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

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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 2 years.

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. Here, 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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8 months. The patient’s parents 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].

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.[9] However, the reported earliest age of onset is 2-week old.[10] 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.[11] Studies have revealed that erosive OLP in children is exceptionally uncommon.[12] 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
| Author          | Year | Number of patients | Age/sex | Site                                      | Features                                           | Clinical type       | Cutaneous/other mucosal/ skin involvement | Systemic pathologies | Diagnosis | Management                                      | Results                        |
|-----------------|------|--------------------|---------|------------------------------------------|---------------------------------------------------|---------------------|------------------------------------------|-----------------------|-----------|------------------------------------------------|--------------------------------|
| Alam F and      | 2001 | 6                  | 8/male  | Buccal mucosa                            | Painful erosions/erythematous plaques, lichenoid striae | Reticular and erosive | Attached gingival and alveolar mucosa | Congenital heart defect | OLP       | Antibiotics + chlorhexidine mouthwash          | Resolved lesion after 2 months |
| Hamburger J     |      |                    |         | Dorsum of tongue                         | Ulceration                                         | Reticular          | NA                                       | NA                    | OLP       | Symptomatic relief                             | Resolved lesion after 2 years |
| Chaitra TR et   | 2012 | 1                  | 9/female| Buccal mucosa                            | Burning sensation                                  | Erosive            | NA                                       | NA                    | OLP       | Topical corticosteroid gel, Analgesics         | Regressed lesion after 1 week |
| Padmini et al.  |      |                    |         | Dorsum of tongue                         | Ulceration and burning sensation                   | Erosive            | NA                                       | NA                    | OLP       | Topical corticosteroid gel, antifungal and anesthetics | Resolved lesion after 1 month |
| 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 |
| Chiyadu Padmini | 2013 | 1                  | 12/male | Dorsum of tongue                         | Burning sensation and pigmentation                 | NA                 | NA                                       | NA                    | OLP       | Topical tretinoin                               | Patient under follow up        |
| Usha Mohan      | 2009 | 1                  | 12/female| Buccal mucosa                           | Burning sensation                                  | Erosive            | NA                                       | NA                    | OLP       | Topical corticosteroid gel                     | Kept under periodic follow-ups |
| Das and Beena JP|      |                    |         | Dorsoventral surface of the tongue, floor of the mouth | Ulceration and erythema                            | Erosive            | NA                                       | NA                    | OLP       | Topical prednisolone mouthwash followed by beclamethasone spray | Patient under follow up        |
| S Patel et al.  | 2005 | 3                  | 15/female| Dorsoventral surface of the tongue, floor of the mouth | Ulceration and erythema                            | Erosive            | NA                                       | NA                    | OLP       | Topical corticosteroid gel                     | Resolved lesion after 3 months |
| GunaShekhar M   | 2010 | 1                  | 7/male  | Dorsum of tongue                         | White patch                                        | LP                  | NA                                       | Austism               | OLP       | Topical corticosteroid cream 0.1%              | Kept under periodic follow-ups |
| Sharma et al.   | 2017 | 1                  | 12/female| Gingiva and buccal vestibule (bilateral) | Burning sensation on taking spicy food             | Reticular          | NA                                       | NA                    | OLP       | Topical corticosteroid cream (0.1%)            | Kept under periodic follow-ups |

