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REPORT

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Oral lichen planus in a young patient: a case report with nine-year follow-up

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ABSTRACT

Introduction: Oral lichen planus is an inflammatory condition that affects the stratified squamous epithelium of the oral mucosa. It occurs more frequently in female patients and it is rarely observed in children, adolescents, or young adults. This study aims to report a case of oral lichen planus in a young patient with a nine-year follow-up. **Case report:** A 19-year-old man reported to the Dentistry Department with a complaint of an asymptomatic white lesion on the dorsum and left lateral border of his tongue, which had appeared a few weeks before. Two weeks later, a second lesion, very similar to the previous one, appeared on the central region of his tongue. An incisional biopsy was performed. The histological slides were stained with hematoxylin-eosin and the expression of interleukin-1beta (IL-1β) and tumor necrosis factor-alpha (TNF-α) was assessed by immunohistochemistry. No pharmacological treatment was prescribed. The clinical and histopathological findings were suggestive of oral lichen planus. The IL-1β/TNF-α expression was low. There was a spontaneous regression of the lesions after approximately one year. The nine-year follow-up showed no signs of recurrence. **Conclusion:** This case presents atypical features such as the age of the patient and the spontaneous remission of the lesions.

Keywords: case reports; lichen planus, oral; young adult; immunohistochemistry; interleukin-1beta; tumor necrosis factor-alpha.

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INTRODUCTION

Lichen planus is a chronic inflammatory condition that affects the skin, nails, scalp, and mucous membranes. It is characterized by pruritic papules usually covered by white scales on their surface[1]. Lesions may appear in one or more sites, with oral lichen planus (OLP) occurring more frequently than cutaneous[2]. OLP is a relatively common inflammatory condition that affects the stratified squamous epithelium of the oral mucosa and presents a prevalence of 0.2% to 5%[3,4]. OLP has a predilection for women and is most commonly observed between the fourth and seventh decades[3].

Although many etiological mechanisms for the such condition have been postulated, including genetic susceptibility, emotional stress, and infections caused by viruses and bacteria, the precise etiology is unknown[5]. However, OLP is considered an immune-mediated disorder[6]. Clinically, six types of lesions may manifest individually or in combination: plaque-like, reticular, papular, bullous, atrophic, and erosive[3]. Lesions can persist for many years, with periods of aggravation and inactivity[7]. Symptoms range in severity from minimal discomfort to difficulty in

speaking, eating, and swallowing[8]. Reticular lesions are generally asymptomatic, while ulcerative and erosive lesions usually result in discomfort or pain[4,7,8].

The histopathological features of OLP are variable, as biopsies from erosive, atrophic, and hypertrophic lesions can present different characteristics, including hyperkeratosis, occasional areas of atrophic epithelium, basal layer degeneration, and a band-like lymphocytic infiltrate[3,9,10]. Since the immunological mechanisms involved in this condition are unclear, several biomarkers, such as IL-4, IL-6, IL-8, IFN-γ, IL-1β, and TNF-α, have been studied to clarify its etiopathogenesis[11]. The diagnosis is based on a combination of clinical findings and histopathologic features[6].

Frequently, oral lesions have unique clinical features and a distinctive distribution, but they may also occur in other forms, simulating other diseases[2]. Differential diagnoses include chronic ulcerative stomatitis, oral lichenoid drug reactions, lichen planus pemphigoid, chronic graft-versus-host disease, oral lichenoid contact hypersensitivity reactions, lupus erythematosus, oral epithelial dysplasia, proliferative verrucous leukoplakia, and mucous membrane pemphigoid[10,12].

OLP requires treatment when symptoms interfere with the daily activities of the patient[8]. Topical corticosteroids are the most common treatment; however, patients with asymptomatic lesions usually do not require active therapy[1,8]. In 1978, the World Health Organization classified OLP as a potentially malignant disorder, given its association with the development of oral squamous cell carcinoma[13]. However, the malignant transformation of OLP is highly controversial in the literature[4,9].

Reports in pediatric patients are rare. The lack of reported cases in young patients may be due to a lack of awareness from patients, their parents, and dentists, and low incidence of triggering factors, such as stress and autoimmune diseases[14]. The purpose of this paper is to report a case of a young patient diagnosed with OLP, who presented spontaneous regression of lesions and no signs of recurrence within nine years of follow-up.

