ORIGINAL ARTICLE

A retrospective study of oral lichen planus in paediatric population

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ABSTRACT

Aim: Well documented cases of oral lichen planus, a cell mediated immune condition is infrequently reported in paediatric population. This study was undertaken to obtain epidemiological data retrospectively and also to explore the possibility of any association that might exist among the clinical and histopathological features in paediatric patients suffering from oral lichen planus. **Subjects and Methods:** A retrospective study was carried out on 22 patients, younger than 18 years with clinical and histopathological diagnosis of oral lichen planus over a period of 14 years. The clinical characteristics and histopathological features were observed. The statistical analysis of the data was performed using Statistical Analysis Software (SAS), Version 9.1. **Results:** Analysis of data of 22 patients revealed that the average age of patient is 15.18 years with equal male and female predilection. The most common site is buccal mucosa (50%) and most frequent clinical form is erosive (63.64%). Focusing on the histopathological findings, parakeratosis was found in 86.36% of the cases, acanthosis in 63.64% of cases, moderate basal cell degeneration was identified in 63.64% of cases and dense lymphocytic infiltration at juxtaepithelial connective tissue region was found in 59.09% of cases. **Conclusions:** Oral lichen planus in paediatric population is rare and appeared between 8 to 18 years of age. There is no significant gender predominance. The most common clinical form is erosive, manifesting mainly in buccal mucosa. Histopathological findings characteristic of oral lichen planus in paediatric patients include parakeratosis, acanthosis, liquefaction degeneration of basal cells and lymphocytic infiltration in the subepithelial layer.

Key words: Basal cell degeneration, lichen planus, lichenoid reaction, pediatric

INTRODUCTION

Lichen planus (LP) is a chronic inflammatory, autoimmune disease, affecting a wide variety of sites, including skin and mucous membranes.[1-3] In contrast to dermal LP, oral lichen planus (OLP) demonstrates clinical variability. The prevalence of OLP ranges between 0.5% and 3% and may occur in 70% to 77% of patients with cutaneous LP. However, some studies report that OLP, without cutaneous involvement occurs more commonly.[4]

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It is currently considered a disease of unknown aetiology and with multifactorial pathogenesis. Most studies suggest that LP is a CD8+ T cell-mediated autoimmune disease. It is believed that the CD8+ T lymphocytes induce keratinocyte apoptosis and cause epithelial basal cell layer damage.[2,4] Possible causes of OLP such as an allergy to dental restorative materials (amalgam, gold), local trauma (Koebner phenomenon) and hepatitis B or C virus infection have been reported.[5] Moreover, genetic factors, lifestyle and emotional stress may be contributing factors in its pathogenesis. Many drugs are capable of producing a lichenoid drug eruption.[5]

OLP is most frequently manifested during the fifth and sixth decades of life.[2] It is rare in children, with the youngest case documented in a child aged 3 months.[4] Paucity of reported cases of juvenile OLP may be due to lack of patient and parent awareness of lesions, lack of recognition by practitioners, low incidence of autoimmune diseases and precipitating

factors such as stress.[3] Of all the cases of OLP reported in children, there is a higher prevalence in the Indian population suggesting probable differences in the genetic background and / or environmental triggers.[4] In adults, women are affected more commonly than men.[4] Some researchers have found an equal sex distribution in children.[6] Although OLP is usually a sporadic disorder there is also a familial form. Familial OLP is very rare but more prevalent in paediatric population. It is usually a disseminated type of LP having a prolonged course with relapses.[6]

OLP commonly affects buccal mucosa, although tongue, gingiva and palatal mucosa may also be affected.[2] In both adults and children cutaneous LP is characterised by extremely pruritic eruption of flat-topped, polygonal and violaceous papules with overlying fine reticular striations known as Wickham Striae.[6] OLP presents in mainly three different clinical forms. In order of advancing severity and symptomatology, they are reticular, plaque and erosive types.[7] Reticular OLP typically presents as asymptomatic, greyish white, lacy, interlacing striae. The plaque form is characterised by slightly elevated, irregular white patch while the erosive form is erythematous and frequently painful[3,7]. Histopathology of OLP reveals hyperkeratosis, acanthosis, liquefaction degeneration of basal cells and existence of a band of lymphocytic infiltrate in close proximity to surface epithelium.[3,7,8]

The objective of treating lichen planus is to control the episodic outbreaks that occurs, given that the lesions are usually not completely cured.[2] Reticular and plaque forms usually do not require treatment other than reassurance and follow up. The mild cases of erosive lichen planus are treated with topical corticosteroids combined with antifungal agent. In cases where such treatment proves to be inefficient, intralesional injection of corticosteroids may be prescribed. Most erosive LP requires systemic corticosteroid regimen.[2,9,10] Evolution of LP varies according to the clinical type. The reticular lesions cure spontaneously in 40% of the cases. Plaque and erosive lesions do not usually disappear and tend to recur in the majority of the cases.[2]

The objective of this article is to highlight the most characteristic histopathological findings of OLP in paediatric population and to explore their correlation with the most frequent clinical manifestations and forms in the sample.