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                                                | NA                                     | Vaccination against Japanese encephalitis | NA                             | OLP                            | Topical antifungal mouth paint – clotrimazole                                | Resolved lesion after 2 weeks                                                 |
| 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                                           | NA                                     | OLP with cutaneous involvement | NA                             | NA                             | Topical 0.1% triamcinolone acetonide combined with 1% clotrimazole, topical anesthetic was given for palliation Corticosteroids for cutaneous lesions as directed by dermatologist | Lesion subsided after biopsy Kept under periodic follow-ups Resolved oral and cutaneous lesions after 1 month Kept under Periodic follow-ups |
| Cascone M et al.| 2017 | 8                  | Mean age 13.5±2.73 years | Tongue affected in 6 patients                                      | Oral burning sensations                                                  | Reticular in 6 cases                              | NA                                     | NA                                   | NA                             | NA                             | NA                             | NA                                                                                     |
| Pandhi D        | 2014 | 1                  | NA      | 10.28 years (range 2-14 years)                                      | NA                                                                      | Reticular in 54% cases                             | Skin involvement in 96% cases                   | NA                                   | Topical and systemic steroids+dapsone | Excellent response in 28.8% Resolution of lip lesion Asymptomatic lichenoid lesion after 3 years follow up | NA                                                                                     |
| Moreas D        | 2011 | 1                  | 7-year old girl | Upper lip                                                                  | Oral burning sensations                                                  | NA                                                 | NA                                     | OLP                                   | NA                             | Topical and intralesional steroids | NA                                                                                     |
| Laeijendecker R | 2005 | 3                  | 11 years girl | Oral cavity                                                              | NA                                                                      | Hyperkeratotic variant 11 years girl Hyperkeratotic in 14 years boy       | NA                                     | NA                                   | NA                             | NA                             | NA                             | NA                                                                                     |
| Basak PY        | 2002 | 1                  | 9-year old boy | Oral mucosa 39% of cases in Oral mucosa                                | NA                                                                      | Nail involvement                                  | NA                                     | NA                                   | NA                             | OLP                            | Dapsone                        | Topical steroids were the mainstay of treatment, also dapsone, UVB phototherapy Responded well to treatment | NA                                                                                     |
| Nanda A         | 2001 | 23                 | 52% boys and 48% girls | Oral mucosa                                                              | NA                                                                      | Nail involvement                                  | NA                                     | NA                                   | NA                             | OLP                            | NA                             | NA                                                                                     |
| Scully C et al. | 1994 | 3                  | Young girls | Buccal and lingual mucosa                                               | NA                                                                      | NA                                                 | NA                                     | NA                                   | OLP                            | NA                             | NA                             | NA                                                                                     |
| Khandelwal et al.| 2013 | 1                  | 10-year-old girl | Buccal mucosa and retromolar region                                 | Oral burning sensations                                                 | Reticular OLP                                     | NA                                     | NA                                   | OLP                            | NA                             | NA                             | 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                                                                      | Limbs mostly affected; palmo-planter, and nail involvement in 7.8%, and 15.7% cases | NA                                     | NA                                   | 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                                                                      | Skin LP in 69% cases; nail in 42.85%, and scalp in 7.1% cases              | NA                                     | NA                                   | NA                             | NA                             | NA                             | NA                                                                                     |
| 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         | NA                                      | 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 6-year-old female | Buccal mucosa (50%) most common site Dorsum of the tongue and buccal mucosa Tongue and buccal mucosa | NA | Erosive OLP | NA | NA | OLP with skin involvement | Topical steroids | NA |
| Gopal KS              | 2016 | 1                  | 5-year-old girl  | Buccal mucosa and tongue         | Asymptomatic                  | Reticular OLP         | NA                                      | NA                   | OLP       | No active treatment                                   | Regular follow up                                          |
| Kelner N              | 2012 | 1                  | 12-year-old girl | Buccal mucosa and tongue         | Oral burning sensations       | Atrophic LP           | NA                                      | NA                   | OLP       | Topical steroids                                   | NA                                                          |
| Reddy et al.          | 2014 | 1                  | 13-year-old boy  | Gingival and buccal mucosa       | Oral burning sensations       | Ulcerative OLP        | NA                                      | NA                   | OLP       | Topical steroids                                   | NA                                                          |
| Morankar R            | 2016 | 1                  | 11-year-old boy  | Labial and buccal mucosa         | Oral burning sensations       | Ulcerative OLP        | NA                                      | NA                   | OLP       | Topical 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                   | OLP       | Topical steroids                                   | 1 year 16 months follow up                                  |
|                       |      |                    | 11-year-old boy   | Dorsum of the tongue Buccal mucosa | Oral burning sensations       | NA                      | NA                                      | NA                   | OLP       | Topical steroids + multivitamins                   | NA                                                          |
|                       |      |                    | 11-year-old boy   | Dorsum of the tongue 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
OLP are asymptomatic, another possible reason for the misdiagnosis by the practitioner. Most studies on childhood OLP have shown female predilection, although, in some studies, no significant gender predominance was identified.

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.

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.

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.

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. Topical corticosteroids are primarily used as the treatment modality for erosive OLP; however, few cases may also require therapy with systemic and intralesional steroids.

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.

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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