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Lichen Planus in Children

Abstract: Lichen Planus (LP) is a chronic, inflammatory disease of the skin and mucous membranes. It is more frequently seen in the middle-aged and elderly population but can be present in children, although this is relatively rare. This paper describes the presentation and management of lichen planus in children, illustrated by seven cases seen within the Paediatric Dentistry Unit. Dentists should be aware of the condition and understand when referral to a specialist centre is required and the need for multidisciplinary management of complex cases.

CPD/Clinical Relevance: Although oral lichen planus is rare in children, it is important that dentists are able to identify its clinical presentation and abnormal changes to the oral mucosa, as well as being aware of possible local and systemic causes of the condition so that reassurance and correct management pathways can be implemented in primary care practice.

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Lichen planus is a chronic inflammatory disease associated with a cell-mediated immune reaction affecting the skin and mucous membranes.[1] Wickham, in 1895, described the characteristic appearance of skin lesions as, 'white striae that develop atop the flat surfaced papules'.[2]

Up to six clinical appearances of oral lichen planus (OLP) have been described in the literature.[3] The characteristic sites involved are the buccal mucosa, lateral borders of the tongue and, less frequently, the gingival tissues. Table 1 describes the different types of oral lichen planus seen.[4]

Oral lichen planus is relatively rare in children, so few reports on this subject are present in the literature.[5,6] This paper reports seven different cases of oral lichenoid disease in children with the aim of supporting clinicians in recognizing the presentation of OLP and its subsequent management.

We present a series of cases seen on the joint Oral Medicine and Paediatric dentistry clinic of the Charles Clifford Dental Hospital. This demonstrates the ages and gender of the patients seen, along with a brief description of the presenting complaint, clinical presentation and the management plan.

Demographic factors

Lichen planus is a chronic inflammatory mucocutaneous disease. It commonly involves the oral mucosa but other extra-oral sites may be affected including the skin, scalp, genital area and the nails. Oral lichen planus (OLP) affects between 0.1% and 2.2% of the adult population.[7] It is seen most frequently in the middle-aged and elderly population,[8] with females accounting for approximately 60–65% of patients.[9] The incidence rates for children are limited to case reports and case series, owing to the small number of cases that present clinically. There is no apparent gender predilection in children.[10] OLP affects all races, although reports suggest that childhood lichen planus is more common in the tropics, especially in Indian populations.[11] OLP has been reported in six boys aged 6 to 14 years over a 20-year period in 2001[12] and, in another case series, reported in three girls.[6]

Aetiology

The exact aetiopathogenesis of lichen planus is not completely understood, but a T-lymphocyte infiltrate suggests cell-mediated immunological damage to the epithelium.[13,14]

Lichen planus has been associated with genetic predisposition, diabetes, hypertension, infections including hepatitis C, autoimmune liver disease and dental restorations.[9,13,15]

Childhood lichen planus has been documented as a complication of Hepatitis B vaccinations (HBV) where the recombinant proteins of the HBV vaccine, especially the viral S-epitope, may trigger a cell-mediated auto-immune response targeted at keratinocytes giving rise to a lichenoid reaction.[16,17] It is also found in association with predisposing conditions such as Graft versus host disease (GvHD) and chronic active Hepatitis C.[18] Genetic factors and lifestyle have also been cited as aetiological factors. More recent studies suggest that at least 50% of cases reported had a familial history of lichen

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Lichen Planus	Clinical Presentation
Reticular	Most common type of LP Characteristic interlacing white keratotic lines
Papular	Small lesions (0.5–1.0 mm diameter) – often overlooked during clinical examination Raised papules may be seen with another type of LP
Plaque-like	Plaques can be smooth, flat areas or irregular, elevated area
Erythematous/ Atrophic	Second most common type of oral LP Presents as areas of erythema and may be surrounded by fine keratotic striae
Ulcerative	Variant of the erosive type (Figure 1)
Bullous	Variant of the erosive type Bullae form and rupture very soon after appearing, resulting in the classic appearance of erosive LP

Table 1. Clinical presentations of oral lichen planus.[3,4]

Figure 1. Erosive oral lichen planus.

planus.[19] In the seven cases presented in Table 2, there was no family history of lichen planus, which is consistent with an earlier case series.[20]

Reports in the literature describing children with cutaneous lichen planus have highlighted additional oral involvement in 4–39% of cases.[21-24] Interestingly, two of our seven cases had cutaneous and oral mucosal lesions and in both these cases the referral had come from medical colleagues based on the initial cutaneous lesions.

