Received: 10 November 2022 | Accepted: 25 March 2023

DOI: 10.1111/pde.15318

Pediatric Dermatology WILEY

RESEARCH IN BRIEF

Oral lichen planus in children: An Italian case series

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Abstract

Oral lichen planus usually occurs in adults; there are no clear data regarding the incidence and the clinical features of oral lichen planus in children. This paper reports clinical findings, treatments, and outcomes of 13 Italian patients with oral lichen planus in childhood diagnosed between 2001 and 2021. The most common finding was keratotic lesions with reticular or papular/plaque-like patterns, confined to the tongue in seven patients. Although oral lichen planus in childhood is rare and the malignant transformation index is unknown, specialists must be aware of its characteristics and oral mucosal lesions must be correctly diagnosed and managed.

KEYWORDS

case series, childhood, lichen ruber planus, oral lichen planus, pediatric oral lichen planus

1 | INTRODUCTION

Lichen planus (LP) is an autoimmune chronic inflammatory disorder affecting the skin and mucous membranes in which autocytotoxic T-lymphocytes trigger apoptosis of epithelial cells.[1] The clinical features of oral lichen planus (OLP) are varied and six different patterns occur: reticular, plaque-like, atrophic, papular, erosive, and bullous.[1] The etiology remains unclear and related to several potential local and systemic triggers that activate T cell-mediated auto-cytotoxicity.

OLP usually occurs in the fourth decade with a prevalence rate of 0.1%–2.2%, with a female-to-male ratio of 1.4:1.[1] There are no clear data regarding the incidence of OLP in children (OLPc), although according to some authors the prevalence in children is <2%–3% of total cases. According to a recent systematic review, the oral mucosa is involved in 22% of pediatric patients, compared to 30%–70% of adult patients.[2] The low incidence of OLPc seems to be related to factors such as low incidence of systemic and autoimmune diseases, low levels of stress (considered a exacerbation trigger factor in adults), less usage of amalgam and orthodontic materials, which were recognized precipitating factors[3,4] in adults, as well as no tobacco consumption, which is a known risk factor for oral LP,[5] and fewer symptoms, leading to a number of undiagnosed cases.[6–9] This study reports clinical

TABLE 1 Clinical characteristics of the 13 patients with OLPc.

	Sex	Age (years)	Site	Lesions morphology	Pain	Concomitant autoimmune diseases	Histopathology	Treatment	Outcomes	Follow up (months)
1	F	6	Bilateral buccal mucosae	Papules with reticular pattern	Yes	No	Focal hyperkeratosis with irregular acanthosis, liquefaction of the basal membrane, and a dense band-like lymphocytic infiltrate in the superficial connective tissue	Topical corticosteroids[a]	Symptom remission (5 weeks)	12
2	F	13	Dorsum linguae	Plaque	Yes	No	Hyperkeratosis and hypergranulosis of the epithelium with a band-like inflammatory infiltrate in the superficial dermis, Civatte bodies associated with vacuolar degeneration in the basal cells layer	Topical corticosteroids[a]	Symptom remission (2 weeks) with 2 acute recurrences	24
3	M	10	Dorsum linguae	Ulcer surrounded by a thin white patch with reticulated borders	Yes	No	Dense lymphocytic infiltrate associated with degeneration of the epithelial basal cells and with a moderate degree of acanthosis and papillomatosis	Topical corticosteroids[a]	Symptom remission (3 weeks)	18
4	M	13	Dorsum linguae + Lingual margin	Papules with reticular and plaques	Yes	No	Hyperkeratotic epithelium with eosinophilic colloid bodies in the lower epidermis and superficial dermis	Topical corticosteroids[a]	Symptom remission (6 weeks)	15
5	F	14	Dorsum linguae + Bilateral buccal mucosae + Palatal	Papule with reticular pattern	No	Rheumatoid arthritis	NR	None[b]	Reduction of the extent of lesions	14
6	M	10	lingual margin + Bilateral buccal mucosae	Papule with reticular pattern	No	Autoimmune thyroiditis	NR	None	Reduction of the extent of lesions	24
7	M	6	Dorsum linguae	Blisters and plaques	Yes	No	NR	Topical corticosteroids[a]	Symptom remission (4 weeks)	36
8	M	13	Dorsum linguae	Papules	No	No	Hyperorthokeratinized stratified squamous epithelium with mild dysplasia	Topical corticosteroids[a]	No reduction of lesions	12
9	M	16	Dorsum linguae	Plaque	No	No	Hyperplastic squamous epithelium, acanthosis, hyperparakeratosis, edema, basal hyperplasia, mild dysplasia, intraepithelial lymphocytic exocytosis with inflammatory lichenoid infiltrate in subepithelial band arrangement and vacuolization of basal keratinocytes	None	No reduction of lesions	12
10	F	15	Dorsum linguae + Bilateral buccal mucosae	Papule with reticular pattern (buccal mucosa), plaque (dorsum linguae)	Yes	No	Variably thickened squamous epithelium, with focal papillomatosis, acanthosis, parakeratosis, and mild dysplasia. Lymphocyte infiltrate, fibrosis, vascular neoformation, and stromal activation present in the subepithelial chorion	Topical corticosteroids[a]	Partial symptom remission (4 weeks)	12

