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Oral lichen planus in children: A systematic review

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Abstract

Background: Oral Lichen Planus is a common chronic inflammatory disease of the oral mucosa. The prevalence in adults ranges between 0.5% and 2%, while in children is reported to be about 0,03%. Clinical features of Oral Lichen Planus could be variable in both adults and children, ranging from painless white hyperkeratotic lesions to painful erythematous atrophic ones.

Actually, there are no systematic reviews in the literature on OLP in children, whereby this paper aims to summarize all the pathophysiological aspects and identify all cases described in the literature of Oral Lichen Planus in children, reporting their clinical characteristics.

Material and Methods: A systematic review of the literature was performed in online databases including PubMed, Scopus, Web of Science, Science Direct, EMBASE. In addition, in order to identify reports not otherwise identifiable, an analysis of the gray literature was performed on google scholar and in Open Gray.

Results: By literature analysis, it emerged that most cases were reported from India. The mean age at time of diagnosis of the disease was 11 years, ranging from 3 to 17 years. The most frequent pattern was the reticular pattern followed by plaque-like, erosive, atrophic, sclerosus, and bullous. The buccal mucosa was the most involved oral site, followed by the tongue, lips and gingiva.

Conclusions: Although Oral Lichen Planus in children is rare, it may cause oral discomfort and need to be differentiated from other oral white lesions and/or chronic ulcers.

Key words: *Oral lichen planus, childhood, lichen ruber planus, paediatric lichen planus.*

Introduction

Oral Lichen Planus (OLP) is a common chronic inflammatory disease of oral mucosa (1,2). The prevalence in adults ranges between 0.5% and 2% (1), with the highest incidence in the third to sixth decade of life (3), and a male-to-female ratio ranging from 1:2 (4) to 1:3 (2). Disease prevalence in children is actually low: it is reported to be about 0.03% (5); most of the cases are from Asian countries, especially India (6,7).

The aetiology OLP is still unclear. It seems that an auto-

immune response (8) involving antigen-presenting cells and regulatory T-lymphocytes, probably triggered by keratinocytes, plays a key role in the development of the disease (9). Histopathologically, OLP shows typical degeneration of the basal cell layer with 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, occasionally, atrophic areas (10). A number of auto-immune diseases, including lupus erythematous, pemphigus, rheumatoid arthritis, and Sjogren's syndrome, have been reported in association to OLP (5).

For paediatric lichen planus, possible associations with hepatitis B virus (HBV) or hepatitis B vaccination have been described (11). Moreover, genetic factors, lifestyle, and emotional stress might be contributing factors in OLP pathogenesis (12,13). In particular, in patients with paediatric lichen planus a higher incidence of positive family history for the disease is reported (14); on the other hand, genetic linkage studies reported an association between familial lichen planus and HLA B, HLA DR1 and DR10 (15).

Clinical features of OLP could be variable in both adults and children, ranging from painless white hyperkeratotic lesions (frequently symmetrical with papules, plaques, or Wickham striae) to painful erythematous atrophic ones (sometimes with blisters, erosions and ulcerations) (16). Cutaneous lichen planus is often self-limiting, whereas OLP tends to be chronic: spontaneous remission has been rarely reported in OLP (2); in general, OLP prognosis in children seems to be more favourable (12) .

The differential diagnosis is strictly related to clinical features of OLP and includes oral candidiasis, morsicatio buccarum, lichenoid drug reaction, leucoplakia, lupus erythematous, graft versus host disease (GVHD), and others.

A recent review of the literature including case series was carried out by Shikha G. *et al*. (17). The analysis of the literature, even if not carried out in a systematic way, allowed to identify 28 manuscripts in the scientific literature for a total of 42 cases of OLP. From the results it emerges that the first case was reported in 1992 while the patient's age range was 3 to 14 years with a mean age of 9.4 years (males 20 and females 22).

At present there are no systematic reviews in the literature on OLP in children, while reviews on Lichen planus (LP) in pediatric age have been performed; Specifically, a recent systematic review has identified all cases described in the literature of lichen Planus pigmentosum which would represent 2.8% of Children with LP without referring to oral manifestations(18-20), and other reviews have focused on the role of vaccinations for hepatitis B in association with the onset of LP in pediatric age (19,20).

Given the importance of intercepting neoformations and oral precancerous diseases in children, it is essential to understand the real impact of OLP cases in children and its clinical course in terms of follow-up.

With this Review, therefore, we set ourselves the aim of summarizing all the pathophysiological aspects and identifying all cases described in the literature of OLP in children, reporting their clinical characteristics.

- List of abbreviations.

Oral Lichen Planus (OLP); Hepatitis B virus (HBV); Graft Versus Host Disease (GVHD); Lichen Planus (LP).

Material and Methods

- Protocol and registration

The planning of the systematic review was performed according to the recommendations of the Cochrane Handbook. The review was written following the indications of the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analysis) (21), the protocol was registered before to proceed with the drafting of the manuscript on the on the International Platform of Registered Systematic Review and Meta-analysis Protocols (INPLASY), with registration number INPLASY202290106 and DOI number 10.37766/inplasy2022.9.0106

- Eligibility criteria

All clinical studies (retrospective, prospective and randomized trials) case series and case reports reporting cases of OLP in children were considered potentially admissible, while systematic reviews on OLP were only consulted in order not to repeat the review work, and as a source of bibliographic references. The review question asked is to identify all clinical cases described in the literature of OLP in children, reporting their clinical and pathophysiological characteristics.

