INPLASY PROTOCOL

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None declared.

Oral lichen planus in children: systematic review

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Review question / Objective: **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.**
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.**

INPLASY registration number: This protocol was registered with the International Platform of Registered Systematic Review and Meta-Analysis Protocols (INPLASY) on 22 September 2022 and was last updated on 22 September 2022 (registration number INPLASY202290106).

INTRODUCTION

Review question / Objective: **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.**

Rationale: 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, while other reviews have focused on the role of vaccinations for the hepatitis B in association with the onset of LP in pediatric age . Given the importance of intercepting neoformations and oral precancerous diseases in children, it is essential to understand what the real impact in terms of OLP cases is in children and its clinical course in terms of follow-up.

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

The etiology OLP is still unclear. It seems that an autoimmune response involving antigen-presenting cells and regulatory T-lymphocytes, probably triggered by keratinocytes, plays a key role in the development of the disease. 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. A number of auto-immune diseases, including lupus erythematous, pemphigus, rheumatoid arthritis, and Sjogren's syndrome, have been reported in association to OLP.

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. In particular, in patients with paediatric lichen planus a higher incidence of positive family history for the disease is reported; on the other hand, genetic linkage studies reported an association between familial lichen planus and HLA B, HLA DR1 and DR10.

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). Cutaneous lichen planus is often self-limiting, whereas OLP tends to be chronic: spontaneous remission has been rarely reported in OLP; in general, OLP prognosis in children seems to be more favourable.

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.

METHODS

Search strategy: 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 (F.S., M.A.). 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 22 September 2022.

Participant or population: Children.

Intervention: Oral lichen Planus.

Comparator: Patients with Oral Lichen.

Study designs to be included: case report, case series, RCT, retrospective study, prospective study.

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.

Information sources: PubMed, Scopus, Web of Science, Science Direct, EMBASE; Gray literature :google scholar and Open Gray (DANS EASY Archive).

Main outcome(s): 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.

Quality assessment / Risk of bias analysis: 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 . The evaluation was performed by a researcher (M.D.) after the data extraction and inclusion phase of the studies.

Strategy of data synthesis: 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.

Subgroup analysis: No subgroup analysis.

Sensitivity analysis: No Sensitivity analysis.

Country(ies) involved: Italy.

Keywords: Oral Lichen Planus, Childhood, Lichen Ruber Planus, paediatric lichen planus.

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