

Case Report

Diagnostic and Management Approach towards Oral Lichen Planus in Adolescents: Report of Two Cases and Review of Current Concepts

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Abstract

Oral lichen planus (OLP), is a chronic inflammatory disease that mainly affects women and is rarely observed in children and adolescents. We report here two cases of OLP in adolescents and review the cases published in the literature. First one, a 10-year-old boy exhibited asymptomatic lesion in the vermilion border of the lower lip and white lesions on the dorsum of the tongue. The second one, a 15-year-old boy presented asymptomatic white striations and plaques in the tongue and buccal mucosa. Incisional biopsies were performed under the clinical hypothesis of OLP. Microscopically, it was observed fragment of oral mucosa lined by orthokeratinized stratified squamous epithelium. Areas of basal cell degeneration and a band-like infiltrate of inflammatory cells predominantly lymphocytic around the basement membrane region. Based on clinical and histopathological features, the histopathological diagnosis of OLP was rendered. In children and adolescents, OLP clinically resembles the disease seen in adult patients but the prognosis is more favorable. Physicians and dentists should be aware of the occurrence of OLP in childhood and adolescence since it is often not included in the differential diagnosis of white lesions in this age group.

ABBREVIATIONS

LP: Lichen Planus; OLP: Oral Lichen Planus; HLA: Human Leukocytes Antigen

INTRODUCTION

Lichen planus (LP), is a chronic inflammatory mucocutaneous disorder that can affect the skin, scalp, nails, and oral mucosa. In about 70 to 77% of patients, oral and skin lesions occur simultaneously. Although its etiology remains unknown, LP is considered a T-cell-mediated autoimmune disease in which helper CD4⁺ lymphocytes produce proinflammatory cytokines such as interleukin 1 and interferon-gamma. These cytokines activate cytotoxic CD8⁺ lymphocytes that trigger the apoptosis of basal epithelial cells [1-4].

Oral lichen planus (OLP) can affect any site in the mouth, with the buccal mucosa and tongue being the most common sites [2,4]. Women in their fifth and sixth decades of life are the most affected group, while the condition is rarely observed in children and adolescents [2,4,5]. The clinical presentation of OLP is variable

and six types have been reported: reticular, erosive, atrophic, papular, plaque like, and bullous. Among these types, the erosive and atrophic forms are frequently symptomatic, while the other presentations are usually asymptomatic [4,5].

There are few studies of OLP in young patients and the literature reports an incidence of this disease of 0.03% in children [1,5,6]. Within this context, the aim of this study was to report two cases of OLP in adolescents and to discuss the features and clinical management of this disease in these patients.

CASE PRESENTATION**Case 1**

A 10-year-old boy was referred to the Stomatology Clinic of the Federal University of Rio Grande do Norte (UFRN) for assessment of a 4-month history of an asymptomatic lesion on the lip. Extraoral physical examination revealed the presence of a lesion in the vermilion border of the lower lip. The lesion measured approximately 2 cm and had a brownish color and white striations (Figure 1A). No skin lesions were observed.

