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Report

Oral lichen planus in childhood: a case series

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Abstract

Background Although the exact incidence of pediatric oral lichen planus (OLP) is unknown, the oral mucosa seems to be less commonly involved, and the clinical presentation is often atypical. The aim of the study is to present a case series of OLP in childhood.

Methods From our database, we retrospectively selected and analyzed the clinical data of OLP patients under the age of 18 where the diagnosis had been confirmed by histopathological analysis.

Results The case series from our database shows eight patients, four males and four females. The mean (±SD) age at the time of diagnosis of the disease was 13.5 (±2.73) years, ranging in age from 9 to 17. Clinically, a reticular pattern was present in six patients (75%), and the tongue was the most commonly involved oral site (six cases, 75%). We also report the first case of OLP in a 9-year-old girl affected by autoimmune polyendocrinopathy-candidiasis-ectodermal dystrophy.

Conclusions We report the largest case series of pediatric OLP published in literature thus far. Differences in the disease between adults and pediatric patients have been detected, but further investigation and a larger case series are needed to establish any detailed differences in clinical outcomes.

Introduction

Lichen planus (LP) is a common, chronic inflammatory disorder that affects the skin, mucous membranes, nails, and scalp; the etiology of this condition is still unknown, although an immune-mediated pathogenesis has been hypothesized.[1] The age at onset is usually between the third and sixth decade of life and it is predominantly seen in females.[2]

Although the exact incidence of LP is unknown, it seems to vary between 0.1 and 1.2%. Children represent only 1–4% of patients with LP, and the clinical presentation is often atypical.[3]

Clinically, cutaneous LP is characterized by purple, polygonal, pruritic papules frequently covered by a lacy network of white scales on their surface, known as Wickham striae. The flat-topped papules are often located on the flexor surface of the wrist, the shins, the trunk, and the medial thighs, subdivided into one of the following variants: linear, hypertrophic, annular, follicular, actinic, vesiculobullous, and pemphigoid-like. The disease often resolves within 8–12 months of treatment, and it is not believed to be capable of malignant transformation.[4,5]

Any nail involvement may appear as a thinning of the nail plate, longitudinal fissuring, or distal splitting. Any hair follicle involvement is called lichen planopilaris and if untreated can lead to scarring alopecia. Any involvement of the mucous membranes can affect the oral mucosa, conjunctivae, larynx, esophagus, tonsils, bladder, vaginal vault, vulva, and anus.[5]

In contrast to skin LP, oral LP (OLP) demonstrates a clinical variability,[6] and the oral manifestation in adults is more frequently resistant and persistent than the cutaneous type.[7]

The oral lesions are categorized as reticular, papular, plaque-like, atrophic, erosive, or bullous.[8]

The hyperkeratotic variants are commonly asymptomatic, while the atrophic/erythematous variant, the erosive/ulcerative variant, and the bullous type often have persistent symptoms of pain or stinging aggravated during eating and drinking.[9,10]

The clinical differential diagnosis depends on the age of the patient, the clinical form of OLP, and the severity and persistence of the lesions and includes: lichenoid drug reaction, leukoplakia, lupus erythematosus, candidiasis, graft-versus-host disease (GVHD), frictional keratosis, autoimmune bullous diseases, erythema multiforme, allergic gingivostomatitis, and gluten sensitivity enteropathy.[10–12]

In challenging cases, more sensitive diagnostic techniques could be useful to achieve a diagnosis such as direct and indirect immunofluorescence.[13]

The aim of this study is to provide an update of the oral involvement of the disease in children through the report of a retrospective analysis of pediatric patients referred to our department during the last 4 years for whom LP has presented in the oral cavity as the single or as an additional site of involvement. We also conducted a literature review of the topic in order to highlight the similarities and differences between our data and the previously published clinical cases.