The selection of the studies was based on the following inclusion criteria; 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: Graft Versus Host Disease lichenoid lesions; No histology; Oral lichenoid drug reaction; No data about clinical form and/or oral sites involved.

No language restrictions were applied, therefore all studies and reports were taken into consideration, even in a language other than English, for which at least one English translation of the abstract was available.

- Sources of information, research and selection

Before proceeding with the research and selection phase of the studies, the 3 researchers assigned to carry out the research and selection were identified, the first 2 with the task of identifying the articles and the 3 to decide in doubtful situations.

The research and selection phase of the articles therefore took place in the following phases:

1) Choice of inclusion and exclusion criteria, databases

and keywords to be used and the time period in which to conduct the research;
2) Search and selection of records on databases carried out independently;
3) Removal of overlaps manually and using reference management software such as EndNote 8.0;
4) Choice of studies to be included (carried out independently by the 2 researchers);
5) Comparison of the included studies and resolution of any conflicts between the 2 reviewers with the help, if necessary, of a 3rd reviewer.
A systematic review of literature was performed in the online databases including PubMed, Scopus, Web of Science, Science Direct, EMBASE on 31 May 2022, in addition, an analysis of the gray literature was performed on google scholar and in Open Gray (DANS EASY Archive) in order to identify reports not otherwise identifiable.
All the relevant papers and reports published in English from January 1966 through may 2022 were extracted. Several combinations of keywords were used in the following orders to conduct the search strategy: 1) "Lichen Planus'' OR "Oral Lichen Planus'' AND 2) "Children" OR ''Child'' OR "Childhood" OR "Paediatric" OR ''Pediatric".
Specifically we report from PubMed, in detail all the combinations of keywords used by the portal:
Search: Lichen Planus AND (Children OR pediatric)
Sort by: Most Recent
("lichen planus"[MeSH Terms] OR ("lichen"[All Fields] AND "planus"[All Fields]) OR "lichen planus"[All Fields]) AND ("child"[MeSH Terms] OR "child"[All Fields] OR "children"[All Fields] OR "child s"[All Fields] OR "children s"[All Fields] OR "childrens"[All Fields] OR "childs"[All Fields] OR ("paediatrics"[All Fields] OR "pediatrics"[MeSH Terms] OR "pediatrics"[All Fields] OR "paediatric"[All Fields] OR "pediatric"[All Fields]))
Translations; Lichen Planus: "lichen planus"[MeSH Terms] OR ("lichen"[All Fields] AND "planus"[All Fields]) OR "lichen planus"[All Fields]
Children: "child"[MeSH Terms] OR "child"[All Fields] OR "children"[All Fields] OR "child's"[All Fields] OR "children's"[All Fields] OR "childrens"[All Fields] OR "childs"[All Fields]
pediatric: "paediatrics"[All Fields] OR "pediatrics"[MeSH Terms] OR "pediatrics"[All Fields] OR "paediatric"[All Fields] OR "pediatric"[All Fields]
Two independent investigators retrieved the studies that were the most relevant by titles and abstracts. Subsequently, the full text of the retrieved papers was reviewed, and the most relevant papers were chosen according to the eligibility criteria. Duplicate results were removed using the EndNote 8 software, the overlaps of studies that could not be uploaded to EndNote were manually removed after the screening phase.
In addition, an update of the research on bibliographic sources was carried out on 01 September 2023.

- Data collection process, Data characteristics.
The data to be extracted from the included articles were decided in advance by the 3 researchers and concerned: the first Author, the country where the study was conducted, the year of publication, the patient's age, sex, lesion morphology, the presence of pain, the cutaneous involvement of the lesions, the type of treatment and the follow-up. The data were reported by the 2 researchers in 2 different tables and subsequently compared and checked by the 3 researchers in order to minimize the error in reporting the data in a single table.
Then, the relevant data were extracted and organized in Table 1.
- Risk of Bias.
The risk of bias was evaluated using a tool relating to case reports. The tool used for case reports is the JBI critical appraisal checklist for case reports (22). The evaluation was performed by a researcher after the data extraction and inclusion phase of the studies.

