

## Case Report

---

### Benign lymphoepithelial cyst

Saequa Habib<sup>1</sup>, Md. Mustafizur Rahman<sup>2</sup>, A Allam Choudhury<sup>3</sup>, Mohammed Kamal<sup>4</sup>

#### Abstract:

*Benign lymphoepithelial cyst, also known, as branchial cyst is an infrequent lesion usually occurs in the parotid gland or the lateral cervical area including lymph nodes. It occurs due to the process of lymphocyte-induced cystic ductular dilatation and is always diagnosed postoperatively by histopathological examination. The cyst is usually lined by squamous epithelium surrounded by lymphoid stroma. Although unusual, two cases diagnosed within two months are reported here. Both patients were male but the locations of the lesions were different. One lesion was within cervical lymph node and the other was near the parotid gland. Both the lesions were surgically excised and diagnosed histopathologically as benign lymphoepithelial cyst. Its frequent association with HIV infected patients since last two decades has made it a significant lesion now-a-days. In this regard along with reported two cases brief literature reviews including origin, diagnosis and treatment are discussed.*

**Key words:** Benign lymphoepithelial cyst, parotid, lymph node, HIV infected patients.

#### Introduction:

Benign lymphoepithelial cysts are lesion of the parotid or upper cervical lymph nodes characterized by multiloculated cyst lined by glandular or squamous epithelium surrounded by a florid lymphoid hyperplasia with prominent germinal centers which may even be infiltrated into the lining epithelium<sup>1</sup>.

Developmentally these are examples of branchial cyst most often found in the lateral cervical area just below the angle of the mandible, anterior to the sternocleidomastoid muscle<sup>2</sup>. It also may occur in the oral cavity beneath the tongue and even in pancreas<sup>1,3</sup>.

Cystic lesions within the parotid glands are uncommon comprising 5% of all salivary gland tumors and many of them represent cystic component of neoplasm<sup>4</sup>. Sometimes some solid lymphoepithelial lesions of parotid gland like adenolymphoma, lymphoepithelioma and Mikulicz's disease or Sjogren's syndrome may have cystic areas mimicking benign lymphoepithelial cyst<sup>5</sup>. Benign lymphoepithelial cysts present as a painless swelling in the parotid area without attachment to the facial nerve or in the lateral cervical area<sup>6</sup>. A few lymphoepithelial cysts occur in children, but the peak incidence is in the 4<sup>th</sup> through 7<sup>th</sup> decades of life. The male to female ratio is 2:1<sup>7</sup>.

1. Asst. Professor, Department of Pathology, BSM Medical University, Dhaka, Bangladesh.
2. Resident, Department of Pathology, BSM Medical University, Dhaka, Bangladesh.
3. Asst. Professor, Department of Otolaryngology-Head and Neck Surgery, BSM Medical University, Dhaka, Bangladesh.
4. Chairman and Professor, Department of Pathology, BSM Medical University, Dhaka, Bangladesh.

**Address of Correspondence:** Dr. Saequa Habib, Asst. Professor, Department of Pathology, BSM Medical University, Dhaka.

Two cases of benign lymphoepithelial cyst were histopathologically diagnosed in the department of pathology at Bangabandhu Sheikh Mujib Medical University (BSMMU) within two months. One case was a lesion within cervical lymph node and another case was located near parotid gland.

### Case studies:

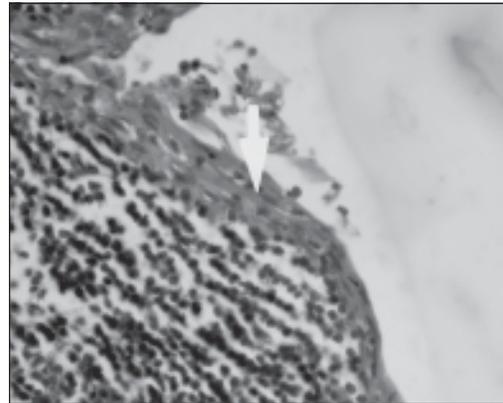
#### Case 1:

An 18-year-old male patient, presented with a painful swelling in the left posterior cervical region for one and half months. The clinical impression was tubercular lymphadenitis. Then the patient was admitted in the Department of Otolaryngology-Head and Neck Surgery of BSMMU. Magnetic resonance imaging (MRI) was done and it revealed a hypo- dense area with regular margin within the lymph node. Other routine investigations were also done which revealed normal findings. The lesion was surgically excised and sent for histopathological examination. On gross examination the specimen was a 4cm lymph node. The cut surface was mostly solid and a cystic area measuring 1 cm in diameter containing whitish material was also found (Fig 1). Histopathology of the nodule showed a lymph node containing a cyst lined



**Fig.- 1:** Gross photograph showing a lymph node containing cyst (Case no 01)

by squamous epithelium with surrounding areas densely infiltrated with mostly lymphocytes. No evidence of malignancy was seen. The diagnosis was benign lymphoepithelial cyst (Fig 2).



**Fig.-2:** Photomicrograph showing benign lymphoepithelial cyst wall lined by squamous epithelium with lymphocytic infiltration in the wall (H & E x 400, Case no. 01)

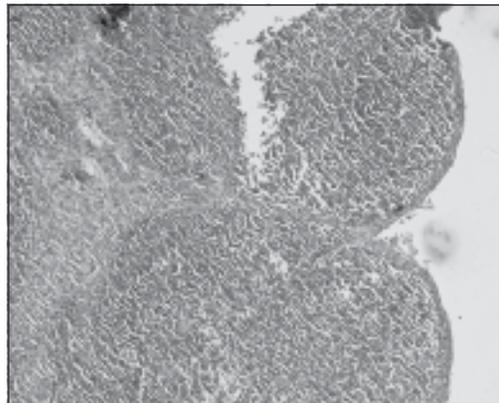
#### Case 2:

A 42-year-old male was admitted in the Department of Surgery of BSMMU with a soft swelling involving the entire left parotid for three months. The right parotid was normal. He had no history of upper respiratory tract infection or trauma to the parotid area. He was stable and afebrile. On examination he revealed 4 cm diffuse soft fluctuant swelling in the left pre and inferior auricular area. There was associated regional lymphadenopathy. The ultrasound was then performed and revealed a well-defined hypo-echoic area (2.91x 1.77cm) with multiple tiny echogenic debris noted in the lower part of the left parotid gland. Fine needle aspiration cytology (FNAC) was also done. On aspiration fluid came out and cytology showed some group of acinar cells, many macrophages and few lymphocytes but no malignant cell. Other routine investigations were done which

revealed normal findings. Surgical specimens were comprised of a collapsed cyst with fragmented pieces of salivary gland tissue and one lymph node (fig 3). The inner surface of cyst wall was rough. Histopathologically the cyst was lined by squamous epithelium with dense infiltration of lymphocytes in the wall forming follicles at places (Fig 4). Salivary gland tissue and lymph node did not show any significant change. No evidence of malignancy was seen. The diagnosis of benign lymphoepithelial cyst was made.



**Fig.-3:** Gross photograph showing cyst and parotid gland (case no. 2)



**Fig.-4:** Photomicrograph of benign lymphoepithelial cyst containing squamous lining with lymphoid follicles (H&E x 200, case no.2).

#### Discussion:

The majority of these cysts occur in young adults<sup>3</sup>. The age of patients ranges from 16-

69 years with the mean age of 44 years old. The distribution was found to be three times more frequent in males than in females<sup>2</sup>. Both the patients reported were male and the ages were also found to be consistent with the literature reviews.

These cysts are slow growing. They originate through cystic transformation of epithelium in lymph node or salivary gland or epithelium of the branchial apparatus<sup>3</sup>.

Godwin J.T. mentioned that many observers (Nicholson, Löwenstein, Lubarsch, Bairati and Bunting et al.) had demonstrated parotid inclusions in lymph nodes<sup>5</sup>. Even in tissue removed from the parotid and contiguous areas for cancer of head and neck in memorial hospital the presence of parotid ducts or acini with lymphoid aggregates or lymph nodes were frequently noted<sup>5</sup>. On the other hand, aggregates of lymphocytes within parotids were also found by Neisse, Thompson and Bryant stated by Godwin J.T.<sup>5</sup>. The first case of branchial cyst in the parotid gland was reported by Hildebrant in 1895. In 1990, Camilleri and Lloyd also reported 70 cases of this type of cysts and Fujibayashi found another 33 cases through the review of 5 publications<sup>8,9</sup>.