SUBJECTS AND METHODS

A retrospective, nonrandomized study was carried out on 22 patients, over a period of 14 years, from 1997 to 2010, at a private oral and maxillofacial pathology clinic in Kolkata, India. The diagnosis of OLP was based on history, clinical findings and histopathological features. Inclusion criteria were an age younger than 18 years and without any history of drug allergy or contact allergy. Patients with amalgam restorations were also excluded from this study. Investigations for possible signs of extraoral lichen planus, routine blood examination including hepatitis B and C serology were performed.

The clinical characteristics such as age, sex, topography, symptoms, clinical pattern of lesion, family history, preexisting medical conditions were documented. Punch biopsy was taken from most representative area of the lesion of each patient. Specimens from four patients having extensive erosive lesions were sent for direct immunofluorescence. Histopathologic features were observed at epithelial level, at epithelial connective tissue interface and at connective tissue level. Histopathologic criteria for OLP consisted of type and degree of keratosis, thickness of epithelium (acanthosis or atrophy), type of rete peg (wavy, saw tooth or bulbous), degree of liquefaction degeneration of basal cells (mild or moderate) and density of lymphocytic infiltration (mild, moderate or severe) at subepithelial lamina propria. Patients having reticular, pigmented and hypertrophic form of OLP were treated with topical corticosteroid or tacrolimus ointment. 13 cases with erosive form of OLP were treated with systemic corticosteroids in tapering dosage. Only one patient having extensive erosive OLP with repeated recurrence was treated with intralesional corticosteroid.

The statistical analysis of the data was performed using the Statistical Analysis Software (SAS), Version 9.1. Fisher's exact Test was employed to explore the association between the clinical pattern and histopathological features. The results obtained are considered significant and highly significant when *P* values were less than 0.05 and 0.01 respectively.

RESULTS

We analysed a total 22 patients, which included 11 males (50%) and 11 females (50%). The average age of the patients was 15.18 years ranging from 8 years to 18 years. The mean duration of OLP in our cases prior to seeking consultations was about 14 months. The mean follow up period after diagnosis of OLP was three years. The most common site was buccal mucosa. 11 patients (50%) revealed OLP of buccal mucosa. five patients (22.73%) had OLP of tongue, three patients (13.64%) had lesions both on the buccal mucosa and tongue, two cases (9.09%) were on both buccal mucosa and lip and only one patient (4.05%) was affected on buccal mucosa, tongue and lip. The most significant finding while comparing the site with age and sex [Figure 1], we observed that seven male patients(63.63%) and four female patient (36.36%) had OLP on buccal mucosa. Out of seven male patients, one was from 8-9 years age group, four patients in 13-15 years age group and two patients in 16-18 years age group. Out of four females all are from 16-18 years age group. In case of tongue lesions three patients were from 16-18 age group. Combined lesions occurred more in females belonging to older age group.

The most frequent clinical form of OLP was erosive type

which manifested in 14 patients (63.64%), followed by hypertrophic type, found in five patients (22.73%). Reticular type was present in two patients (9.09%) and only one case (4.05%) of pigmented variety was reported. Comparing clinical type with site [Figure 2] it is evident that erosive type [Figure 3] exclusively occurred in buccal mucosa of nine patients (64.29%), buccal mucosa and tongue in two patients (14.29%), lip and buccal mucosa in two patients (14.29%) and only tongue in one patient (7.14%). The hypertrophic (plaque) type was found mostly on tongue, being reported in eight patients (80%) and concomitantly in lip, tongue and buccal mucosa in one case (20%). The reticular type is evenly distributed over the sites buccal mucosa [Figure 4] and both buccal mucosa and tongue, being reported in one patient each case. Cutaneous involvement was noted in two patients. Routine hemogram was within normal limits for all the cases and serological testing for Hepatitis B and C were not significant.

