

# Lymphoepithelial Cyst, A Diagnostic Dilemma: Case Report

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## ABSTRACT

Lymphoepithelial cysts are benign, slowly growing unilocular or multilocular lesions that appear in the head and neck region. They are also called branchial cysts and occur due to lymphocyte induced cystic ductular dilatation. The confirmatory diagnosis is always made on histopathological examination after resection.

A 29 years old female patient presented to ENT department, Pt B. D. Sharma PGIMS, Rohtak with a soft to firm, non-tender, swelling in the submandibular region since 2 -3 days and was referred to Department of Pathology for FNAC on which a differential diagnosis of infected epidermoid cyst or squamous cell carcinoma was made and excision biopsy for confirmation and categorization was advised. The swelling was then excised and sent for histopathological examination and a final diagnosis of Lymphoepithelial cyst was made. The confirmatory diagnosis is always made postoperatively by histopathological examination. The treatment of a lymphoepithelial cyst is the surgical approach, which includes complete enucleation of the cyst. This is a case report of a lymphoepithelial cyst diagnosed on histopathology specimen for which differential of Infected cyst or Squamous cell carcinoma was given. The authors intend to highlight the importance of early histopathological diagnosis of Benign LEC as it has been reported to undergo malignant transformation

**Keywords:** Lymphoepithelial cyst, Branchial epithelium, Squamous cell carcinoma, HIV Infection

## Introduction

Bernier and Bhaskar coined the term lymphoepithelial cyst (LEC), and defined them as solitary or multiple cysts within the lymph nodes associated with salivary glands.<sup>[1]</sup> The LEC accounts for 0.09% - 0.18% of all the lesions occurring in oral cavity.<sup>[2]</sup> and commonly occurs in adults with 60-80% cases being in females.<sup>[3]</sup> According to the authors benign lymphoepithelial cysts (BLC) arise from cystic degeneration of salivary gland inclusion in the lymph node. They emphasized that these cysts are distinct entities from lymphoepithelial lesions.<sup>[1]</sup> Other authors including Stramandinoli-Zanicotti et al believe it to be probably dysontogenetic benign cyst arising from the epithelial remnants retained in lymphoid tissues during the embryogenesis or from branchial cleft epithelium.<sup>[4]</sup>

## Case Report

A 29 years old female patient presented to the ENT department, PGIMS, Rohtak with the chief complaint of swelling in the submandibular region for 2-3 days and was referred to Department of Pathology for FNAC. Patient had a history of right tympanoplasty 1 year back and ear ache for which she was taking treatment. Her medical, dental and personal history was non-contributory.

On examination the swelling was present over the left upper cervical region measuring 2 x 2 cm. on palpation the swelling was soft to firm in consistency. Radiological details

and images were not available with the patient. The patient was referred to our department for FNAC of the swelling. Informs consent was taken from the patient for FNAC. FNAC yielded a dirty white aspirate. Smears prepared and examined showed nucleated and anucleate squames in a background of cystic macrophages, degenerated and intact neutrophils and amorphous material. Also seen were few singly scattered cells showing thick cytoplasm and irregular hyperchromatic nucleus possibly due to degenerative changes (Figure 1a & 1b). A cytological possibility of cyst of developmental origin (infected epidermal cyst) or a well differentiated squamous cell carcinoma was given.

The patient was advised excision of the cyst for confirmation of diagnosis. An excision biopsy of the cyst was performed and sent to the Department of Pathology, Pt B. D. Sharma PGIMS, Rohtak for histopathological examination. On gross examination globular cystic mass measuring 3.5 x 2 x 2 cm was received. Cut surface was yellow in color and firm to cystic in consistency. Also received was an attached grey brown soft tissue piece measuring 0.8 x 0.5 x 0.2 cm. The representative sections passed from cystic structure showed histopathological features of lymphoepithelial cyst (Figure 2a & 2b). Also, sections passed from attached soft tissue showed structure of multiple lymph nodes revealing reactive hyperplasia. No salivary gland tissue was seen. Patient was followed up and showed improvement with no local recurrence of the lesion.

