Oral Lichen Planus in Childhood associated with Cutaneous Lichen Nitidus: Case Report

Tushar Phulambrikar, Nikita Goyal, Manasi Kode, Shali P Magar

1Professor & Head, Department of Oral Medicine & Radiology, Sri Aurobindo College of Dentistry & PG Institute, Indore, Madhya Pradesh, India,
2Post Graduate Student, Department of Oral Medicine & Radiology, Sri Aurobindo College of Dentistry & PG Institute, Indore, Madhya Pradesh, India,
3Associate Professor, Department of Oral Medicine & Radiology, Sri Aurobindo College of Dentistry & PG Institute, Indore, Madhya Pradesh, India,
4Reader, Department of Oral Medicine & Radiology, Sri Aurobindo College of Dentistry & PG Institute, Indore, Madhya Pradesh, India

INTRODUCTION

Oral lichen planus (OLP) is a common chronic immunologic inflammatory mucocutaneous disorder that varies in appearance from keratotic (reticular or plaque like) to erythematous and ulcerative forms.¹

OLP is a common disease in middle aged and elderly population and has a prevalence of about 0.5-2%. OLP is rare in children with few reports available in the literature.²

The etiology of lichen planus remains uncertain, but many factors have been implicated. Such factors include the genetic predisposition, infective agents, systemic diseases, graft versus host disease, drug reactions, and hypersensitivity to dental materials, vitamin deficiencies,³ and psychological factors.⁴,⁵

Baccaglini et al. 2013 reports indicate that hepatitis C virus is associated with lichen planus and might be involved in pathogenesis of OLP.⁶ Lichen planus has been associated with several autoimmune diseases, including lupus erythematosus, pemphigus, Sjogren syndrome, autoimmune liver disease, autoimmune thyroiditis, myasthenia gravis, alopecia areata, thymoma, autoimmune polyendocrinopathy.⁷-¹²

In addition to that, several studies have found an association of lichen planus with atopic dermatitis.¹³,¹⁴ Although the association of lichen planus and lichen nitidus is controversial, however it has been documented in the literature. In this case report, we are presenting a case of OLP in childhood with associated lichen nitidus.

CASE REPORT

A 14-year-old boy was referred to Department of Oral Medicine, and Radiology with the complaint of burning sensation in the mouth (Figure 1). He reported that burning sensation had been present for preceding 1-year, which aggravates on consuming spicy food. Because of the disability, he had not been to the school for many days. His family history and medical history were unremarkable at initial presentation. Routine blood investigation was normal. General examination did not reveal any findings.

Skin examination showed multiple tiny papules which were of skin color, shiny, non-pruritic and measuring about 0.3-0.7 mm in dimension present on extensor
surface of elbow of left hand and on index finger of left hand (Figures 2 and 3). Intraoral examination revealed fine white radiating striae was present on lower labial mucosa, bilateral buccal mucosa, floor of mouth, and attach gingiva bilaterally, few erythematous macules were present on right and left buccal mucosa (Figures 4-6). His oral hygiene was good without any dental restoration. Diagnosis of OLP and cutaneous lichen nitidus was made.

Histopathological examination of the oral lesion showed parakeratotic and focally atropic stratified squamous

---

**Figure 1:** Extraoral picture of 14-year-old boy

**Figure 2:** Multiple, tiny, skin color, shiny papules present on extensor surface of elbow of left hand

**Figure 3:** Multiple, tiny color, shiny papules present on index finger of left hand

**Figure 4:** Fine, faint white radiating striae on left buccal mucosa

**Figure 5:** Fine, faint white radiating striae with few erythematous patches on right buccal mucosa

**Figure 6:** Fine faint white radiating striae extending from gingiva to buccal sulcus
epithelium and focal areas of basal cells liquefaction degeneration and increase intraepithelial lymphocytes. There was dense juxtaepithelial band of inflammatory cells the connective tissue shows dense collagen bundles, dilated blood vessels, muscle tissue, adipocytes. Both clinical and histopathological features were consistent with lichen planus (Figure 7).

