BANGLADESH JOURNAL OF
OTORHINOLARYNGOLOGY
DOI: https://doi.org/10.3329/bjo.v31i1.84294
www.banglajol.info

Article info
Received : 17.01.2025
Accepted : 29.03-2025
No. of Tables : 0
No. of Figure : 03
No. of References : 20

Case Report

Parotid Gland Lymphoepithelial Cyst: A Case Report

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Abstract:

Benign lymphoepithelial Cyst (BLEC) is one of the rare benign pathology of parotid gland. These painless cysts grow slowly which may mimic haemangioma, Warthin'stumour and lymphoma. Now a days BLEC incidence has increased due to increasing numbers of AIDS patients but it is very much rare in non HIV infected patients. In case of non HIV positive it is embryonic in origin. So surgeons should have concern about HIV infection when patient presents with lymphoepithelial cyst. In our case, patient is HIV negative.

Keywords*: Parotid Gland, Parotidectomy, Lymphoid Hyperplasia, Lymphoepithelial Cyst, Branchial Cleft Cyst.*

Cite the Article: *Sarker DK, Rahman DMA, Haque Z, Mahalam M. Parotid Gland Lymphoepithelial Cyst: A Case Report. Bangladesh J Otorhinolaryngol 2025; 31(1): 47-51.*

Introduction:

Salivary glands are common sites of benign pathology. There is a multiplicity of tumor type with a variable and diverse histological pattern. It may be very difficult to distinguish between benign and malignant pathology on the basis of fine needle aspiration or Imaging. Many salivary malignancies are characterized by an indolent growth pattern with a high tendency to recur locally or give rise to distant metastases. Such recurrences may appear after many years of apparent disease-free survival. So, the nature of salivary neoplasms should be thoroughly understand by the head and neck surgeon.

Benign lymphoepithelial cyst (BLEC) of the parotid gland, which is also known as branchial cleft cyst, is a rare benign cystic neoplasm of embryonic dysplasia. It usually occurs in the anterolateral region of the neck but has been reported in the oral cavity or parotid gland in rare cases[1,2]. HIV-associated BLECs are often bilateral and with multicystic appearance. Unilateral BLECs are more

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common in HIV negative individuals and rare in HIV positive ones[3]. Bernier and Bhaskar introduced the term "lymphoepithelial cyst" to stress that this lesion is not an embryologic remnant and defined it as solitary or multiple cysts within lymph nodes associated with salivary glands[1]. In literature review it is found that in the last few years, such cysts have been found in increasing numbers in AIDS patients as well as in the patients belonging to the AIDS risk groups[4]. The etiology of these cysts is controversial but the treatment is not.

Case Report:

The patient of this case report had admitted in department of ENT, TMSS medical college and hospital in 15.07.2024. A 35 years old female presented with swelling below and in front of her right ear for 1 year which was gradually increasing in size. There was no pain but mild discomfort. No history of repeated fever, dry mouth, weight loss, other systemic infection. There were no history of trauma, parotid or dental surgery. On inspection overlying skin was normal, no scar mark, no engorged vein was found. On palpation it was about 1.5 cm x 1.5 cm in size, non-tender, soft to cystic in consistency, slightly mobile, free from underlying structure and overlying skin. No audible bruit was detected. There was no sign of facial nerve palsy. Ear, Throat and neck examination revealed no abnormality. There was no associated regional lymph-adenopathy. On intra oral examination, there was free flow of saliva from the right parotid duct (Stensen's duct) and bimanual palpation revealed no abnormality.In CT scana fairly large cystic mass having few foci of calcification was noted in superficial and deep part of parotid gland. The mass was measuring about 1.5 cm x 2.1 cm x 1.9 cm, encasing external carotid artery. Post contrast scan showed marginal enhancement of lesion. FNAC showed benign cystic lesion of parotid. We excised the cyst along with surrounding normal parotid tissue by preserving the facial nerve. Post-surgery there was no facial nerve palsy or wound infection. Histopathology report showed benign lymphoepithelial cyst composed of lymphocytes of different stages of maturation with associated squamous lining of the cyst focally lymphocytes permeate the cyst lining epithelium. No malignancy or granuloma was seen.