So the origin and development of branchial cysts (lymphoepithelial cyst) is a controversial subject, and many theories have been suggested. At least four theories have been put forward to explain the origins of branchial cysts depending on embryology, histology and clinical findings by Camilleri and Lloyd, (1990) which are: 1. Branchial apparatus theory. 2. Cervical sinus theory. 3. Thymus-pharyngeal duct theory. 4. Inclusion theory. The first two theories are also known as classical theory which holds that the cysts develop from the remnant of the branchial cleft because it occurs in the area of the embryonic gill apparatus<sup>8</sup>. The inclusion

theory or so-called recent theory described by Bhashkar and Bernier in 1959 considers that the cysts arise from cystic changes in parotid epithelium that become entrapped in the upper cervical lymph node during embryonic life which is also the most feasible explanation for our reported cases<sup>10</sup>.

This type of lesion may be present for a long time without giving rise to severe or serious manifestation. Discomfort, occasional pain or enlargements of the cervical region unilaterally or bilaterally are the usual presenting symptoms<sup>5</sup>. Usually HIV- infected patients have bilateral parotid lymphoepithelial cysts<sup>11</sup>.

The most common site for the lymphoepithelial cyst is in the lateral cervical area. Bashkar and Bernier in 1959 mentioned from the file of the Armed Forces Institute of Pathology, that only 5 cases were located in the parotid area among 468 cases of branchial cyst<sup>10</sup>. Another study by Rickles and Little in 1967 showed that only 14 cases out of 149 cases of branchial cysts were found in the upper part of neck along the angle of the mandible<sup>12</sup>. Our first case was in the left posterior triangle of neck and the second case was in left parotid area.

The true nature of this lesion is not known, though likely inflammatory. However in some instances the lesion presents as a neoplastic mass<sup>5</sup>.

In 1988, Wymen et al concluded that ultrasound scanning was a simple investigation by which cystic and solid areas could be easily distinguished<sup>13</sup>. Usually lymphoepithelial cyst is unilocular but in HIV infected patients it is multiloculated<sup>7</sup>. Ultrasound was done in our second case which also revealed a unilocular cyst containing cellular debris. Besides that FNAC was used to identify the nature of cystic content which usually yielded clear watery or straw color fluid mentioned by Skouteris

et al in 1989<sup>14</sup>. The cytology of these lesions consists of anucleate squamous and squamous cells or cuboidal cells in a reactive lymphoid background with scattered histiocytes<sup>11, 15</sup>. FNAC was also done in our second case. According to Altman and Bailey in 1994. Magnetic resonance imaging (MRI) can give the clear images of this lesion and its anatomical relations than CT scanning<sup>4</sup>. In our first case MRI was done and revealed a cyst within a lymph node. It has also been suggested that a patient with sufficiently characteristic CT appearance like multiple parotid cysts and cervical adenopathy should be screened for HIV exposure<sup>11</sup>.

In the review of literatures reported that more than 90% of lymphoepithelial cysts were lined by squamous epithelium might or might not be keratinized consistent with our both cases. However there were also reported cases where the cysts were lined by respiratory epithelium, columnar or cuboidal epithelium<sup>2</sup>. The cyst is surrounded by aggregates of mature lymphocytes which have a variable number of germinal centers<sup>1</sup>. Both the cases also showed dense infiltration of lymphocytes in the cyst wall.

This combination of epithelium lined cyst with lymphoid aggregates is unique enough to make the diagnosis an easy one, but the pathologist must differentiate it from Warthin tumor, metastatic squamous cell carcinoma, sialolithiasis and others.

In early 1980s fairly rare lesion lymphoepithelial cysts of the parotid, were representing a manifestation of Sjogren's syndrome<sup>15</sup>. However in the last few years, lymphoepithelial cysts have been found in increasing number in AIDS patients as well in the patients belonging to the AIDS risk groups<sup>16</sup>. Allen et al identified 27 cases of lymphoepithelial cyst of which 18 were found in HIV patients<sup>15</sup>. Even patients infected with

HIV infrequently develop benign lymphoepithelial lesion of the parotid gland in the course of disease<sup>17</sup>. Although investigation for HIV infection was not done in our cases.