Subjects and methods

From our database, we retrospectively selected and analyzed the clinical data of pediatric OLP patients in the outpatient clinic of the Department of Neurosciences, Reproductive and Odontostomatological Sciences, Federico II University of Naples. The selection was based on the following inclusion criteria:

- age <18 years old at the time of diagnosis
- a clinical and histological diagnosis of OLP

The exclusion criteria were:

- GVHD lichenoid lesions
- the lack of a confirmatory histology
- oral lichenoid drug reaction
- the lack of any results of routine hematological testing including tests for hepatic and kidney functionality, markers of hepatitis A, B, and C viruses, and a red and white blood cell count and platelet count

From our database, we collected the following data: age at time of diagnosis, gender, preexisting medical conditions, presence of a positive family history of immunological disorders, concomitant or previous assumption of drugs, concomitant oral predisposing or iatrogenic factors, confirmatory histology, clinical pattern, oral sites involved, oral symptoms reported, extraoral sites involved, and the treatment and resolution of oral lesions.

Literature review

A PubMed search was carried out of articles published between 1966 and 2015 using the keywords “lichen” OR “lichenoid” alternatively matched with “oral” OR “lip” AND “juvenile” OR “child*” OR “familial” OR “pediatric”.

The selection of the studies was based on the following inclusion criteria:

- the English language
- a case series or case reports
- age <18 years old at the time of diagnosis
- clinical and histological diagnosis of OLP
- an accurate description of the oral sites and clinical features

The exclusion criteria were:

- lack of clarity in reporting data about the clinical form(s) of OLP and/or the oral site(s) involved
- GVHD lichenoid lesions
- lack of confirmatory histology
- oral lichenoid drug reaction

The study was approved by the Ethics Committee of the University of Naples “Federico II” in July 2014, and it conforms to the provisions of the Declaration of Helsinki (as revised in Tokyo 2004).

Results

The case series from our database shows eight patients, four males and four females. The mean (±SD) age at the time of diagnosis of the disease was 13.5 (±2.73) years, the patients ranging in age from 9 to 17.

Table 1 Flow chart

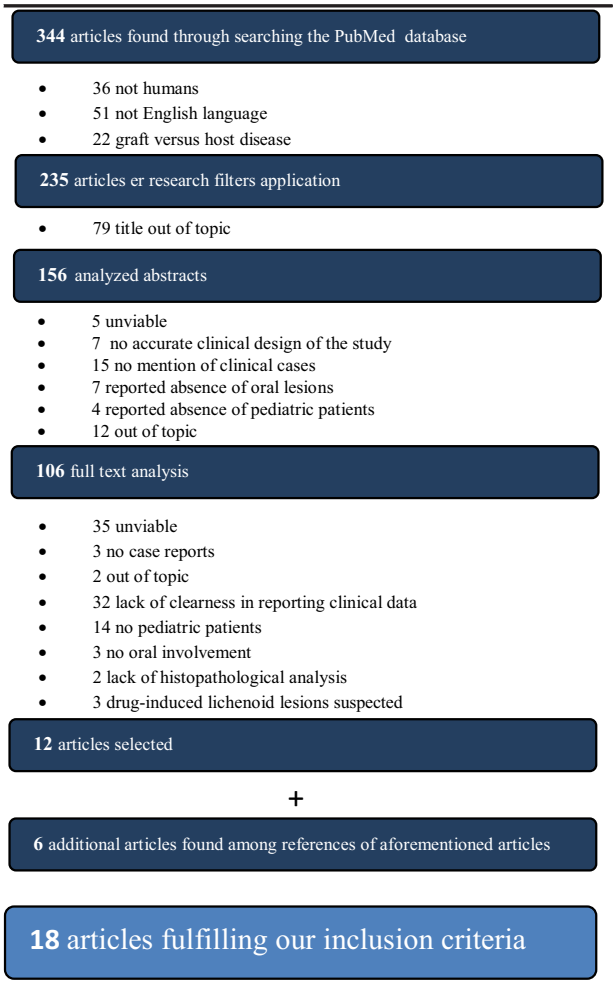

Table 2 Data on the eight patients from our case series: epidemiology, predisposing factors, clinical features, diagnosis, and treatment