REPORT

A Brazilian 19-year-old male patient, caucasian, attended the Stomatology Clinic in 2011, complaining of a white lesion in the tongue, with an evolution period of three weeks. In the anamnesis, the patient reported good general health and no use of any medication. He was experiencing a period of great emotional stress and denied smoking and alcoholism. In his medical history, there was an episode of atopic dermatitis that affected his feet and legs, without significant improvement with the treatments used at the time (topical corticosteroids, and phytotherapeutic ointments, among others). Atopic dermatitis was diagnosed at 5 years of age and regressed spontaneously after seven years. In his family history, there was no significant data to aid in diagnosis.

During the extraoral examination, no alterations were observed. At the intraoral examination, a whitish plaque measuring about 2 cm in diameter with a striated appearance and smooth surface was identified, involving the dorsum and lateral border of the tongue. The lesion had normal consistency, poorly defined borders, was asymptomatic and was not removable by scraping. The patient had excellent oral hygiene, had been in orthodontic treatment for about five months, reported no trauma, and had no metallic restorations. After two weeks of follow-up, a second lesion, similar to the first one, appeared in the central region of the tongue (Figure 1).

The diagnostic hypotheses considered were focal hyperkeratosis, oral lichen planus, migratory glossitis, and leukoplakia. Complementary exams were performed before the biopsy and the results are presented in Table 1. An incisional biopsy was performed on the first lesion. The histological slides were stained with hematoxylin-eosin. Hyperparakeratosis, areas of atrophic epithelium with saw-tooth rete pegs, and liquefactive degeneration of the basal cell with a dense band-like lymphocytic infiltrate at the interface between the epithelium and the connective tissue were observed, with no signs of dysplasia (Figure 2). To verify the presence of chemical mediators related to the inflammatory process, the IL-1β and TNF-α expression was analyzed by immunohistochemistry. Only a few cells of the basal layer were IL-1β/TNF-α positive (Figure 3).

The clinical and histopathological findings were suggestive of OLP. Since there were no symptoms, no pharmacological treatment was prescribed. After one year, the lesions showed spontaneous regression and there were no signs of recurrence during nine years of follow-up (Figure 4).

DISCUSSION

Although OLP is rare in young patients, the clinical and histological characteristics of this case were in accordance to what has already been established in the literature, especially the presence

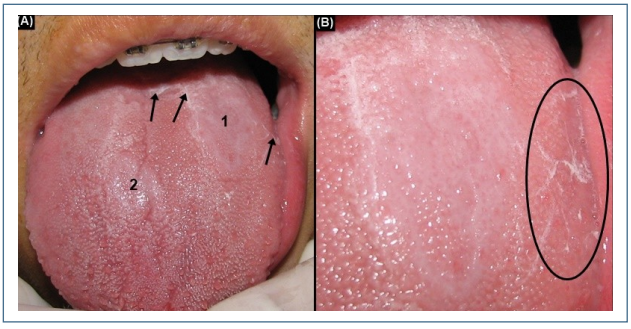

Figure 1: (A): Clinical aspect of the lesions; (1): the first lesion, a whitish plaque with the presence of Wickham striae (arrows), which appeared in the dorsum and lateral border of the tongue; (2): the second lesion, which appeared in the central region of the tongue a few weeks after the first one. (B): A closer look at the Wickham striae.

of Wickham striae, hyperparakeratosis, saw-tooth rete pegs, basal cell degeneration, and dense lymphocyte infiltrate[10,12]. The diagnosis was hard to establish due to some unusual factors: the age and gender of the patient, and the location of the lesions. OLP usually occurs in female patients of middle age, with symmetrical lesions on both sides of the buccal mucosa[3].

The differential diagnoses of OLP include drug-induced lichenoid reactions. In most cases, lichenoid reactions are indistinguishable from OLP given the histological and clinical similarities[4]. This hypothesis was discarded because no factor could have triggered this reaction. The patient was not taking any of the medications known to cause this reaction (psychotropic agents, antidiabetics, statins, antimalarials, antifungals, and antiretrovirals, among others)[9]. Lichenoid reactions can also be the result of contact hypersensitivity to dental materials (amalgam, nickel, mercury, copper, or gold), but the patient did not have any metallic restoration and the lesions regressed three months before the removal of the orthodontic appliance. There were no changes in the location and size of the lesions, a typical finding in migratory glossitis, over the course of one year before they disappeared. Therefore, this hypothesis was discarded as well.

