Background
Although the exact incidence and prevalence of oral lichen planus (OLP) is unknown, many previous studies have reported that there is no gender or age-specific predilection for pediatric patients. The oral mucosa seems to be less commonly involved with a prevalence of approximately 0.03% compared with 1% to 2% of general population and the clinical presentation is often atypical.

Aim of the study
The aim of this study is to perform an update of the oral involvement of the disease in children through a review of the literature. We also report a retrospective analysis of pediatric patients referred to our Department during the last four years in whom lichen planus has been presented in the oral cavity as the single or as an additional site of involvement.

Subjects and methods
A PubMed search was carried out from 1966 to 2014 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
- graft versus host disease (GVHD)
- lack of confirmatory histology
- oral lichenoid drug reaction

The analysis was completed reporting retrospectively from our database the cases with OLP aged <18 at the time of diagnosis, referred to our Department in the last four years.

Table 1: Flow chart.

Table 2: comparison between our cases and the review of the literature.

Results
The analysis of the literature yielded 315 articles published between 1966 and 2014. After the application of the inclusion criteria, 9 articles were included in our study. We also studied among the references of the aforementioned articles and found a further six articles making a total of 15 articles, so fulfilling our inclusion criteria for a total of 25 patients described.

The flow chart is reported in Table 1. The case series from our database shows eight patients, which so increases the total number of pediatric patients in the literature to thirty-one. A comparison between our cases and the literature is provided in Table 2.

The details about the epidemiology, predisposing factors, diagnosis and treatment in our case series are listed in Table 3.

The analysis of these epidemiological data showed no gender predilection (M:F = 1:1.16) but revealed evidence of immunological disorders and family history of 38.7% and 12.9% of cases respectively. Clinically, a predominance for the reticular form (67.7%) and a localization at the buccal mucosa (56.5%) has been reported. However, among our patients the tongue (75%) was the most commonly involved oral site.

Conclusions
In summary our case series mostly mirrors previous epidemiological, clinical and therapeutic knowledge about pediatric OLP but a different oral site predilection between adults and children has been suggested by our findings. Clinicians must be aware that OLP in childhood also may have a different clinical and/or future involvement of the skin and other mucosal sites and, due to 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 pediatricians agree that the scheduling of follow-up of pediatric OLP should be one to two visits per year as long as the OLP persists even if the prognosis seems to be more favorable. We also report the first case of an oral lichenoid lesion related to autoimmune polyendocrine-candidiasis-ectodermal dysplasia-c zonderwitz disease (APS-1) also known as autoimmune syndrome (PAPA) type alone in a 9 year-old girl. Up to now there has been only one other case of literature in OLP associated autoimmune polyendocrine syndrome type II reported by our research group in a 42 year-old woman, reinforcing the suggestion of a common immune-mediated pathogenesis between OLP and PAPA. Differences in OLP between adults and pediatric patients have been detected but further investigation and larger case series are needed to establish detailed differences in clinical characteristics.

References: