Unusual presentation of pediatric lichen planus with cutaneous, oral and nail involvement: a case report

Nairica E Rebello[1], MDS, **Anita Spadigam**[1], MDS, **Anita Dhupar**[1], MDS, **Jochima Cota**[1], MDS

[1]Department of Oral and Maxillofacial Pathology, Goa Dental College and Hospital, Bambolim, Goa, India

Corresponding author: Dr. Nairica Eurico Rebello

Email- nairicarebello@gmail.com

Phone- +919823221509

ABSTRACT

Introduction: Lichen planus (LP) is a chronic, immune-mediated mucocutaneous disorder with rare oral involvement in pediatric patients. Oral lichen planus (OLP), a recognized oral potentially malignant disorder, poses diagnostic and therapeutic challenges in children due to its rarity. **Case Presentation:** A 16-year-old male presented with a one-year history of burning sensation in the oral cavity, aggravated by spicy food. Intraoral examination revealed bilateral greyish-white reticular plaques with peripheral striae on the buccal mucosa and tongue. Violaceous papules on extremities and toenail trachyonychia were noted. Histopathology was consistent with OLP. The patient was treated with triamcinolone. **Discussion:** Reported pediatric LP cases show varied lesion patterns and age inclusion resulting in underestimation of the actual burden of the condition. Nail involvement, which is particularly rare, was noted in the present case. **Conclusion:** This case highlights the need for age standardization, early recognition, and long-term follow-up of pediatric LP to prevent potential complications.

Keywords: histopathology; mucocutaneous lesions; oral potentially malignant disorder; pediatric lichen planus; triamcinolone

CDHA Research Agenda category: risk assessment and management

CONTEXT/INTRODUCTION/BACKGROUND

Lichen planus (LP) is a widespread, chronic inflammatory disorder with variable mucocutaneous manifestations. Though the exact cause remains unidentified, an immune-mediated mechanism is suspected.[1] A majority of cases have been reported in middle-aged adults and prevalence in the pediatric population is merely 1%-4%.[2] Oral lichen planus (OLP) is considerably less common in children, with an incidence of merely 0.03%. Clinical presentation is variable and there exists a lack of consensus regarding treatment strategies specific to pediatric patients.[2] Oral lichen planus has been categorized as an oral potentially malignant disorder[3] with a malignant transformation rate of 0.44%-2.28% in adults.[4] Despite malignant transformation not being reported in children, likelihood cannot be entirely excluded.[2] Nail involvement is rare in pediatric LP with a prevalence of less than 10% and clinical presentation similar to adults.[2, 5] Lack of clarity around clinical presentation and course may render diagnosis challenging, resulting in underreporting of the condition, specifically in pediatric patients. We present the case of a 16-year-old male who reported with the chief complaint of burning sensation in the oral cavity.

CASE PRESENTATION

A 16-year-old male presented with a one-year history of oral burning sensation, exacerbated by spicy food and characterized by intermittent remission and exacerbation. Medical and dental histories were non-contributory. Patient reported prolonged exposure to the sun and soil when participating in sports. Extraoral examination revealed flat topped violaceous papules on the fingers, palms, forearms, feet, and toes, with a history of pruritus. Toenails exhibited longitudinal ridging, splitting, and trachyonychia with spontaneous chipping. Intraorally, bilateral greyish-white reticular plaques with peripheral striae were observed on the buccal mucosa, along with a linear plaque on the lateral tongue. Lesions were non-scrapable and non-tender. Differential diagnoses included lichen planus, psoriasis and lupus erythematosus (Figure 1). Values of hematological investigations including CBC, hemoglobin, liver function, renal function, and viral markers were within normal limits. Histopathological examination of an incisional biopsy of the buccal mucosa revealed parakeratinized stratified squamous epithelium with focal hyperkeratosis, saw-tooth rete pegs, basal cell liquefaction, and a subepithelial band of lymphocytic infiltration (Figure 2). A definitive diagnosis of oral lichen planus was established based on the clinicopathological findings.

Management

The patient was prescribed topical corticosteroid cream containing 0.1% triamcinolone acetonide (Kenacort), as the first line of treatment for the oral and nail lesions. Systemic corticosteroid therapy was unwarranted, since the patient reported relief. Patient was counselled about trigger avoidance. Intraoral examination during the follow-up after one month revealed partial regression of lesions. The patient has been advised follow-up every 6 months to evaluate disease progression.

DISCUSSION

The definition of "pediatric" varies significantly across literature, with upper age limits ranging from 12 to 21 years. The American Academy of Pediatrics sets this limit at 21 years.[6] Such inconsistencies can contribute to misclassification and underestimation of disease burden in younger populations, particularly in rare conditions like pediatric OLP, thereby complicating diagnosis and delaying tailored therapeutic approaches. Pediatric lichen planus has been described under several terms—"childhood lichen planus," "juvenile lichen planus," and "lichen planus in childhood"—without uniform age-based criteria.[1, 7–5] Cascone et al. and Chatterjee et al. conducted two independent studies on pediatric OLP and included individuals under 18 years.[1,9] Consequently, we have classified our 16-year-old patient under the pediatric age group.

The etiopathogenesis of OLP remains elusive (Figure 3). Contributing factors may include hypersensitivity reactions to dental materials, chronic mechanical irritation, and viral infections such as hepatitis B and C.[1] Pediatric OLP has been associated in some cases with hepatitis B vaccination.[10] Our patient reported frequent exposure to dust and soil during outdoor sports, which could represent a potential environmental trigger. However, this relationship is speculative and yet to be established.

Cutaneous LP typically manifests as purple, polygonal, pruritic papules with a lace-like network of white striae, most often affecting the flexor surfaces, trunk, and thighs.[1] Pediatric cases often present with a linear distribution of skin lesions;[2] however, in this case, the distribution was non-linear.

The nail changes observed were consistent with previously reported findings. Oral involvement is less common in pediatric LP than in adults, but it can present with significant clinical

heterogeneity.[9] Oral lesions are classified into six clinical types—reticular, atrophic, erosive, papular, plaque-like, and bullous—with a bilateral, symmetrical predilection for the buccal mucosa, tongue, and gingiva.[12] Our patient presented with the reticular variant, the most frequently reported subtype in pediatric cases.[13]

Familial OLP is rare but more frequently reported in pediatric populations and is often characterized by a chronic and recurrent course.[9,14] Although our patient had no family history of OLP, Wang et al. reported a pediatric case with familial predisposition and long-term follow-up.[15] While malignant transformation in pediatric OLP is exceedingly rare, the potential risk cannot be entirely excluded. Long-term follow-up is therefore recommended. Alrashdan et al. suggest periodic monitoring every two months to annually, depending on disease severity and progression.[16]

CONCLUSION

It is essential for dental practitioners to be well-informed about the clinical presentation, diagnostic considerations, and management strategies of pediatric oral lichen planus. Comprehensive examination should be performed to identify rare manifestations like nail involvement. Emphasis should also be placed on regular follow-up visits to monitor disease progression and manage potential recurrences effectively. Additionally, a standardized definition of the pediatric age group in the context of oral lichen planus is essential to ascertain the actual burden of the disease.

Practice Implications:

- Standardizing age criteria for the paediatric category is essential for accurate disease prevalence estimates and subgroup-specific management.
- Scheduling regular follow-up is crucial to monitor progression and mitigate malignant risk in pediatric LP.

REFERENCES

1. Cascone M, Celentano A, Adamo D, Leuci S, Ruoppo E, Mignogna MD. Oral lichen planus in childhood: a case series. *Int J Dermatol*. 2017;56(6):641–52.
2. Merhy R, Sarkis AS, Assaf J, Afiouni R, Zeinaty P, Kechichian E, et al. Pediatric lichen planus: a systematic review of 985 published cases. *Int J Dermatol*. 2022;61(4):416–21.
3. Joshi, Arun; Rathi, Sanjay Kumar; Manchanda, Yashpal. Childhood Lichen Planus. *Indian J Paediatr Dermatol*. 2021; 22(4):p 306–15.
4. Warnakulasuriya S, Kujan O, Aguirre-Urizar JM, et al. Oral potentially malignant disorders: a consensus report from an international seminar on nomenclature and classification, convened by the WHO Collaborating Centre for Oral Cancer. *Oral Dis*. 2021;27(8):1862–80.
5. González-Moles MÁ, Warnakulasuriya S, González-Ruiz I, Ayén Á, González-Ruiz L, Ruiz-Ávila I, et al. Dysplasia in oral lichen planus: relevance, controversies and challenges. A position paper. *Med Oral Patol Oral Cir Bucal*. 2021;26(4):e541–8.
6. Sawyer SM, McNeil R, Francis KL, Matskarofski JZ, Patton GC, Bhutta ZA, et al. The age of paediatrics. *Lancet Child Adolesc Health*. 2019;3(11):822–30.
7. George S, John SA, Anandaraj S, Issac JS, Harris A, Reshmi J. Childhood oral lichen planus: report of two cases. *J Dent (Tehran)*. 2015;12(5):374–8.
8. Chaitra TR, Telgi RL, Kishor A, Kulkarni AU. Juvenile oral lichen planus: a clinical rarity. *BMJ Case Rep*. 2012;2012:bcr2012006192.
9. Chatterjee K, Bhattacharya S, Mukherjee CG, Mazumdar A. A retrospective study of oral lichen planus in paediatric population. *J Oral Maxillofac Pathol*. 2012;16(3):363–7.
10. Pendyala G, Joshi S, Kalburge J, Joshi M, Tejnani A. Oral lichen planus: a report and review of an autoimmune-mediated condition in gingiva. *Compend Contin Educ Dent*. 2012;33:e102–8.
11. Woo VL, Manchanda-Gera A, Park DS, Yoon AJ, Zegarelli DJ. Juvenile oral lichen planus: a report of 2 cases. *Pediatr Dent*. 2007;29(6):525–30.
12. De Moraes PC, Teixeira RG, Tacchelli DP, Bönecker M, Junqueira JLC, Oliveira LB. Atypical case of oral lichen planus in a pediatric patient: clinical presentation and management. *Pediatr Dent*. 2011;33(5):445–7.

13. Cheng YS, Gould A, Kurago Z, Fantasia J, Muller S. Diagnosis of oral lichen planus: a position paper of the American Academy of Oral and Maxillofacial Pathology. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2016;122(3):332–4.
14. Wang F, Tan YQ, Zhang J, Zhou G. Familial oral lichen planus in a 3-year-old boy: a case report with eight years of follow-up. *BMC Oral Health*. 2020;20(1):341.
15. Alrashdan MS, Cirillo N, McCullough M. Oral lichen planus: a literature review and update. *Arch Dermatol Res*. 2016;308(8):539–51.

FIGURES

Figure 1. Photograph showing (a) and (b) white non scrapable plaque on the left buccal mucosa and right buccal mucosa, respectively; (c) violaceous papules on the hands and (d) violaceous papules on the feet with toenail trachyonychia