Table 1: Complementary exams performed before the biopsy

Complementary exam	Results	Reference ranges
Red blood cells	5.4 million/mm³	4.4-6 million/mm³
Hemoglobin	16 g/dL	12.5-18 g/dL
Hematocrit	46.4%	40-54%
MCV	85.93 fL	79-98 fL
MCH	29.63 pg	25-35 pg
MCHC	34.48 g/dL	31-36 g/dL
RDW	12.40%	10-15%
Platelets	220,000/mm³	150,000-400,000/mm³
MPV	8.5 fL	7-11 fL
Leukocytes	5,900/mm³	4,500-11,000/mm³
Eosinophils	59/mm³	0-714/mm³
Basophils	0/mm³	0-204/mm³
Typical lymphocytes	2,065/mm³	1,150-4,590/mm³
Atypical lymphocytes	0/mm³	0/mm³
Monocytes	354/mm³	0-1,224/mm³
Blast cells	0/mm³	0/mm³
Myelocytes	0/mm³	0/mm³
Metamyelocytes	0/mm³	0-100/mm³
Neutrophils	3,422/mm³	1,800-8,000/mm³
Band neutrophils	118/mm³	150-600/mm³
Segmented neutrophils	3,304/mm³	1,380-6,120/mm³
Glucose levels	91 mg/dL	60-110 mg/dL
Potassium	4 mEq/L	3.5-5.5 mEq/L
Sodium	133 mEq/L	130-146 mEq/L
Thyroid-stimulating hormone	0.739 mUI/mL	0.5-5 mUI/mL
Triglycerides	55 mg/dL	10-200 mg/dL
Creatinine	0.82 mg/dL	0.4-1.4 mg/dL
Urea	29 mg/dL	15-50 mg/dL
Cholesterol	164 mg/dL	>170 mg/dL
HDL cholesterol	50 mg/dL	≥ 35 mg/dL

MCV: Mean Corpuscular Volume; MCH: Mean Corpuscular Hemoglobin; MCHC: Mean Corpuscular Hemoglobin Concentration; RDW: Red cell Distribution Width; MPV: Mean Platelet Volume

The patient also reported a history of atopic dermatitis which persisted for seven years during his childhood and then regressed spontaneously. Although there is still no proven association between atopic dermatitis and lichen planus, there are some reports in the literature involving both diseases. Even though many studies have discussed the influence of stress and anxiety in the onset and evolution of OLP[3,4,7,10], their role is still unclear[12].

Some studies suggest an association between OLP and hepatitis C virus, but this hypothesis was also discarded since the patient was not exposed to the risk factors to acquire this infection: he never shared needles, never underwent hemodialysis nor needed a blood transfusion or organ transplantation, besides living in a non-endemic area. OLP occurs more frequently in young patients of Asian origin, with this being the third case of a young patient reported in Brazil.

The low expression of IL-1β and TNF-α could be justified by the fact that the lesion was asymptomatic and from the plaque-like/reticular forms. Previous studies demonstrated a significantly lower expression of many cytokines, including TNF-α, in the saliva of patients with reticular-type lesions when compared to patients with erosive/ulcerative forms, suggesting an association between the clinical form and the expression of cytokines[11].