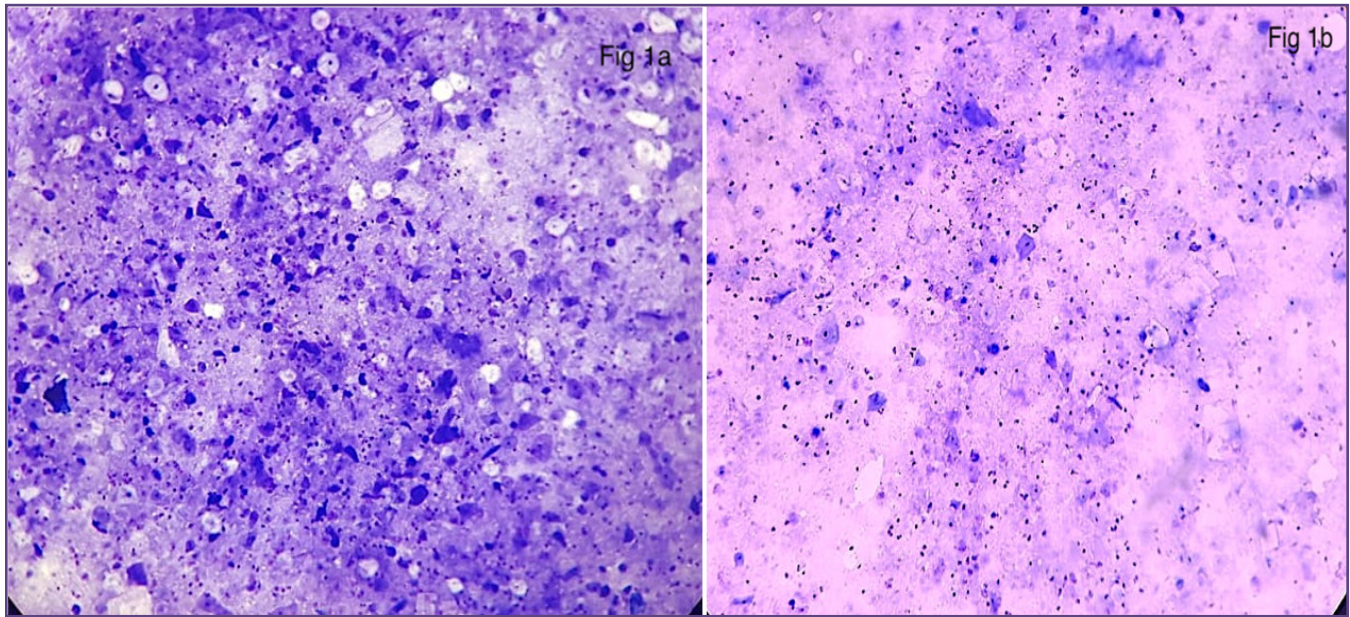


Fig. 1a & 1b: FNAC -Nucleated and anucleate squames in a background of cystic macrophages and amorphous material. (Leishman x400).

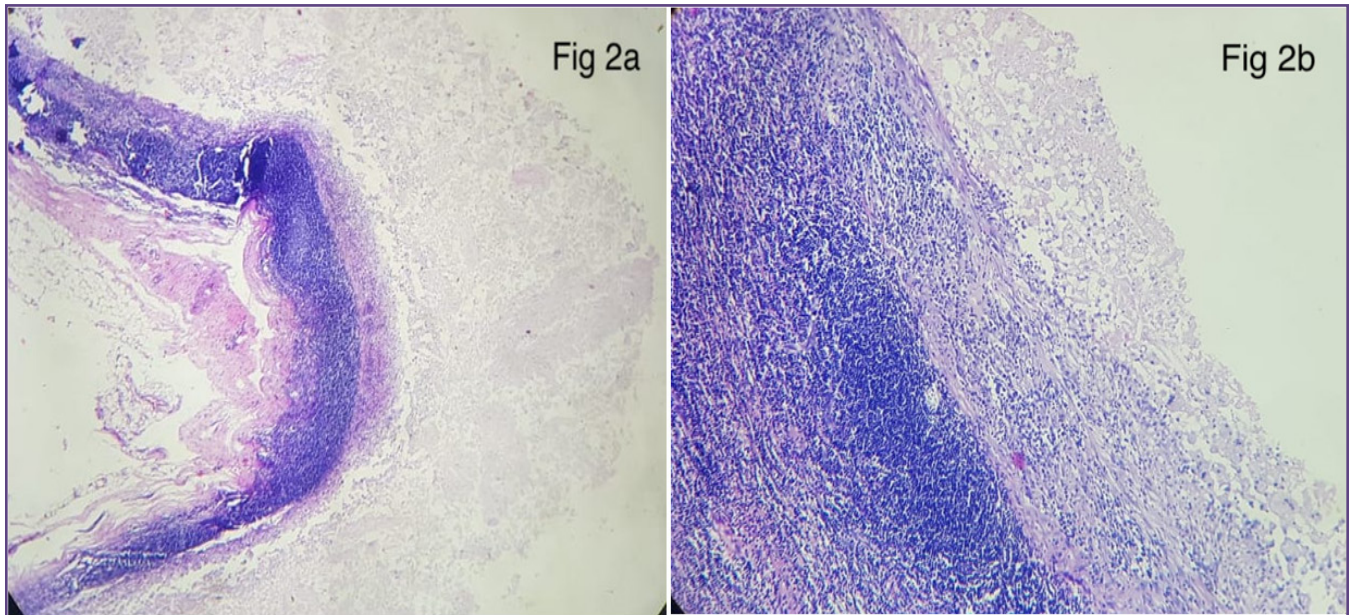


Fig. 2a: Cyst wall lined by squamous epithelium with lymphoid tissue in subepithelium. (H&E - X100), 2b: Cyst wall lined by squamous epithelium with lymphoid tissue in subepithelium (H&E - X400).