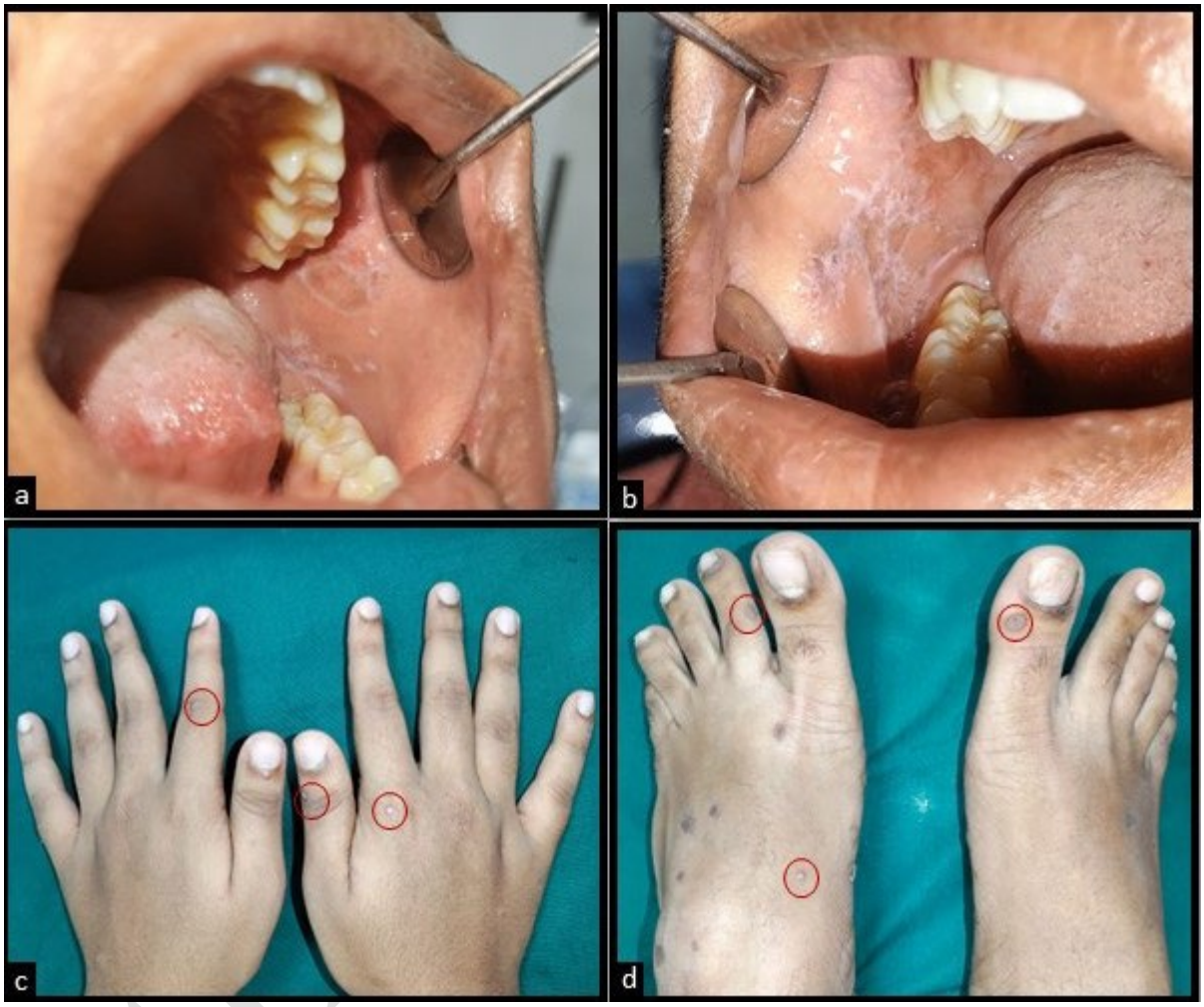


Figure 2. Photomicrograph of incisional biopsy of buccal mucosa showing (a) liquefaction degeneration of the basal cells, saw tooth rete pegs (H & E, 4x) and (b) subepithelial band of chronic inflammatory cell infiltrate (H & E, 10x)

