Case Report

Oral lichen planus in an 8-year-old child: A case report with a brief literature review

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Abstract

Lichen planus (LP) is a chronic autoimmune condition of uncertain etiopathogenesis and usually affects the skin, oro-genital mucosa, nail and scalp appendages. LP is primarily seen in middle-aged individuals, and oral lesions of LP in children are relatively uncommon. Herewith, we report a case of oral LP in an 8-year-old boy, which regressed well with the treatment modality.

Keywords: Children, erosive, lichen planus, reticular

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INTRODUCTION

Lichen planus (LP) is a chronic autoimmune mucocutaneous condition, primarily affecting the oral and genital mucous membrane, skin, nails and scalp. Although, the condition has an obscure etiopathogenesis; however, an underlying immune dysfunction and multifactorial predisposing factors also play a role.[1] Oral lichen planus (OLP) is the mucosal analog of LP of the skin.[2] LP frequently affects middle age and elderly females (F:M ratio of 2:1). The estimated prevalence of OLP in the general population is 0.5%–2%.[3] However, OLP is relatively uncommon in the pediatric population with very few published cases.[4,5]

OLP in children was first described in the early 1920's. The cases reported were found mainly in the regions namely India, Africa, America, United Kingdom, Italy, Mexico and Kuwait. However, the cases reported with skin lesions mainly and rare with oral involvement.[6-8] The overall estimated prevalence of OLP in children comprises <2%–3% of the total.[6,7] The oral lesions demonstrate clinical variability as compared to their cutaneous counterpart and have been categorized as subtypes namely reticular, plaque-like, papular, erosive, atrophic and bullous.[9]

This article presents a rare case of OLP in an 8-year-old boy who responded well to the prescribed treatment.

CASE REPORT

The parents of an 8-year-old boy reported to our department with a chief complaint of oral ulcers and difficulty in eating for the past 2 years. History revealed that the parents of the patient noticed minute ulceration on the tongue and buccal mucosa 2 years back. The patient started experiencing burning sensation which aggravated while eating spicy and hot food for the past

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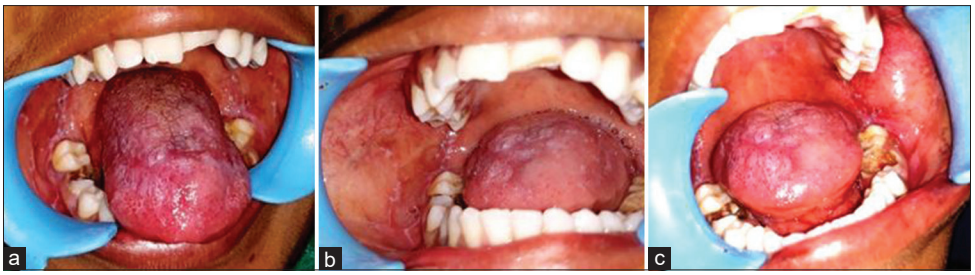

Figure 1: (a) Depapillated tongue with interspersed white striae and melanin pigmentation. (b) Slender radiating white striae on the right buccal mucosa. (c) Erosive lesion on the left retro molar region and Wickham's striae

8 months. The patient's patents had consulted local practitioners for the same, but he did not respond well to the treatment provided (no previous medical prescriptions were available). His medical and family history was nonsignificant. There was no associated history of vesicular/bullous eruptions. Physical examination did not reveal any evidence of cutaneous, genital, scalp or nail involvement. Oral examination revealed white interlacing striae (Wickham's striae) extending from commissural to the retromolar region on both sides of the buccal mucosa along the level of the occlusal plane. The lesion on the left buccal mucosa presented with a 2 cm × 3 cm erosive lesion localized in the retromolar region in relation to 37, while a reticular type variant of OLP was evident on the right buccal mucosa presenting with the peculiar slender radiating white striae (Wickham's striae). Area of depapillation interspersed with radiating slender white striae and melanin pigmentation was appreciated on the dorsum of the tongue. The lesion was flat, nontender, nonindurated and nonscrappable on palpation [Figure 1a-c]. Oral hygiene was fair without any restorations. Asymptomatic deeply carious first molars were seen bilaterally, although both the patient and the parents denied any restorative treatment done in those teeth. Based on the chronicity of the lesions and the characteristic clinical appearance, OLP, oral lichenoid reactions (OLR) and discoid lupus erythematosus (DLE) were considered as the differential diagnosis. OLR was ruled out based on a negative history of drug intake and the clinical absence of any amalgam restorations in the deeply carious teeth. The interlacing radiating white striations in DLE are much finer and subtle in contrast to OLP.