Results

- Selection of studies
The analysis of literature between 1966 and 2023 allowed the selection of 7447 papers (Science Direct, SCOPUS, PubMed, EBSCO, Web of Science). After the application of the inclusion/exclusion criteria, only 36 papers (5,6,12,16,17,23-53) were included in the present systematic review (Fig. 1) with 65 cases (Table 1).
Furthermore, the analysis of the gray literature (http://www.opengrey.eu DANS EASY Archive and Google Scholar) and previous systematic reviews did not allow the identification of further OLP cases to be included in the qualitative assessment. The entire procedure of identification, selection and inclusion of the studies has been indicated in the flowchart of Fig. 1.
The risk of bias was assessed as acceptable for all included studies (Table 2)
- Data characteristics
First of all, if we consider the geographical location, the most interested country is India (23/65, 35.4%), followed by Italy (17/65, 26.1%), UK (7/65, 10.7%), USA (3/65, 4.6%), France (3/65, 4.6%) and Brazil (3/65, 4.6%). The mean age at time of diagnosis of the disease was 11 years, ranging from 3 to 17 years (Fig. 2). The most frequent pattern was the reticular pattern (28/65, 43%), followed by plaque-like (15/65, 23%), erosive/ulcerative (12/65 18.4%), papular (10/65, 15.3%), atrophic (8/65, 12.3%), sclerosus (3/65, 4.6%) and bullous (1/65, 1.5%) patterns. The buccal mucosa was the most commonly involved oral site (38/65, 58.4%), followed by tongue (27/65, 41.5%), lips (8/65, 12.3%) and gingiva (9/65, 13.8%). 32/65 patients (59.2%) were symptomatic. 12/65 (18.4%) patients showed skin involvement. 33 patients were male (50.7%) and 32 were female (49.2%). The treatment strategy are described in Table 1.

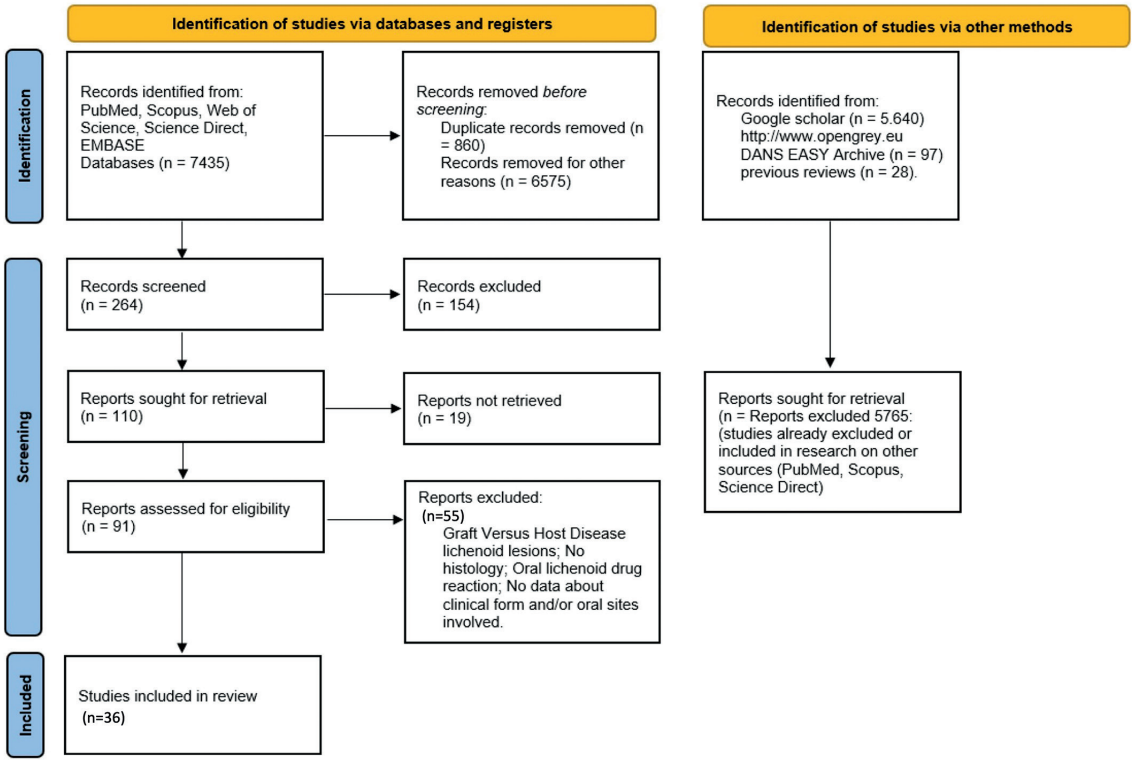

Fig. 1: Flow diagram for the selection process of identified articles.