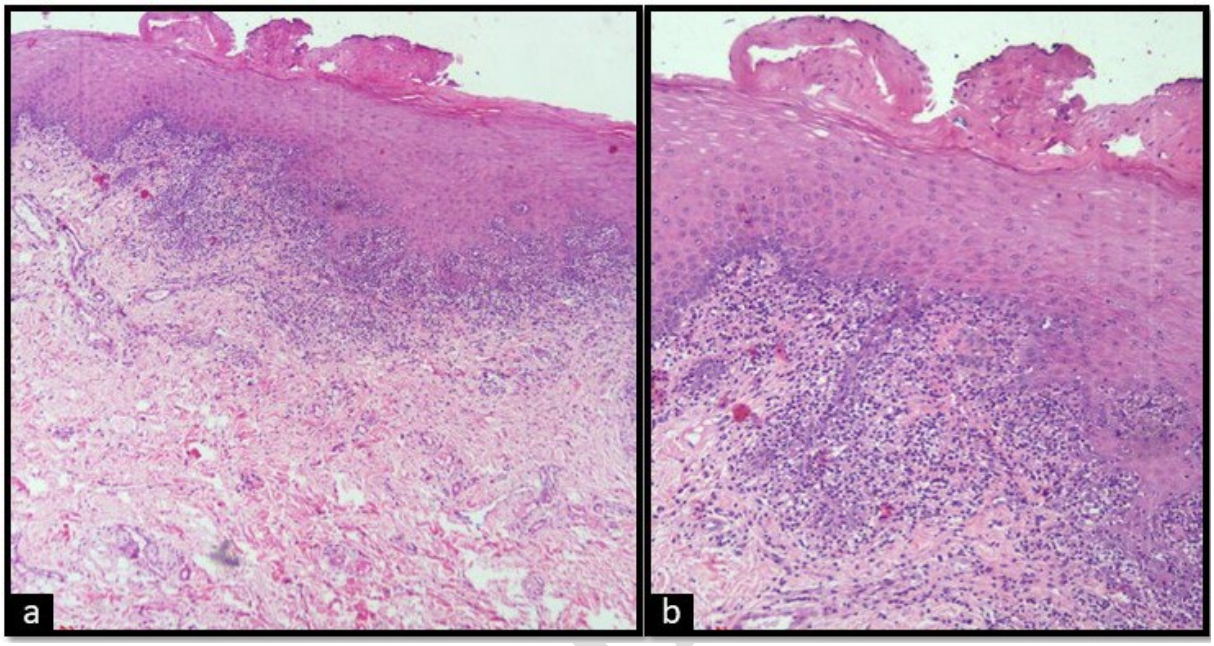
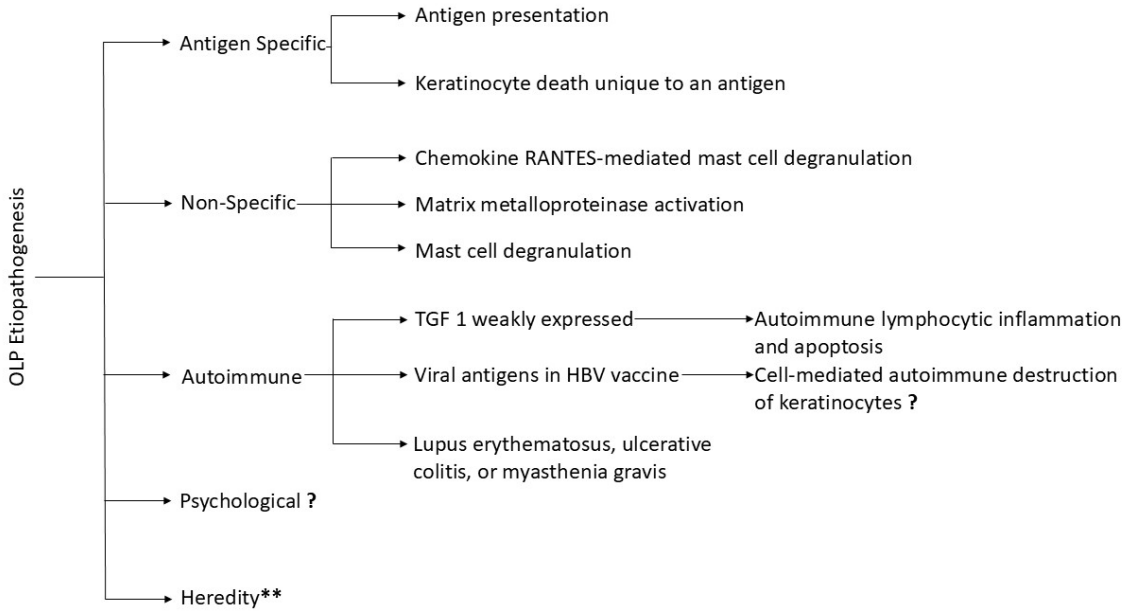


Figure 3. Proposed ethiopathogenesis of oral lichen planus

.



?: questionable role; **: specific to pediatric oral lichen planus