TABLE 1 (Continued)

	Sex	Age (years)	Site	Lesions morphology	Pain	Concomitant autoimmune diseases	Histopathology	Treatment	Outcomes	Follow up (months)
11	F	16	Ventrum linguae + Bilateral buccal mucosae	Papules and erythematous lesions	No	ANA+	NR	None	No reduction of lesions	18
12	M	15	Dorsum linguae + Lingual margin	Atrophic lesions and papules with reticular pattern	Yes	No	NR	Topical corticosteroids[a]	Symptom remission (6 weeks)	15
13	F	12	Dorsum linguae + Bilateral buccal mucosae	Papules	No	No	Stratified squamous epithelium with papillomatosis, acanthosis, parakeratosis, and subepithelial lymphocyte infiltrate	None	No reduction of lesions	11

[a]Clobetasol propionate 0.05% mixed with 4% hydroxyethyl cellulose gel to obtain a final concentration of 0.025% once or twice a day.
[b]Patient was under systemic steroid treatment for rheumatoid arthritis (Prednisone 0,25 mg/kg die).

findings, treatments, and outcomes of 13 Italian patients with OLPc diagnosed between 2001 and 2021.

2 | MATERIALS AND METHODS

A retrospective study of OLPc patients diagnosed between 2001 and 2021 was conducted at the Oral Medicine units of five Italian Universities. Patients younger than 18 years were enrolled in the study. Diagnosis of OLPc was confirmed by clinical findings, history, or histopathology when useful for differential diagnosis.

3 | RESULTS

Thirteen patients with OLPc were managed with a mean age at the time of diagnosis 12.2 years, (range 6–16 years). (Table 1). In no case was history of lichen planus, cutaneous lesions, or predisposing conditions such as active hepatitis identified. The most frequent pattern was the reticular pattern (7/13, 53.8%), followed by plaque-like (5/13, 38.4%), papular (3/13, 23%), erythematous/atrophic (2/13, 15.3%), bullous (1/13, 7.6%) and ulcerative (1/13, 7.6%) (Figure 1A,B) patterns. The tongue was the most involved oral site (12/13, 92.3%), followed by the buccal mucosa (6/13, 46.1%) and the palate (1/13, 7.6%). Seven patients (53.8%) were symptomatic. All patients were evaluated by dermatology and none showed skin involvement. When histological examination was necessary, it revealed the typical features of lichen planus (Figure 2). Two patients presented with concurrent autoimmune disorders (juvenile rheumatoid arthritis and autoimmune thyroiditis) (15.3%). Eight patients (61.5%) were treated with topical medications (clobetasol propionate 0.05% mixed with 4% hydroxyethyl cellulose gel to obtain a final concentration of 0.025% once or twice a day) and one with systemic steroids (prednisone 0,25 mg/kg/day) for concomitant systemic autoimmune disease (juvenile rheumatoid arthritis) (1/13, 7.6%). Six patients (46.1%) showed complete remission of the oral lesions, while three patients (23%) showed a reduction of clinical manifestations.

4 | DISCUSSION

Although OLPc was initially described in 1920s, there are few reports, and oral involvement has been often described in association with predisposing conditions, such as hepatitis and vaccination.[6,7,10,11] The frequency of OLPc is unknown, but it is thought to be higher in Asians[11,12] suggesting a genetic susceptibility, which could contribute to the high occurrence of LP in Asians.[7,10]

Several case studies have shown that a family history of lichen planus in children is unusual, occurring in only 1%–2% of cases.[13–15] In our cases, no family history of lichen planus, cutaneous lesions, or predisposing conditions such as active hepatitis were identified. In one case a family history for autoimmune diseases and allergic diathesis was found, whereas two patients presented with concurrent

