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**Case Report**

# A rare presentation of benign lymphoepithelial cyst in laryngopharynx

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## ABSTRACT

Lymphoepithelial cysts are rare benign lesions with uncertain etiopathogenesis, mainly affecting salivary glands and oral mucosa. These most commonly present as lesions within the parotid gland and is typically associated with HIV infection in immunocompromised patients. These lesions also present in oral mucosa where the most common sites are floor of mouth and tongue. This case report presents an unusual presentation of a benign lymphoepithelial cyst (BLEC) in an immunocompetent patient arising from the palatine tonsil and extending up to laryngopharynx. A middle-aged male patient presented with symptoms of a swelling in the throat, and clinical examination as well as imaging features were similar to a mucocele, but on surgical excision and histopathological examination, it turned out to be a benign lymphoepithelial cyst. Even though these lesions exhibit indolent clinical behavior, with no tendency to recur after conservative surgical management, these are frequently misdiagnosed clinically due to the similarity of presentation to other common oral mucosal lesions. Hence an accurate clinical diagnosis requires histopathological correlation, especially in those cases which present in unusual locations.

**Keywords:** Lymphoepithelial cyst, Laryngopharynx, Rare benign lesion, Cyst of pharynx, Blec

## INTRODUCTION

Lymphoepithelial cysts are uncommon benign lesions which arise within the lymphoid tissues, particularly in salivary glands, and in oral mucosa, commonly floor of mouth and posterior aspect of tongue. These presenting in tonsils are extremely rare and there are 2 reported cases in literature of solitary LEC of tonsil in 2010 and 2013.[1,2] There is another rare case of multiple bilateral lymphoepithelial cysts of the palatine tonsils in a 72-year-old patient, reported in 2013.[3] Multiple lymphoepithelial cysts are reported within the parotid gland of individuals associated with HIV infection, Sjogren's syndrome and other autoimmune diseases. There is also report of an incidentally found lymphoepithelial cyst in the laryngeal vestibule during an autopsy on an 82-year-old female in literature, published in 2015.[4]

These constitutes to less than 1% of all mucosal lesions affecting the oral cavity. Most of these occur in adult females, presenting as painless well circumscribed yellowish submucosal nodules. Multiple theories are proposed in the pathogenesis of lymphoepithelial cysts, one of which is that these represent true cysts from ectopic glandular epithelium trapped in the normal lymphoid tissue during embryological development.[5] Another theory proposed is that these are pseudocysts due to obstruction of tonsillar crypts.[6] Microscopically these are lined by stratified squamous epithelium, with dense lymphoid infiltrates, containing lymphoid follicles with germinal centres.[7]

Primary diagnostic tests included imaging like CT scan, MRI and PET-CT. Different treatment modalities included surgical as well as nonsurgical options like sclerotherapy, radiotherapy and simple clinical observation. The

treatment should be selected depending on multiple factors like patient choice, treatment costs, severity of symptoms, location of lesion.[10]

## CASE REPORT

40-year-old male patient presented to ENT outpatient department with persistent foreign body sensation of throat and voice change manifesting as heaviness and muffling of voice for 3 months duration. Symptoms were gradually progressing. A flexible laryngoscopy was done as outpatient procedure which revealed a globular, mucosa covered lesion arising with a pedicle from lower pole of tonsil and reaching up to the level of arytenoid. A provisional diagnosis of pharyngeal mucocele was made and patient was subjected to imaging. CT scan of neck revealed an elongated smooth ovoid hypodense lesion of approximately 32×18×15 cm is noted along the right posterolateral wall of hypopharynx. Lesion was abutting pharyngeal wall posterolaterally, and right aryepiglottic fold anteriorly.

Patient underwent excision biopsy of the lesion with bipolar cauterization of the base of the lesion under general anesthesia; and the specimen was sent in toto for histopathological examination. Gross findings were single piece of tissue, 3×2×1 cm in greatest dimensions, greyish white in color, soft, cut surface shows pale yellow jelly like consistency. Microscopic sections showed a cystic lesion lined by stratified squamous epithelium. The underlying stroma exhibited dense lymphoid aggregates with prominent germinal centers. The cyst wall was composed of fibro collagenous tissue with mild chronic inflammation. No evidence of dysplasia, malignancy, cilia, goblet cells or granulomas identified. Histopathological features were consistent with benign lymphoepithelial cyst. HIV serology was done and was reported negative.

The patient is asymptomatic and on regular 6 monthly follow up with office laryngoscopy for last 1 year with no evidence of recurrence so far.