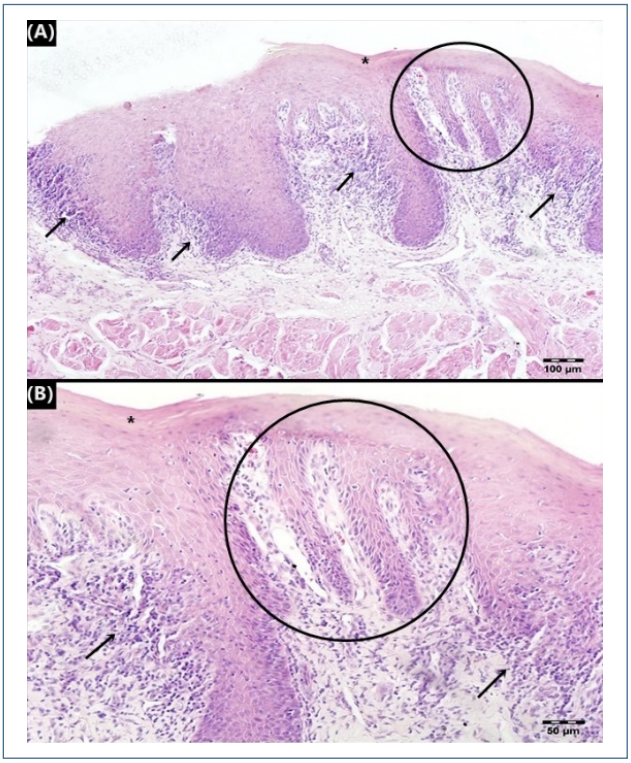

Figure 2: H.E. photomicrographs showing hyperparakeratosis (*), areas of atrophic epithelium with saw-tooth rete pegs (circled area), and liquefactive degeneration of the basal cell with a dense band-like lymphocytic infiltrate at the interface between the epithelium and the connective tissue (arrows) (Magnification of 100x (A) and 200x (B)).

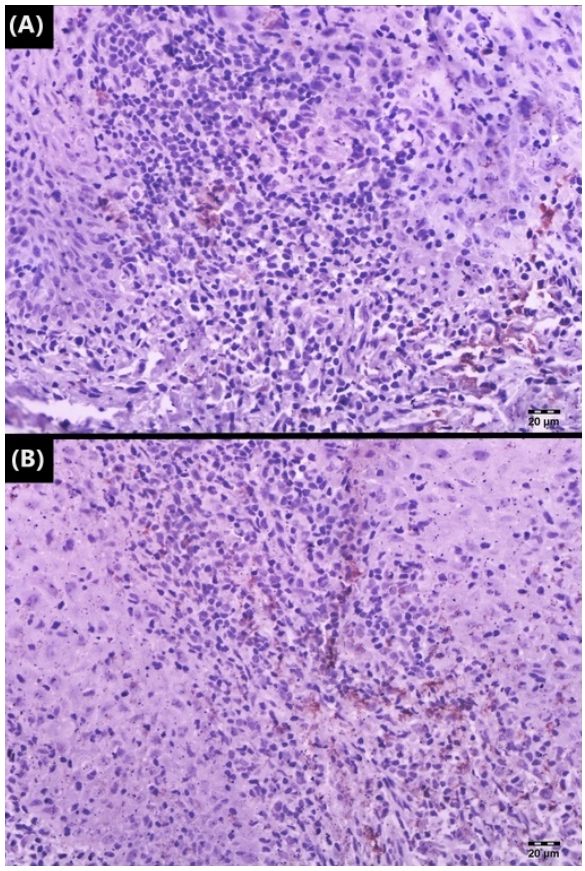

Figure 3: Photomicrographs showing the expression of IL-1β (A) and TNF-α (B) in the basal layer of epithelium (Magnification of 400x).

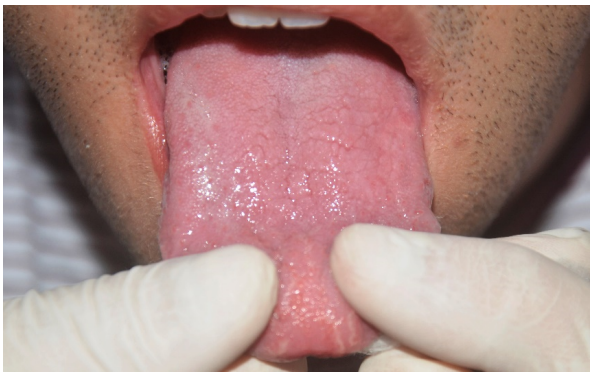

Figure 4: Clinical aspect of the lesions after nine years of follow-up, showing no signs of recurrence.

Oral lichen planus is a rarely observed condition in children and youth, and the mechanisms by which it is established remain unclear. In this report, there was spontaneous remission of both lesions one year after diagnosis with no signs of recurrence during the nine-year follow-up. OLP has a much fairer prognosis in young patients[15]. The present case highlights the importance of considering oral lichen planus in the diagnosis of hyperkeratotic lesions of the oral mucosa of young patients.

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