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# Oral lichen planus in childhood: a report of three cases

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**Summary.** Lichen planus is a common mucocutaneous condition in which the occurrence of oral lesions has been cited as between 0·5 and 1·9% of the adult population. It is rare in childhood. Here we report three cases of children with intra oral lesions of lichen planus. Lichen planus, although reportedly rare in childhood, should be considered in the diagnosis of hyperkeratotic or erosive lesions of the oral mucosa in children.

## Introduction

Lichen planus is a common chronic inflammatory disease of skin and mucous membranes. It is observed most frequently in patients of middle age or older, with females accounting for approximately 60–65% of patients [2,3]. The aetiology of lichen planus remains uncertain but many factors have been implicated. Such factors include genetic predisposition, infective agents, systemic diseases, graft-vs.-host disease, drug reactions, and hypersensitivity to dental materials and vitamin deficiencies [1]. Lichen planus has been associated with several auto-immune diseases, including lupus erythematosus, pemphigus, Sjögren's syndrome and autoimmune liver disease [3,4]. The pathogenesis of lichen planus is not completely understood but a T-lymphocyte infiltrate suggests cell-mediated immunological damage to the epithelium [2]. Modified Langerhans' cells and keratinocytes possibly trigger an immune response and the recruitment of T lymphocytes, encouraged by expression of cell-surface adhesion molecules [1,4]. Both CD4 (helper) and CD8 (cytotoxic) cells are present but increasing numbers and activation of the CD8 cells is thought to contribute to the characteristic damage to the basal epithelium [1,2].

Up to six clinical appearances of oral lichen planus have been described [3], including reticular, atrophic, plaque-like, papular, erosive and bullous types. The characteristic sites involved are the buccal mucosae, dorsum of the tongue and less frequently the gingivae.

There is very little literature on oral lichen planus occurring in childhood [4,5]. This paper reports three cases of oral lichen planus in children.

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## Case report

### *Case one*

A 15-year-old Caucasian girl was referred to the Charles Clifford Dental Hospital with 4-month history of persistent oral ulceration affecting the tongue bilaterally. The lesions were painful with no relieving or aggravating factors. Her medical and family histories were unremarkable at initial presentation.

Oral examination showed ulceration and erythema with a surrounding margin of hyperkeratosis on both left and right lateral borders of the tongue extending onto the dorsal and ventral surfaces and into the floor of the mouth (Fig. 1). The dental state was excellent and there were no amalgam restorations. No other mucosal or skin surfaces showed lesional changes at the initial presentation.

A differential diagnosis of erosive lichen planus or discoid lupus erythematous was made. Investigations included full blood count, haematinics, biochemistry and immunology screen. The results were uniformly normal other than a borderline low serum ferritin of 8 μg/L (normal range 7–140) and a slightly reduced IgA of 0·60 g/L (normal range 0·8–2·8). An incisional biopsy of the lateral margin of the tongue showed histopathological features consistent with lichen planus.