Histopathologically [Figures 5 and 6] we observed that parakeratosis was found in 19 cases (86.36%) and orthokeratosis in three cases (13.64%). Acanthotic epithelium was found in 14 cases (63.64%) and atrophic epithelium in eight cases (36.36%). Wavy rete peg was most common and was found in 14 cases (63.64%). Saw tooth rete peg was present in six patients (27.27%) and bulbous rete peg in two cases (9.09%). Moderate basal cell degeneration was found in 14 cases (63.64%) and mild basal cell degeneration in eight cases (36.36%). Severe inflammatory infiltrate at subepithelial layer was detected in 13 cases (59.09%), moderate infiltration in three cases (13.64%) and mild in 6 cases (27.27%). Direct immunofluoroscence of lesional tissues performed in four equivocal cases were negative for any vesicullobulous lesions.

A cross tabulation of histopathological findings and clinical forms was performed [Table 1]. We observed orthokeratosis was found in a total three patients of which two were of erosive form and one of reticular form. Parakeratosis was found in 19 patients of whom 12 were from erosive type. Atrophic epithelium was found in eight patients of which five belonged to erosive variety, two were hypertrophic variety and one was reticular variety. Acanthotic epithelium was identified in a total 14 patients of which nine were of erosive form, three of hypertrophic form, one of pigmented form and one of reticular form. Wavy rete pegs were evident in 14 sections in which ten were erosive type, three hypertrophic form and one reticular form. Saw tooth rete peg was present in six sections (27.27%), of which three were of erosive form, one of the hypertrophic form, one of pigmented

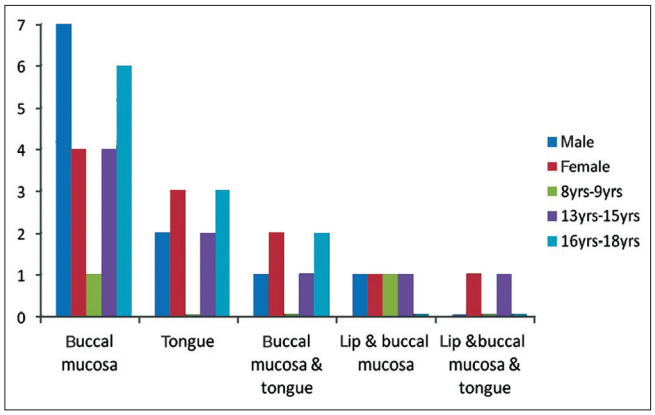

Figure 1: Comparing site with age and sex of oral lichen planus

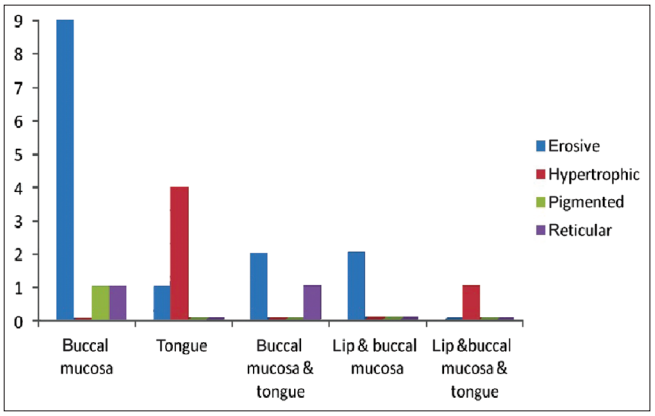

Figure 2: Comparing clinical form and site of oral lichen planus

Table 1: Percentage distribution of histological parameters and in various clinical forms of lichen planus

Histological findings	Clinical forms			
	Erosive %	*Hypertrophic %*	*Pigmented %*	*Reticular %*
Orthokeratosis	66.67	0	0	33.33
Parakeratosis	63.16	26.32	5.26	5.26
Atrophic epithelium	62.50	25	0	12.5
Acanthotic epithelium	64.29	21.43	7.14	7.14
Wavy rete-pegs	71.43	21.43	0	7.14
Bulbous rete-pegs	50	50	0	0
Saw tooth rete-pegs	50	16.67	16.67	16.67
Mild basal cell degeneration	62.50	25	0	12.5
Moderate basal cell degeneration	64.29	21.43	7.14	7.14
Severe infiltrate density	53.85	38.46	7.69	0
Mild infiltrate density	83.33	0	0	16.67
Moderate infiltrate density	66.67	0	0	33.33

