Case Report

Atypical Case of Oral Lichen Planus in a Pediatric Patient: Clinical Presentation and Management

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Abstract: *Lichen planus (LP) is a mucocutaneous disease of unknown etiology that is relatively common in adults but rarely present in childhood. LP has been documented in dental and medical literature; however, there are few cases with oral involvement in children. The purpose of this paper was to report an unusual case of oral lichen planus involving the upper lip in a 7-year-old girl. A diagnosis was made based on clinical examination and histopathology features. The treatment consisted of topical corticosteroid and intralesional injection. After treatment with an intralesional corticosteroid, a complete remission of lesions involving the lip was observed. The 3-year follow-up, however, revealed asymptomatic lichenoid bilaterally affecting the buccal mucosa. The patient is currently under regular review. (Pediatr Dent 2011;33:445-7) Received March 15, 2010 / Last Revision April 29, 2010 / Accepted April 29, 2010*

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Lichen planus (**LP**), first described by Erasmus Wilson in 1869, is a pruritic dermatosis[1,2] and a mucocutaneous disease. Although the etiology of LP remains unknown,[1,3,4] an immunopathogenic basis has been proposed as the most favored etiology for LP.[5-8] LP occurs in association with other autoimmune diseases such as: lupus erythmatosus; pemphigus; Sjogren's syndrome and autoimmune liver disease; rheumatoid arthritis; and dermatomyositis.[3,6] According to Nnoruka,[5] a hepatitis C virus has been proposed as the likely antigen that triggers the LP reaction in predisposed individuals. Tasanen et al.[9] recently reported a rare case of LP after simultaneous measles-mumps-rubella and diphtheria-tetanus-pertussis-polio vaccinations in a 6-year-old boy. The skin symptoms in this patient were long-lasting and severe. LP was also associated with the Koebner phenomenon,[5] which refers to the development of isomorphic lesions in the traumatized area of normal skin in certain skin diseases.

LP can affect the skin, scalp, nails, and mucous membranes and predominately affects adults.[10] It is worldwide in distribution, and the general LP prevalence rate varies from 0.3% to 0.8% in adults.[11] Overall LP equally affects both sexes.[12] Nnoruka[5] mentioned that childhood LP is unusual; the author found a prevalence of less than 0.1%.

The oral manifestation in adults is more frequently resistant and persistent than the cutaneous type. Three varieties are recognized: reticular, plaque-like, and atrophic/erosive lesions.[4] The typical age of presentation is between 30 and 60-years-old, and the condition is more frequently seen in women.[13] Recently, oral lichen planus (**OLP**) has a prevalence rate reported between 0.1% and 4%.[14] OLP occurs mostly on the buccal mucosa, but the gingivae, tongue, floor of the mouth, and retromalar pads also may be affected.[7] Approximately 30% to 50% of patients with oral lesions also have cutaneous disease.[15]

According to Patel et al.,[16] reports in the literature describing children with LP have tended to show the majority as having cutaneous manifestations and a low incidence of oral involvement. Handa and Sahoo[2] found approximately 14% with oral involvement in a study of 87 children with LP; however, Sharma and Maheswari[17] observed a 30% incidence of OLP in children in a report of 50 cases. Scully et al.[18] reported 3 10- to 11-year-old females with OLP. Woo et al.[6] documented cases in the literature of juvenile LP, which is defined in patients younger than 20-years-old. The authors reported 2 children who presented clinically and microscopically with OLP.

A diagnosis of OLP lesions can be made based on clinical features if they are sufficiently characteristic, particularly when presenting in the "classic" reticular form; however, a biopsy is recommended to confirm the diagnosis.[4,13] The classic histopathologic features of OLP show: liquefaction of the basal cell layer accompanied by apoptosis of the keratinocytes; a dense, band-like lymphocytic infiltrate at the interface between the epithelium and the connective tissue; focal areas of hyperkeratinized epithelium; and occasional areas of atrophic epithelium where the rete pegs may be shortened and pointed.[15]

Several therapies have been investigated for the treatment of OLP, with varying results. The classic treatment is based on topical or intralesional injection of corticosteroids. Other

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alternatives to the treatment include: cyclosporine; creams based on retinoids; anti-inflammatories or antipruritics; use of tacrolimus and ultraviolet light therapy[4]. According to Choonhakarn et al.,[19] aloe vera gel can be considered a safe alternative treatment for OLP patients.

The purpose of this paper was to present an unusual case of oral lichen planus involving the upper lip in a 7-year-old girl and highlight the therapeutic management and a 3-year follow-up. Early diagnosis and treatment of OLP by pediatric dentists can have a significant impact on the oral health of affected children.