Case	Age	Gender	Preexisting medical conditions	Family history	HBV vaccination status	Drugs	Concomitant oral factors	Confirmatory histology	Clinical pattern	Oral sites	Oral symptoms	Extra-oral sites	Treatment	Resolution of oral lesions
1	17	F	Allergy to NSAIDs	Atopy (mother)	Vaccinated	None	Parafunction	HE	Plaque-like + erosive plaque-like + erosive	Margins of tongue bilaterally ventrum of tongue bilaterally	Burning with spicy foods	None	Topical steroids topical antimycotics	None
2	17	M	Atopy, ↓ Hb	OLP (father)	Vaccinated	None	None	HE	Reticular plaque-like	Hard palate retromolar fossae bilaterally	Dryness	None	Topical antimycotics	None
3	14	F	None	None	Vaccinated	None	None	HE	Plaque-like plaque-like + atrophic plaque-like + atrophic plaque-like + pigmentation	Buccal mucosa right side margins of tongue bilaterally ventrum of tongue bilaterally lower lip	None	None	Topical antimycotics	Complete
4	14	M	Hg (NH_2) C patch test ++ IgG-EBV 159 U/l erythema dyschromicum perstans	None	Vaccinated	None	Orthodontics	HE	Reticular reticular reticular + atrophic reticular + atrophic	Buccal mucosae bilaterally gingiva dorsum of tongue bilaterally margins of tongue bilaterally	None	None	Topical antimycotics	None
5	15	M	Autoimmune thyroiditis atopy	Autoimmune thyroiditis (mother)	Vaccinated	None	None	HE	Reticular reticular	Buccal mucosae bilaterally ventrum of tongue bilaterally	None	None	Topical antimycotics	Complete

Table 2 Continued

Case	Age	Gender	Preexisting medical conditions	Family history	HBV vaccination status	Drugs	Concomitant oral factors	Confirmatory histology	Clinical pattern	Oral sites	Oral symptoms	Extra-oral sites	Treatment	Resolution of oral lesions
6	9	F	APECED	N/A	N/A	N/A	None	HE	Bullous + atrophic + reticular bullous + atrophic + reticular reticular bullous + atrophic + reticular	Dorsum of tongue margins of tongue bilaterally gingival buccal mucosae bilaterally	Pain	None	Topical steroids topical antimycotics	None
7	11	F	Atopy	OLP (father)	Vaccinated	None	Parafunction	HE	Reticular	Gingiva	None	None	Topical antimycotics	None
8	11	M	Atopy immune carrier for Mediterranean anemia	Crohn's disease rheumatoid arthritis	Vaccinated	None	None	HE	Reticular + atrophic	Dorsum of tongue	Roughness	None	Topical steroids topical antimycotics	None

APECED, autoimmune-polyendocrinopathy-candidiasis-ectodermal dystrophy; F, female; Hb, hemoglobin; HE, Hemotoxylin and eosin; IgG-EBV, Immunoglobulin G versus Epstein-Barr virus; M, male; N/A, not available, NSAIDs, nonsteroidal anti-inflammatory drugs.

Table 3 Data on the 24 patients from the PUBMED search: epidemiology, predisposing factors, clinical features, diagnosis, and treatment