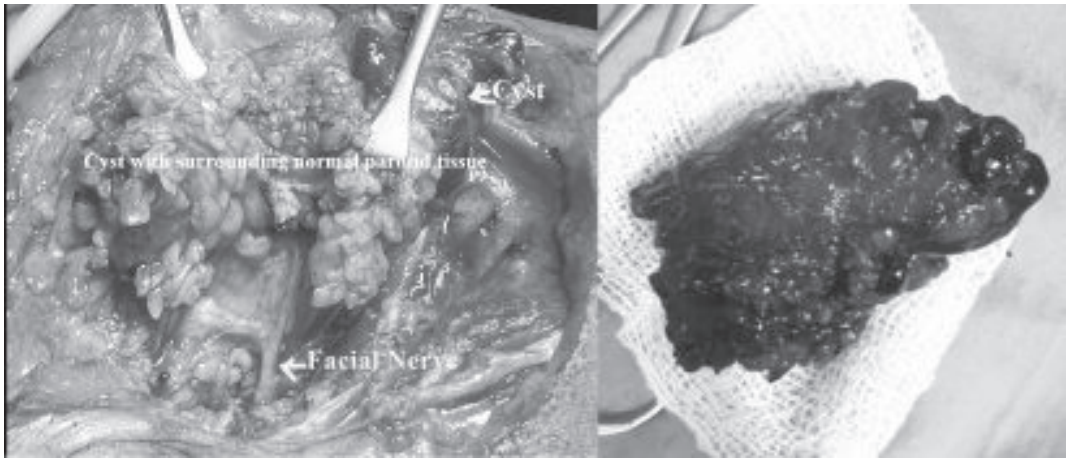

Fig.-1 :

Discussion:

BLEC is a rare cystic neoplasm with benign embryonic dysplasia. It might probably derive from the cystic degeneration of salivary gland inclusions within lymph nodes[3]. The first case of BLEC was reported by Hildebrandt in 1895.Lymphoepithelial cystsare usually found overthe lateral aspect of neck but can also be found in salivary glands and floor of mouth.[5] It presents as gradual, painless swelling of one or both parotid glands. BLECs should be considered in the differential diagnosis if cystic or mixed parotid lesions are discovered[3].

Generally Branchial Cleft Cysts affect the lateral side of neck deep to the sternocleidomastoid muscle. However, any site with lymphoid tissue component, lymphoepithelial cysts can occur. The parotid gland has intraparotid lymph nodes embedded within thus, being a possible site for development of this type of cysts. These cysts are not locally invasive but may grow to reach considerable sizes enough to cause facial deformities[6,7].

With the emergence of the HIV epidemic, the incidence of BLEC in the parotid gland gradually increased, and researchers found that BLECs were closely related to HIV infection in most cases. In sharp contrast, BLECs are rarely found in non-HIV-infected patients, and the exact prevalence of BLEC in this population has not been reported.[1,2,8]

Differential diagnosis of parotid lymph-oepithelial cyst include: Warthin's Tumor, Pleomorphic Adenoma, Hemangioma, Lymphoma[7].Diagnosis is done after studying the case history, detailed examination and histopathological examination. Noninvasive diagnostic modalities are ultrasonography, computed tomography scan and magnetic resonance imaging. Invasive modalities are fine needle aspiration cytology and biopsy of the glandular tissue[6]. Imaging findings include multiple variable-sized cysts, solid and mixed cystic/solid masses within the gland, reactive cervical lymphadenopathy, tonsillar hypertrophy, or intraparotid node enlargement. The differential diagnosis for these imaging findings includes Sjögren syndrome, Warthin's tumor, and metastases[9]. In our case, CT scan shows a cystic mass having few foci of calcifications in superficial and deep part of rightparotid gland measuring about 1.5 x2.1 x 1.9 cm encasing external carotid artery. FNAC shows benign cystic lesion. (Fig.- 2).

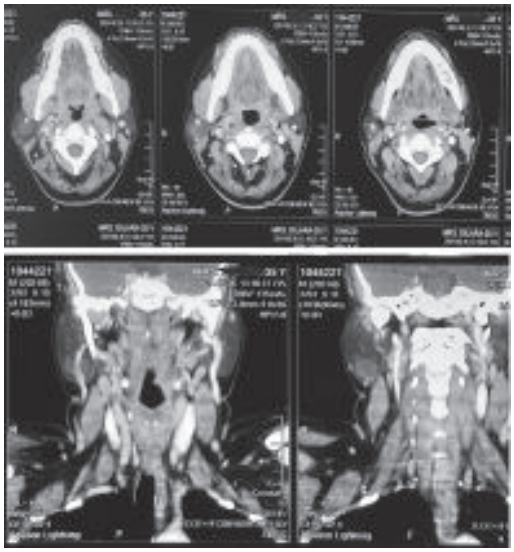

Fig.-2

Histopathology is always confirmatory. It reveals epithelial cysts lined by squamous, columnar, cuboidal, or mucin secreting epithelium and abundant lymphoid tissue with germinal center formations underneath. BLEC can neither recur nor metastasize; hence, a definite histological diagnosis is necessary for exclusion of malignant lesions speciallylow grade mucoepidermoid carcinoma and metastatic squamous cell carcinoma[10]. In our case, histopathology shows cyst composed of lymphocytes of different stages of maturation with associated squamous lining. No granulation or malignancy is seen. (Fig.-3)

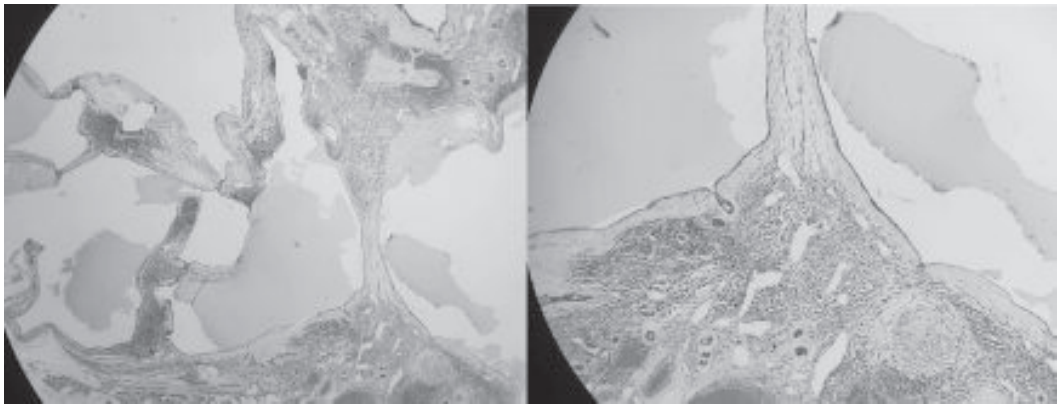

Fig.-3

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[11]. However, the exact incidence of malignant transformation is unknown[5].

Management of Lymphoepithelial cyst includes both conservative as well as surgical approach. The conservative approach includes decompression of the cyst by aspiration and drainage of fluid. Such a procedure should be considered in immunodeficient patients. The other treatment is external radio-therapy,sclerotherapy, anti-viral therapy. However, the definitive treatment is surgical management by complete enucleation of the cyst along with excision of the involved gland and may not get a recurrence in their lifetime[5,6,7]. In this case we have done excision of cyst with surrounding normal parotid tissue under general anesthesia.

Take home message is that BLECs should always raise concern of HIV infection and can present as cystic parotid swellings even in children with a poor immune status. These are rarely malignant, but they can aggressively turn so. For a child with any neck mass, human immunodeficiency virus test should be done to know if it is the underlying cause of lymphoid hyperplasia. In case of adult when BLEC diagnosis is confirmed, clinicians need to check the patient's HIV status, in order to timely perform the appropriate treatment. Excision of cyst with a portion of normal tissue should be appropriate management in these patients[3,6].

Conclusion:

In the absence of HIV infection BLEC is a rare disease condition of parotid gland which is an embryonic dysplasia. Diagnosis and management of patients with non HIV infected BLEC are poorly understood. In this case report wehave highlighted all the differential diagnosis in mind and our suggestion is to rely only on the histopathology of the excised cyst for a confirmatory diagnosis.

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