It is unusual for children to develop an oral lichenoid reaction in association with dental amalgam.[25] Hypersensitivity to dental amalgam is rare and, according to Holmstrup,[26] is due to corrosion products of amalgam restorations. In almost all cases it seems to be related to mercury, with only a few cases implicating silver, copper, or tin.[26,27] These oral lichenoid lesions (OLL) represent a contact allergy to dental amalgam which may develop after sustained contact for several years.[28,29] Resolution of oral lesions after removal of amalgam restorations in such patients has been reported.[29] This improvement was found within one week to three months of replacing amalgam restorations, with greatest improvement when the lesion is in close contact with OLLs.[25]

There are three case reports in the literature stating lichen planus in association with Turner's syndrome.[30-32] Previous associations of lichen planus with other medical disorders have been described, however, its occurrence with Turner's syndrome is yet unrecognized.

Diagnosis

The clinical presentations of oral lichen planus are diverse, ranging from the classical white symmetrical reticular network found characteristically bilaterally on the buccal mucosa, to widespread and debilitating ulcerative lesions. LP may be diagnosed readily by its clinical presentation alone, particularly if the classic reticular form is noted.[15] However, in most cases histological evaluation of tissue from a biopsy is required for a definitive diagnosis to be made.[3]

The differential diagnosis of lichen planus includes chronic candidosis, chronic cheek chewing, lichenoid reaction to dental amalgam or drugs, Graft versus host disease and possible vesiculobullous conditions in children presenting with bullae or ulcers.[3]

Management

Children affected with LP are often asymptomatic or minimally symptomatic but it is important that parents or carers are aware that there is currently no cure for the condition. Asymptomatic LP (generally the reticular and plaque forms) does not usually require treatment or intervention. Diagnosis and providing the patient with information about the condition will provide reassurance.

Patients will need to be encouraged to maintain excellent oral hygiene as this is believed to be contributory in reducing symptoms.[3] However, this is likely to be difficult during periods of disease activity.

Since LP is relatively uncommon in children, there is very little information in the literature relating specifically to the treatment of the condition in this age group. The concepts of treatment of LP are the same as those for adults, with attention to the recommended age-specific dosages for topical or systemic medication used for children.

Treatment is based around removal or avoidance of aggravating factors and management of symptoms. Symptoms may include burning sensation or discomfort eating certain foods and so avoidance of those foods/flavours should be encouraged. Any amalgam restoration in close proximity to a lesion may need to be replaced. Patch testing may help to diagnose a lichenoid reaction.

Topical analgesia such as Benzydamine hydrochloride (Difflam®, 3M Health Care Limited, UK) mouthrinse or spray is an anti-inflammatory agent that can provide symptomatic relief in cases where pain is experienced during speech and eating. Antiseptic mouthrinses, such as chlorhexidine gluconate (Corsodyl®, GlaxoSmithKline, UK) may also be used[13] and is available in gel form (1.1% w/w) or as a spray. Topical preparations, such as Gengigel® (Ricerfarma SRL, Milano) are also used. The active ingredient in Gengigel® is hyaluronic acid, which promotes tissue healing and provides pain relief. It is available as a gel or mouthrinse.

Topical corticosteroids, such as betamethasone sodium phosphate (Betnesol® UCB Pharma) are indicated in the treatment of LP. Betnesol® is available as a 0.5 mg tablet to be dissolved in water and used as a mouthrinse. Similarly, fluticasone propionate preparations (Flixonase®, Allen & Hanburys

Ltd, UK) and Beclometasone spray (Clenil Modulite®, Chiesi Ltd UK – 50 micrograms) may be used to help alleviate symptoms.[13] Fluticasone propionate drops (Flixonase® Nasule® Drops 400 micrograms (1mg/ml)) are mixed with water and used as a mouthrinse. No single successful treatment regimen has been identified, although betamethasone sodium phosphate mouthrinse and fluticasone propionate preparations are widely used.[33] The risk of topical and systemic steroid use in children is an important consideration.