Authors	Age	Gender	Preexisting medical conditions	Family history	Drugs	Concomitant oral factors	Confirmatory histology	Clinical pattern	Oral sites	Oral symptoms	Extra-oral sites	Treatment	Resolution of oral lesions
Chandna *et al.*[14] (2014)	7	F	None	None	None	None	HE (oral)	Reticular papular + reticular + pigmentation	Buccal mucosae bilaterally upper and lower lips	None	Skin	Oral hygiene topical steroids	Partial
Khandelwal *et al.*[15] (2013)	10	F	↓ Hb (10.2 g)	None	None	N/A	HE + DIF (oral) (oral)	Reticular + pigmentation reticular + pigmentation	Buccal mucosae bilaterally retromolar fossae bilaterally	Burning with spicy foods	None	Topical retinoids	None
Moger *et al.*[16] (2013)	7	F	None	None	None	Caries at 36, 63, 65, 74	HE + DIF (oral) (oral)	Reticular + atrophic + erosive + papular + pigmentation	Buccal mucosae bilaterally	Burning with spicy and hot foods	Skin	Topical antimycotic topical steroids topical anesthetic	Partial
Padmini *et al.*[17] (2013)	12	M	None	N/A	None	None	HE (oral)	Erosive	Dorsum of tongue	Burning with spicy foods	None	Topical steroids topical anesthetic topical antimycotic	Complete
Chaitra *et al.*[18] (2012)	9	F	None	N/A	None	None	HE (oral)	Reticular + erosive + pigmentation	Buccal mucosae bilaterally	Burning with spicy foods	N/A	Topical steroids topical anesthetic	None
Pendyala *et al.*[11] (2012)	17	M	None	None	None	None	HE (oral)	Bullous	Gingiva	Burning exacerbated by acid and spicy foods	None	Topical steroids topical NSAIDs antioxidants Chlorhexidine	Complete
De Moraes *et al.*[7] (2011)	7	F	None	N/A	None	Parafunction	2 × HE (oral)	Reticular reticular + atrophic	Buccal mucosae bilaterally upper lip	Discomfort (upper lip)	N/A	Topical steroids topical CNIs	Complete

Table 3 Continued

Authors	Age	Gender	Preexisting medical conditions	Family history	Drugs	Concomitant oral factors	Confirmatory histology	Clinical pattern	Oral sites	Oral symptoms	Extra-oral sites	Treatment	Resolution of oral lesions
Anuradha *et al.*[19] (2011)	9	F	None	None	N/A	N/A	HE + DIF (oral) (oral)	Reticular reticular	Buccal mucosae bilaterally retromolar fossae bilaterally	Burning with spicy foods	None	N/A	None
Sanjaya *et al.*[12] (2011)	9	M	None	None	None	N/A	2 × HE (oral + skin)	Reticular reticular reticular reticular	Buccal mucosae bilaterally hard palate lower and upper lips tongue	Burning with spicy foods	Skin	Systemic steroids	Complete
GunaShekar *et al.*[9] (2010)	7	M	None	None	None	Caries at 36, 37	HE (oral)	Reticular + atrophic reticular + atrophic reticular + atrophic reticular + atrophic reticular + pigmentation	Buccal mucosa right side margins of tongue bilaterally dorsum of tongue floor of mouth upper and lower lips	Soreness with spicy foods (tongue)	None	Chlorhexidine topical steroids	None
Mohan Das & Jp[20] (2009)	12	F	↓ Hb (9 g)	None	None	None	HE (oral)	Reticular + erosive + pigmentation	Buccal mucosae bilaterally	Burning on consuming food	None	Topical retinoids	None
Woo *et al.*[6] (2007)	9	F	ANA+	None	None	None	HE (oral)	Reticular + papular + erosive reticular + papular + erosive	Margins of tongue bilaterally ventrum of tongue	Dryness	None	Topical steroids systemic steroids	N/A
Woo *et al.*[6] (2007)	11	F	None	None	None	Orthodontics	HE (oral)	Reticular reticular	Buccal mucosae bilaterally ventrum of tongue bilaterally	None	None	Orthodontics removal	None