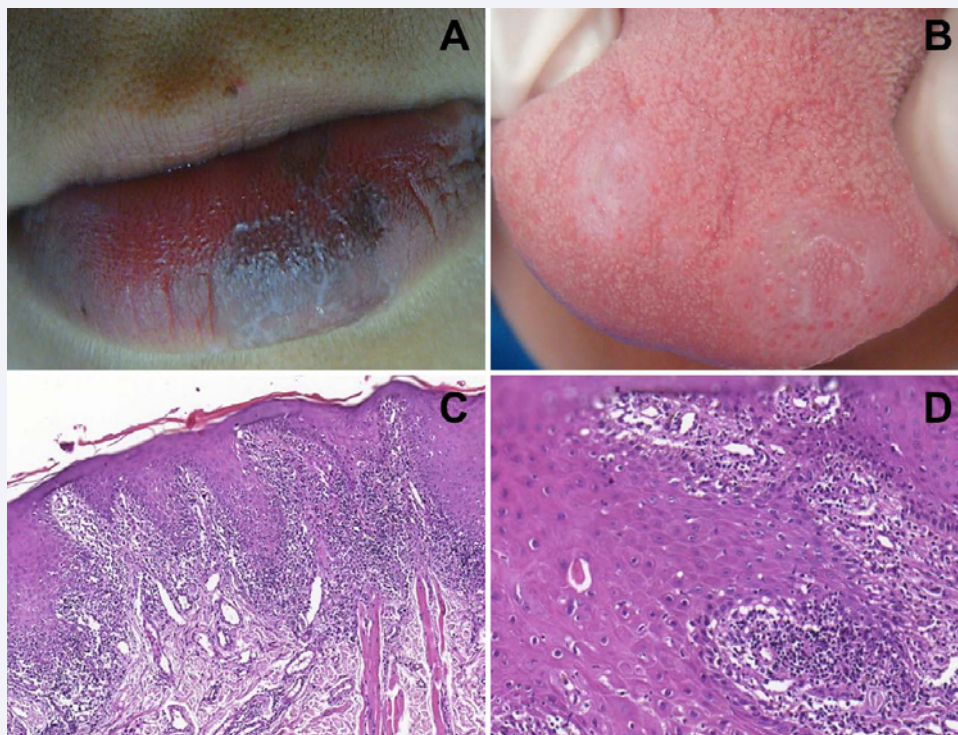


Figure 1 (A) Brownish plaque associated with white striations on the vermillion of the lower lip. (B) White plaques on the dorsum of the tongue. (C) Fragment of oral mucosa lined by orthokeratinized stratified squamous epithelium. Areas of basal cell degeneration, and presence of inflammatory infiltrate in the subepithelial region were observed (1,000 µm; H / E). (D) Presence of apoptotic body (Civatte) in the middle third of the lining epithelium (500 µm; H / E).

Intraoral examination showed the presence of asymptomatic white lesions on the dorsum of the tongue (Figure 1B). Based on the clinical diagnosis of OLP, an incisional biopsy of the lip lesion was performed, and histopathological analysis confirmed the diagnostic hypothesis of OLP (Figure 1C and 1D). Follow-up of the patient showed no change in the clinical presentation.

Case 2

A 15-year-old Afro-descendant boy without comorbidities presented to the Stomatology Clinic of UFRN for assessment of asymptomatic white lesions located bilaterally on the buccal mucosa. The duration of the lesions was unknown (Figure 2A and 2B). The patient also had white and pigmented lesions on the lateral border and dorsum of the tongue (Figure 2A, 2B, 2C, and 2D). No lesions were observed in other regions of the body. During anamnesis, the patient reported having recently experienced moments of emotional stress. An incisional biopsy of the buccal mucosa lesion was performed, and the histopathological diagnosis confirmed the clinical hypothesis of OLP (Figure 2E and 2F). The patient is currently under follow-up and there is no change in the clinical presentation.

DISCUSSION

Oral lichen planus is uncommon in young patients, especially children and adolescents. Only seven studies reporting its occurrence in this group of patients have been published in the last 5 years (Table 1).

Although the etiology of OLP remains unclear, it is known

that the immune response plays an important role in its development [5,7], with evidence even pointing to the possibility of autoimmunity [6,8]. Autoimmune diseases are less common in young patients, a fact that may explain the low incidence of OLP in these patients. Furthermore, emotional factors such as stress, anxiety and depression have been associated with the occurrence of OLP [8]. In the present case 2, the patient reported episodes of stress but no association with the emergence of the lesions could be established.

A rare family condition of LP characterized by autosomal dominant inheritance and variable penetrance has also been described. This condition is associated with alleles HLA-B7 and HLA-BR10, and is frequent in pediatric patients. This type of LP is characterized by the early onset of severe lesions in the oral mucosa [2,5,7,9]. In the cases reported here, there was no evidence of an association of the lesions with this genetic condition considering the absence of a history of OLP in other family members. Likewise, no family history was observed in the other clinical cases reported in the literature, highlighting the rarity of OLP, especially of the familial form, and demonstrating a higher frequency of conventional OLP in this age group.

