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Case Report

Simplifying the diagnosis of benign lymphoepithelial cyst: A case report

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ABSTRACT:

Benign lymphoepithelial cyst (BLC), also called as branchial cyst, is a rare lesion usually occurs in the parotid gland or sometime lateral cervical area. Most likely the etiology could be due to the process of lymphocyte-induced cystic ductular dilatation and the final diagnosis can be made postoperatively by histopathological examination (HPE), but here in this article we try to give some clinical characteristics which can be helpful to arrive at a diagnosis. 55-year-old male reported to our department 1 year ago, with a soft, nontender, gradually increasing, sessile localized swelling involving the right parotid gland since 3 to 4 months of size $3.5 \text{ cm} \times 3.5 \text{ cm} \times 2 \text{ cm}$ at the time of presentation with normal Stensen's duct and facial nerve function. Biochemistry indicated high protein content of aspirated fluid. Cyst enucleation was done with nonincidental healing. HPE of excisional tissue revealed it to be BLC.

Keywords: Benign lymphoepithelial Cyst, Parotid lesion, Diagnosis.

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INTRODUCTION

Lymphoepithelial cyst was coined by Bernier & Bhaskar in 1958¹. They said that benign lymphoepithelial cysts (BLCs) results from the cystic degeneration of salivary gland within lymph nodes. Some also support that remnants of brachial arch could be also be the etiology behind BLC 2 . BLC also is linked to patients infected with HIV, so during investigation HIV should also be ruled. Cystic lesions of the parotid gland are infrequent and comprises only around 3% of all salivary gland tumors ^{3,4}. Many cysts of the salivary glands may be because of an obstructive process, which can occur as a result of obstruction of salivary gland ducts, partial or complete blockage of the excretory ducts, or stasis of salivary flow in ducts. Incidence of BLCs most commonly seen in the fifth decade. Most lesions are slowly enlarging painless swellings affecting a single gland. As pleomorphic adenoma is most frequent of all salivary gland tumor ⁵. Hence, the diagnosis is seldom made preoperatively and superficial parotidectomy done which has increased risk of injury to facial nerve⁶. This article however highlights

conservative approach in treating such lesion and avoiding injury to facial nerve.

CASE REPORT

A 55 years old male with the big swelling in the right parotid region was referred to Oral & Maxillofacial Department of Terna Dental College. Patient started to notice his swelling 3 to 4 months before his presentation to us. Since then it gradually increased in size, however it was entirely asymptomatic. He did not have any history of trauma to the parotid area. Patient had no relevant medical history.

On extra oral examination revealed $3.5 \text{ cm} \times 3.5 \text{ cm} \times 2 \text{ cm}$ that localized soft fluctuant swelling in the right inferior auricular area with elevation of lower ear lobe. The overlying skin noted to be normal in color and freely moveable over the mass. The function of facial nerve was normal. The swelling appeared to be fluid filled, with no audible bruit was detected. There was no associated regional lymphadenopathy or thyroid enlargement. Intra oral examination revealed a free flow of saliva from the right parotid (Stensen's duct) opening.

Further investigations were done including plain radiograph of orthopantomographic radiograph (OPG), which revealed normal conditions. Ultra sound however showed a hypoechoic with posterior enhancement which suggestive for cystic lesion in the inferior aspect of right parotid gland. The content of the cyst aspirated to look for the nature of the fluid, in which was found to be a straw color containing whitish cheesy materials. The aspirate was then sent Pathology Department for cytology the to examination. The result showed no malignant cells suggesting parotid cyst as diagnosis. After skin preparation with povidone solution, assess to the cyst was made through the post ramal incision.

This will help to minimize the post operation scarring and to avoid puncturing the cyst. The incision was then performed layer by layer until it reached the platysma muscle. This muscle was dissected to expose the underneath cyst capsule. Once the plane between cyst capsule and platysma muscle was identified the blunt dissection was carried out carefully separating the cyst from parotid capsule preserving the facial nerve. However, the upper part of the cyst was found embedded in the parotid tissue, so part of the parotid tail was excised together with the cyst. The specimen was then sent for histopathology examination. After the hemostasis was achieved, the surgical area was sutured in layers with the help of 3-0 vicryl and skin suturing with 5-0 prolene was done. The patient recovered from surgery uneventful. There was no evidence of facial nerve palsy and wound infection.

The histopathology examination showed a collapsed cyst wall with adjacent salivary gland and small reactive lymphoid tissue. The cyst was lined by columnar epithelium with abundant, granular cytoplasm and densely eosinophilic. However, no nuclear atypia was seen. The appearances were those of a lymphoepithelial cyst.

Fig 1: Post ramal incision taken



Fig 2: Cystic lesion exposed



Fig 3: Closure done



Fig 4: Extraoral photos



Preoperative



Postoperative after 1 year





DISCUSSION

Parotid cyst often diagnosed as a slowly enlarging painless lump in the parotid region. Reported cases most commonly presented as slow growing, painless swelling with normal movable overlying skin^{7,8}. Lymphoepithelial cyst were most frequently found in the lateral cervical area. The parotid gland is the commonly occurred site for these cysts because of the presence of intraparotid lymph nodes in the glands unlike other salivary glands^{2,4,8}. Lymphoepitelial (socalled branchial) cysts within the parotid gland are rare. The first reported case of branchial cyst in the parotid gland was in 1895 by Hildebrant⁹. The most common sites for the lymphoepitelial cysts were found to be in the lateral cervical area¹. However, the parotid lymphoepitelial cysts have also been associated with human immunodeficiency virus (HIV) infected as well as HIV-high risk patients, which are probably related to intraparotid lymphadenopathy associated with HIV infection¹⁰. Our aim was to establish a systematic approach towards swelling in the neck and simplifying the diagnosis of lymphoepithelial cyst. After detailed case history, clinical examination we normally advice OPG to rule out odontogenic cause of swelling. USG done to rule out sialolith. Wyman et al. explained that ultra sound scanning was a simple investigation that was recommended for pre-operative diagnosis of cervical swelling. Here, the nature of echoes could be used to distinguish between cystic and solid tissue. After which, aspiration was also useful in identification of the nature of the cystic content. The aspirate usually shows clear watery or straw colour fluid¹². The differential diagnosis of parotid LEC includes Warthin's tumor, intramuscular benign haemangioma (IMH), branchial cleft cyst and lymphoma. Treatment plan for such swelling in parotid region is mostly superficial parotidectomy which is very invasive for BLC. Whereas, surgical excision and periodic follow ups is sufficient for BLC.



CONCLUSION

In conclusion, the definitive diagnosis of BLC is challenging owing to its rarity. Hence a detailed case history, clinical examination and proper diagnosis can make surgery less invasive and can decrease the patient morbidity. Also enhances patients postoperative cosmetics and quality of life.

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