Histopathological features

After unremarkable hematological investigations and obtaining informed and written parent's consent, an incisional biopsy was taken from the perilesional left buccal mucosa region. Histopathology showed typical features of LP, i.e., acanthotic epithelium with dense band-like of lymphocytic infiltration (ruling out OLR in which infiltrates are composed of plasma cells and eosinophils) and irregular saw tooth rete pegs. There were no atypical/dysplastic changes evident histopathologically [Figure 2a and b].

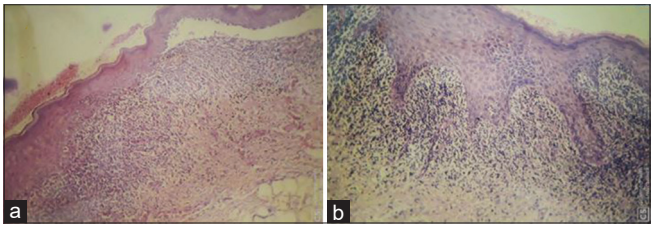

Figure 2: (a) Acanthotic epithelium with lymphocytic infiltration. (b) Saw tooth rete pegs with dense band of lymphocytic infiltration

Treatment

After meticulous oral prophylaxis, the patient was educated and motivated for oral hygiene maintenance. Root canal treatment was done for the deeply carious first molars. Based on histopathological absence of dysplastic features and taking into consideration, the patient's age, steroid mouth rinses (betnesol 0.5 g, swish and spit three to four times daily × 15 days) was prescribed to the patient. The patient was also advised to avoid the consumption of spicy food. The patient was reviewed after 15 days and the lesions showed marked resolution with steroid rinses [Figure 3a-c]. The patient was then reviewed at an interval of 1-month for consecutive 3 months and did not report any recurrence.

DISCUSSION

OLP is uncommonly encountered in children, i.e., <2%–3% with very few cases documented in the literature.[6-8] Several studies have reported the mean age of onset being 7.1–8.4 years. The youngest case has been reported in a 3-month-old child.[10] However, the reported earliest age of onset is 2-week old.[11] Handa and Sahoo reported that the lesions appear earlier in boys than in girls with the age of onset being 5–9 years of age.[12] Studies have revealed that erosive OLP in children is exceptionally uncommon.[4] Children usually do not have associated systemic and autoimmune pathologies, medications and dental restorations, and these could possibly contribute for the uncommon occurrence of childhood OLP. Furthermore, most of the childhood