Table 3 Continued

Authors	Age	Gender	Preexisting medical conditions	Family history	Drugs	Concomitant oral factors	Confirmatory histology	Clinical pattern	Oral sites	Oral symptoms	Extra-oral sites	Treatment	Resolution of oral lesions
Singal[21] (2005)	11	M	N/A	OLP (father) OLP (grandmother)	N/A	N/A	HE (oral)	Plaque-like + pigmentation	Dorsum of tongue	Burning with hot and spicy foods	None	Topical steroids	Complete
Laeijendecker *et al.*[10] (2005)	11	F	Fragrance mix 8% pet patch test++	None	None	Four amalgam fillings	HE (oral)	Reticular	Buccal mucosae bilaterally	None	None	None	Complete
Laeijendecker *et al.*[10] (2005)	16	M	None	None	None	Eight amalgam fillings, poor oral hygiene	HE + DIF (oral)	Erosive erosive	Buccal mucosae bilaterally gingiva	Pain and stinging	None	Topical steroids topical anesthetic topical CNIs systemic steroids systemic CNIs	Complete
Laeijendecker *et al.*[10] (2005)	14	F	Atopy nickel sulfate 5% pet patch test++	None	Cetirizine	Orthodontics	Yes	Reticular reticular	Buccal mucosae bilaterally margins of tongue bilaterally	Soreness	None	Systemic steroids systemic CNIs	Complete
Patel *et al.*[1] (2005)	15	F	↓ferritin (8 μg/L) ↓IgA (0.60 g/L) hypothyroidism	None	None	None	HE (N/A)	Erosive + reticular erosive + reticular erosive + reticular	Margins of tongue bilaterally dorsum of tongue floor of mouth	Pain	Skin	Topical steroids	None
Patel *et al.*[1] (2005)	6	M	Autism	None	None	N/A	HE (oral)	Plaque-like	Dorsum of tongue	None	None	None	None
Alam & Hamburger[22] (2001)	6	M	None	None	None	N/A	HE (oral)	Reticular reticular	Buccal mucosa right side dorsum of tongue	N/A	None	None	Complete
Alam & Hamburger[22] (2001)	7	M	None	None	None	Poor oral hygiene	HE (oral)	Reticular + atrophic erosive	Buccal mucosae bilaterally gingiva	Soreness	None	Chlorhexidine topical steroids	None

Table 3 Continued

Authors	Age	Gender	Preexisting medical conditions	Family history	Drugs	Concomitant oral factors	Confirmatory histology	Clinical pattern	Oral sites	Oral symptoms	Extra-oral sites	Treatment	Resolution of oral lesions
Alam & Hamburger[22] (2001)	14	M	Asthma	N/A	Salbutamol and beclomethasone inhaler	N/A	HE (oral)	Reticular plaque-like	Buccal mucosae bilaterally margins of tongue bilaterally	Pain with spicy foods	N/A	None	Complete
Alam & Hamburger[22] (2001)	14	M	None	None	None	N/A	HE (oral)	Reticular + atrophic + pigmentation	Buccal mucosa left side	None	N/A	None	None
Scully *et al.*[23] (1994)	11	F	None	None	None	N/A	HE (oral)	Erosive	Margin of tongue	Soreness	None	Topical steroids	None
Scully *et al.*[23] (1994)	10	F	None	None	None	N/A	HE (oral)	Erosive erosive erosive	Buccal mucosae bilaterally ventrum of tongue floor of mouth	N/A	None	Topical steroids topical CNIs	Complete
Borrego Hernando *et al.*[24] (1992)	10	F	IgG anti-BMZ[i] 1:80 LPP[i]	N/A	N/A	N/A	HE + DIF (skin)	Reticular	Buccal mucosae bilaterally	N/A	Skin, vulvar mucosae	Systemic steroids	Complete

ANA, antinuclear antibodies; BMZ, basement membrane zone; CNIs, calcineurin inhibitors; DIF, direct immunofluorescence; F, female; Hb, hemoglobin; HE, Hemotoxylin and eosin; IgA, immunoglobulin A; LPP, lichen planus pemphigoides; M, male; NSAIDs, nonsteroidal anti-inflammatory drugs; N/A, not available.