Case report

A 7-year-old Caucasian girl was referred to the Stomatology Department of São Leopoldo Mandic Dental School, Campinas, Brazil, regarding discomfort in her upper lip. Her parents reported a completely negative medical history of diseases related to her hypersensitivity. The patient had received hepatitis B vaccinations at 1-year-old. The hepatitis B vaccination protocol adopted in Brazil recommends the first, second, and third vaccinations, respectively, at 1, 2, and 3-month-old. The patient reported that she used to bite her oral mucosa.

A clinical examination revealed atrophy and labial disfigurement on the vermilion of the upper lip (Figure 1). An oral examination showed reticular white plaque and hyperkeratotic striaes, measuring approximately 1 cm x 1 cm involving the upper lip (Figure 2). Our hypothesis was atrophic LP. An incisional biopsy confirmed the clinical diagnosis of LP (Figures 3 and 4).

The condition was initially treated with a topical clobetasol propionate gel (0.05%) 4 times per day. No resolution occurred after 1 month. After this, intralesional corticosteroid was indicated, and the patient received 3 injections of aqueous triamcinolone acetonide (40 mg/ml with applications of 1 ml/cm^2, 1 injection per week over 3 weeks). After this treatment, the symptoms of discomfort experienced previously had been reduced, and the clinical aspect was resolved (Figure 5). Follow-up 3 years later, however, revealed asymptomatic lichenoid bilaterally affecting the buccal mucosa (Figures 6 and 7). A new incisional biopsy of the right buccal mucosa confirmed a diagnosis of LP again. Because these lesions are asymptomatic, no active treatment was provided, and the patient is currently under regular review.

Discussion

LP, an inflammatory mucocutaneous disease, first described by Erasmus Wilson in 1869, is a very common disease in adults and rare in pediatric patients.[3,5,10,11,17,20] LP's etiology remains obscure,[1-4] but recently the most accepted pathogenic mechanisms probably had an immune-mediated basis. LP has been reported as a complication of hepatitis B vaccination in both children and adults.[9,20,21] There are no adequate epidemiologic data, however, to demonstrate a comparison of its prevalence before and after the introduction of large-scale recombinant hepatitis virus (**HBV**) immunization.[21] Reactions to MMR (measles-mumps-rubella) and DTaP-IPV (diphtheria-tetanus-pertussis-polio) vaccinations can be related to this disease.[9] Koebner phenomenon could also be associated with LP's