Table 1: Review of literature: case reports of lichen planus in children/series

Author	Year	Number of patients	Age/sex	Site	Features	Clinical type	Cutaneous/other mucosal/skin involvement	Systemic pathologies	Diagnosis	Management	Results
Alam F and Hamburger J	2001	6	8/male	Buccal mucosa	Painful erosions/ erythematous plaques, lichenoid striae	Reticular and erosive	Attached gingival and alveolar mucosa	Congenital heart defect	OLP (reticular and erosive)	Antibiotics + chlorhexidine mouthwash	Resolved lesion after 2 months
			6/male	Dorsum of tongue	Ulceration	Reticular	NA	NA	OLP (reticular)	Symptomatic relief	Resolved lesion after 2 years
			7/male	Buccal mucosa (right and left), retromolar fossa	Swollen lips	Atrophic	Desquamative gingivitis	NA	OLP (atrophic)	Chlorhexidine gluconate and prednisolone mouthwashes	Resolved lesion after 4 years
			14/male	Buccal mucosa and tongue	Painful/burning sensation	Lichenoid reaction	NA	Asthma (salbutamol and beclomethasone inhaler)	Lichenoid reaction	No active treatment	Resolved lesion after 2 years
			14/male	Buccal mucosa	Asymptomatic	Atrophic	NA	NA	OLP	No active treatment	Patient kept under review
			11/male	Buccal mucosa and tongue	Asymptomatic	Reticular (buccal mucosa, dorsum of the tongue) and papular (lateral border of the tongue)	NA	NA	OLP	No active treatment	Patient kept under review
Neena *et al.*	2015	1	9/male	Buccal mucosa	Itching and burning sensation	Reticular	NA	NA	OLP	Topical corticosteroid gel	Resolved lesion after 3 months
Chaitra TR *et al.*	2012	1	9/female	Buccal mucosa	Burning sensation	Erosive	NA	NA	OLP	Topical corticosteroid gel and Analgesics	Regressed lesion after 1 week
Chiyadu Padmini *et al.*	2013	1	12/male	Dorsum of tongue	Ulceration and burning sensation	Erosive	NA	NA	OLP	Topical corticosteroid gel, antifungal and anaesthetics	Resolved lesion after 1 month
Usha Mohan Das and Beena JP	2009	1	12/female	Buccal mucosa	Burning sensation and pigmentation			NA	OLP	Topical tretinoin	Patient under follow up
S Patel *et al.*	2005	3	15/female	Dorsal and ventral surface of the tongue, floor of the mouth	Ulceration and erythema	Erosive	Cutaneous lesions on the neck and upper trunk	Idiopathic hypothyroidism	OLP	Topical prednisolone mouthwash followed by beclamethasone spray	Patient under follow up
			6/male	Dorsum of tongue	White patch	LP	NA	Austism	OLP	No active treatment required	Kept under periodic follow-ups
			9/female	Buccal mucosa, dorsum and lateral border of the tongue, floor of the mouth	Soreness	Reticular	NA	Mitral valve atresia/cardiac transplant awaited	OLP	Topical beclamethasone spray when symptomatic	Resolved lesion. Kept under Periodic follow-ups
GunaShekhar M	2010	1	7/male	Buccal mucosa, lateral border of the tongue, floor of the mouth, upper and lower lip	Burning sensation on taking spicy food	Reticular	NA	NA	OLP	Topical corticosteroid cream 0.1% triamcinolone acetonide (kenacort)	Resolved lesion after 3 months. periodic follow-ups
Sharma *et al.*	2017	1	12/female	Gingiva and buccal vestibule (bilateral)	Burning sensation on taking spicy food		NA	NA	OLP	Topical corticosteroid cream (0.1%) triamcinolone acetonide followed by topical retinoids	Kept under periodic follow-ups

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Table 1: Contd...