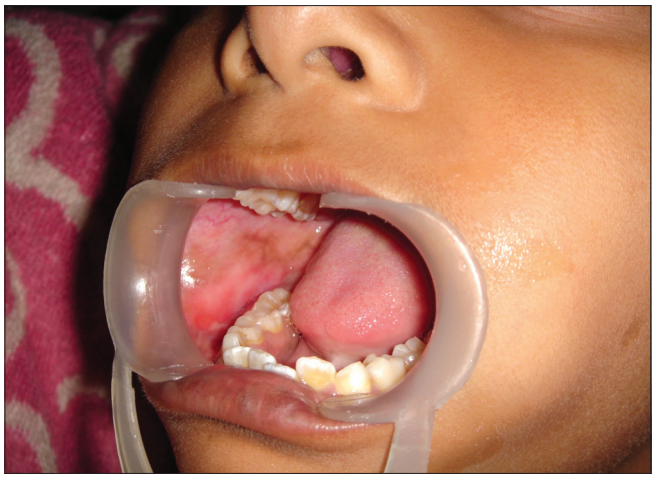

Figure 3: Clinical photograph showing erosive lichen of right buccal mucosa in a 9 year old male child

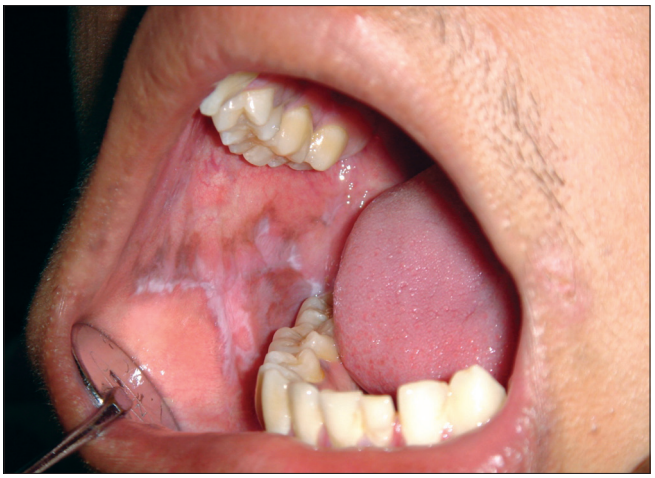

Figure 4: Clinical photograph showing reticular lichen planus of right buccal mucosa in a 14 year old male patient

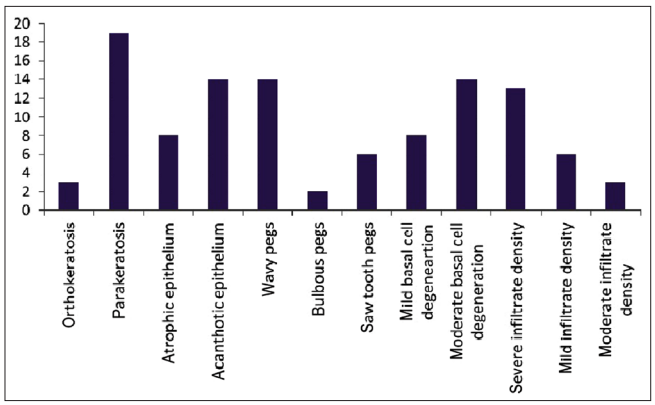

Figure 5: Distribution of histological features seen in pediatric oral lichen planus

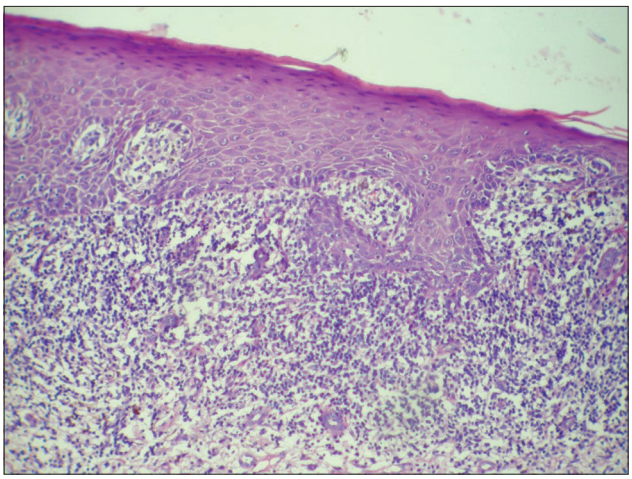

Figure 6: Photomicrograph of oral lichen planus showing basal cell degeneration and dense lymphocytic infiltration at subepithelial connective tissue.(H and E stain, 10× magnification)

form and one of the reticular form. Mild basal cell degeneration was identified in eight patients (36.36%) of which five were of erosive variant. Moderate basal cell degeneration was found in 14 patients (63.64%), nine of which were of erosive form, three hypertrophic form, one pigmented form and one reticular form. Regarding subepithelial infiltrate density, severe infiltration was identified in 13 cases (59.09%), mild in six cases (27.27%) and moderate in three cases (13.64%).