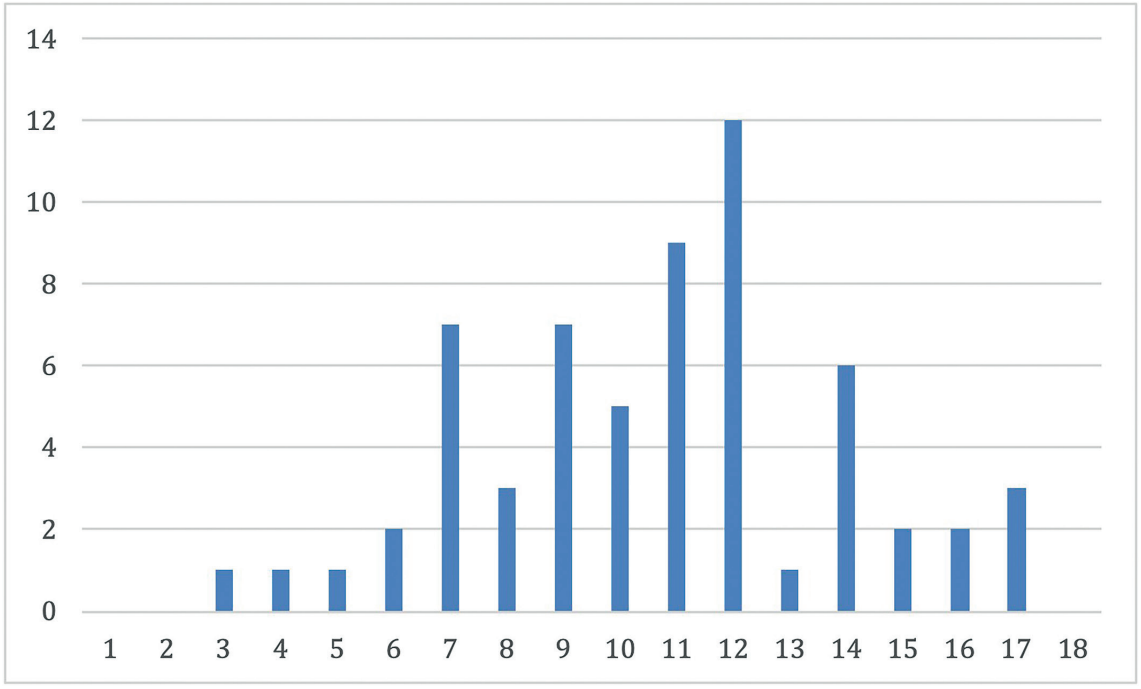

Fig. 2: Case distribution according to age.

Table 1: Studies of childhood LP in literature from 1980 to 2023.

Author	Country	Year	Age (years)	Sex	Lesions morphology	Pain	Skin involvement	Treatment	Follow up
Malhotra Y.K. *et al.* (case 1) (23)	Libya	1980	16	F	Multiple whitish streaks (buccal mucosa)	No	No	None	6 months
Silverman R.A. *et al.* (24)	USA	1984	9	F	Small groups of white papule and plaques on buccal mucosa	No	Yes (20-nail dystrophy)	None	2 years
Berbis P. *et al.* (25)	France	1987	12	M	Erosive OLP (tongue)	Yes	Yes (trunk, wrist, hands and feet)	Topical steroids + etretinate	7 years
Kanwar, A. *et al.* (6)	India	1991	9	F	Violaceous papules coalescing at places to form plaques over the lips	No	Yes (legs, face)	Topical corticosteroids + antihistamines	2 years
Borrego, H. *et al.* (26)	Spain	1992	10	F	Reticular OLP (buccal mucosa)	Not reported	Yes	Systemic steroids	Not reported
Scully, C. *et al.* (case 2) (27)	France	1994	11	F	Erosive lichenoid lesion on the lateral margin of the tongue	No	No	Topical corticosteroids	Not reported
Scully, C. *et al.* (case 3) (27)	France	1994	10	F	Erosive lesions in the floor of the mouth, extended to the tongue	Yes	No	Triamcinolone acetonide	Not reported
Bettoli, V. *et al.* (28)	Italy	1997	10	M	Groups of round yellowish papules located at the lower alveolar margin	No	Yes (all body)	None	8 months
Alam F. *et al.* (case 2) (5)	UK	2001	6	M	White patch on the tongue	No	No	None	2 years
Alam F. *et al.* (case 3) (5)	UK	2001	7	M	White striae were present in the right and left buccal mucosae extending into the retromolar fossae + desquamative gingivitis	No	No	Prednesol mouthwashes	4 years
Alam F. *et al.* (case 4) (5)	UK	2001	14	M	White striae on the right and left buccal mucosa and bilateral white patches on the margin of the tongue.	No	No	None	2 years
Alam F. *et al.* (case 5) (5)	UK	2001	14	M	Asymptomatic white lesion on his buccal mucosa. This was approximately 6 mmx12 mm in size, with faint white striae on a slightly atrophic base	No	No	None	Not reported
Alam F. *et al.* (case 6) (5)	UK	2001	11	M	Bilateral white lesions on his buccal mucosa (Reticular OLP)	No	No	None	Not reported
Sandhu K. *et al.* (case 1) (29)	India	2003	12	F	Whitish streaks in the oral mucosa	No	No	None	6 months
Laeijendecker M.D. *et al* (case 1) (12)	The Netherlands	2005	11	F	Reticular OLP (buccal mucosa)	No	No	None	3 years
Laeijendecker M.D. *et al* (case 2) (12)	The Netherlands	2005	16	M	Erosive OLP (buccal mucosa, gingiva)	Yes	No	Topical steroids (class II and IV) lidocaine gels, tacrolimus ointment; Systemic corticosteroids and systemic cyclosporine	4 years
Laeijendecker M.D. *et al.* (case 3) (12)	The Netherlands	2005	14	F	Reticular OLP (buccal mucosa, lateral part of the tongue)	Yes	No	Topical tretinoin 0.05% topical corticosteroids (class II)	2 years
Patel S. *et al.* (case 1) (30)	UK	2005	15	F	Ulceration and erythema with a surrounding margin of hyperkeratosis on both left and right lateral borders of the tongue extending onto the dorsal and ventral surfaces (Erosive OLP)	Yes	Yes (neck)	Steroids	Not reported
Patel S. *et al.* (case 2) (30)	UK	2005	6	M	White patch on the dorsal surface of the tongue. (Plaque-like OLP)	No	No	None	Not reported
Singal A. *et al* (case 3) (31)	Saudi Arabia	2005	11	M	Erythematous lesion with a violaceous hue, measuring 1.0 × 1.5 cm in size, on the right half of the tongue (Erosive OLP	Yes	No	Topical betamethasone dipropionate 0.05% in gel formulation	6 months
Woo V.L. *et al.* (case 1) (16)	USA	2007	9	F	Bilateral and symmetric ulcers were evident involving the right and left lateral ventral tongue surfaces (Erosive OLP)	Yes	No	Beclomethasone dipropionate inhaler and dexamethasone elixir	Not reported
Woo V.L. *et al.* (case 2) (16)	USA	2007	11	F	White, lacy striations were noted on the patient's right and left lateral tongue surfaces and left posterior buccal mucosa (Reticular OLP)	No	No	None	1 year

