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Benign lymphoepithelial cyst of parotid region-A case report

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Bernier and Bhaskar introduced the term "lymphoepithelial cyst". It is an embryogenic remanent, which is a solitary or multiple cysts in salivary gland associated with lymph nodes results from cystic degeneration of salivary gland inclusions or may be due to cystic dilation of ducts within lymph nodes.[1]

Clinically it is unilocular seen in parotid and submandibular gland and most commonly seen in cervical area just below mandible. It is commonly seen in middle aged to older age with equal sex distribution. It is painless, slow growing cyst which can be unilateral or bilateral, with or without cervical lymphadenopathy.[2]

These lesions are most commonly associated with HIV positive patients and mostly bilateral among them. It is also seen in autoimmune diseases like Sjogren's disease, Mikulicz's disease.[3] When associated with HIV, it is due to hyperplasia of lymph nodes present in the salivary gland and associated with ductal obstruction. Sjogren's associated lymphoepithelial cyst is due to secondary to infiltration of B cells into the ductal epithelial and expands within the striated ducts and later causes ductal cell hyperplasia. Radiologically it is a well-defined unilocular cyst. It has an excellent prognosis; recurrence is extremely very rare.

Treatment for lymphoepithelial cyst is repeated fine needle aspiration and drainage, sclerotherapy, radiotherapy and surgery. But in cases of HIV associated, surgery is not indicated only conservative treatment is indicated. Lymphoepithelial cyst are mostly benign but if not treated can transform into malignant lesions like lymphomas, adenocarcinomas, mucoepidermoid carcinomas. As these are uncommon lesions, their histopathologic characteristics often are incompletely understood and rarely described in literature and textbooks.[4-6]

Case Report

We reported a case of 70 years old female who came with complaints of swelling in the left parotid region for 10 days. The swelling was not associated with pain and was gradually increasing in size. On examination, the swelling was noted in the left parotid region. The skin over the swelling was normal; no discharge or discoloration was noted. On palpation, single swelling of size measuring 3x3cm, well defined, firm in consistency, mobile, skin pinchable with no local rise of temperature was found. No regional lymph nodes were palpable. Intraoral examination findings were unremarkable.

Fig 1: Swelling of left parotid gland

Ultrasonography was done which showed the hypoechoic area with well-defined borders with focal granules and with no vascularity suggestive of lymphoepithelial cyst. No cervical lymphadenopathy was present.

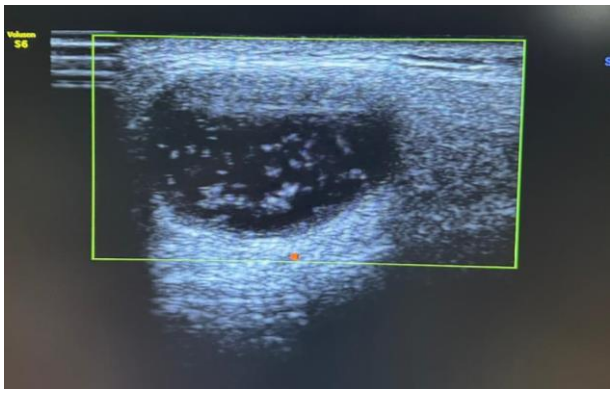

Fig 2: Ultrasonography suggestive of lymphoepithelial cyst

Fine needle aspiration was done from the swelling twice with wide bore needle which yielded 1ml of straw-colored serous fluid in 1st attempt and 5ml of serous blood-tinged fluid in 2nd attempt. The swelling was reduced completely after the aspiration. The fluid was centrifuged and 5 pap-stained smears were made which revealed predominantly of cyst macrophages with lymphocytes and scattered squamous epithelial cells in a proteinaceous background.
Cytological features were suggestive of Lympho-epithelial cyst of left parotid region.

Fig 3: Received a specimen of left parotid gland measuring 6X3.5X1.5cm.

Cut section shows a unilocular cyst of size 3.5x2.5x2cm filled with hemorrhagic material without any solid areas.

