vaccination, (2) Liver disease, including chronic active hepatitis, (3) Genetic predisposition, such as in familial LP.

A familial history of LP deserves brief discussion, as it has been regarded as a relevant predisposing factor in pediatric patients. Milligan and Graham-Brown reported a family history of LP in 1% to 2% of their juvenile OLP patients, whereas Cottonie et al found that 1 of 5 (20%) of their juvenile OLP cases had a positive family history. In contrast the present case has no related family history.

Handa and Sahoo reported 87 patients with childhood LP in India. Out of which only seven patients showed concomitant involvement of the oral mucosa and only one patient had isolated OLP. Kumar et al reported involvement of the oral mucosa in only one of the 25 children with cutaneous lesions. However, Sharma and Maheswari reported 50 children with LP and with concomitant oral lesions in 15 of them. Generally the oral mucosa seems to be less common in children with LP than adults.
Eisen reported that patients with juvenile OLP were initially misdiagnosed and treated incorrectly for other oral conditions, such as herpes simplex, candidiasis, and recurrent aphthous stomatitis. Based on the initial clinical presentation, the present case was also suspected as vesiculobullous disorder.

Treatment of juvenile OLP does not differ significantly from treatment of adult OLP. Pharmacologic treatment is often unnecessary in asymptomatic patients. For symptomatic lesions, topical corticosteroids are the most commonly used agents. The patient and parents should be informed about the chronic use of topical steroids which can lead to oral candidiasis. Systemic steroid therapy typically reserved for refractory and recurrent cases. Extreme caution also should be taken when these agents are used, as significant long-term effects are of concern in this young patient population.

Periodic follow-up is required in all OLP patients. This is especially important in the pediatric population as malignant transformation has been described in a small percentage of adult OLP cases in follow-up studies. Skin lesions may develop in these patients and appropriate referral is necessary. Additionally, it is advisable to rule out lichenoid mucosal reaction due to systemic medication or dental restorative materials.

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. Many authors are of opinion that prognosis and effect of treatment of OLP in children seems to be more favourable than OLP in adults.

**Conclusion**

OLP is rare in paediatric patients. However, it is important to diagnose OLP in children to prevent the morbidity and to avoid unnecessary pharmacotherapy. This case shows that OLP should be included in the differential diagnosis of oral mucosal ulcerative diseases in children. Mucosal lesions in paediatric patients may be misdiagnosed by practitioners. It is essential that dentists should be aware of such existence so that the patients can be referred to specialist management without undue delay.

**References**


Figure 1: white reticular lesion and black pigmentation in both upper and lower lips

Figure 2: cleft formation along the basal cell layer with dense band of lymphocytes admixed with plasma cells in the upper corium associated with basal cell destruction and apoptosis. (H & E x200)