Table 4 Comparison between our case series and the cases from the PubMed search

	Case series (n = 8)		Review cases (n = 26)		Total (n = 34)	
Age						
Overall age range (years)	9–17		6–17		6–17	
Mean (±SD) age	13.5	±2.73	10.41	±3.24	11.18	±3.40
Gender						
Males	4	50%	11	42.3%	15	46.8%
Females	4	50%	15	57.7%	19	55.8%
Familial OLP	3	37.5%	1	3.8%	4	11.7%
Preexisting medical conditions	7	87.5%	9	34.6%	16	47%
Immunological disorders	7	87.5%	5	19.2%	12	35.2%
Confirmatory histology	8	100%	26	100%	34	100%
Concomitant oral factors	3	37.5%	8	30.7%	11	32.3%
OLP clinical pattern						
Reticular	5	62.5%	19	79.1%	24	70.5%
Papular	0		3	11.5%	3	8.8%
Plaque-like	3	37.5%	3	11.5%	6	17.6%
Atrophic	3	37.5%	5	19.2%	8	23.5%
Erosive	1	12.5%	10	38.4%	11	32.3%
Bullous	1	12.5%	1	3.8%	2	5.8%
Mixed	5	62.5%	10	38.4%	15	44.1%
OLP site involvement						
Tongue	6	75%	13	50%	19	55.8%
Buccal mucosae	4	50%	19	73%	23	76.4%
Gingiva	3	37.5%	3	11.5%	6	17.6%
Retromolar fossae	1	12.5%	2	7.6%	3	8.8%
Lips	1	12.5%	4	15.4%	5	14.7%
Palate	1	12.5%	1	3.8%	2	5.8%
Floor of mouth	0		3	11.5%	3	8.8%
Extraoral involvement	0		5	19.2%	5	14.7%
Symptoms referral	4	50%	18	69.2%	22	64.7%
Complete resolution of oral lesions	2	25%	11	42.3%	13	38.2%

A positive familial history of immunological disorders was found in seven cases (87.5%). Seven patients had been submitted to hepatitis B virus (HBV) vaccination, and three (37.5%) patients presented concomitant oral factors. Findings and/or a history of an immune disorder were present in seven patients (87.5%).

For each patient, a confirmatory histology was obtained, and in no case was dysplasia reported.

A reticular pattern was the one most frequently reported, present in six (75%) patients, followed by the atrophic (50%), plaque-like (37.5%), erosive (12.5%), and bullous (12.5%) patterns; no patient showed a papular pattern. A simultaneous multiple clinical pattern was observed in six cases (75%), and in one patient, a mucosal pigmentation was detected.

The tongue was the most commonly involved oral site (six cases, 75%), followed by the buccal mucosa (four cases, 50%), gingiva (three cases, 37.5%), retromolar fossae (one case, 12.5%), palate (one case, 12.5%), and lip (one case, 12.5%). No patient showed any floor of the mouth or extraoral involvement. Four patients (50%) were symptomatic.

The most commonly used drugs were topical antifungal medications prescribed in order to avoid overlapping fungal overgrowth. Topical steroids were also associated in three cases.

Two patients (25%) showed complete disappearance of the oral lesions.

The analysis of the literature yielded 344 articles published between 1966 and 2015. After the application of the inclusion criteria, 12 articles were included in our study. We also searched among the references of the aforementioned articles and found a further six articles, making a total of 18 articles that fulfilled our inclusion criteria, which described a total of 26 patients.

The flow chart is reported in Table 1.

Data from our cases concerning the epidemiology, predisposing factors, clinical features, diagnosis, and treatment are recorded in Table 2.

Data from the PubMed search concerning the epidemiology, predisposing factors, clinical features, diagnosis, and treatment are recorded in Table 3.

A comparison between our cases and the review of literature is provided in Table 4.

Discussion

Many previous studies have reported that in LP among pediatric patients, the oral mucosa seems to be less commonly involved with a prevalence of approximately 0.03%[17] compared with 1–2% of the general population.[2]

The present case series confirms the epidemiological data previously collected concerning the pediatric LP population with a balanced M : F ratio[25] and a greater prevalence for familial LP in children (25%) than in adults. In fact, although LP is usually a sporadic disorder, there is a rare familial form more prevalent in the pediatric population ranging from 1 to 4.3%, with childhood familial LP considered to occur at an earlier age and with a greater severity.[3] An autosomal dominant mode of inheritance with a variable penetration has been suggested, and a linkage of familial LP with HLA-B7 and HLA-BR10 has been observed.[3]

The exact etiology of LP is unknown, but it appears to be complex and multifactorial. Possible cofactors of OLP, such as a hypersensitivity to dental restorative materials (e.g. amalgam and gold), local trauma (the Koebner phenomenon), and several kinds of infections (plaque-causing microorganisms and hepatitis B or C virus infection) have been reported.[10,25]

