CASE REPORT

Juvenile oral erosive lichen planus - A rare case report

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Abstract

Lichen planus (LP) is a chronic autoimmune mucocutaneous disease that predominantly affects adults and occurs less frequently in the pediatric patients. Oral LP in childhood is a rare entity with very few cases cited in the literature. The objective of this paper is to contribute a clinically and histologically documented case of juvenile oral LP to the literature. Despite the fact of rare occurrence, early identification and diagnosis of this condition by dental specialists can have a huge impact on the oral health of affected pediatric patients.

Keywords: Erosive, juvenile, oral lichen planus, pediatric

Introduction

Lichen planus (LP) is a common chronic inflammatory mucocutaneous disease.[1] It is estimated that approximately 50-70% of adult LP patients have both skin and oral lesions and only 25% of patients present with predominant oral lesions.[2] By contrast, oral LP (OLP) in childhood is extremely rare.[3] The exact incidence of pediatric LP is ambiguous, but various retrospective analysis has reported the incidence of 1-16% of LP in patients younger than 15 years old.[4] Juvenile OLP, is defined as OLP in patients younger than 20 years old, has barely been documented in the medical/dental literature. This article presents a brief review of a case report of juvenile erosive OLP and emphasizes the importance of considering OLP in the differential diagnosis of hyperkeratotic lesions affecting the oral mucosa in children.

Case Report

A 6-year-old male patient reported with the chief complaint of burning sensation in the mouth since 4 months. Burning sensation was only during consumption of hot and spicy food. Medical and dental history and review of systems were noncontributory. On examination, he appeared to be a healthy with no skin lesions. Nails, scalp, and genitalia were normal. Oral examination showed an irregular erythematous patch with surrounding hyperkeratotic margins of size approximately 3 × 2.5 cm, in the right buccal mucosa. A similar but ovoid brownish patch with a central grey area of approximately 1 cm was noticed in the left buccal mucosa (Figures 1 and 2). The lesions were tender and non-scarable.

Dental hard tissue examination revealed dental caries in 51 and 52. The periodontal status appeared to be normal. Based on the above history and clinical findings of the patient, a provisional diagnosis of oral erosive LP was considered. Routine hematological and biochemical investigations were negative. An incisional biopsy of the right buccal mucosa was done, histopathological reports confirmed the lesion to be erosive LP showing hyperparakeratinized epithelium with basal cell degeneration and dense lymphocytic infiltration (Figure 3). Based on these findings the clinical differential diagnosis of erythematosus candidiasis was ruled out. As the clinical and histopathological features were consistent with WHO criteria (2003)[5] the final diagnosis of oral erosive LP was arrived at.

The patient was advised to undergo all indicated dental treatments such as oral prophylaxis and restoration of carious teeth. General measures for management of the oral LP (OLP) included meticulous oral hygiene and avoidance of any form of physical injury to the oral mucosa. The patient was instructed to have a diet rich in fresh vegetables and fruits. Specific treatment for OLP prescribed was topical 0.1% triamcinolone acetonide combined with 0.03% tacrolimus and topical anesthetic as a palliative for a week to apply thrice daily.

The first review of the patient after one week showed a significant reduction of about 40-50% in both symptoms and signs of the oral lesions. At second follow-up after 16 days,
there was almost 80% improvement in the condition. 2 months later oral lesions had completely healed, leaving asymptomatic reticular areas (Figures 4 and 5). The patient was on periodic follow-up for 24 months.

Discussion

The term LP was first described by Erasmus Wilson in 1869. Though this condition was considered rare in children in older days, it is now being frequently reported in recent literature. Sharma et al., 1991 reported 50 children with LP, out of which 15 children were noticed with concomitant oral lesions. Kumar et al., 1993 reported 24 children with cutaneous lesions and only one child with both oral and cutaneous lesions. Handa et al., 2002 noticed 87 children with LP in India. Out of which only seven children had a concomitant involvement of the oral mucosa and only one patient had oral lesions without cutaneous involvement. A retrospective study by Chatterjee et al., 2012 described twenty-two children with OLP and the most common clinical form was found to be an erosive type, manifesting mainly in buccal mucosa. Juvenile LP is seen more common in tropics. The largest cases reported of childhood LP originate from India. In adults, OLP occurs more frequently in females, but reports of OLP in children showed discrepancies. Alam et al., 2001 reported six cases OLP in male children with age range of 6-14 years. Based on their series, the authors proposed a possible male predilection for juvenile OLP. Although reporting bias could be a contributing factor, an increased incidence of juvenile OLP reports is from India, China, United Kingdom, and Italy. The conditions commonly associated juvenile OLP include (1) previous history of hepatitis B vaccination; (2) liver disease and (3) genetic predisposition, as in familial LP. Alterations in clinical features of adult and juvenile LP have been noticed. Several authors have reported that LP in children could exhibit atypical features, like “linear” pattern uncommon seen in adults. In our patient, the clinical differential diagnosis of lichenoid reaction and discoid lupus erythematosus was considered. Treatment of juvenile OLP does not vary much from the treatment of adult OLP. Topical corticosteroid therapies are commonly given for symptomatic lesions. But, oral candidiasis can result due to chronic use of topical steroids. Systemic steroid therapy and dapsone are reserved for refractory and recurrent

Figure 1: Right buccal mucosa with erosive reticular margins

Figure 2: Left buccal mucosa with brownish patch surrounding central grey area

Figure 3: Photomicrograph depicting hyperparakeratinized epithelium with basal cell degeneration and dense lymphocytic infiltration

Figure 4: Post treatment - right buccal mucosa
cases. However, these group of drugs should be prescribed with extreme caution, as major long-term effects are of concern in this younger population. A thorough follow-up is vital in all patients with OLP. Malignant transformation of ulcerative OLP in adults is 0.07-5%; however, malignant transformation of OLP in children is not documented in the literature till now.

Conclusion

The case described in this paper documents the clinical and microscopic features of juvenile erosive OLP in a 6-year-old boy, with lesions predominantly of erosive OLP in the right and left buccal mucosa which is rarely seen in children. Our patient responded well to the combination treatment of topical 0.1% triamcinolone acetonide and 0.03% tacrolimus. Due to the lacunae in clinical trials, no consensus exists regarding standardized treatment regimens and the prognosis for LP in the pediatric population. On comparing the clinical features presented by this patient with the available literature review was found to be similar. As these mucosal lesions are unusual in the pediatric population, it is often misdiagnosed by practitioners. A better understanding and awareness of different clinical forms of juvenile OLP could help to make an early diagnosis and management of such rare lesions in children.

References