Tacrolimus or ciclosporin are topical immunomodulators that may be used as second-line treatment in persistent OLP.[33] In very severe cases of OLP, systemic corticosteroids are considered, particularly if the OLP is widespread and involving other body sites that are non-responsive to the first-line topical therapies.[7]

Because of the paucity of randomized controlled clinical trials to evaluate therapies for children, there is a lack of strong evidence supporting the effectiveness of any palliative therapy for OLP in this age group. Recent systematic reviews of therapies for symptomatic OLP concluded that topical corticosteroids are considered to be first-line treatment[33] and evaluated interventions for treating erosive LP, which failed to show superior effectiveness of any specific treatment.[34]

Clinical cases (Table 2)

Case one

Presenting complaint

A 15-year-old Caucasian girl was referred by her dentist to the joint Paediatric Oral Medicine clinic at Charles Clifford Dental Hospital, with a 6-month history of a burning sensation affecting the oral mucosa when consuming spicy foods. Her medical and family histories were unremarkable.

Clinical findings

Extra-orally, no abnormalities were detected. Intra-oral examination revealed white striae bilaterally in the left buccal sulcus and on the right dorsum of the tongue. No other mucosal or skin surface lesions were noted.

Diagnosis

An incisional biopsy of the left buccal mucosa was carried out and the histopathology revealed hyper-parakeratinized stratified squamous epithelium overlying fibrous connective tissue with adipose tissue at the deep aspect and thickening of the basement membrane. A definitive diagnosis of oral lichen planus was made.

Management

The management consisted of avoidance of irritating foods and Difflam® spray to be used when symptomatic. Periodic review showed an improvement in both symptoms and severity of the lichen planus. The lesions are currently present but quiescent and no topical therapy is needed.

Case two

Presenting complaint

A 13-year-old Caucasian girl was referred regarding a 6-month history of pain in the mouth which was aggravated on food intake. The past medical history was unremarkable.

Clinical findings

Oral examination revealed a white, translucent lesion with exaggerated fissuring on the left lateral border of the tongue, measuring approximately 20 mm long x 8 mm wide. Large amalgam restorations in UL6 and LL6 were present (Figure 2).

Diagnosis

Routine haematology and biochemistry were unremarkable. An incisional biopsy revealed features consistent with a lichenoid tissue reaction. As a result patch testing to the dental series was requested and showed a positive reaction for both amalgam alloy and mercury.

Management

The management included replacement of the amalgam fillings in both UL6 and LL6 with glass ionomer cement. The lesion subsequently resolved and was not discernible six months later.

Case three

Presenting complaint

A 13-year-old Caucasian girl was referred by her Consultant Paediatrician with a chief complaint of red and painful gums with small blisters. Medically the patient had been diagnosed with Turner's syndrome at birth and she also suffered from eczema. The patient was originally referred to Sheffield Children's Hospital regarding rashes on the genital area, which had been diagnosed by the Paediatric Physician as lichen planus.

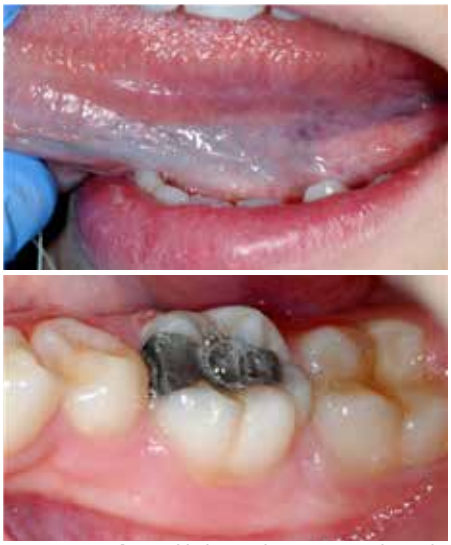

Figure 2. (a, b) Oral lichenoid reaction on lateral border of the tongue associated with large amalgam restoration (Case 2).