As the patient did not respond to topical steroids, the patient was treated with a tablet betnesol forte 0.5 mg, tacrolimus ointment and isotretinoin 0.5 mg was started. Patient was followed up periodically every 15 days on commencement of treatment then after 2 months. Marked clinical Improvement followed after 6 weeks of treatment (Figures 8-11).

DISCUSSION

Lichen planus was first described in the literature by Erasmus Wilson in 1869 as predominantly a disease of middle-aged or older. OLP has been described in children in 1920s. The literature available reporting the occurrences of lichen planus in pediatric age is low, and that of oral involvement is extremely rare.

Woo et al. 2007 did the literature review on childhood OLP from 1990 to 2005 and found the slight male predilection and common age of occurrence were 11 and 15 years with no ethnic predilection. Buccal mucosa was
the commonly affected site, and most patients were with reticular pattern.\textsuperscript{15}

We have further reviewed literature from 2007 to 2014 using the same criterias as used by Woo \textit{et al} and descriptive statistical analysis was performed.

We found an almost equal predilection for male (52.2\%) and female (47.8\%) unlike Woo \textit{et al} review who had slight male predilection. This is in contrast with lichen planus occurring in elders who have female predilection. Mean age of occurrence was 10.39 years with common age group ranging from 7-11 years. There was racial predilection, majority were of Asian origin unlike Woo \textit{et al} which showed no racial predilection. Buccal mucosa was the common site, followed by tongue, and reticular pattern was common.

Some of our findings were consistent with Chatterjee \textit{et al} 2012 who have studied 22 OLP cases, like equal predilection for male and females, buccal mucosa as common site, however the mean age reported was 15.8 years and most common form was erosive\textsuperscript{6} which is in contrast with Woo \textit{et al} 2007, Walton \textit{et al} 2010 and our study.

Majority of the case reports and studies are in Asian patients suggesting the geographic predilection. It may be due to environmental factors and/or genetic influence disease evolution\textsuperscript{15} (Table 1).

Balasubramaniam \textit{et al} 2008 studied lichen planus from outside the Indian subcontinent and found that of their 26 patients, 21 (80.8\%) were from the Indian subcontinent but only 28\% of the city’s general child population was from the region.\textsuperscript{17} However, Walton \textit{et al} 2010 found statistically significant greater number of African Americans with childhood lichen planus compared with the control population.\textsuperscript{18}

OLP is a disease of older age as many illnesses and conditions are associated with lichen planus occur in older patients.\textsuperscript{15}

Therefore, for explaining the occurrence of lichen planus in pediatric age group, various factors have been determined which includes previous hepatitis B vaccination, liver diseases such as chronic active hepatitis and genetic predisposition as in familial lichen planus.\textsuperscript{15}

Woo \textit{et al} 2007 reported two cases of OLP in childhood, out of which one had a history of hepatitis B vaccination.\textsuperscript{15} Kanwar \textit{et al} 2010, studied on 100 cases of childhood lichen planus and found that 16 had been vaccinated against hepatitis B.\textsuperscript{19} Familial pattern has also been reported.\textsuperscript{20,21}

However, in the current case report neither familial pattern nor hepatitis B vaccination was documented.

Moreover, recently published study had also not found the association of hepatitis B and C with OLP. Chatterjee \textit{et al} 2012 conducted a retrospective study for 13 years and had not found positive serology for hepatitis B and C.\textsuperscript{16} Kumar \textit{et al} 2013 studied on correlation of hepatitis virus (B and C) and OLP and had found no serology positive concluding that other factors could be there in underlying disease.\textsuperscript{22}

Childhood lichen planus commonly occurs on the skin, and oral involvement is rare.