Table 1 cont.: Studies of childhood LP in literature from 1980 to 2023.

Azevedo R.S. *et al*. (case 3) (32)	Brazil	2009	11	F	Single white lesion on the upper lip, extending from the labial mucosa to vermillion of the lip (Lichen sclerosus)	No	No	None	No
Mohan Das U. and Jp, B. (33)	India	2009	12	F	Bilaterally bluish-purple striations in the posterior buccal sulci extending onto the buccal mucosa	Yes	No	Topical application of 0.05% Tretinoin cream for the first month + systemic steroid therapy	No
GunaShekar M. *et al*. (34)	India	2010	7	M	White striae affecting the posterior buccal mucosa, dorsal surface and lateral borders of the tongue, and the floor of the mouth	Yes	No	Topical corticosteroid cream containing 0.1 percent triamcinolone acetonide (Kenacort), when symptomatic.	3 months
De Moraes P.C. *et al*. (35)	Brazil	2011	7	F	Reticular white plaque and hyperkeratotic striaes, measuring approximately 1 cm x 1 cm involving the upper lip (atrophic LP)	Yes	No	Intralesional corticosteroid: 3 injections of aqueous triam-cinolone acetonide (40 mg/ml with applications of 1 ml/cm2, 1 injection per week over 3 weeks)	3 years
Chaitra T.R. *et al*. (36)	India	2012	9	F	White reticular lines and dark red and white patches on both the left and right buccal mucosa (Erosive OLP)	Yes	No	Topical corticosteroid (triamcinolone acetonide 0.1%) and analgesic (2% lidocain gel)	Not reported
Pendyala G. *et al*. (37)	India	2012	17	M	Red erosion of the connective tissue involving the entire maxillary and mandibular buccal and lingual/palatal surfaces was observed (Erosive OLP)	Yes	No	Triamcinolone acetonide 0.1% three times a day	1 year
Khandelwal V. *et al*. (38)	India	2013	10	F	Greyish brown patch with white striae were observed bilaterally in the posterior buccal mucosa extending into the retromolar fossae. (Reticular OLP)	Yes	No	Topical application of 0.05% Tretinoin cream	Not reported
Moger G. *et al*. (39)	India	2013	7	F	Grayish-white patches with white striae on both right and left buccal mucosa on an erythematous background. (Atrophic OLP)	Yes	Yes (back, hands)	Topical 0.1% triamcinolone acetonide combined with 1% clotrimazole. Topical anesthetic was given for palliation.	Not reported
Padmini C. *et al*. (40)	India	2013	12	M	A single irregular red and white ulcerative lesion measuring approximately 2.5 × 1.0 cm in size with granulation tissue at the centre surrounded by an inflammatory red border on the dorsum of the tongue. (Erosive OLP)	Yes	No	Topical 0.1% triamcinolone acetonide combined with 1% clotrimazole 3- 5 times per day for a duration of one week. Topical anesthetic was given for the pain relief.	Not reported
Chandna P. *et al*. (41)	India	2014	7	F	White lesions bilaterally on the buccal mucosa and purplish lesions on the lips and neck.	No	Yes (neck)	Topical corticosteroids (0.1% triamcinolone acetonide) applied on the lesions three times a day.	2 months
George S. *et al*. (case 1) (42)	India	2015	8	M	White striae were observed on the buccal mucosa bilaterally, on the ventral tongue mucosa and on the labial mucosa.	Yes	No	Topical anti- fungal treatment (clotrimazole mouth paint twice daily)	Not reported
George S. *et al*. (case 2) (42)	India	2015	8	M	White interlacing striae were noted on the left buccal mucosa (unilaterally)	Yes	No	None	Not reported
Zychowska M. *et al*. (43)	Poland	2015	10	M	White streaks on buccal mucosa	No	No	Intramuscular depot injections of methylprednisolone 0.8 mg/kg every 4 weeks for 12 weeks	10 months
Gupta C. *et al*. (44)	India	2016	12	M	White ill-defined plaques over the tongue and hard palate along with a few ulcers	No	Yes	Oral prednisolone, 1 mg/kg and topical triamcinolone pastem + dapsone, 50 mg was added as maintenance therapy	3 months
Cascone M. *et al*. (case 1) (45)	Italy	2017	17	F	Plaque-like + erosive OLP (margins of tongue bilaterally + ventrum of tongue bilaterally)	Yes	No	Topical steroids + Topical antimycotics	Not reported
Cascone M. *et al*. (case 2) (45)	Italy	2017	17	M	Reticular OLP (hard palate, retromolar fossae bilaterally)	No	No	Topical antimycotics	Not reported
Cascone M. *et al* . (case 3) (45)	Italy	2017	14	F	Plaque-like + atrophic OLP (buccal mucosa, right side of tongue bilaterally, lower lip)	No	No	Topical antimycotics	Not reported

