Case Report
A rare occurrence of lymphoepithelial cyst in the palatine tonsil: a case report and discussion of the etiopathogenesis

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Received February 6, 2015; Accepted March 30, 2015; Epub April 1, 2015; Published April 15, 2015

Abstract: Lymphoepithelial cysts are uncommon benign lesions that present as painless yellowish nodules arising from various sites in the oral cavity and other parts of the body. Their etiopathogenesis is controversial, but most authors have assumed that they develop from obstruction of crypts in oral lymphoid aggregates, thus they are not true cysts but pseudocysts of retention. This paper describes a case of a large lymphoepithelial cyst located in the tonsil of a 21-year-old man complaining of a lump in the throat for four months. The patient underwent excisional biopsy, and the histopathological features showed squamous epithelium surrounded by lymphoid tissue, which were characteristically consistent with a lymphoepithelial cyst. We discuss the etiopathogenesis of these lesions and treatment modalities, which can consist of conservative surgery or only follow-up examination.

Keywords: Lymphoepithelial cyst, palatine tonsil, pseudocysts, etiopathogenesis

Introduction
Lymphoepithelial cysts (LECs) are uncommon benign lesions arising from entrapment or proliferation of epithelium in association with lymphoid tissue [1-3] that may occur in various sites, including the pancreas [4], thyroid [5] and mediastinum [6]. In the head and neck region, LECs have been described in lateral areas of the neck (known as branchial cyst), the parotid gland, and the oral cavity [1-3].

Oral LECs present as painless yellowish submucosal nodules, usually smaller than 1 cm and located on the floor of the mouth and ventral or posterolateral surface of the tongue [7]. Demographic data are controversial. In a recent clinical analysis of 120 cases, Yang et al found female predilection, with a male-to-female ratio of 1:2 and ages ranging from 2 to 75 years with a mean of 44.1 years [8].

Microscopically, oral LECs are characterized by a cystic cavity containing keratin that is lined with a parakeratinized stratified squamous epithelium, exhibiting a smooth interface with the lamina propria. Surrounding the epithelial lining, the cyst capsule is usually thick and presents intensively infiltrated by lymphocytes arranged as germinal centers [3, 7, 9].

There is no consensus regarding etiopathogenesis of oral LECs, and Knapp’s theory is the most accepted [7, 8, 10]. Supporting this classic theory, Buchner and Hansen [7] stated that oral LECs are not true cysts resulting from epithelial proliferation but rather pseudocysts originating from a dilated, obstructed crypt of the oral tonsil in which either purulent material or desquamated epithelial lining accumulates [7, 10].

The occurrence of LEC in the palatine tonsils is extremely rare, as there are few cases described in the medical literature [11]. Herein, we report a case of LEC arising from the right palatine tonsil and discuss its histogenesis.

Case presentation
An otherwise healthy 21-year-old man was referred to the Oral Medicine Center at the School of Dentistry, Federal University of Goiás, with a 4-month history of a painless lump in the
A lymphoepithelial cyst in the palatine tonsil

throat. His medical and dental histories were unremarkable. There were no findings on extraoral examination. The intraoral inspection revealed a well-circumscribed, yellowish, soft nodule that measured approximately 1.5 cm in diameter arising from the right palatine tonsil (Figure 1A). A closer examination showed numerous small vessels over the surface of the lesion. The clinical diagnosis was LEC. Excisional biopsy was performed under local anesthesia, and no complications were observed during the surgery. The specimen obtained was sent to the Oral Pathology Laboratory of the Federal University of Goiás.

Microscopic examination showed a ruptured cystic cavity without keratin that contained rare epithelial cells scattered in the lumen (Figure 1B). The cavity was lined by thin, parakeratinized, stratified squamous epithelium, showing a flat interface with the underlying connective tissue and lacking rete pegs (Figure 1C). The subepithelial cyst wall was thick with very scarce connective tissue and densely infiltrated by lymphocytes arranged as typical germinal centers (Figure 1B, 1C). In some areas, intraepithelial infiltration of lymphocytes could be seen (Figure 1D). The histopathological characteristics confirmed the diagnosis of LEC. The 1-week postoperative period was uneventful, but the patient was lost on subsequent follow-up.

Discussion

LEC are lesions that can be found in different parts of the body and are characterized by the presence of squamous epithelium surrounded...
Table 1. Case series of lymphoepithelial cysts published in the literature

<table>
<thead>
<tr>
<th>Author(s)</th>
<th>Year</th>
<th>N</th>
<th>Location (Number of cases)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bhaskar &amp; Colonel</td>
<td>1966</td>
<td>24</td>
<td>FM (15), T (8), P (1)</td>
</tr>
<tr>
<td>Knapp</td>
<td>1970</td>
<td>13</td>
<td>FM (7), T (2), P (4)</td>
</tr>
<tr>
<td>Acevedo &amp; Nelson</td>
<td>1971</td>
<td>9</td>
<td>FM (9)</td>
</tr>
<tr>
<td>Giunta &amp; Cataldo</td>
<td>1973</td>
<td>21</td>
<td>FM (17), P (2), BM (1), RM (1)</td>
</tr>
<tr>
<td>Buchner &amp; Hansen</td>
<td>1980</td>
<td>38</td>
<td>FM (19), T (14), P (4), RM (1)</td>
</tr>
<tr>
<td>Toto et al</td>
<td>1982</td>
<td>6</td>
<td>FM (2), T (3), P (1)</td>
</tr>
<tr>
<td>Chaudhry et al</td>
<td>1984</td>
<td>24</td>
<td>FM (24)</td>
</tr>
<tr>
<td>Nonaka et al</td>
<td>2011</td>
<td>10</td>
<td>FM (2), T (4), P (1), OF (2)</td>
</tr>
<tr>
<td>Yang et al</td>
<td>2012</td>
<td>120</td>
<td>FM (46), T (60), P (6), BM (3), Others (5)</td>
</tr>
</tbody>
</table>

FM: floor of mouth; T: Tongue, P: Palate; BM: Buccal Mucosa; RM: Retromolar area; OF: Oropharynx.

by lymphoid tissue [1-7]. Although the histopathological scenario suggests the involvement of both epithelium and lymphocytes in the genesis of these cysts, the exact mechanism of pathogenesis has not yet been completely elucidated [3, 4].