Kanwar \textit{et al} 1991 presented report of seventeen patients involving the legs, arms, trunk, neck, nails, scalp, and face. Oral involvement was seen in one patient.\textsuperscript{23} Kumar 1993 \textit{et al} reported 1 out of 25 (4\%) patient with OLP.\textsuperscript{24} Sharma and Maheswari 1999 reported OLP in 15 of 50 (30\%) children.\textsuperscript{14} Nanda \textit{et al} 2001 reported 9 of 23 (39\%) children with oral lesions.\textsuperscript{25} Handa and Sahoo 2002 reported 87 patients with childhood lichen planus in India 8 (9\%) patients showed involvement of oral mucosa.\textsuperscript{26} Montoya \textit{et al} 2005 reported 1 of 16 (6\%) children with oral lesions.\textsuperscript{27} Laeijendecker \textit{et al} 2005 conducted a 10 years retrospective study comprised of 10,000 patients below 18 years, showed the prevalence of 0.03\% (3 patients) with oral involvement.\textsuperscript{27} Recently, two large studies have been published reporting the incidence of OLP in childhood as 17\% and 18\% out of 100 and 316 children respectively.\textsuperscript{19,28}

There is greater variability in prevalence of OLP in childhood ranging from 0.03\% to 39\% maximum depending upon the region involved. This strongly indicates the geographic or racial predilection suggesting the genetic influence.

In addition, lichen planus has been found to be in association with other systemic diseases. In our case, we found it to be associated with cutaneous lichen nitidus. To our best knowledge, only single case report has been found which showed a correlation of lichen nitidus and OLP.\textsuperscript{29}

\textbf{CONCLUSION}

Lichen planus in childhood is uncommon, and the oral involvement is extremely rare. However, there is evidence of increased documentation of case reports and studies in recent years suggesting increase in the number of cases of childhood OLP indicating the rise of environmental factors.
Table 1: Summary of juvenile oral lichen planus (1990-2014)