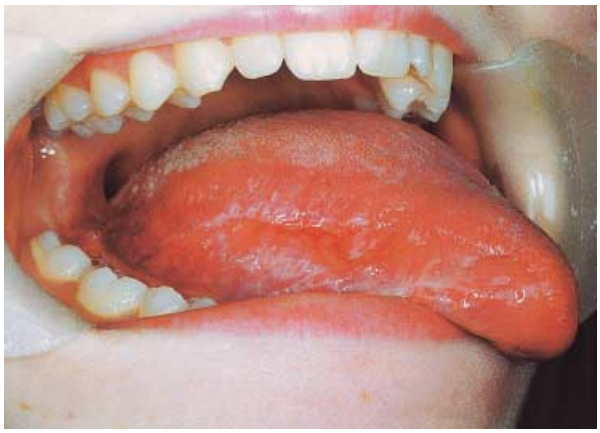

**Fig. 1.** Case 1: erosive lichen planus of the right lateral border of the tongue.

The patient was initially treated with a topical Prednisolone mouthwash of 5 mg four times per day, but response to therapy was slow. As this treatment regime proved difficult to maintain in a school setting the prescription was changed to topical Beclamethasone spray; the ulceration gradually resolved. The borderline low serum ferritin (owing to dietary factors) returned to normal following therapy with iron supplements. Six months following presentation the patient was diagnosed with idiopathic hypothyroidism and placed on Thyroxine; currently the dosage is 100 μg per day. This is under follow up by the endocrinologists. Two years after the occurrence of oral lesions, skin lesions of lichen planus occurred on the neck and upper trunk and were successfully treated with topical Betamethasone ointment. The oral lichen planus lesions occasionally show flares of activity and are successfully controlled by the topical Betamethasone spray when required. The patient is currently under regular review.

*Case two*

A six-year-old Caucasian boy was referred with a 2-year history of an asymptomatic white patch on the dorsal surface of the tongue. The patient was autistic but otherwise fit and healthy. The family history was unremarkable.

Examination revealed two areas of hyperkeratosis on the dorsal surface of the tongue, measuring approximately $4 \times 2$ cm and $1 \times 0{\cdot}5$ cm in size (Fig. 2). No other mucosal or skin lesions were present.

The incisional biopsy confirmed features consistent with lichen planus. Routine haematology and biochemistry were unremarkable. No active treatment was provided and the patient is being reviewed periodically.

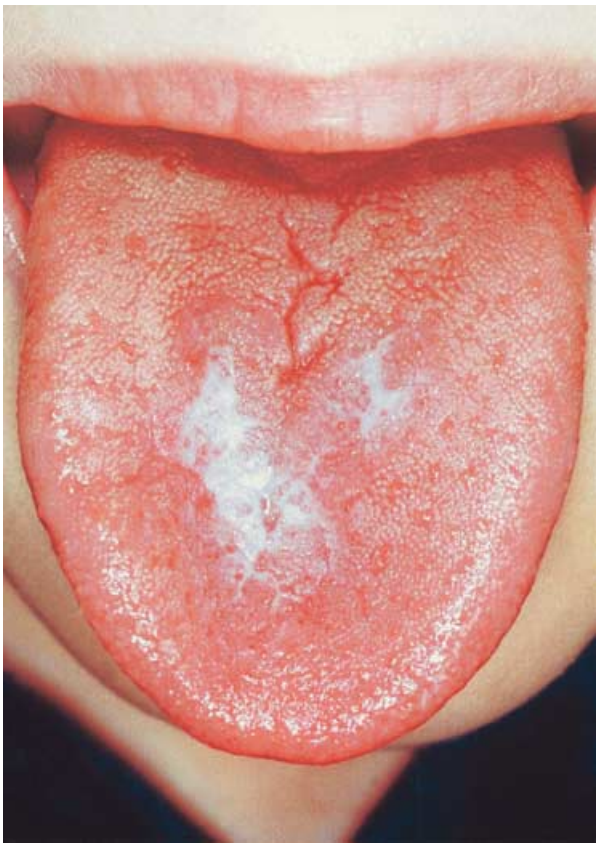

**Fig. 2.** Case 2: plaque-like lichen planus of the dorsum of the tongue.

*Case three*

A nine-year-old Caucasian girl was referred with a 6-month history of increasing soreness affecting both keratinized and nonkeratinized oral mucosa. No other mucosal or skin surface was affected. Medical history revealed the patient to have mitral valve atresia and to be awaiting a cardiac transplant. She was allergic to penicillin and was currently taking Enalapril on presentation. There was no relevant family history.

