Lymphoepithelial Cyst in the Sublingual Region: Report of a case and review of literature

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Oral lymphoepithelial cyst occurring in the sublingual region of a 30-year-old Japanese man is reported. This is a rare lesion that develops within oral lymphoid tissue. Clinical and pathological findings of our case show almost the same appearance as the cases which were reported previously. Serial sectioning was done in this case. We observed the salivary duct structure in the lymphoid tissue. We concluded that the histopathogenesis of this lesion is Bhaskar’s “enclavement theory”.

Key words: lymphoepithelial cyst, oral, enclavement theory

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Introduction

Lymphoepithelial cyst is a relatively rare lesion that develops within lymphoid tissue. These cystic lesions in the lateral aspect of the neck were first described by Bhaskar and Bernier (1) as “branchial cyst” in 1959. In a review of the English literature during the last 37 years, 145 cases of the oral lymphoepithelial cyst were noted (1, 3-20). Bhaskar (2) described that about 60 % occurred in the floor of the mouth and 40 % on the lateral and ventral surfaces of the tongue. Rarely, the cysts occur in the soft palate or palatoglossal area. All of these locations represent sites of normal or accessory oral lymphoid tissue. It is usually 0.3 x 0.3 cm to 1.5 x 1.5 cm in size, freely movable, with the duration of a few months to many years.

This article describes a case of lymphoepithelial cyst in the sublingual region with review of the English literature.

Case Report

A 30-year-old man visited the clinic of the Department of Oral and Maxillofacial Surgery, Mori-machi Public Hospital on July 2, 1999, with a chief complaint of slight contact pain on the right sublingual area, which he had first noticed about one month previously. The patient’s past medical history was non-contributory. On physical examination, an elastic hard nodule with small surface erosion, measuring 0.8 cm in diameter was observed. The color of overlying mucosa was normal (Fig. 1). The clinical diagnosis was “Soft fibroma, suspected”.

Under local anesthesia, the nodular lesion was excised with surrounding normal tissues and sent for histopathologic examination.

Macroscopically, the color was yellowish-white and cut surface was smooth. In the cut surface, the cystic cavity was not remarkable (Fig. 2). Microscopically, the cystic cavity was lined by thin flattened layer of keratinized stratified squamous epithelium. Desquamated cells, a few neutrophils and lymphocytes were present in the lumen. The stratified squamous epithelium was thin and rete ridge of it was flattened. The lining squamous epithelium was surrounded by well-circumscribed mass of lymphoid tissues (Fig. 3). The flattened rete ridge was considered to be pressure atrophy due to the surrounding lymphoid tissue. In the lymphoid tissues, germinal cen-
ters were present. The lymphoid tissue contained the
typical follicular arrangement, with densely packed cen-
trocytes surrounding the centroblasts and reticuloendo-
thelial cells comprising the germinal center (Fig. 4). The
tissue resembled the so-called “oral tonsils”.

Serial sectioning was done in this case. We ob-
served the salivary duct structure in the lymphoid tissue.
No direct communication between the surface epithelium
and lining epithelium was observed (Fig. 5).

The pathological diagnosis was lymphoepithelial
cyst in the sublingual region.

One year after surgery, postoperative course was
uneventful.

Discussion

In 1959, Bhaskar and Bernier (1) demonstrated the
468 cases of branchial cysts of the neck and recom-
mended the term of lymphoepithelial cyst for them. Bran-
chial cleft cyst arise from incomplete embryological de-
velopment. In 1962, Gold and Levittown (3) described the
oral lymphoepithelial (Branchial cleft) cyst as a case re-
port, which is the first one in the world.

As previously reported cases (Table 1&2), the char-
acteristic features of the oral lymphoepithelial cysts are;
(1) Predominance in male: Male 78 cases
   Female 54 cases
   (except 13 unknown cases)
(2) Size: 3×3 mm to 15×15 mm
(3) Color: yellowish -white
(4) The age of occurrence: Mean 32.6 years
   Range 7-81 years
   (except 13 unknown cases)
(5) Site: Almost all the lesions are located in the floor of
   the mouth and tongue (89.7%).

The clinical diagnosis had been made variously.
Buchner and Hansen (10) described that the most two
common diagnoses were mucocele and lipoma. Others
were irritation fibroma, sialadenitis, inclusion cyst, hy-
perplastic lymphoid tissue, papilloma, and chronic ab-
scess. In our case, we made a clinical diagnosis of so-
called fibroma due to its size and induration.