Furthermore, childhood LP has been documented as a complication of HBV vaccination, where the recombinant proteins of the HBV vaccine – specifically the viral S epitope – may trigger a cell-mediated autoimmune response targeted at the keratinocytes.[11]

For these reasons, in Tables 2 and 3 we have recorded the presence of any concomitant systemic and oral factors that could have had a role in the OLP pathogenesis or in its exacerbation; however, to date, these associations are still unclear.[7]

The medical histories collected from our cases confirm the presence of an increased association between OLP and autoimmune diseases,[1,3,7,10,11,22] with seven of our cases showing associated immunological disorders. Among our patients, we also report the first case of an oral lichenoid lesion related to autoimmune polyendocrinopathy-candidiasis-ectodermal dystrophy (APECED) also known as polyglandular autoimmune (PGA) syndrome type I, in a 9-year-old girl. Until now, there has been only one other case in the literature of OLP-associated autoimmune polyendocrine syndrome type II[26] in a 42-year-old woman, reinforcing the suggestion of a common immune-mediated pathogenesis between OLP and PGA (Fig. 1).

From the reported data in our case series, the reticular pattern appears to be the most common in childhood followed by the erosive one, in accordance with the literature. Interestingly, we instead recorded a clear difference, when comparing our eight cases to the literature (Table 4), concerning the oral site predilection with 75% of our eight pediatric patients showing lesions on the tongue. Previous studies have reported the buccal mucosa as the most commonly involved oral site in pediatric OLP with the next most common location being the tongue.[25] Finally, confirming the hypothesis of a less common oral involvement in pediatric patients with LP[25] previously reported at a rate of 12.6%,[27] none of our eight cases showed any extraoral involvement.

The histology of OLP has revealed that parakeratosis is the most frequent type of keratosis, while the erosive variety has involved the acanthotic epithelium in more than 50% of cases; the rete pegs are predominantly of a wavy pattern, while basal cell degeneration and band-like subepithelial lymphocytic infiltration seem to be present in all cases. Our data are consistent with the previous literature with basal cell degeneration and band-like subepithelial lymphocytic infiltration present in all of our eight cases.[25]

The treatment of juvenile OLP does not differ significantly from the treatment of adult OLP and is often unnecessary in asymptomatic patients. Oral symptoms are relatively frequent

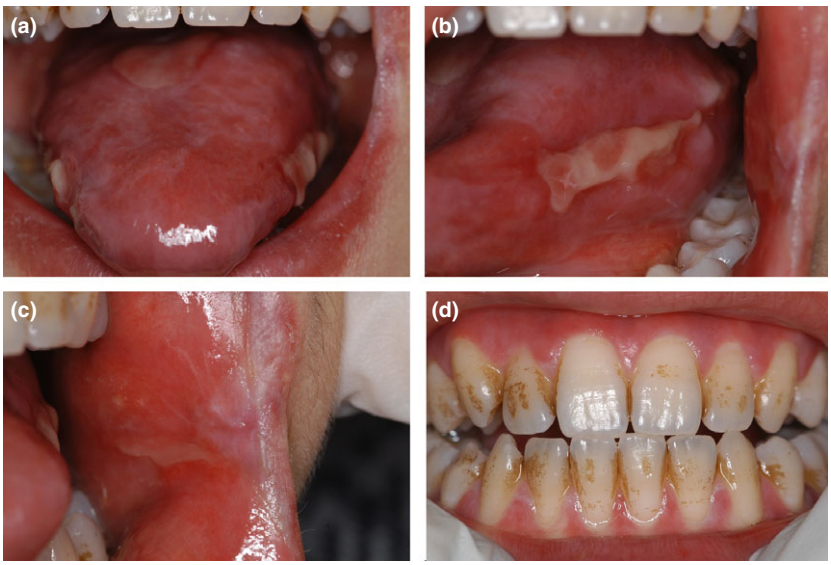

Figure 1 Case no. 9, a 9-year-old patient affected by APECED. (a) Bullous lesions involving the dorsum and margins of the tongue bilaterally and interlaced by reticular keratotic, erythematous, and atrophic aspects of the epithelium; (b) bullous lesion involving the left margin of the tongue surrounded by reticular keratotic, erythematous, and atrophic aspects of the epithelium; (c) bullous lesion involving the left buccal mucosa surrounded by reticular keratotic, erythematous, and atrophic aspects of the epithelium; (d) reticular keratotic lesions of the upper and lower gingiva