Four months before presentation to the Oral Medicine Department the patient had seen her General Medical Practitioner (GMP) with an 'ulcer' affecting her left buccal mucosa. The GMP wondered if it was an infected ulcer, prescribed a week's course of erythromycin and withdrew the previous captopril therapy, as he was concerned that there might be a possible drug-related aetiology. No resolution occurred

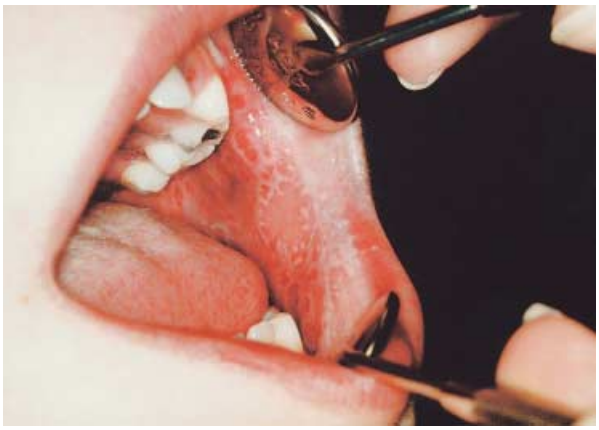

**Fig. 3.** Case 3: reticular lichenoid pattern of the left buccal mucosa.

and the area on the buccal mucosa remained sore. A course of nystatin provided little improvement in symptoms. Just prior to her referral to the Oral Medicine Department, corlan pellets were prescribed and the cardiac physicians started enalapril therapy to replace the previous captopril. Captopril has an established history of inducing lichenoid reactions.

At presentation the 9-year-old girl was obviously cyanotic. Intra-oral examination revealed widespread reticular lichen planus affecting the buccal mucosae bilaterally, the palate, dorsal surface and lateral borders of the tongue and the floor of the mouth (Fig. 3). There was no evidence of ulceration, but some erythema was present amidst the reticulation. The symptoms of discomfort previously experienced had reduced, but were still exacerbated by favoured foods such as potato crisps. In light of the patients underlying medical problems and diagnostic presentation, routine haematological investigations and biopsy were not performed.

Treatment consisted of avoidance of irritating foodstuffs and topical Beclamethasone spray to be used when symptomatic. Periodic reviews showed an improvement in both symptoms and severity of the widespread nature of the reticular lichen planus. The lesions are currently present but quiescent and no topical therapy is needed.

## Discussion

Lichen planus was first described in the literature by Eramus Wilson in 1869 [3], as predominately a disease of the middle aged or older. There is limited literature available reporting the occurrences of oral lichen planus in children [4–11,14,15,18].

Cutaneous lichen planus in childhood is an uncommonly encountered dermatosis [3,6–12,20] and is extremely rare in infancy [3,19]. Childhood lichen planus has been documented as a complication of Hepatitis B vaccinations (HBV) where the recombinant proteins of the HBV vaccine, specifically the viral S epitope, may trigger a cell-mediated autoimmune response targeted at keratinocytes giving rise to a lichenoid reaction [16,17]. It is also found in association with predisposing conditions such as graft-vs.-host disease and chronic active hepatitis C [18]. In the majority of cases reported cutaneous involvement was the only manifestation of the lichenoid lesions. Cottoni *et al*. reported oral lichen planus involvement in one patient affected by autoimmune chronic active hepatitis [18], and Agrawal *et al*. [17] reported oral lesions in one patient after HBV vaccination. Limas *et al*. [16] did not identify oral lichen planus involvement in their study of five children following HBV vaccination but a review of their literature mentioned two children with mucosal involvement following vaccination.

Familial lichen planus has been reported as being uncommon [5,12–15]. Milligan [12] reported a family history present in 1–2% of cases. Childhood familial lichen planus is said to occur at an early age [5] and with greater severity [12]. Mahood reported that 12% of his patients with familial lichen planus presented before the age of 10 years [14].

Reports in the literature describing children with lichen planus have tended to demonstrate the majority as having cutaneous manifestations and a low incidence of oral involvement. Kumar *et al*. [11] and Kanawar *et al*. [6] reported one out of 25 (4%) and one out of 17 (6%) cases, respectively, showing mucosal involvement. More recent studies have revealed a greater incidence of children developing oral lichen planus. Nanda *et al*. [7] reported nine out of 23 (39%) of their childhood cases presented with mucosal lesions, and Sharma *et al*. [8] described 15 out of 50 cases (30%) with mucosal lichen planus involvement. The most recent study of 87 cases of childhood lichen planus reported 12 of the patients (13·8%) to have oral mucosal involvement [9].

