

CASE REPORT

Lichen Planus In A Three Year Old - A Case Report

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ABSTRACT

Introduction: Lichen planus is a very common chronic inflammatory mucocutaneous disorder and the etiopathology of this disease remains obscure. Lichen planus is usually seen to occur commonly in the middle aged and postmenopausal females where as a few cases have been reported in children in the past ten years.

Case report: This is a case report of the youngest child patient reported in the outpatient clinic of the Department of Oral Medicine and Radiology in Government Dental College, Kozhikode. The chief complaint was burning sensation on intake of food for a period of two months. The child presented with Lichen planus involving the oral cavity, skin and nails. There was no scalp and genital involvement. The child was treated with topical steroids and symptomatic relief was present. There were no new lesions on the follow up visits.

Conclusion: The classic picture of presentation of both adult and childhood. Lichen planus more or less remains the same. The prognosis and the treatment outcome of childhood lichen planus is better when compared to the adult.

Keywords: lichen planus, oral, children, burning sensation

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INTRODUCTION

Lichen planus is a common chronic inflammatory mucocutaneous disorder. Lichen planus (LP) is derived from the Greek 'leichen' meaning 'tree moss' and the Latin 'planus' meaning 'flat'. Erasmus Wilson first described LP in 1869, as a chronic disease affecting the skin, scalp, nails, and mucosa, with possible rare malignant degeneration. The prevalence rate varies based on different geographic regions but a worldwide prevalence rate is about 1-2%.¹ The disease is seen more frequently in middle aged and elderly patients and shows a female predilection.² The true cause of the disease is still obscure whilst the immunologic basis for the disease has been proved beyond question.³ Other predisposing factors that play a role in the disease are genetic predisposition, infective agents, systemic diseases, graft-vs.-host disease, drug reactions, and hypersensitivity to dental materials and vitamin deficiencies. OLP constitutes 9% of all white lesions affecting the oral cavity.¹ Oral LP appears in several forms, including atrophic, bullous, erosive, papular, pigmented, plaque-like, and reticular. Reticular lesions are the most common, but patients with reticular lesions are generally asymptomatic. Atrophic, bullous, and erosive lesions cause discomfort, ranging from mild to severe pain. The posterior buccal mucosa is most frequently involved, followed by the tongue, gingiva, and vermilion of the lower lip. Francois Henri Hallopeau reported the first oral lichen planus (OLP) related carcinoma in 1910.⁴ Oral lichen planus in childhood (OLPc) is rare and only a few reports are available in the literature. The family history of LP is more commonly positive in patients with LP in childhood than in adulthood.⁵ The childhood LP prevalence rate varies from 0.56 to 13% and occurs predominantly in the Asian population. There is controversy in the gender predilection of LPc and the largest series have reported male preponderance.⁶⁻⁸ The oral and scalp involvement

of LPc is rare compared to the adult. Isolated nail and skin involvement have also been reported.⁹ In Children Classic LP was most prevalent (53.8%), followed by eruptive (16.5%), hypertrophic (8.2%), linear (6.9%), and lichen planopilaris (6.3%). LP pigmentosus, annular, and atrophic variants were encountered infrequently.¹⁰ Buccal mucosa, gingiva and lateral part of the tongue are most commonly affected sites in OLPC.¹¹

CASE REPORT

A 3 year old female child was brought to the outpatient clinic, Department of Oral Medicine and Radiology, Government Dental College, Kozhikode, Kerala. Her parents had noticed white lesions on the inner aspect of upper and lower lip approximately 2 months back. They had also noticed small white raised areas of varying sizes on the skin of the knee at the same time. The patient complained of burning sensation of oral cavity for the last 2 months. They thought the lesion might subside on its own and waited for 2 months. The child was brought to Dental College for thorough investigation and treatment because the lesions were not subsiding and the complaint was persisting.

The child appeared to have normal growth and a detailed history revealed normal achievement of milestones and there was no significant drug or medical history. There was no relevant family history. No similar white lesions in any family members were reported. The vaccination schedule of the patient was strictly followed and parents had reported an oral polio vaccination about 6 months back. The detailed inspection of the patient revealed ridging and splitting of both thumb nails and nails of both great toe (Fig-1). There were small 2-3mm sized round white papules distributed diffusely over both the knees and legs (Fig-2). Involvement of scalp and genital region was not seen. Extraoral Inspection revealed continuous white linear striae on the upper lip involving the vermilion border (Fig-3). Intraorally striae in the form of reticular pattern on the mid third of the upper and lower labial mucosa and on the lower mucobuccal sulcus were present bilaterally (Fig- 5,6). White nonscrapable striae were seen on the dorsal and ventral surface

of the tongue (Fig-4,7). Although in gingiva and buccal mucosa, lesions were not found. The lesions were nonscrapable and rough on palpation.

Routine blood examination was performed and the results were within normal limits. Incisional biopsy was performed under local anesthesia after obtaining consent from the parents and sent for histopathological evaluation and direct immunofluorescence (Fig-5 and Fig-6). The results obtained were consistent with the clinical diagnosis of oral lichen planus.

The patient was prescribed topical steroids and was reviewed after two weeks. There was dramatic improvement in the appearance of the lesions and the patient remained asymptomatic. In 6 months follow up the white lesions decreased in severity and were seen to involve the upper and lower labial mucosa (Fig- 4, 5, 6, 7 and 8).