Table 5 Comparison between OLP in childhood and adulthood

Topic	Childhood	Adulthood
OLP frequency	0.03%[17]	1–2%[2]
Familial OLP	1–4%[3]	1.5%[21]
Most common clinical pattern	Reticular erosive	Reticular (83.5%)[2] erosive (15–39%)[2,6]
Most common oral sites involved	Buccal mucosae and tongue[25]	Buccal mucosae (88%) tongue and gingiva (18.7%)[2]
Involvement of both skin and oral mucosae	12.6%[27]	20–34%[2]
Histology	No dysplasia, basal cell degeneration and band-like subepithelial lymphocytic infiltration[25]	No dysplasia, basal cell degeneration and band-like subepithelial lymphocytic infiltration[13]
Treatment	Symptomatic[6]	Symptomatic[28]
Resolution	More frequent than in adults[17]	2–5%[29,30]
Malignant transformation of OLP	Never reported[10]	0.4–5.3%[2]

when the erosive and/or atrophic pattern occurs; although patients with a keratotic form can report a roughness, treatment is rarely necessary. In the present study, we have considered prescribing topical antifungal medications to all patients in order to avoid overlapping fungal overgrowth in patients undergoing contemporary topical steroidal therapy. The analysis of previously published studies confirms that topical corticosteroid therapy is the most commonly used treatment in symptomatic OLP, reported in connection with 12 of the 18 symptomatic pediatric patients (66.6%), even if the chronic use of topical steroids can lead to oral candidiasis; an association with retinoid therapy and a plaque control regimen in children has shown favorable responses.[11] Systemic steroid therapy and dapsone are typically reserved for refractory and recurrent cases; extreme caution is employed because significant long-term effects are of concern in this young patient population. Of note, tacrolimus ointment, topical tretinoin, and topical cyclosporine have also been used with success in some cases,[6] but the safety of any long-term continuous use of some of these drugs in pediatric patients has not been adequately evaluated.[25] The effect of the treatment of OLP in children seems to be more favorable than in adults for whom the symptoms usually persist for many years in spite of intensive treatment and a thorough investigation of any associated factors.[17] Considering our case series and the previously reported papers, a complete resolution of oral lesions has been observed in 38.2% of cases (Table 4).

In Table 5, we show a summary of the most important similarities and differences in OLP between children and adults so far reported in literature.

In conclusion, our case series mostly mirrors previous epidemiological, clinical, and therapeutic knowledge about pediatric OLP, but a larger case series is needed to confirm the possibility of a different oral site predilection between adults and children as suggested by our findings. Clinicians must be aware that OLP in childhood may also have a simultaneous or future involvement of the skin and other mucosal sites[6] and, because of a more frequent positive familial history of LP in childhood, close relatives should be examined. Although there have been no OLP-related malignancies described to date in the pediatric population, most previous studies suggest that the schedule of follow-up of pediatric OLP should be of at least one or two examinations per year as long as the OLP persists[25] even if the prognosis seems to be more favorable.[19]

What is new:

- the present study represents the largest case series so far published
- we report the first case of an oral lichenoid lesion related to APECED in a 9-year-old girl
- our cases suggest a different site predilection in OLP between children and adults
- the accurately tabulated review of the literature relating to pediatric OLP may facilitate further studies on the topic

Limitations of the study

Even if the present study represents the largest case series so far published, further studies are needed to establish the epidemiological and clinical features in this population of patients. The review of the literature conducted may be conditioned by previous biases in reporting exceptional cases on PUBMED, and therefore the results of the present comparison with previous studies should be critically considered.

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