The diagnosis of lymphoepithelial cyst is seldom made preoperatively. So regardless of possible origin, histology and biological behavior many authors suggested that this is treated by thorough surgical excision<sup>2, 3</sup>. But in case of HIV-patient this lesion may be treated by radiotherapy<sup>17</sup>.

#### References:

1. Juan Rosai. Rosai and Ackerman's. Surgical Pathology, Ninth edition, vol. 1, London: Elsevier Inc., 2004; 262: 875-76.
2. Rahman S., Shaari R., Hassan R. Parotid Lymphoepithelial Cyst: A Case Report. Archives of Orofacial Sciences, 2006; 1: 71-75.
3. Kandalkar B.M., Kandalkar M. G., Rao S. S. Benign Cervical Lymphoepithelial Cyst – Rare in the Elderly. Bombay Hospital Journal, 2008; 50(1): 92-93.
4. Altman K., Bailey M.W. Parotid cyst: a case report. Int J Oral Maxillofac Surg, 1994; 23: 165-166.
5. Godwin J.T. Benign lymphoepithelial lesion of the Parotid gland (Adenolymphoma, Chronic Inflammation, Lymphoepithelioma, Lymphocytic Tumor, Mikulicz Disease): Report of Eleven cases. Cancer, November 1952; 5: 1089-1103.
6. Antoniadis K., Karakasis D., Tzarou V., Skordalaki A. Benign cysts of the parotid gland. Int J Oral Maxillofac Surg, 1990; 19: 139-140.
7. Ellis G.L., Auclair P. L. Tumors of the salivary glands. AFIP Atlas of Tumor Pathology, Series 4, Fascicle 9. Washington, DC: ARP Press, 2008; 481-514.
8. Camilleri A.C., Lloyd R.E. Lymphoepithelial cyst of the parotid gland. Br J Oral Maxillofac Surg, 1990; 28: 329-332.
9. Fujibayashi T., Itoh H. Lymphoepithelial (so-called branchial) cyst within the parotid gland. Report of a case and review of the literature. Int J Oral Surg, 1981; 10: 283-292.
10. Bhaskar S.N., Bernier J.L. Histogenesis of Branchial cysts: A report of 468 cases. Am J Pathol, 1959; 35: 407-423.
11. Holliday R. A., Cohen W. A., Schinella R.A., Rothstein S. G., Persky M. S., Jacobs J. M., Som P. M. Benign Lymphoepithelial Parotid Cysts and Hyperplastic Cervical Adenopathy in AIDS- Risk Patients: A New CT Appearance. Radiology, 1988; 168(2): 439-441.
12. Rickles N.H., Little J.W. The histogenesis of the branchial cyst II. A study of the lining epithelium. Am j Pathol, 1967; 50: 765-773.
13. Wyman A., Dunn L.K., Talati V.R., Rogers K. Lympho-epithelial 'branchial' cysts within the parotid gland. Br. J Surg, 1988; 75(8): 818-819.
14. Skouteris C.A., Patterson G.T., Sotereanos G.C. Benign cervical lymphoepithelial cyst: Report of cases. J Oral Maxillofac Surg, 1989; 47: 1106-1112.
15. Allen E. A., Ali S.Z., Mathew S. Lymphoid lesions of the Parotid.

- Diagnostic Cytopathology 1999; 21(3): 170-173.
16. Marmary Y., Gomori J.M., Nitzan D.W. Lymphoepithelial parotid cysts as presenting symptom of immunodeficiency virus infection: Clinical, sialographic and magnetic resonance imaging findings. J Oral Maxillofac Surg 1990; 48: 981-984.
  17. Kooper D. P., Leemans C. R., Hulshof M.C.C.M. Claessen F. A. P., Snow G. B. Management of benign lymphoepithelial lesions of the parotid gland in human immunodeficiency virus-positive patients. Eur Arch Otorhinolaryngol 1998; 255: 27-429.