Clinical findings

Extra-oral examination revealed dry skin on the face, neck and hands with small and dry nails. Oral examination showed a depapillated red patch on the right side of the dorsum of the tongue with three ulcers on the ventral surface of the tongue. It was also noted that there were white striae and plaque-like patches in the lower right and left lingual sulci.

Diagnosis

A diagnosis of mucocutaneous lichen planus, with lesions affecting the oral mucosa and the genital area, was made based on the patient's history and clinical examination.

Management

The oral lichen planus lesions were successfully controlled using Gengigel® and betamethasone sodium phosphate mouthrinse. The patient is currently under regular review.

Case four

Presenting complaint

A 14-year-old Caucasian girl was referred from the Oral & Maxillofacial Surgery department regarding a white patch on the buccal mucosa. Medical history revealed the patient to have Irritable Bowel Syndrome and an allergy to penicillin.

Case	Gender	Age	Presenting Complaint	Clinical Presentation	Medical History	Diagnosis	Treatment
1	F	15	Burning sensation on consuming spicy food	White striae in buccal sulcus and dorsum of tongue	Nil	Lichen Planus	Difflam® mouthwash
2	F	13	Pain on eating	White translucent lesion with fissuring on lateral border of tongue adjacent to a large amalgam restoration	Nil	Lichenoid tissue reaction associated with amalgam restoration	Replace amalgam with glass ionomer cement
3	F	13	Painful gums and blisters	Intra-oral depapillated red patch on right side of dorsum of tongue with 3 ulcers on ventral surface of tongue White striae and plaque-like patches in lingual sulcus	Turner's syndrome Eczema	Lichen Planus in association with Turner's syndrome	Gengigel® Betnesol® mouthwash
4	F	14	Nil	Bilateral white striae on buccal mucosa	Irritable Bowel Syndrome Penicillin allergy	Lichen Planus	Nil
5	M	8	Nil	Bilateral white striae on gingivae, buccal and palatal mucosa and lateral and ventral surfaces of the tongue with erythema	VACTERL association T-cell lymphopenia Developmental delay	Lichen Planus associated with T-cell immune deficiency	Gengigel®
6	F	12	Nil	Small, reticular, white patches on buccal mucosa and retromolar area	Nil	Lichenoid tissue reaction	Nil
7	M	14	Nil	Unilateral white striae on buccal mucosa and lateral border of tongue	Growth and muscular problems related to meningococcal Septicaemia in infancy	Lichenoid tissue reaction	Replace amalgam with glass ionomer cement

Table 2. Case series of patients with oral lichen planus seen within the unit of Paediatric Dentistry in Charles Clifford Dental Hospital.

Clinical findings

Intra-oral examination revealed white striae on the buccal mucosa bilaterally. No other mucosal or skin lesions were present.

Diagnosis

Investigations included a full blood count, haematinics and biochemistry screen. The results were uniformly normal. An incisional biopsy of the right buccal mucosa was performed under local anaesthesia and the histopathology revealed a hyperplastic stratified squamous epithelium overlying a fibrovascular connective tissue with adipose tissue at the deep aspect. Thickening of the basement membrane was also seen. A definitive histopathological diagnosis of oral lichen planus was made.

Management

As the patient was asymptomatic no active treatment was provided and the patient is under periodic review.

Case five

Presenting complaint

An 8-year-old boy of South Asian ethnicity was referred by his Consultant

Paediatrician to the joint Paediatric Oral Medicine clinic regarding asymptomatic white lesions on the oral mucosa. The patient's medical history included; VACTERL association which consists of anomalies affecting Vertebral, Anal, Cardiovascular, Tracheo-Esophageal, Renal/Radial systems and the Limbs. Additionally, he had T-cell lymphopenia and developmental delay. The patient was taking Co-trimoxazole prophylaxis. He was under the care of Dermatology, Paediatric surgery, Infectious diseases and Immunology specialists at Sheffield Children's Hospital.

Clinical findings

Extra-oral examination showed papular lesions affecting the neck, trunk and legs. Intra-oral examination revealed bilateral white striations on the gingivae, buccal and palatal mucosa and the lateral and ventral surfaces of the tongue with a background of erythema (Figure 3).