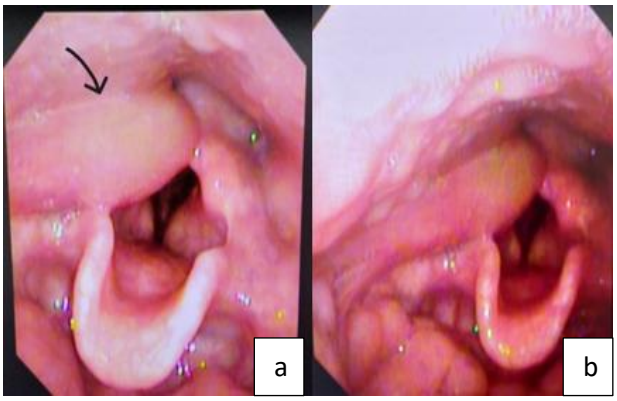


**Figure 1 (a and b): Flexible laryngoscopy images showing the mucosa covered lesion arising with a pedicle from lower pole of tonsil and reaching up to the level of arytenoid.**

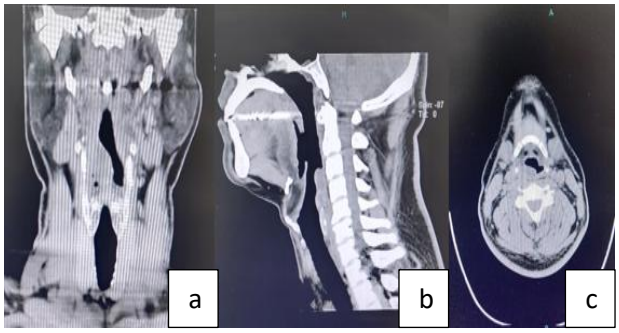


**Figure 2 (a-c): CT images (coronal, sagittal and axial) showing the location of the lesion.**

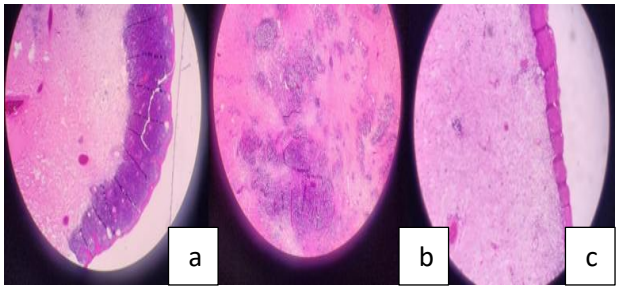


**Figure 3 (a-c): Histopathology slides showing lesion lined by stratified squamous epithelium, underlying stroma exhibiting dense lymphoid aggregates with prominent germinal centers and the cyst wall composed of fibro collagenous tissue.**

## DISCUSSION

Herein reported above is a case of benign lymphoepithelial cyst, presenting with the clinical features of a laryngeal lesion, primarily arising from the lower pole of right palatine tonsil extending to the laryngopharynx. The term lymphoepithelial cyst was first recommended by Bhaskar and Bernier in 1959 and the first case in oral cavity was described by Gold and Lewittown in 1962.[3,8] In this case the lesion presented in an immunocompetent patient, whereas they were commonly described in literature as lesions within the parotid glands of immunocompromised patients. In oral cavity, the most common sites described are floor of mouth, and ventral and posterolateral aspects of tongue.[1] One more interesting aspect is that the above lesion is larger (more than 3 cm) compared to majority of lymphoepithelial cysts described in literature which are around 1 cm.[1] In our case, an unusually large cyst presenting at an uncommon site underlines the importance of proper clinical examination and need for proper diagnosis. The etiopathogenesis of lymphoepithelial cysts is not clearly understood; possible theories that have been suggested are local trauma, proliferation of trapped epithelial remants, obstructed minor salivary gland ducts and obstructed tonsillar crypts.[7,11] The differential diagnoses to be considered include neoplasms like lipoma or a granular cell tumor, developmental lesions like Fordyce granules, dermoid and epidermoid cysts, other calcified masses like sialolith and tonsillolith, also

reactive and inflammatory conditions, and oral manifestations of systemic diseases.[13] In most of the cases reported in literature, the primary diagnosis was not lymphoepithelial cyst, which highlights the unfamiliarity among clinicians and need for more awareness regarding these lesions.

Some lymphoepithelial cysts are found to regress spontaneously.[9] Hence asymptomatic lymphoepithelial cysts at typical locations maybe managed with reassurance and wait and watch policy. Treatment plans should be selected according to personal situation and patients' own choice, and options include surgical treatment and non-surgical treatments such as sclerotherapy, puncture treatment, radiotherapy and dynamic observation.[12] When surgery is considered, conservative simple surgical excision or marsupialization is found to offer excellent results. A retrospective case analysis of 201 patients in December 2023 reported no recurrence after surgical excision upto 24 months follow up.[10]

## CONCLUSION

Even though lymphoepithelial cysts are benign lesions with low recurrence rate, when symptomatic, conservative surgical excision is the treatment of choice. Histopathology is essential to arrive at an accurate diagnosis because of its similarity to other mucosal lesions. The treatment decision should be based on the size and location of the lesion, and the symptoms caused by the lesion. Long term follow up is not indicated unless the patient is symptomatic.

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