<table>
<thead>
<tr>
<th>Case reports</th>
<th>No. of cases</th>
<th>Age/sex</th>
<th>Ethnicity</th>
<th>Site</th>
<th>Skin involvement</th>
<th>Clinical form</th>
</tr>
</thead>
<tbody>
<tr>
<td>Khandelwal et al. 2013</td>
<td>1</td>
<td>10/f</td>
<td>Asian</td>
<td>NS</td>
<td>NS</td>
<td>Reticular</td>
</tr>
<tr>
<td>Moger et al. 2013</td>
<td>1</td>
<td>7/f</td>
<td>Asian</td>
<td>BM</td>
<td>Yes</td>
<td>Erosive</td>
</tr>
<tr>
<td>Padmini et al. 2013</td>
<td>1</td>
<td>12/f</td>
<td>Asian</td>
<td>Tongue</td>
<td>No</td>
<td>Ulcerative</td>
</tr>
<tr>
<td>Sanjaya et al. 2011</td>
<td>1</td>
<td>9/f</td>
<td>Asian</td>
<td>BM, tongue, hard palate, lips</td>
<td>Yes</td>
<td>Reticular</td>
</tr>
<tr>
<td>Demoraes et al. 2011</td>
<td>1</td>
<td>7/f</td>
<td>Caucasian</td>
<td>Lip</td>
<td>No</td>
<td>Reticular</td>
</tr>
<tr>
<td>Anuradha et al. 2011</td>
<td>3</td>
<td>12/f</td>
<td>Asian</td>
<td>BM</td>
<td>Yes</td>
<td>Papular</td>
</tr>
<tr>
<td>Gunashekhar et al. 2010</td>
<td>1</td>
<td>7/m</td>
<td>Asian</td>
<td>BM, FOM, tongue</td>
<td>No</td>
<td>Reticular</td>
</tr>
<tr>
<td>Das et al. 2009</td>
<td>1</td>
<td>12/f</td>
<td>Asian</td>
<td>BM</td>
<td>No</td>
<td>Reticular</td>
</tr>
<tr>
<td>Woo et al. 2007</td>
<td>2</td>
<td>9/f</td>
<td>Caucasian</td>
<td>Tongue</td>
<td>No</td>
<td>Erosive</td>
</tr>
<tr>
<td>Lajijendecker et al. 2005</td>
<td>3</td>
<td>11/f</td>
<td>Asian</td>
<td>BM</td>
<td>No</td>
<td>Reticular</td>
</tr>
<tr>
<td>Patel et al. 2005</td>
<td>2</td>
<td>6/m</td>
<td>Caucasian</td>
<td>Tongue</td>
<td>No</td>
<td>Erosive</td>
</tr>
<tr>
<td>Singhal 2005</td>
<td>1</td>
<td>11/m</td>
<td>NS</td>
<td>Tongue</td>
<td>No</td>
<td>Atrophic</td>
</tr>
<tr>
<td>Sandhu et al. 2003</td>
<td>1</td>
<td>12/f</td>
<td>NS</td>
<td>NS</td>
<td>No</td>
<td>Reticular</td>
</tr>
<tr>
<td>Alam and Hamburger 2001</td>
<td>6</td>
<td>6/m</td>
<td>Asian</td>
<td>BM Tongue</td>
<td>No</td>
<td>Reticular, Erosive</td>
</tr>
<tr>
<td></td>
<td></td>
<td>7/m</td>
<td>Asian</td>
<td>BM</td>
<td>Atrophic</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>8/m</td>
<td>Caucasian</td>
<td>Gingiva</td>
<td>Reticular</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>11/m</td>
<td>Caucasian</td>
<td>BM, Tongue</td>
<td>Reticular, Papular</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>14/m</td>
<td>Asian</td>
<td>BM, Tongue</td>
<td>Reticular, Plaque</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>14/m</td>
<td>Asian</td>
<td>BM</td>
<td>Atrophic</td>
<td></td>
</tr>
<tr>
<td>Scully et al. 1994</td>
<td>2</td>
<td>10/f</td>
<td>Caucasian</td>
<td>FOM</td>
<td>Erosive</td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>11/f</td>
<td>Caucasian</td>
<td>Tongue</td>
<td>Erosive</td>
<td></td>
</tr>
<tr>
<td>Review studies</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pandhi et al. 2014</td>
<td>57/316</td>
<td>&lt;14/a</td>
<td>Asian</td>
<td>NS</td>
<td>Yes</td>
<td>NS</td>
</tr>
<tr>
<td>Zheng-Yu Shen et al. 2012</td>
<td>5/518</td>
<td>&lt;20</td>
<td>Asian</td>
<td>NS</td>
<td>NS</td>
<td>NS</td>
</tr>
<tr>
<td>Chatterjee et al. 2012</td>
<td>22</td>
<td>&lt;18/b</td>
<td>Asian</td>
<td>BM&gt;tongue</td>
<td>Yes</td>
<td>Erosive</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Walton et al. 2010</td>
<td>8/36</td>
<td>&lt;18/c</td>
<td>African american and other races</td>
<td>BM</td>
<td>Yes</td>
<td>Reticular&gt;erosive</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Xue et al. 2005</td>
<td>4/674</td>
<td>10-13</td>
<td>NS</td>
<td>NS</td>
<td>NS</td>
<td>Reticular (2), Erosive (2)</td>
</tr>
<tr>
<td>Eisen 2002</td>
<td>5/723</td>
<td>&lt;15</td>
<td>NS</td>
<td>NS</td>
<td>Yes</td>
<td>Erosive, atrophic</td>
</tr>
<tr>
<td>Handa et al. 2002</td>
<td>12/87</td>
<td>7-11.5</td>
<td>NS</td>
<td>BM (5), Lips (3)</td>
<td>Yes</td>
<td>Reticular</td>
</tr>
<tr>
<td>Sharma and Maheshwari 1994</td>
<td>15/50</td>
<td>14</td>
<td>NS</td>
<td>BM</td>
<td>Yes</td>
<td>Reticular (skin)</td>
</tr>
</tbody>
</table>

*Familiar lichen planus, *a*Mean age 10.28 years, *b*Mean age-15.18 years, *c*Two patient had skin involvement, *d*Mean age 11.8 years, FOM: Floor of mouth, BM: Buccal mucosa, NS: Not specified

REFERENCES

Oral Lichen Planus in Childhood Associated with Cutaneous Lichen Nitidus

Phulambrikar, et al.


Source of Support: Nil, Conflict of Interest: None declared.