Another important consideration is that, since OLP is uncommon in young patients, it is often not included in the differential diagnosis of white lesions in this age group. Furthermore, a biopsy of these lesions is not always obtained, a fact that makes it difficult to distinguish them from other similar conditions such as lichenoid reactions, leukoplakia and

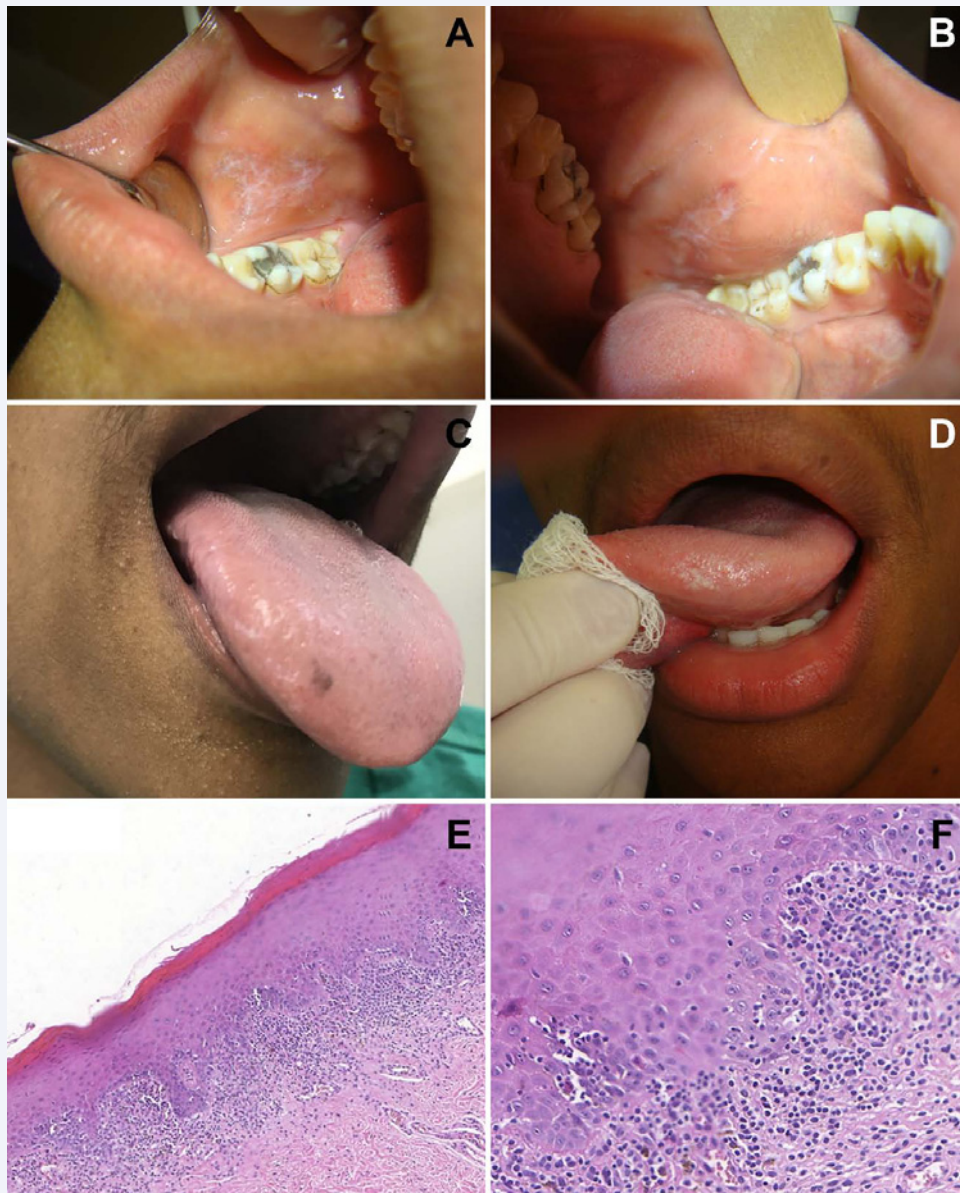


Figure 2 White reticular lesions on the right (A) and left (B) buccal mucosa. Brownish patch and white plaque on the dorsum (C) and left lateral border (D) of the tongue, respectively. (E) Mucosal fragment lined by parakeratinized stratified squamous epithelium. Basal cell degeneration, and a band-like infiltrate of inflammatory cells predominantly lymphocytic around the basement membrane region (1,000 μm ; H / E). (F) Subepithelial lymphocytic inflammatory infiltrate (500 μm ; H / E).

hyperkeratosis, and can lead to the underreporting of cases [2,6]. A biopsy is therefore imperative so that histopathological analysis can assist in establishing the definitive diagnosis. In the two cases reported here, the clinical presentation was suggestive of OLP because of the white striations and microscopic examination confirmed the clinical suspicion.

A literature search over the last years identified seven studies that reported OLP in children and adolescents, totaling 17 cases including the two cases reported here. Four of the seven studies analyzed were conducted in India. Although there are no data on a racial predilection, most of the published reports are from India. This fact may suggest that children from this country are genetically more susceptible or are more exposed to certain

environmental factors that predispose to the development of OLP [2]. A male predominance of OLP cases exists in this age group. In contrast, in adults, the disease more commonly affects females [5,8]. Although case reports and case studies have demonstrated characteristics of some specific groups, the higher incidence of OLP observed in male children might be related to polymorphisms in the human leukocytes antigen (HLA) system, which are frequently observed in boys with autoimmune diseases such as juvenile idiopathic arthritis [10,11].

The mean age of the patients in the identified studies was 11.8 ± 3.09 years (range: 7-17) and the lesions involved multiple oral sites, particularly the buccal mucosa, tongue and lip. The lesions mainly exhibited features of the reticular type (Table 1). Similar

Table 1: Demographic and clinical data on oral lichen planus pediatric cases reported in the literature (Pubmed, 2014-2019 - acesso em 14/10/2019).

AUTHORS	GENDER/ AGE (YEARS)	LOCATION	CLINICAL ASPECTS/ SYMPTOMATOLOGY	DURATION	EXTRAORAL LESIONS
Present cases, 2020 (Brazil)	Case 1 - M/10	Lower lip ¹ and dorsum of the tongue ²	Interlacing white striae on a brownish background ¹ and white plaque ²	04 months	None
	Case 2 - M/15	Buccal mucosa (bilaterally) ¹ ; lateral borders (bilaterally) ¹ and dorsum ² of the tongue	White reticular striaes ¹ and brownish plaque ²	Not reported	None
Jonas et al. [13] (Germany)	Case 1 - M/11	Lips ¹ and buccal mucosa (bilaterally) ²	Lichenoid plaques ¹ and multiple reticular white stripes ²	03 months	Trunk, neck, arms, and legs
Cascone et al. [5] (Italy)	Case 1 - F/17	Margins of tongue and ventral surface of the tongue (bilaterally)	Plaque-like and erosive pattern/Burning sensation with spicy food	Not reported	None
	Case 2 - M/17	Hard palate and retromolar fossae (bilaterally)	Reticular plaque-like/Dryness	Not reported	None
	Case 3 - F/14	Buccal mucosa, margins, and ventral surface of the tongue (bilaterally), and lower lip	Atrophic plaque like and pigmentation	Not reported	None
	Case 4 - M/14	Buccal mucosa (bilaterally), gingiva, dorsum, and lateral borders of the tongue (bilaterally)	Reticular and atrophic pattern	Not reported	None
	Case 5 - M/15	Buccal mucosa (bilaterally) and ventral surface of the tongue	Reticular pattern	Not reported	None
	Case 6 - F/9	Dorsum and margins of tongue, gingiva, and buccal mucosa (bilaterally)	Bullous, atrophic, and reticular pattern/Pain	Not reported	None
	Case 7 - F/11	Gingiva	Reticular pattern	Not reported	None
	Case 8 - M/11	Dorsum of the tongue	Roughness, reticular and atrophic pattern	Not reported	None
Thomas; Betsy [14] (India)	M/14	Upper labial mucosa extending onto the gingival margin	White reticulate in a linear pattern/Asymptomatic	Not reported	Skin and nasal mucous membrane
Sharma et al. [15] (India)	F/12	Gingiva and vestibular area of the permanent mandibular molars (bilaterally)	Grayish white patches with peripherally radiating white striae/Burning sensation with spicy food	03 months	None
George et al. [7] (India)	Case 1 - M/8	Buccal mucosa (bilaterally), ventral surface of the tongue; and labial mucosa	Interlacing white striae (corrugated)/Pain and discomfort with spicy food	Not reported	Fluid-filled blisters on feet
	Case 2 - M/8	Left buccal mucosa (unilaterally)	Interlacing white striae and pigmentation/Burning sensation with spicy food	03 months	None
Zychowska et al. [16] (Poland)	M/10	Buccal mucosa	White linear lesions	18 months	Nails
Chandna et al. [1] (India)	F/7	Buccal mucosa (bilaterally) ¹ and vermilion border of the lip ²	White patches ¹ and purplish lesions ²	06 months	Purplish lesions on the neck