Diagnosis

A skin biopsy taken of lesions on patient's back under general anaesthetic by paediatric dermatology confirmed the diagnosis of lichen planus, negating the need for oral biopsy.

Management

No treatment was required as the lesions were asymptomatic. The patient remains under regular review.

Case six

Presenting complaint

A 12-year-old Caucasian girl was referred by her dentist to the Oral Medicine Clinic at Charles Clifford Dental Hospital, with a 3-month history of an asymptomatic, white lesion on the left buccal mucosa and right retromolar region. Her medical and family histories were unremarkable.

Clinical findings

Extra-oral examination showed no skin lesions. However, the patient gave a 3-month history of previous red, itchy and dry rashes on her hands and feet. The lesions had been treated with steroid cream and were completely resolved. Intra-oral examination showed small, reticular, white patches on the left buccal mucosa and the right retromolar area.

Diagnosis

An incisional biopsy of the left buccal mucosa was undertaken and histopathology showed hyperplastic and atrophic parakeratinized stratified squamous epithelium overlying fibrous connective tissue, confirming a diagnosis of a lichenoid tissue reaction.

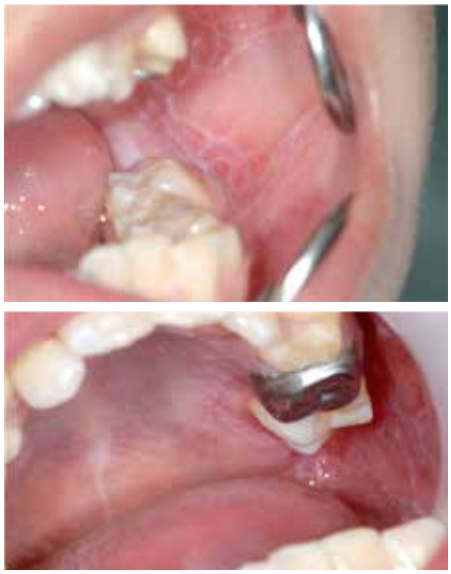

Figure 3. (a, b) Reticular oral lichen planus (Case 5).

Management

No active treatment was provided and the patient is under 6-month review.

Case seven

Presenting complaint

A 14-year-old Caucasian boy was referred by his dentist to the joint Paediatric Oral Medicine Clinic at Charles Clifford Dental Hospital regarding an asymptomatic white patch on the buccal mucosa. The medical history revealed that the patient had growth and muscular problems relating to meningococcal septicaemia in infancy. The patient's regular medications included Naproxen and Codeine pain-killers. He had no known allergies.

Clinical findings

Extra-orally no abnormalities were detected. Intra-oral examination revealed unilateral white striae affecting the buccal mucosa and lateral border of the tongue. Amalgam restorations were present on UR6 and UL6. The patient recently completed a course of orthodontic treatment with removable appliances (Figure 4).

Diagnosis

An incisional biopsy was carried out and revealed features consistent with a lichenoid tissue reaction. Patch testing for the constituents of amalgam filling material and stainless steel wire were requested. The results revealed the patient had an allergic contact reaction to mercury and vanadium.

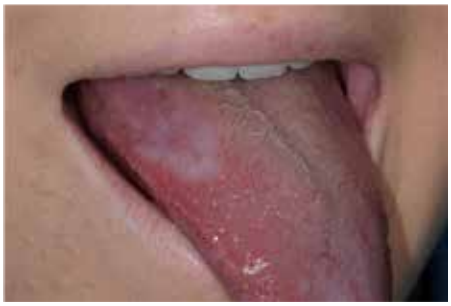

Figure 4. Plaque-like oral lichen planus of the tongue (Case 7).

Management

The amalgam restorations were replaced with glass ionomer cement.

Conclusion

Lichen planus is rare in children and oral mucosal involvement is rarer still. The cases described in this paper demonstrate the aetiopathogenesis and presentation of oral LP, highlighting that this should be considered in the differential diagnosis of oral mucosal lesions in children. The general dental practitioner may be the first healthcare professional to identify such lesions in paediatric patients and should know when to refer to a specialist for appropriate assessment and management.

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