Several theories have been proposed to explain the etiopathogenesis of LEC. In their classic study, Bhaskar and Bernier sustained that LEC arose from proliferation of glandular epithelium within lymph nodes of the parotid and cervical regions [1]. Strengthening this assumption, Vickers and Von Der Muhll demonstrated cystic proliferation within lymph nodes of hamsters after surgical autogenous transplantation of bucal epithelium [12]. In addition, Adsay et al affirmed that the abdominal lymph nodes might be the source for the development of pancreatic LEC [13], in accordance with other studies [14, 15]. Although not completely rejecting the role of lymph nodes, Wu et al proposed that LECs of the parotid would start from the dilatation of ducts due to epithelial hyperplasia, which might be stimulated by lymphocytic infiltration in focal sialadenitis [2].

Regarding the etiopathogenesis of oral LECs, Knapp's theory remains the most broadly accepted. Knapp claimed that these oral cysts, actually pseudocysts, arise not from lymph nodes but from submucosal lymphoid aggregates located on the floor of the mouth, the ventral surface of the tongue, and the soft palate. Knapp coined these aggregates of ectopic lymphoid tissue of oral tonsils [10]. These “new tonsils” were added to the previously known “regular tonsils” placed at the posterior part of the oral cavity, oropharynx, and nasopharynx, which together form the lymphatic Waldeyer's ring. The ring includes the nasopharyngeal tonsil (adenoid), the paired tubal tonsils, the paired palatine (faucial) tonsils, and the lingual tonsil [7, 16].

Knapp's theory, endorsed by Buchner and Hansen [7] and Giunta and Cataldo [17], assumed that LECs, which are actually, would arise from obstruction of the crypt of an oral or palatine tonsil, resulting in a dilated cavity lined by epithelium with communication breaches with the external environment and containing keratin and desquamated cells in the lumen. However, such communication with the oral cavity has not been easily demonstrated. Acevedo and Nelson [9] do not find any continuity in nine reported cases. Giunta and Cataldo [17] and Buchner and Hansen [7] found such continuity in a few cases. Consequently, Buchner and Hansen proposed the possible existence of an additional mechanism in the pathogenesis of oral LECs [7].

It is worth noting that the great majority of intraoral LECs have been encountered on the floor of the mouth and on the ventral/posterolateral surface of the tongue [7-9, 17], which are exactly the same locations as oral tonsils [10]. In 2012, Kashima et al reported an oral tonsil in the floor of the mouth mimicking clinically a benign neoplasm or lymphoepithelial cyst. The microscopic aspect of the tonsil was very similar to that of an LEC, but without a cystic cavity. Thus, the authors postulated that oral tonsils would be earlier stages of LECs, corroborating Knapp's theory [18].

Assuming that LECs develop from oral lymphoid aggregates or tonsils, the challenge is to explain the paucity of case reports of LECs in the palatine tonsil [11] and other tonsils [19] of the lymphatic Waldeyer's ring, since all tonsils have the same histological architecture. Palatine tonsils were not cited as a location for LECs in the case...
A lymphoepithelial cyst in the palatine tonsil

series published in the literature, as we can see in Table 1. [7-10, 17, 20-23]. This observation makes our case report a rarity in the medical literature.

However, in 1987, Giunta reported 11 cases of yellow nodules located in the tongue and palatine tonsils. According to clinical criteria, the author categorized the lesions in two types, bacterial plugs and pseudocysts. Of the 11 cases described, 3 were presumably diagnosed as pseudocysts of the palatine tonsil and had spontaneous resolution over a period of 3 to 6 months [24]. The author stated these pseudocysts were the same type of lesions called lymphoepithelial cysts in other studies. Giunta affirmed that, in his experience, the occurrence of such pseudocysts in the palatine tonsils was not rare, but as lesions are small, they tend to undergo spontaneous regression before the patient seeks medical care. Spontaneous regression of a yellow swelling on the floor of the mouth, presumably a pseudocyst or LEC, was also reported by McDonnell [25].

In this paper, we reported a case of LEC arising from the palatine tonsil with four months of evolution and large size, considering the majority of intraoral LEC are less than 1 cm [7]. Based on Knapp, Buchner and Hansen, and Giunta, our case must be more properly diagnosed as a pseudocyst of the palatine tonsil that resulted from the obstruction of the tonsillar crypt, generating the accumulation of keratin within the lumen. Indeed, extravasation of keratin could be seen intraoperatively.

The tendency is for this type of lesion to resolve in a few months when a rupture occurs in the obstructed portion of the crypt. However, as it was a larger lesion that was causing discomfort to the patient, the minor surgery under local anesthesia produced a quicker resolution. In addition, the patient expressed some concern about his lesion, and the excisional biopsy allowed histopathology and patient reassurance.

Conclusions

Yellowish nodules located in the palatine tonsil usually represent lymphoepithelial cysts, better termed as pseudocysts with retention of keratin. Depending on the lesion size and discomfort reported by the patient, these nodules may be treated with a conservative approach, consisting only of a follow-up examination. When surgery is selected, a simple excision or marsupialization produces excellent results and a low recurrence rate.

Disclosure of conflict of interest

None.

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References

A lymphoepithelial cyst in the palatine tonsil