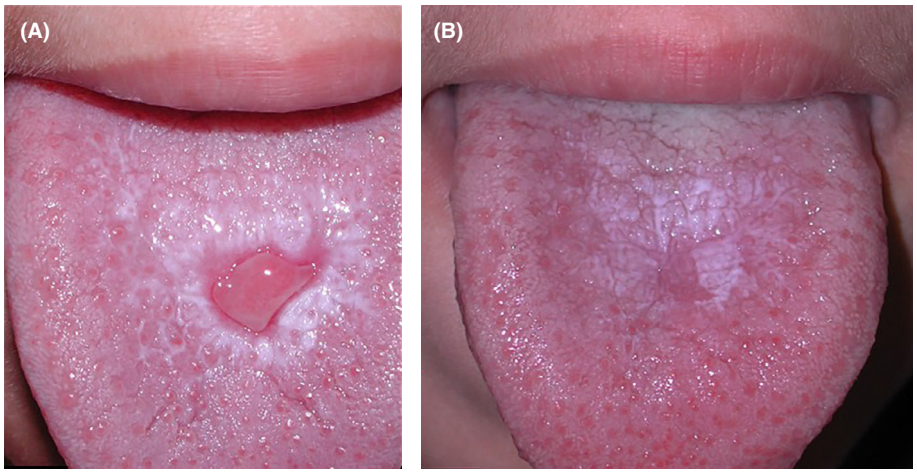

FIGURE 1 (A) Ulcer surrounded by hyperkeratotic white mucosa presenting plaques with interlacing lines, prior to treatment; (B). White asymptomatic areas remaining after complete healing of the ulcer after 3 weeks of topical treatment.

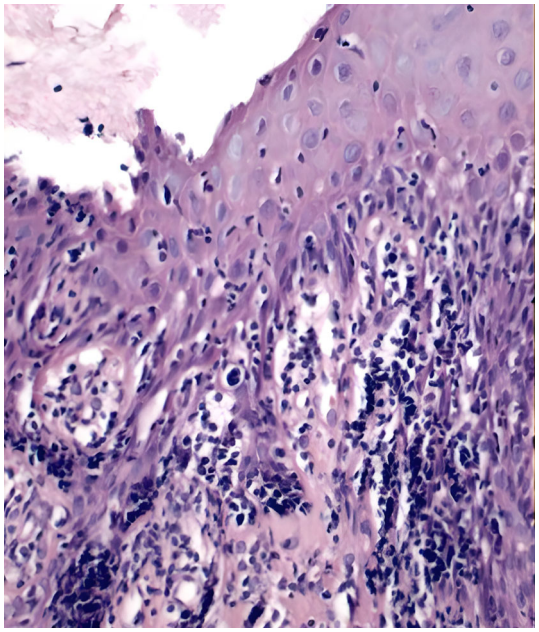

FIGURE 2 Histopathology showing dense lymphocytic infiltrate associated with degeneration of the epithelial basal cells and with a moderate degree of acanthosis and papillomatosis (H&E, ×200).

autoimmune disease (juvenile rheumatoid arthritis and autoimmune thyroiditis). Literature suggests that children with cutaneous LP had oral involvement in 12.6%–39% of cases.[2,16–18]

Among rare presentations of OLP in children, the most frequent patterns observed are keratotic lesions with reticular or papular/plaque-like patterns.[19] It has been suggested that childhood LP often shows atypical clinical features,[10,11] with buccal mucosa followed by tongue reported to be the most involved sites in childhood OLP.[7] This seems to be confirmed also in our series, with 7 of 13 patients having lesions confined to the tongue; this is uncommon in adults, who typically exhibit multiple oral sites of involvement.[20] Histologically, OLPc features do not differ from adult disease.

OLPc treatment outcomes seem to be more favorable than adult OLP.[21] In the absence of OLPc trial or consensus data, many different treatment modalities have been reported, ranging from local corticosteroids to systemic immunosuppression.[7,22] Herein, eight patients received treatment; topical corticosteroid for an average of 4.75 weeks was successful in obtaining a stable remission of symptoms in 7/8 patients. Only symptomatic patients were treated since the therapeutic regimen for oral lichen planus recommends starting therapy only when symptoms are present; otherwise, a 6-month follow-up is recommended. With an average of 17.8 months of follow-up, no patients experienced malignancy. Although OLPc is rare and the risk of malignant transformation is unknown, specialists must be aware of its characteristics and oral mucosa lesions must be correctly diagnosed and managed.

AUTHOR CONTRIBUTIONS

Conceptualization: Francesca Spirito and Lorenzo Lo Muzio. *Validation*: Lorenzo Lo Muzio and Eleonora Lo Muzio. *Data curation*: Giuseppina Campisi, Andrea Santarelli, Lucio Lo Russo, Corrado Rubini, Gianfranco Favia, Luisa Limongelli, Noemi Coppola, Stefania Leuci. *Writing—original draft preparation*: Francesca Spirito, Vito Carlo Alberto Caponio. *Writing—review and editing*: Francesca Spirito, Lorenzo Lo Muzio. *Supervision*: Stefania Leuci and Eleonora Lo Muzio. All authors have read and agreed to the published version of the manuscript.

FUNDING INFORMATION

This study received no external funding.

CONFLICT OF INTEREST STATEMENT

The authors declare that they have no competing interests.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

PATIENT CONSENT STATEMENT

Consent for publication was obtained from every patient involved in this study.

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How to cite this article: Spirito F, Caponio VCA, Lo Muzio E, et al. Oral lichen planus in children: An Italian case series. *Pediatr Dermatol*. 2023;1-5. doi:10.1111/pde.15318