Abbreviations: M – Male; F – Female

to the cases shown in Table 1, the present patients were 10 and 15 years old and had reticular and plaque-like lesions on the lip, tongue and buccal mucosa. Since autoimmune diseases generally show a sex predilection, it is believed that sex hormones, particularly estrogen, play an important role in the development of these diseases [10]. One study indicated that estrogen is responsible for reinforcing the physical barrier of oral epithelial cells by modulating the nuclear estrogen receptor-dependent upregulation of tight junction proteins [12]. This might explain why menopausal middle-aged women are more affected by LP. In contrast, in young individuals especially those in the pubertal phase, elevated hormone levels may act as a protective factor, a fact that may explain the rarity of this disease in this group of patients.

The treatment of children and adolescents with OLP does not differ significantly from that of adults. Topical or systemic corticosteroids are recommended for symptomatic cases. These drugs can be combined with antifungal agents, when necessary, to prevent overlapping candidiasis [5,7]. However, pharmacological therapy is not indicated in asymptomatic patients, such as the cases reported here [5,6]. Follow-up of the patient is established as clinical management in this case. This is an important aspect of the treatment of patients with OLP because of the known dynamic nature of oral lesions which, at a certain time point, may appear as asymptomatic white striations and may become atrophic, ulcerated, and symptomatic at other times.

Controversies exist regarding the malignant transformation

of OLP since it remains unknown whether malignancy would be related to the immune response inherent to the pathogenesis of OLP or whether it would be associated with genetic factors and/or intrinsic factors of these lesions. Oral LP is described by the World Health Organization as a potentially malignant disorder and its estimated transformation potential ranges from 0.4 to 5%; however, there are no reports of malignant transformation in children or adolescents [4]. However, clinical follow-up is recommended for all patients with OLP, regardless of age. Furthermore, it is important to mention that spontaneous resolution within a few weeks can occur in young and elderly patients and that only 10-20% of cases develops intermittent recurrences [4-6], facts that highlight the importance of long-term follow-up.

In conclusion, the present article agrees with the main demographic and clinical features of OLP described in the literature. In young patients, especially children and adolescents, OLP clinically resembles the disease seen in adult patients but the prognosis is more favorable. It is therefore important that physicians and dentists are aware of the occurrence of OLP in childhood and adolescence, especially in the case of a positive family history, and that oral lesions often precede skin lesions. Additionally, we emphasize the importance of case studies and case reports demonstrating the occurrence of this disease in young patients. In addition to providing knowledge about the characteristics of OLP in this age group, such studies will allow long-term follow-up and thus contribute to the understanding of the clinical behavior of this disease.

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