Although the histogenesis of the oral lymphoepithe-
lial cyst is unknown, there are several theories about the
histogenesis of this lesion. In 1962, Gold and Levittown
stated that the location of this developmental defect in the floor of the mouth suggests that it originated from the epithelium entrapped between the branchial arches. In 1966, Bhaskar (4) described the “enclavement theory”. According to this theory, this lesion arises from the epithelium which, during embryogenesis, becomes included within the lymphoid aggregates in the mucosa of the oral cavity. Vickers and Muhll (5) certified the “enclavement theory” experimentally by autogenous epithelial (buccal mucosa) transplantation into the submandibular lymph-node in hamsters. On the other hand, Knapp (6, 7) described that the lymphoepithelial cysts arises through an obstruction of the crypt of the oral tonsils. They are “pseudocysts”, not true cysts. Toto and coworkers (8), Iwase and coworkers (9) studied the histomorphology of the lymphoepithelial cyst employing special stains, and the identification and distribution of immunoglobulins G, M, A, and C’. They supported the theory of Knapp. Buchner and Hansen (10) stated that they found only 2 cases in which the lining and surface epithelium were continuous, but in other 36 cases, did not find such a finding. So the pathogenesis of the oral lymphoepithelial cysts cannot always be explained by his “obstruction theory”. Chaudhry and coworkers (11) proposed that these lesions may originate from the excretory duct of sublingual gland or ectopic minor salivary glands. They stated that this would explain the most exclusive occurrence in the floor of the mouth and in other part of the oral cavity.

We performed serial sectioning in this case. It showed the salivary duct structure in the lymphoid tissue. No direct communication between the surface epithelium and lining epithelium was observed. We might concluded that the histopathogenesis of this lesion is Bhaskar’s “enclavement theory” (4).

Table 1: The cases of lymphoepithelial cyst described in the literature

<table>
<thead>
<tr>
<th>Authors</th>
<th>Year published</th>
<th>Gender</th>
<th>Age (Range)</th>
<th>Site</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gold C. et al (3)</td>
<td>1962</td>
<td>1</td>
<td>32</td>
<td>F.M.</td>
</tr>
<tr>
<td>Calman HJ. (12)</td>
<td>1963</td>
<td>1</td>
<td>40</td>
<td>T.</td>
</tr>
<tr>
<td>Bhaskar SN. (4)</td>
<td>1966</td>
<td>17</td>
<td>36.4 (15-65)</td>
<td>S.P.</td>
</tr>
<tr>
<td>Young WG. et al (13)</td>
<td>1967</td>
<td>1</td>
<td>42</td>
<td>A.P.P.</td>
</tr>
<tr>
<td>Knapp MJ (6, 7)</td>
<td>1970</td>
<td>unknown</td>
<td>unknown</td>
<td>R.M.P.</td>
</tr>
<tr>
<td>Acevedo A. et al (14)</td>
<td>1971</td>
<td>9</td>
<td>27.6 (20-46)</td>
<td>B.V.</td>
</tr>
<tr>
<td>Merchant NE (15)</td>
<td>1972</td>
<td>1</td>
<td>21</td>
<td>M.L.V.</td>
</tr>
<tr>
<td>Guinter J. et al (16)</td>
<td>1973</td>
<td>9</td>
<td>32.0 (7-65)</td>
<td>P.G.F.</td>
</tr>
<tr>
<td>Budner A. et al (10)</td>
<td>1980</td>
<td>23</td>
<td>39.8 (14-81)</td>
<td></td>
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<tr>
<td>Toto PD. et al (8)</td>
<td>1982</td>
<td>3</td>
<td>43.2 (25-60)</td>
<td></td>
</tr>
<tr>
<td>Sakoda S. et al (17)</td>
<td>1983</td>
<td>1</td>
<td>19</td>
<td></td>
</tr>
<tr>
<td>Chaudhry AP. et al (11)</td>
<td>1984</td>
<td>9</td>
<td>39.5 (12-74)</td>
<td></td>
</tr>
<tr>
<td>Iwase T. et al (9)</td>
<td>1985</td>
<td>1</td>
<td>34</td>
<td></td>
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<tr>
<td>McDonell D. et al (18)</td>
<td>1990</td>
<td>1</td>
<td>5</td>
<td></td>
</tr>
<tr>
<td>Kumara GR. et al (19)</td>
<td>1995</td>
<td>1</td>
<td>25</td>
<td></td>
</tr>
<tr>
<td>Ahn SK. et al (20)</td>
<td>1996</td>
<td>1</td>
<td>56</td>
<td></td>
</tr>
<tr>
<td>Suzuki H. et al (21)</td>
<td>2000</td>
<td>1</td>
<td>30</td>
<td></td>
</tr>
</tbody>
</table>


*Including the cases in the tongue

Table 2: Site distribution and frequency of oral lymphoepithelial cyst in various studies

<table>
<thead>
<tr>
<th>Site</th>
<th>No. of cases</th>
<th>Percentage (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Floor of the Mouth &amp; Tongue</td>
<td>130</td>
<td>89.7</td>
</tr>
<tr>
<td>Soft Palate</td>
<td>9</td>
<td>6.2</td>
</tr>
<tr>
<td>Anterior Palatine Pillar</td>
<td>2</td>
<td>1.3</td>
</tr>
<tr>
<td>Retromolar Pad</td>
<td>1</td>
<td>0.7</td>
</tr>
<tr>
<td>Buccal Vestibule</td>
<td>1</td>
<td>0.7</td>
</tr>
<tr>
<td>Mandibular Labial Vestibule</td>
<td>1</td>
<td>0.7</td>
</tr>
<tr>
<td>Palatoglossal Fold</td>
<td>1</td>
<td>0.7</td>
</tr>
</tbody>
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Acknowledgement

We thank Mr. Masaaki Kaneda (The Second Department of Pathology, Hamamatsu University School of Medicine) for technical assistance.

References


(Accepted for publication October 12, 2000)