Presently there are no published figures on the prevalence of childhood oral lichen planus. Scully *et al*. [5] reported three females with an age range of 10–11 years with oral lichen planus All three children had no relevant underlying medical or family histories. Alam *et al*. [4] documented six cases of oral lichen planus in male patients between the

ages of 6–11 years. Of these, one was an asthmatic; otherwise all medical and family histories were non-contributory. Four of the six patients were of Asian origin. It has been documented that childhood lichen planus is more common in the tropics [11] and that children of Asian origin may be more prone to the condition [4,9,12]. None of the children presented in this case report was of Asian origin.

Some authors [12,18] have suggested that there is a difference between childhood and adult lichen planus in that the presentation is often of prolonged atypical skin lesions, which are sometimes recalcitrant to topical therapy. Also, of those cases reported a significant proportion had a family history of lichen planus. Handa *et al*. [9] in a study of 87 cases concluded that the natural history of lichen planus in children was essentially similar to that in adults. The majority of children affected with oral lichen planus, however, have been asymptomatic or complain of slight dryness of the mouth [9,10].

Erosive oral lichen planus tends to affect approximately 40% of adult patients [4]. In children, of the cases reported, it is rare. Alam *et al*. [4] reported one case of erosive lichen planus from their study of six cases. Howard and Tsuchiya [20], reviewing paediatric cutaneous disorders, reported one 16-year-old with erosive oral lichen planus. In this report of three cases of childhood lichen planus one patient presented with erosive lesions.

In the three cases presented in this report there was no family history of lichen planus. All three children presented with an associated medical condition, which is of interest, as few of the previously reported cases are similar. Alam *et al*. [4] reported one child who suffered from asthma, and Cottoni *et al*. documented one child with oral lichen planus affected by antiliver-kidney microsome-positive chronic active hepatits [18].

## Conclusion

Although oral lichen planus is considered rare in childhood, the presence of often asymptomatic oral lesions should alert the clinician to such a diagnosis. The cases described in this paper highlight the importance of considering lichen planus in the differential diagnosis of hyperkeratotic and erosive lesions of the oral mucosa in childhood.

**Résumé.** Le lichen plan est une pathologie cutanéo-muqueuse commune dont l'occurrence buccale va de 0,5 à 1,9% dans la population adulte [1], mais est rare chez l'enfant. Trois cas chez l'enfant sont rapportés dans cet article. Le lichen plan, bien que rarement répertorié chez l'enfant, devrait être évoqué lors du diagnostic des lésions hyperkératosiques ou érosives de la muqueuse buccale chez l'enfant.

**Zusammenfassung.** Lichen planus ist eine verbreitete mukokutane Veränderung, die Häufigkeit von oralen Veränderungen wurde angegeben mit 0.5% bis 1.9% der erwachsenen Bevölkerung. Im Kindesalter beobachtet man solche Veränderungen dagegen selten. Hier werden drei Fälle von oralem Lichen planus bei Kindern berichtet. Auch wenn entsprechende Fälle bei Kindern selten dokumentiert sind, sollte man bei hyperkeratotoschen oder erosiven Läsionen auch im Kindesalter an Lichen planus denken.

**Resumen.** El liquen plano es una alteración mucocutánea común en la que la aparición de lesiones bucales se ha situado entre el 0,5 y el 1,9% de la población adulta (1), es raro en la infancia. Aquí señalamos tres casos de niños con lesiones orales de liquen plano. El liquen plano, aunque se ha informado que es raro en la infancia, debería ser considerado en el diagnóstico de lesiones erosivas o hiperqueratósicas de la mucosa oral en niños.

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