Table 1 cont.: Studies of childhood LP in literature from 1980 to 2023.

Cascone M. *et al* . (case 4) (45)	Italy	2017	14	M	Reticular + atrophic OLP (Buccal mucosae bilaterally, gingiva, dorsum of tongue bilaterally, margins of tongue bilaterally)	No	No	Topical antimycotics	Not reported
Cascone M. *et al*. (case 5) (45)	Italy	2017	15	M	Reticular OLP (buccal mucosae bilaterally, ventrum of tongue bilaterally)	No	No	Topical antimycotics	Not reported
Cascone M. *et al*. (case 6) (45)	Italy	2017	9	F	Bullous + atrophic + reticular OLP (dorsum of tonbgue, margins of tongue bilaterally, gingival buccal mucosae bilaterally)	Yes	No	Topical steroids + Topical antimycotics	Not reported
Cascone M. *et al*. (case 7) (45)	Italy	2017	11	F	Reticular OLP (gingiva)	No	No	Topical antimycotics	Not reported
Cascone M. *et al*. (case 8) (45)	Italy	2017	11	M	Reticular + atrophic (dorsum of tongue)	No	No	Topical steroids + Topical antimycotics	Not reported
Sharma G. *et al*. (46)	India	2017	12	F	Hirregular 2×2cm grayish white patches with peripherally radiating white striae on the gingiva and vestibular area of the permanent mandibular molars bilaterally (Plaque-like OLP)	Yes	Yes	Topical steroids + topical antifunginals	Not reported
Thomas M.G *et al*. (47)	India	2018	14	M	White reticulate pattern along the upper labial mucosa extending onto the gum margin of the upper central incisor teeth (Reticular OLP)	No	Yes	Topical tacrolimus 0.1%	Not reported
Meng W. *et al*. (48)	China	2019	4	F	A clear, atrophic, white plaque with sclerosis on the right upper lip with an area of 1.5 cm in diameter extending to vestibular gingival of these teeth (Lichen Sclerosus)	No	No	The patient was treated with 1% pimecrolimus cream twice daily for 4 weeks	3 years
Simonato L.E. *et al*. (49)	Brazil	2019	12	F	Whitish macule on the lower lip vermillion, extending to the labial mucosa, buccal sulcus and gingiva in the region of teeth 21 and 32. (Lichen Sclerosus)	No	No	Intralesional triamcinolone acetonide 10 mg/mL was injected in decreasing dosage at 2-month intervals (0.3, 0.2, and 0.1 mL)	2 months
Chinnasamy N.K. *et al*. (50)	India	2020	7	F	Multiple diffuse grayish white raised linear lesions seen on the right and left buccal mucosa, which extended to upper labial mucosa (Reticular OLP)	Yes	No	Topical 0.1% triamcinolone acetonide ointment once daily.	1 year
Hasan S. *et al*. (51)	India	2020	8	M	2 cm × 3 cm erosive lesion localized in the retromolar region in relation to 37, while a reticular type variant of OLP was evident on the right buccal mucosa presenting with the peculiar slender radiating white striae (Wickham's striae). (Erosive OLP + Reticular OLP)	Yes	No	Steroid mouth rinses (betnesol 0.5 g)	3 months
Wang F. *et al*. (52)	China	2020	3	M	Whitish reticular and papular lesions were widespread on the lips, the dorsum of the tongue, and bilateral buccal mucosa (Reticular OLP)	Yes	Yes (back and right arm)	Topical treatment with recombinant bovine basic fibroblast growth factor (rbbFGF)	8 years
Shikha G. *et al*. (case 1)(17)	India	2022	12	M	white plaque like lesion measuring 2 × 2 cm in size on right and left buccal mucosa adjacent to commissure of mouth. White patch of size 2 × 2 cm with reticular striations were also present on anterior dorsal surface of tongue	No	No	None	Not reported
Shikha G. *et al*. (case 2)(17)	India	2022	12	M	white striations with surrounding erythema on right buccal mucosa extending up to the retromolar region. Faint white striations were also seen on the left buccal mucosa posteriorly in the retromolar region. A 1×1 cm patch with white striations was seen on the dorsum of the tongue on the left side. A diagnosis of reticular and plaque-like OLP was made.	Yes	No	topical application of 0.1% triamcinolone acetonide 3-4 times daily for 4 weeks. The patient responded well to treatment with marked resolution of lesions in the buccal mucosa with little resolution on the tongue.	Regular follow-up at an interval of 1 month.

Table 1 cont.: Studies of childhood LP in literature from 1980 to 2023.

Shikha G. *et al.* (case 3)(17)	India	2022	11	M	white striations surrounded by pigmented areas on bilateral buccal mucosa. Faint white patch was also evident on anterior dorsal surface of tongue and labial gingival mucosa in relation to right mandibular premolars: reticular OLP.	No	No	None	Not reported
Shikha G. *et al.* (case 4)(17)	India	2022	12	M	reticular white striae present in a symmetrical fashion over bilateral buccal vestibule and mucosa in relation to mandibular posterior teeth: Reticular OLP	Yes	No	topical application of 0.1% triamcinolone acetonide 3-4 times daily. After 6 months follow-up, there is marked resolution of lesions bilaterally	Regular follow-up at an interval of 1 month.
Shikha G. *et al.* (case 5)(17)	India	2022	13	M	an erythematous area with surrounding white striae and pigmentation was seen on right buccal mucosa corresponding to mandibular first and second molar teeth, on left buccal mucosa, areas of grayish pigmentation were seen along occlusal plane posteriorly in relation to mandibular molar teeth: reticular and atrophic OLP	yes	no	topical application of 0.1% triamcinolone acetonide 3-4 times daily with regular follow-up at an interval of 1 month. At 6 months follow up, there is marked resolution of erythema on right buccal mucosa and complete resolution of lesion on left buccal mucosa.	Regular follow-up at an interval of 1 month
Shikha G. *et al.* (case 6)(17)	India	2022	12	F	Difuse white papules with peripheral radiating striae on bilateral buccal mucosa extending posteriorly from retromolar area anteriorly to region of premolar teeth: Reticular & Papular OLP	Yes	No	Topical application of 0.1% triamcinolone acetonide 3-4 times daily	The patient did not follow-up
Spirito F. *et al.* (case 1) *(53)*	Italy	2023	6	F	Papules with reticular pattern on bilateral buccal mucosae	Yes	No	Topical application of clobetasol propionate 0.05% mixed with 4% hydroxyethyl cellulose gel to obtain a final concentration of 0.025% once or twice a day	12 months
Spirito F. *et al.* (case 2) *(53)*	Italy	2023	13	F	Plaque on dorsum linguae	Yes	No	Topical application of clobetasol propionate 0.05% mixed with 4% hydroxyethyl cellulose gel to obtain a final concentration of 0.025% once or twice a day	24 months
Spirito F. *et al.* (case 3) *(53)*	Italy	2023	10	M	Ulcer surrounded by a thin white patch with reticulated borders on dorsum linguae	Yes	No	Topical application of clobetasol propionate 0.05% mixed with 4% hydroxyethyl cellulose gel to obtain a final concentration of 0.025% once or twice a day	18 months
Spirito F. *et al.* (case 4) *(53)*	Italy	2023	13	M	Papules with reticular pattern and plaques on dosum linguae and lingual margin	Yes	No	Topical application of clobetasol propionate 0.05% mixed with 4% hydroxyethyl cellulose gel to obtain a final concentration of 0.025% once or twice a day	15 months
Spirito F. *et al.* (case 8) *(53)*	Italy	2023	13	M	Papules on dorsum linguae	No	No	Topical application of clobetasol propionate 0.05% mixed with 4% hydroxyethyl cellulose gel to obtain a final concentration of 0.025% once or twice a day	12 months
Spirito F. *et al.* (case 9) *(53)*	Italy	2023	16	M	Plaque on dorsum linguae	No	No	None	12 months
Spirito F. *et al.* (case 10) *(53)*	Italy	2023	15	F	Papules with reticular pattern on buccal mucosa, plaque on dorsum linguae	Yes	No	Topical application of clobetasol propionate 0.05% mixed with 4% hydroxyethyl cellulose gel to obtain a final concentration of 0.025% once or twice a day	12 months
Spirito F. *et al.* (case 13) *(53)*	Italy	2023	12	F	Papules on Dorsum linguae and Bilateral buccal mucosa	No	No	None	11 months

Table 2: Risk of bias: JBI critical appraisal checklist for case reports.

Author, data	Were patient's demographic characteristics clearly described?	Was the patient's history clearly described and presented as a timeline?	Was the current clinical condition of the patient on presentation clearly described?	Were diagnostic tests or assessment methods and the results clearly described?	Was the intervention (s) or treatment procedure (s) clearly described?	Was the post-intervention clinical condition clearly described?	Were adverse events (harms) or unanticipated events identified and described?	Does the case report provide takeaway lessons?
Malhotra Y.K. *et al*. (23) [1]	n	n	y	y	n	n	n	Y
Silverman R.A *et al*.(24) [1]	y	n	y	y	n	y	n	y
Berbis P. *et al*. (25)	n	y	y	y	y	y	y	y
Kanwar, A. *et al*. (6)[1]	y	n	u	u	y	u	u	y
Borrego, H. *et al*. (26)	n	y	y	u	y	n	n	y
Scully, C. *et al*. (27) °	n	y	y	y	n	n	n	y
Bettoli, V. *et al*. (28)	n	y	y	n	n	n	n	y
Alam F. *et al*. (5) °	y	n	y	n	n	n	n	y
Sandhu K. *et al*. (29) [1]	n	n	y	n	y	n	n	y
Laeijendecker M.D. (12) °	y	y	y	n	n	n	n	y
Patel S. *et al*. (30) °	y	y	y	n	y	y	n	y
Singal A.(31) [1]	y	y	y	u	n	n	n	y
Woo V.L. *et al*. (16) °	y	y	y	y	n	n	u	y
Azevedo R.S. *et al*. (case 3) (32)	n	n	y	y	y	n	n	y
Mohan Das U. and Jp, B. (33)	y	y	y	n	y	u	y	y
GunaShekar M. *et al*. (34)	u	n	y	y	n	y	n	y
De Moraes P.C. *et al*. (35)	y	y	y	y	y	n	n	y
Chaitra T.R. *et al*. (36)	n	y	y	y	u	u	u	y
Pendyala G. *et al*. (37)	n	y	y	y	y	y	y	y
Khandelwal V. *et al*. (38)	n	y	y	y	n	n	n	y
0Moger G. *et al*. (39)	n	y	y	y	y	n	n	y
Padmini C. *et al*. (40)	y	y	y	y	y	y	y	y
Chandna P. *et al*. (41)	y	y	y	y	y	y	u	y
George S. *et al*. (42) °	n	y	y	y	n	n	y	y
Zychowska M. *et al*. (43)	y	n	y	n	n	n	n	y
Gupta C. *et al*. (44)	n	y	y	y	y	y	y	y
Cascone M. *et al*. (45) °	y	y	y	y	y	u	u	y
Sharma G. *et al*. (46)	n	y	y	y	y	y	y	y
Thomas M.G *et al*. (47)	n	u	y	y	n	n	n	y
Meng W. *et al*. (48)	u	n	y	y	y	y	y	y
Simonato L.E. *et al*. (49)	n	n	y	y	y	y	y	y
Chinnasamy N.K. *et al*. (50)	y	y	y	y	y	n	n	y
Hasan S. *et al*. (51)	y	y	y	y	y	y	y	y
Wang F. *et al*. (52)	y	u	y	y	y	u	y	y
Shikha G. *et al*.(17) °	y	u	y	y	y	u	u	Y
Spirito *et al.(53)*	y	n	y	y	y	y	n	y

Legend n: no, y: yes, u: unclear; ° more cases of OLP in the child are described within a study with multiple cases or a case series; [1] Only one case of LP in the child is described in a study with multiple cases or in a case series.

Discussion

This systematic literature review was reported following the PRISMA guidelines, and at the end of the selection process we included in our review 36 manuscripts including 22 case reports, the other manuscripts were reported as case series on Oral Lichen with pediatric cases or described family cases, with a total of 65 pediatric OLP cases.

Although OLP in children was initially described in 1920s, just few cases have been so far reported (51). In most of the studies mucocutaneous lichen planus has been addressed, while oral involvement has been often

described in association with predisposing conditions, such as hepatitis and vaccination (54). In Table 1, 65 cases of oral childhood lichen planus published are detailed. The most interested country was India. The mean age at time of diagnosis of the disease was 11 years, ranging from 3 to 17 years (Fig. 2). The most frequent patterns were the reticular and plaque-like patterns. The buccal mucosa was the most involved oral site, followed by tongue. 33 patients were male (50.7%) and 32 were female (49.2%).

It has been suggested that childhood LP often shows atypical clinical features; this seems to be confirmed also in our series. In fact, four of the seven patients here presented had lesions confined to the tongue; this is uncommon in adults, in whom most of the times patients had multiple oral sites of involvement, whereas single sites of involvement are uncommon (4). On the other hand, we found that OLPc histological features are not different from OLP in adults.

OLPc treatment outcomes seems to be more favourable than adulthood OLP (40). To date, neither trials, nor consensus about standardized therapy for OLPc exists; so many different treatment modalities have been reported in literature (ranging from local corticosteroids to immunosuppressive drugs) (3,12). In a recent case series of 13 OLPs in the child described by our research group, when a treatment was needed, a local therapy based on low-dose corticosteroids (clobetasol propionate 0.05% mixed with 4% hydroxyethyl cellulose gel to obtain a final concentration of 0.025% once or twice a day), was successful in obtaining a stable remission of symptoms (53).

A limitation to this systematic review of the literature is the inclusion of studies and reports that describe only case reports or case series, in fact the absence in the literature of observational studies that clearly identify and with precise clinical stratification, the real incidence of Pediatric OLP.

OLPc is an extremely uncommon occurrence. Majority of the childhood OLP cases are not reported due to misdiagnosis by the physicians (51). Any mucosal lesion in children should be referred to the specialist for an early and precise diagnosis and treatment protocol (51). General OLPc usually has a much fairer prognosis and responds well with therapy. In conclusion, though lichen planus in children is uncommon and oral mucosal involvement quite rare, clinicians should be aware of its existence and management and its differential diagnosis in children presenting oral white lesions and/or chronic ulcers.

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Conflict of interest
The authors declare that they have no competing interests.

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