

## CASE REPORT

# Lymphoepithelial Cyst of Tonsil

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## ABSTRACT

The tissue interactions between epithelium and mesenchyme are few of the events which result in pathological processes. One such interaction of epithelium and lymphoid tissue may result in lymphoepithelial lesions in the oral cavity. Lymphoepithelial cyst (LEC) is a rare developmental cystic lesion of the oral cavity with various histogenetic implications. Documented cases of LEC of the tonsil are low in number in the literature. We are hereby reporting a case of LEC of the tonsil in a male patient with review of literature.

**Keywords:** Cyst, Tonsil, Oral cavity.

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## INTRODUCTION

A 48 years old male patient presented with discomfort in the throat of 1 month duration which was mainly on the right side. He incidentally noticed a swelling in the right side of the throat, which did not change much with time. There was no dysphagia or dyspnea.

On examination, a well-circumscribed swelling arising from the right tonsil extending till the midline was observed. Surface of the swelling was smooth and pale pink in color (Fig. 1). There was no cervical lymphadenopathy. Hematological investigations were within normal limits. Patient was assessed for HIV status and was found to be seronegative for HIV.

Under general anesthesia, the oropharynx was exposed with a mouth gag. The cyst was found to arise from the medial border of the entire length of right tonsil. There was minimal tonsillar tissue beyond the cyst. As the cyst had a wide attachment to the tonsil, the cyst along with the tonsil was excised by dissection method. Complete hemostasis was achieved.

Gross examination showed a cystic mass measuring  $3 \times 2 \times 1$  cm surrounded by tonsillar tissue (Fig. 2). Microscopic examination of H and E stained sections showed cystic lining of keratinized stratified squamous epithelium. The cyst wall was dense with lymphoid stroma containing lymphoid follicles. Germinal center formation was seen within the follicle (Figs 3 and 4). Few mucous minor salivary gland lobules were seen. Cystic cavity was surrounded by normal tonsillar tissue.

A diagnosis of lymphoepithelial cyst (LEC) of tonsil was made. The uneventful healing of the fossa was noted by one week. Patient was followed up for a year with no evidence of recurrence.



Fig. 1: A swelling arising from the right tonsil



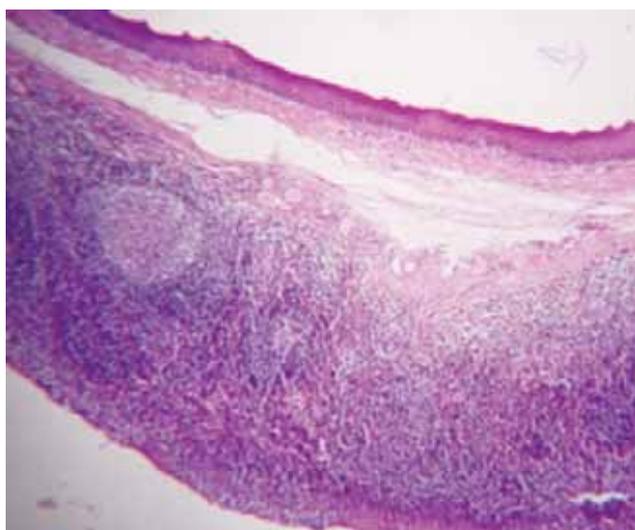
Fig. 2: Gross specimen of cyst with tonsillar tissue

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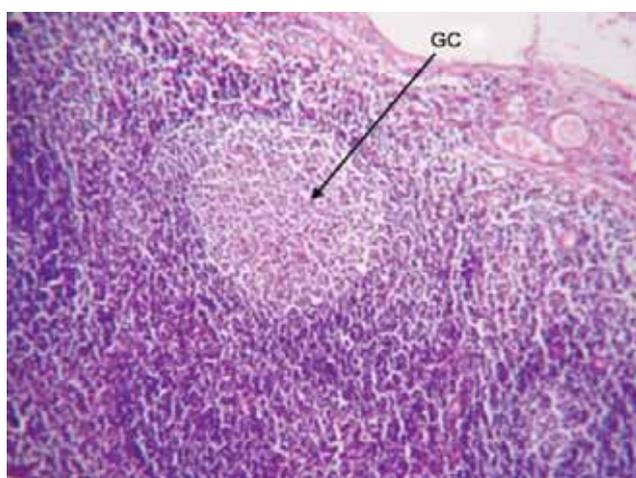
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**Fig. 3:** Cystic epithelium with lymphoid tissue in the wall (H&E, x4)