Fisher's exact test was employed to assess the relationship between each of the histopathological findings and clinical forms. The study revealed a significant relationship between the clinical type and site of occurrence (P=0.0112). The erosive type was mostly identified in buccal mucosa (64.29%). The hypertrophic type of OLP was found mostly (80%) on tongue. However no case of hypertrophic OLP was found on buccal mucosa of paediatric patients. While comparing clinical form with histopathological findings of OLP in pediatric patients we found that parakeratosis was most frequent type of keratosis which occurred in 63.16% of erosive lesion. The erosive variety revealed acanthotic epithelium in 64.29% patients. The rete pegs were predominantly of wavy pattern. Significant basal cell degeneration and band like subepithelial lymphocytic infiltration were present in all cases. However moderate basal cell degeneration and mild subepithelial infiltrate density were the common findings.

DISCUSSION

Oral lichen planus is extremely rare in childhood with very few cases cited in the literature. It is suggested that childhood LP is more common in tropics.[5] 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.[11] Kumar *et al.*[12] reported involvement of the oral mucosa in only one of 25 children with cutaneous lesions. However, Sharma and Maheswari[13] reported 50 children with LP and with concomitant oral lesions in 15 of them. Generally the oral mucosa seems to be less commonly involved in children with LP than in adults.[14]

In our study, out of 22 cases of OLP only two patients had concomitant cutaneous involvement.

Several studies concluded that patients with LP have a higher risk of being HCV seropositive and similarly, HCV-infected patients had a higher probability of developing LP.[4,5] According to Tilly *et al.*[6] there is no increase in hepatitis C among Indian childhood LP cases studied to date. Serological testing for hepatitis B and C were also not significant in our cases. According to some studies[1,12,13] predominance of LP was from five to 15 years of age. In our study the average age of patient was 15.18 years. Kumar *et al.*[12] observed more female predominance in his series while male predominated in the studies of Sharma and Maheswari and Woo *et al.*[3]

Some researchers[6,14] have found an equal sex distribution in children and this observation is consistent with our study. In most of the literature buccal mucosa was the most commonly affected site[3,5] with the next most common location being tongue. Our study was also corroborative to the finding as 50% of the cases had involvement of buccal mucosa and 22.73% of cases had OLP in tongue. According to previous studies, most of the paediatric patients[3,10] had reticular OLP. But we found most frequent clinical form of OLP was erosive type which is a rare finding in paediatric population. Focusing on histological findings our study revealed parakeratosis in 86.36% and acanthosis in 63.64% of cases. This data was consistent with that found in the literature.[2,7] We observed wavy rete peg as most common (63.64%) type of rete peg and that was described by other authors as well.[2,7,8] Liquefaction degeneration of basal layer of epithelium and the band-like subepithelial lymhocytic infiltrate were present in all cases and these were corroborative to observations by other authors.[1-3] Most of the authors[3,5] are of opinion that prognosis and effect of treatment of OLP in children seems to be more favourable than OLP in adults. In conclusion, oral lichen planus in childhood is rare. It is more prevalent in 15-16 years of age with no gender predilection. The most common type is erosive form, primarily manifesting in buccal mucosa. Topical corticosteroid therapy and a plaque control regimen in children with symptomatic OLP have shown favourable responses. The use of topical calcineurin inhibitors like tacrolimus is not recommended in patients below 2 years. Safety of long term continuous use of these drugs in pediatric patients has not been adequately evaluated. Although Laeijendecker *et al.*[5] reported no OLP-related malignancies to date in the pediatric population, the schedule of follow up of pediatric OLP should be atleast one or two visits per year as long as OLP in children persists. Paedodontists must be aware of its clinical presentation, diagnosis and management and periodic follow-up visits should be emphasized to control possible recurrence of the disease.

CONCLUSION

Oral lichen planus is rare in pediatric population. This paper contributes 22 pediatric patients of oral lichen planus to the literature. The patients were diagnosed following the WHO diagnostic criteria (1978) of OLP both clinically and histologically. The paper also reveals some significant association between the clinical forms and histopathological findings of OLP. Most often mucosal lesions in pediatric patients are misdiagnosed by practitioners. A better understanding of different clinical forms of oral lichen planus in children would help the pediatric dentists to make an early diagnosis and management of the lesion.

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