## Discussion

Lymphoepithelial cysts are usually found over the lateral aspect of neck but can also be found in salivary glands and floor of mouth. Within the category of BLCs various types of epithelium have been described. The most frequent has been squamous but variable combinations of cuboidal, columnar, ciliated columnar and mucin producing epithelia have also been reported. In addition, rare examples of squamous

epithelium containing sebaceous differentiation have been documented.<sup>[5]</sup> Recurrent infections changes the lining epithelium to fibrous or granulomatous lining. The cases with columnar epithelium are usually dormant while those with squamous epithelium had purulent content and symptoms.<sup>[6]</sup>

The characteristic histopathological pattern is that of glandular or squamous epithelium lined cleft surrounded

by abundant lymphoid tissue with prominent germinal centers, with some specimens even containing salivary gland tissue a duct.<sup>[7]</sup> In 1989 Skouteris et al, reported that more than 90% of these cysts were lined by stratified squamous epithelium that might or might not be keratinized. Some cysts demonstrated respiratory epithelium. They also reported cases with the cysts lined by either columnar or cuboidal epithelium.<sup>[8]</sup>

Four major theories have been put forth and explained by Maran and Buchman in 1974 which are,<sup>[9]</sup> Branchial apparatus theory, Cervical sinus theory, Thymopharyngeal theory Inclusion theory Squamous cell carcinoma occurring in LEC is rare. Martin proposed 4 criteria for provisional diagnosis of such a clinical lesion. They are the tumor should occur in the line extending from anterior border of sternocleidomastoid to the clavicle, the histologic appearance should be consistent with an origin from tissue present in branchial vestigial, the patient must survive at least five years without development of other tumor that be regarded as primary and it must be demonstrated that cancer developed in the wall of an epithelial lined cyst situated in the lateral aspect of the neck.<sup>[7]</sup>

Based on the location there are 4 types of cervical LEC<sup>[6]</sup>

1. Lying superficially beneath platysma and cervical fascia along the anterior border of the sternocleidomastoid.
2. Lies on the great vessels
3. Extends through the carotid bifurcation to the lateral wall of pharynx, occasionally exhibiting a prolongation to the skull base.
4. Cyst usually lined by columnar epithelium and lying against the pharyngeal wall.

The Lymphoepithelial cysts have been associated with HIV infection as part of diffuse infiltrative lymphocytosis syndrome. HIV associated salivary gland disease is a lymphoid hyperplasia. The incidence of HIV associated salivary disease is about 3-10% among the HIV positive patient. It affects all the age groups and both the sexes equally. Salivary gland disease usually develops before AIDS and sometimes it's the first manifestation of HIV infection. It is usually bilateral and is accompanied by cervical lymphadenopathy.<sup>[10]</sup>

The differential diagnosis of cyst include lymphoepithelial sialadenitis (show hyperplastic lymphoid infiltrates with loss of acini of salivary gland), Warthin tumor, (show double layered oncocytic epithelium), extra nodal marginal zone B cell lymphoma (monoclonal with centrocytic or monocytoid appearing cells), retention cyst (mucinous

cystic components will be present), mucosa associated lymphoid tissue lymphoma. Patients with salivary gland LEC are at an increased risk of developing lymphoma and that is the reason such cysts should be treated as early as possible.<sup>[3]</sup> However, the exact incidence of malignant transformation is unknown.

Treatment of Lymphoepithelial cyst includes both conservative as well as surgical approach. The conservative approach includes decompression of the cyst by aspirating the fluid out. Such a procedure should be considered in immunodeficient patients. The other treatment is external radiotherapy. However, the definitive treatment is surgical management by complete enucleation of the cyst along with excision of the involved gland.<sup>[3]</sup>

## Conclusion

Lymphoepithelial cysts are benign and should be treated as early as possible as it can transform into a malignant lesion such as malignant lymphoma, adenocarcinoma, mucoepidermoid carcinoma.<sup>[8]</sup> This case report aims to highlight the importance of keeping all the differential diagnosis in mind when reporting such a case on cytology as well as to rely only on the histopathology of the excised cyst for a confirmatory diagnosis.

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