**Fig. 4:** Lymphoid follicles with germinal center (GC) (H&E, x10)

## DISCUSSION

Lymphoepithelial cyst is a rare lesion composed of an epithelial cyst within the lymphoid stroma. It is commonly found on lateral part of neck where it is called 'branchial cyst'. The term 'lymphoepithelial cyst' was introduced by Bhaskar and Bernier in 1958.<sup>1</sup> LEC has been found in varied anatomical regions like neck, oral cavity, parotid glands, mediastinum, stomach, pancreas and esophagus.<sup>1</sup>

Oral LECs are uncommon constituting about 0.18% of all lesions.<sup>2</sup> The most common location of oral LEC is the floor of the mouth (70.7%) followed by tongue, soft palate, hard palate, retromolar area and oral vestibule.<sup>3</sup> Lymphoepithelial cyst of palatine tonsils is extremely rare.<sup>4</sup>

Lymphoepithelial cyst occurs in all ages and is frequently reported in 3rd and 4th decades of life. Male to female ratio was found to be 1.42:1.<sup>3</sup>

The size ranges from 0.1 to 1.5 cm. Most of the patients of oral LECs are asymptomatic with some presenting with

local discomfort. The color is pink, yellow or white with a soft consistency.<sup>2</sup>

The pathogenesis of LEC remains unclear. There are few theories suggested to understand their histogenesis. The 'enclavement theory' described by Bhaskar suggests that the lesion arises from the epithelium which becomes entrapped in the mucosa of the oral cavity during embryogenesis.<sup>5</sup> Knapp described the 'obstruction theory' where he said that the LECs arise due to an obstruction of the crypts of oral tonsils. He called them 'pseudocysts' and not true cysts.<sup>5</sup>

In our case, the cyst was surrounded by normal tonsillar tissue and the cyst was well encapsulated. The normal surrounding tonsillar tissue could be separated from the cyst wall. This suggested entrapment of epithelium in the tonsillar tissue pointing more toward the 'enclavement theory'. On microscopic examination, the lining epithelium of the cyst was not continuous with the surface epithelium of the tonsil further confirming the enclavement theory rather than the obstruction theory of Knapp.

Histologically, it represents as a cystic lesion lined by stratified squamous epithelium with desquamated epithelial cells seen filling the cyst lumen.<sup>6</sup>

Immunostaining for cytokeratins highlight the epithelial lining of cysts leaving unstained lymphoid component.<sup>7</sup>

In recent years, LEC has gained more attention due to its association with HIV positive patients. HIV associated LEC of the parotid gland has been reported which can sometimes be the primary and main clinical manifestation of HIV infection.<sup>8</sup>

There are no reports of any case of malignant transformation of LEC in English literature.

Treatment of LEC of the tonsil consists of local conservative excision.<sup>9</sup>

Doxycycline sclerosis is a simple, cost-effective office-based nonsurgical therapeutic option. Doxycycline reduces further growth of the cyst.<sup>10</sup>

Some of the other treatment options described include radiation, azidothymidine injection and sodium morrhuate sclerotherapy of the cysts which are used mainly for LEC of parotid gland in HIV-infected patients.<sup>11</sup> Recurrence is not expected.<sup>6</sup>

## CONCLUSION

Lymphoepithelial cysts are rare lesions of the oral cavity with tonsil being the rarest site. At recent times, LECs have gained much attention due to their association with HIV-positive patients, at times even presenting as the early manifestation of HIV infection. Histopathological examination AIDS in accurate diagnosis of LECs. Malignant transformation has not been established. Lymphoepithelial cyst should be kept in mind in the differential diagnosis of tonsillar cysts.

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