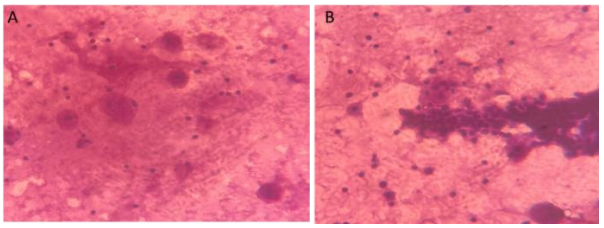

Fig 4: Photomicrograph A(PAP,40x) showing many cyst macrophages and lymphocytes in the proteinaceous background. Photomicrograph B(PAP,40x) showing sheets of squamous epithelial cells in the background of lymphocytes.

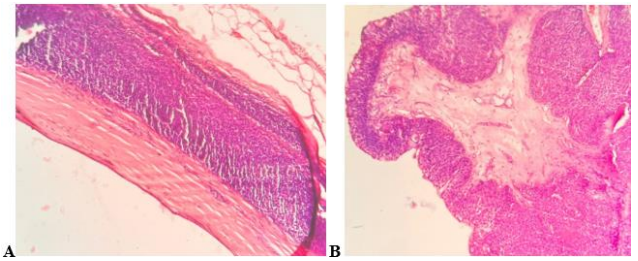

Fig 5: A Photomicrograph 1(H&E4x) showing lymphoid component. **B.** Photomicrograph 2(H&E,40x) showing squamous epithelium with lymphocytic infiltration permeation into epithelium.

Patient was advised for histopathological examination for further confirmation of the diagnosis. Multiple sections studied shows salivary gland tissue with dilated ducts and cyst lined by sheets of polygonal to cuboidal epithelial cells and lymphoid tissue. These findings were also consistent with Lympho-epithelial cyst of left parotid region.

Discussion

The Etiopathogenesis of branchial cysts development is a controversial subject.

There are many theories which are proposed to explain the origins of branchial cysts, which are: Branchial apparatus cleft theory, Cervical sinus theory, Thymus-pharyngeal duct theory and Parotid gland inclusion theory.[7,8] The first two theories are also known as classic theories which hold that the cysts develop from the remnants of the branchial cleft because they occur

in the area of the embryonic gill apparatus. For the present case, the inclusion theory or the so-called recent theory would seem the most feasible explanation for the LEC which was found in the parotid gland, where this theory considers that the cysts arise from cystic changes in parotid gland epithelium that become entrapped in the upper cervical lymph nodes during embryonic life.[9] Hence, it is considered that the epithelial remnants of the parotid gland can give rise to LEC inside the parotid gland and cervical lymph nodes.[10] The age of the patients between 16 to 70 year, with unilateral majority but can be also bilateral. Commonly seen in parotid gland, may be due to presence of intra parotid lymph nodes as the other glands have no lymph nodes. Clinical Presentation of this patient is as a slow growing, painless, movable swelling in the parotid region with normal overlying skin. The swelling was reduced completely for this patient after the aspiration. There are two peculiarities that distinguish the cyst associated with HIV: frequent bilateral occurrence and multi cystic appearance observed in computed tomography (CT) or MRI[11] The greatest problem related to the LEC in Human immune deficiency virus [HIV]-positive patients seems to be the progression to a lymphoma. [12]

In this case, serology for HIV were sent and the patient was non-reactive to HIV. The main types of lymphoepithelial cyst that can be found in the parotid gland are: simple cysts, large LECs [which are mainly congenital, but also reported in AIDS patients] and polycystic disease of the parotid gland. These lesions are not known to recur or metastasize.[13]

Differential diagnosis of LEC is Warthin's tumor, Intra muscular hemangioma, Brachial cleft cyst, Lymphoma. [7,8]

Declaration of Patient Consent

Consent from the patient was taken for the images. Any other information about the patients was not be used for the publication.

Source of Funding

None.

Conflict of Interest

None.

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