Author	Year	Number of patients	Age/sex	Site	Features	Clinical type	Cutaneous/other mucosal/skin involvement	Systemic pathologies	Diagnosis	Management	Results
George S	2015	2	8/male	Labial mucosa and buccal mucosa, ventral tongue	Pain and burning sensation on taking spicy food		NA	Vaccination gainst Japanese encephalitis	OLP	Topical antifungal mouth paint -clotrimazole	Resolved lesion after 2 weeks
			8/male	Buccal mucosa	Burning sensation on taking spicy food		NA	NA	OLP	Incisional biopsy performed	Lesion subsided after biopsy Kept under periodic follow-ups
Moger *et al*.	2013	1	7/female	Lesion labial extending commissure to pterygomandibular region involving buccal mucosa, buccal vestibule, , retromolar region	Burning sensation on taking hot and spicy food	Erosive	Cutaneous lesions were present (skin of back and hands)	NA	OLP with cutaneous involvement	Topical 0.1% triamcinolone acetonide combined with 1% clotrimazole. topical anesthetic was given for palliation Corticosteroids for cutaneous lesions as directed by dermatologist	Resolved oral and cutaneous lesions after 1 month Kept under Periodic follow-ups
Cascone M *et al*.	2017	8 (4 males and 4 females)	Mean age-13.5±2.73 years	Tongue affected in 6 patients	Oral burning sensations	Reticular in 6 cases	NA	NA	NA	NA	NA
Pandhi D	2014	316 (166 boys, 150 girls)	10.28 years (range 2-14 years)	NA	NA	Reticular in 54% cases	Skin involvement in 96% cases	NA	NA	Topical and systemic steroids+dapsone	Excellent response in 28.8%
Moreas D	2011	1	7-year old girl	Upper lip	Oral burning sensations		NA	NA	OLP	Topical and intralesional steroids	Resolution of lip lesion Asymptomatic lichenoid lesion after 3 years follow up
Laeijendecker R	2005	3	11 years girl 16 years boy 14 years girl	Oral cavity	NA	Hyperkeratotic variant 11 years girl Erosive OLP in 16 years boy Hyperkeratotic in 14 years girl	NA	NA	OLP	NA	NA
Basak PY	2002	1	9-year old boy	Oral mucosa	NA	NA	Nail involvement	NA	OLP	Dapsone	NA
Nanda A	2001	23	52% boys and 48% girls	39% of cases in Oral mucosa	NA	NA	NA	NA	NA	Topical steroids were the mainstay of treatment, also dapsone, UVB phototherapy	NA
Scully C *et al*.	1994	3	Young girls	Buccal and lingual mucosa	NA	NA	NA	NA	OLP	Topical and intralesional steroids	Responded well to treatment
Khandelwal *et al*.	2013	1	10-year-old girl	Buccal mucosa and retromolar region	Oral burning sensations	Reticular OLP	NA	NA	OLP	Topical steroids	NA
Ravi Kiran PS *et al*.	2017	76	Mean age - 10.7 years: 42 boys and 34 girls	Oral mucosal involvement in 14.4% cases	NA	NA	Limbs mostly affected; palmo-plantar, and nail involvement in 7.8%, and 15.7%	NA	NA	NA	NA
Kumar A	2018	42	Mean age - 11.6±5.1 years 26 girls and 16 boys	Oral mucosal involvement in 28.6%	NA	NA	Skin LP in 69% cases; nail in 42.85%, and scalp in 7.1% cases	NA	NA	NA	NA

Contd...

Table 1: Contd...

Author	Year	Number of patients	Age/sex	Site	Features	Clinical type	Cutaneous/other mucosal/skin involvement	Systemic pathologies	Diagnosis	Management	Results
Juhi Jahan S	2017	1	7-year-old boy	Buccal mucosa bilaterally	Oral burning sensations	Erosive OLP	NA	NA	OLP	Topical steroids along with topical tacrolimus and anesthetic	40%-50% resolution after 1 week; 80% after 2 weeks and completely healed after 2 months follow up
Hugar MS *et al.*	2015	1	9-year-old girl	Buccal mucosa bilaterally	Asymptomatic	Reticular OLP	White mucocutaneous Patch on the left arm	NA	OLP with cutaneous involvement	Topical steroids + multivitamins	Completely healed after 4 months follow up
Chatterjee K *et al.*	2012	22	Mean age - 15.18; 11 males and 11 females	Buccal mucosa (50%) most common site	NA	Erosive OLP	NA	NA	OLP	NA	NA
Gopal KS	2016	1	6-year -old female	Dorsum of the tongue and buccal mucosa	Ulcerations on the tongue	Erosive OLP	Papular lesions on the wrist, ankle, and knee	NA	OLP	Topical steroids	NA
Kelner N	2012	1	12-year-old girl	Tongue and buccal mucosa	Asymptomatic	Reticular OLP	NA	NA	OLP	No active treatment	Regular follow up
Reddy *et al.*	2014	1	13-year-old boy	Gingival and buccal mucosa	Oral burning sensations	Atrophic LP	NA	NA	OLP	Topical steroids	NA
Morankar R	2016	1	5-year-old boy	Buccal mucosa and tongue	Oral burning sensations	Ulcerative LP	NA	NA	Ulcerative OLP	Topical and systemic steroids	Significant improvement in 6 months
Kapse CS *et al.*	2018	3	12-year-old boy	Labial and buccal mucosa	Oral burning sensations	NA	Skin lesions at the neck and below the navel regions	NA	NA	Topical steroids	1 year 16 months follow up
			11-year-old boy	Dorsum of the tongue	Oral burning sensations	NA	NA	NA	OLP	Topical steroids+multivitamins	NA
			11-year-old boy	Buccal mucosa	Oral burning sensations	NA	NA	NA	OLP	Topical steroids	NA