DISCUSSION

Largest case series on Lichen planus in children is from the Indian subcontinent with the recent studies being conducted in 2009 and 2011. This supports the suggestion by Ramsay and Hurley that the disease is more common in the tropics and reflects the role of genetic factors in addition to the environmental factors in the causation of LP.¹² The study in 2009 by Kanwar and De on 100 patients over a period of 6.5 years in children below 18 years of age have reported predominant involvement of scalp, extremities and nails.⁷ The study in 2011 was a retrospective analysis done on the clinical features and treatment response on 316 patients with lichen planus shows an 18% involvement of oral mucosa.¹⁰ Other studies conducted worldwide had reflected increased incidence in people of Indian origin. Though cutaneous lichen planus is not so rare in India, the oral involvement of lichen planus appears to be quite rare. Familial LP is said to have an earlier age of onset, widespread disease, common mucosal involvement, and frequent relapses.¹³ Childhood familial lichen planus is said to occur at an early age and with greater severity.¹⁴ The age of occurrence of lichen planus has been reported to be as early



Figure-1: Splitting and ridging of nails typical of LP



Figure-2: Raised white papules of diffuse pattern involving the knees and legs.

White nonscrapable striae involving the lips

After 6 months on follow up



Figure-3: OLP involving Lips on initial visit and follow up

White lacy pattern on the ventral surface of tongue

After 6 months on follow up



Figure-4: OLP on the ventral surface of tongue on initial visit and follow up

OLP involving the lower labial mucosa

After 2 weeks follow up

After 6 months follow up



Figure-5: OLP on lower labial mucosa on initial visit and follow up

OLP involving the upper labial mucosa

After 2 weeks follow up

After 6 months follow up



Figure-6: OLP on the upper labial mucosa on initial visit and follow up

OLP involving the tongue



After 6 months follow up



Figure-7: OLP on the dorsal surface of tongue on intial visit and follow up

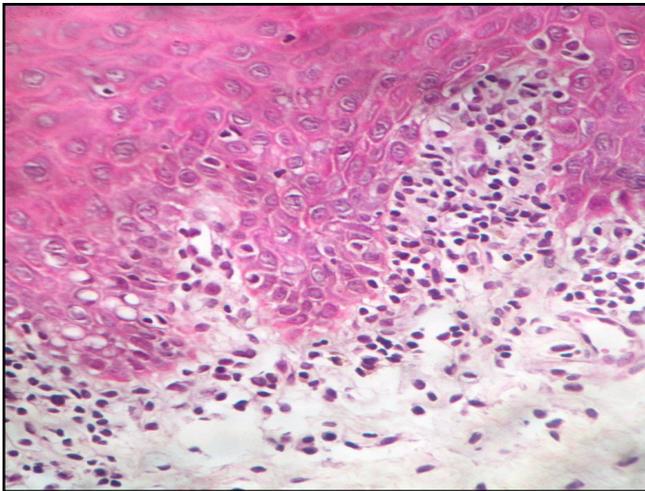


Figure-8: Saw toothed rete pegs with inflammatory infiltrate characteristic of Lichen Planus

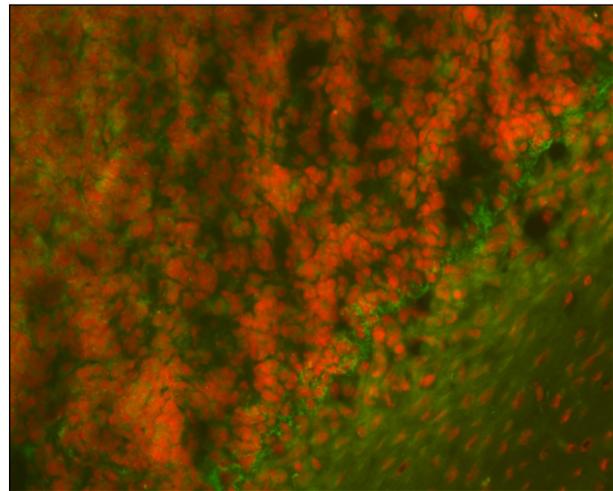


Figure-9: Deposition of immunoglobulin complexes and fibrin in a shaggy patten Direct immunofluorescence

as 2 weeks in one study⁷ and 3 months in another¹⁰ whilst the oral involvement in as early as 3 years should be the first case to be reported. OLP in children had presented as reticular, annular or papular varieties but isolated case reports of ulcerative lichen planus and atypical lichen planus of oral mucosa has also been reported. Lip involvement was seen in about 8.5% in the largest study on childhood lichen planus of 316 patients and an isolated lip involvement of 8% among 87 patients has also been reported.⁸ There is discrepancy regarding the sex predilection of LPc.² The largest case series on LPc by Kanwar et al and Pandhi et al showed an increased number of cases in boys where in case series on 36 patients by Walton et al showed an increased occurrence in girls.^{7,10,15}

Several diseases and conditions had been considered as predisposing factors like hepatitis, autoimmune diseases and there has also been studies proving or challenging the association of childhood OLP with hepatitis B vaccination.^{16,17} There is no consensus regarding the treatment protocol for childhood oral lichen planus. Topical 0.1% triamcinolone acetonide three times daily was tried considering the age, location and size of the lesion in this patient and it showed an improvement in the appearance within a 2 weeks' time. The patient became asymptomatic after 2 weeks and is under constant follow up.

CONCLUSION

The increased occurrence of the disease in child-

ren had made oral lichen planus to be considered in the differential diagnosis of white lesions in children. Due to the atypical presentation of the reported cases of LPc no written protocol has been followed for the treatment. Further researches are awaited for predicting the prognosis and treatment of oral lichen planus in children.

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