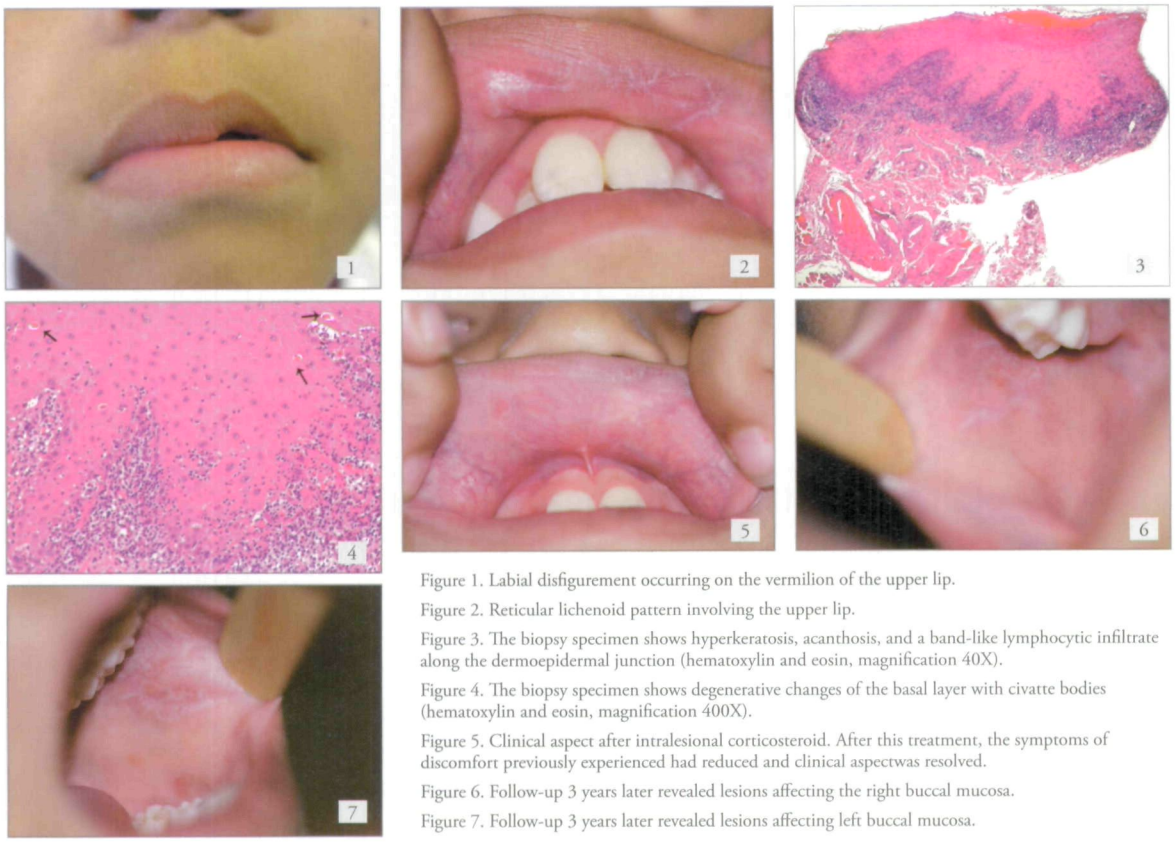

Figure 1. Labial disfigurement occurring on the vermilion of the upper lip.

Figure 2. Reticular lichenoid pattern involving the upper lip.

Figure 3. The biopsy specimen shows hyperkeratosis, acanthosis, and a band-like lymphocytic infiltrate along the dermoepidermal junction (hematoxylin and eosin, magnification 40X).

Figure 4. The biopsy specimen shows degenerative changes of the basal layer with civatte bodies (hematoxylin and eosin, magnification 400X).

Figure 5. Clinical aspect after intralesional corticosteroid. After this treatment, the symptoms of discomfort previously experienced had reduced and clinical aspectwas resolved.

Figure 6. Follow-up 3 years later revealed lesions affecting the right buccal mucosa.

Figure 7. Follow-up 3 years later revealed lesions affecting left buccal mucosa.

etiology.[5] In addition, a history of a pre-existing dermatologic disorder was observed in the literature.[18]

There are no established data for the prevalence of OLP, because reports of it in children consist of isolated cases or small case series.[1,17] Culver[22] reported the first case in a pediatric patient in 1920. Childhood LP is common in the tropics, and children of Asian origin may be more prone to this condition.[16] Previous studies[10,20] reported that children of Indian origin are more susceptible to LP. Handa and Sahoo[2] examined 87 cases of LP in children and observed an incidence rate of approximately 14% with oral involvement. Mucosal lesions frequently have a bilateral, symmetrical distribution and commonly take the form of minute white papules that gradually enlarge and coalesce to form either an annular, reticular, or plaque-like pattern.[13]

In the present case, we report an unusual case of symptomatic LP involving multiple sites in a 7-year-old girl. She had a completely negative medical history, was on no medication, and had no family members with LP. The association with HBV vaccine cannot be attributed in this case, because the patient had received hepatitis B vaccinations at 1-year-old, which eliminated this possibility.[9,21] According to the literature review, this association is unclear. Limas and Limas[21] investigated the possible association of HBV vaccines with childhood LP or LP-like eruptions seen in a hospital over a 3-year period. The data demonstrated that LP rarely occurs in children receiving the HBV vaccine. The HBV vaccination may induce the development of LP-like reactions in a very limited percentage.[21]

As mentioned, the Koebner phenomenon could have triggered development of the lesions. This agrees with Nnoruka,[5] who reported a relationship between this phenomenon and the occurrence of LP.

Use of topical corticosteroid is the most widely accepted treatment for OLP lesions because it modulates the patient's immune response. Several therapies, including intraregional injection, retinoids, dapsona, tacrolimus, and ultraviolet light, have been tried, with varying results.[3,4,8] In the currently reported case, the patient was treated initially with a topical corticosteroid (clobetasol propionate 0.05%). No resolution occurred after 1 month. After this, intralesional corticosteroid was indicated. Marked clinical improvement followed after 1 month of treatment. Previous reports showed a good response after intralesional injection.[4,10,17]

In the present case, however, the 3-year follow-up revealed new lesions bilaterally affecting the buccal mucosa. A new incisional biopsy of the right buccal mucosa confirmed a diagnosis of LP again. The patient has been currently under regular review since periodic follow-up of all OLP patients is recommended. There is controversy in the literature about the possible premalignant character of OLP.

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