LP: Lichen planus, OLP: Oral LP, NA: Not available, UVB: UltraViolet light B

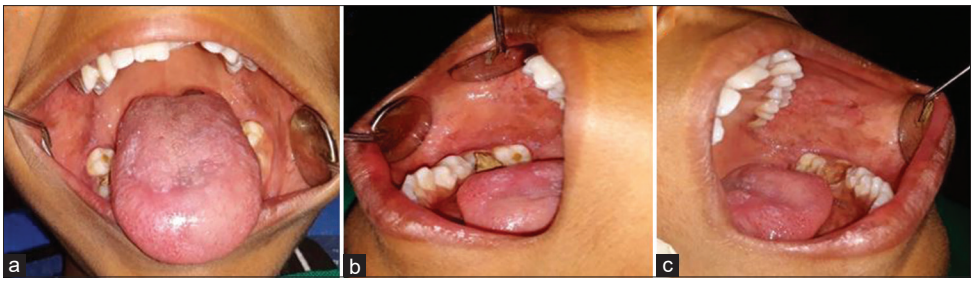

Figure 3: (a) Marked resolution in tongue lesions. (b) Completely resolved lesion on the right buccal mucosa. (c) Completely resolved lesion on the left buccal mucosa

OLP are asymptomatic, another possible reason for the misdiagnosis by the practitioner.[13] Most studies on childhood OLP have shown female predilection, although, in some studies, no significant gender predominance was identified.[14]

According to previous studies, most of the pediatric patients had reticular OLP. But according to few studies, the most frequent clinical form of OLP was the erosive type which is a rare finding in the pediatric population.[11,15]

The exact etiology remains obscure as the condition is complex and multifactorial. In the majority of cases, the condition may be idiopathic, whereas in others, a range of dental materials and medications may serve as a predisposing factor. Viruses, genetic factors, and lifestyle are the other noteworthy causative agents.[16]

In the present case, there was no contributory drug/restoration history; neither there was any associated systemic or family history.

Characteristically bilateral, symmetrical presentation of fine, interlacing reticular pattern is essential for a clinical diagnosis of OLP. A biopsy is helpful not only for the confirmation of the tentative clinical diagnosis but also empowers to rule out cellular atypia and malignant transformation.[17]

The patient presented with the classic bilateral symmetrical appearance of OLP. Erosive and reticular lesions were seen on the left and right buccal mucosa and the dorsum of the tongue, respectively, in the patient.

Acanthotic epithelium with dense band of lymphocytic infiltration and irregular saw tooth rete pegs were seen histologically in the present case.

Currently, the treatment protocol aims at minimizing the mucosal inflammation and ulcerations and resolution of the symptoms possibly enhances the disease remission period.[18] Topical corticosteroids are primarily used as the treatment modality for erosive OLP; however, few cases may also require therapy with systemic and intralesional steroids.[1]

Low-potency topical steroid application (kenacort 0.1% paste three to four times daily) along with chewable Vitamin C (tablet celine BD daily) was prescribed to our patient and was reviewed after a month.

Childhood OLP usually has a much fairer prognosis and responds well with therapy. This is contrast to in OLP in adults, which usually exhibits chronicity despite rigorous therapy and meticulous exploration of predisposing factors. In general, 0.07%–5% cases of erosive OLP in adults undergo malignant transformation. However, childhood OLP case with malignant transformation has yet not been reported.[19]

Table 1 summarizes few reported cases/case series of childhood OLP.

CONCLUSION

Childhood OLP is an extremely uncommon occurrence. Majority of the childhood OLP cases are not reported due to misdiagnosis by the physicians. Any mucosal lesion in children should be referred to the specialist for an early and precise diagnosis and treatment protocol. General childhood OLP usually has a much fairer prognosis and responds well with therapy.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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