

Case Report:

Diagnostic misdiagnosis of lymphoepithelial cyst of the parotid in a HIV patient

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Abstract:

The lymphoepithelial cyst is a benign cyst originates from the epithelial remnants of the lymphoid tissue during the embryogenesis. Lymphoepithelial cyst most commonly occurs in the lateral neck region, less frequently in the oral cavity and in salivary glands. Among the salivary glands, the parotid is the most common site due to the presence of intra-parotid lymph nodes. The parotid gland involvement incidence increased significantly after the discovery of Human Immunodeficiency (HIV) virus infection and the pathogenesis is related to ductal obstruction phenomenon that goes along with a follicular hyperplasia in the periductal and intra-parotid lymph nodes. However, Lymphoepithelial cysts are rare in non-HIV patients. According to literature lymphoepithelial cysts are considered as early or initial indicators of Human Immunodeficiency Virus infection. Basic investigations such as Ultrasound and Fine Needle Aspiration Cytology (FNAC) play an important role in establishing the diagnosis of the lesion but in this case both the investigations gave the diagnosis as Warthin's tumor of the parotid gland. Enucleation of the cyst is the treatment of choice along with Anti-retroviral therapy but more chances of recurrence is associated with enucleation. Complete removal of the parotid gland is the gold standard treatment. This report documents a case of middle aged HIV positive male who was misdiagnosed as Warthin's tumor on Ultrasonography and Fine Needle Aspiration Cytology. The diagnosis of Lymphoepithelial cyst was made on Histopathological examination (HPE) which plays a major role in making definite diagnosis of cystic lesions.

Keywords: Lymphoepithelial Cyst, HIV, FNAC, Warthin's Tumor, Benign Cystic Lesion

INTRODUCTION

Benign lymphoepithelial cyst has been widely recognized as a common cause of parotid gland in the patients infected with HIV. In general, cystic lesions of the parotid gland are uncommon and it comprises approximately 3% of all salivary gland tumors. Cysts of the salivary glands may originate as benign non-neoplastic entities or in association with benign and malignant tumors of the salivary glands. Cyst development as part of specific neoplasm of the salivary glands is well recognized, including those

that occur in the pleomorphic adenoma, Warthin's tumor, mucoepidermoid carcinoma, acinic cell carcinoma, and the adenoid cystic carcinoma⁽¹⁾. The histological features of these neoplasms are sufficiently distinctive; however, non-neoplastic salivary gland cysts do require differentiation from cystadenoma, mucoepidermoid carcinoma, and acinic carcinoma. Many cysts of the salivary glands may be generically attributed to an obstructive process⁽¹⁾. They can occur as a result of traumatic severance of salivary gland ducts, partial or complete blockage of

the excretory ducts, or stasis of salivary flow in ducts. Many of them represent cystic components of neoplasm. Patients most commonly present in the fifth decade and although duct obstruction appears to be the cause, the source of the obstruction is often not apparent. Most lesions are slowly enlarging painless swellings affecting a single gland. Hence, the diagnosis is seldom made preoperatively and sometimes a superficial parotidectomy is needed ⁽²⁾.

CASE REPORT

A 39 year-old HIV positive male patient presented with complaints of swelling in the left side of the neck since 2 years. Patient revealed his HIV status. On general physical examination, patient was mild pallor, with swelling in the left side of the neck. Soft in consistency, with no clear cut borders. Routine investigations were done and specific investigations such as ultrasound neck and FNAC were advised.

Hemoglobin -9.3 Gm %, peripheral blood lymphocytes (65 %), HIV – Reactive. Ultraso-

nography diagnosis given was benign cystic lesion – Warthin’s tumor. FNAC revealed cyst macrophages, lymphocytes in a dirty background. Final FNAC diagnosis was benign cystic lesion- possibility of Warthin’s tumor.

Patient was posted for surgery and the enucleation of the cyst was done through a pre-auricular incision, which confirmed the cyst-like aspect, intimately related to the parotid duct. Gross findings show a cystic mass measuring 3.5 x 3 x 1 cm. (Fig. 1) Cut section shows 1.5 cc of brownish fluid. Histopathological examination demonstrated a cyst covered by flattened epithelium with multiple lymphoid follicles with germinal centers and few congested blood vessels. (Fig. 2, 3, 4)



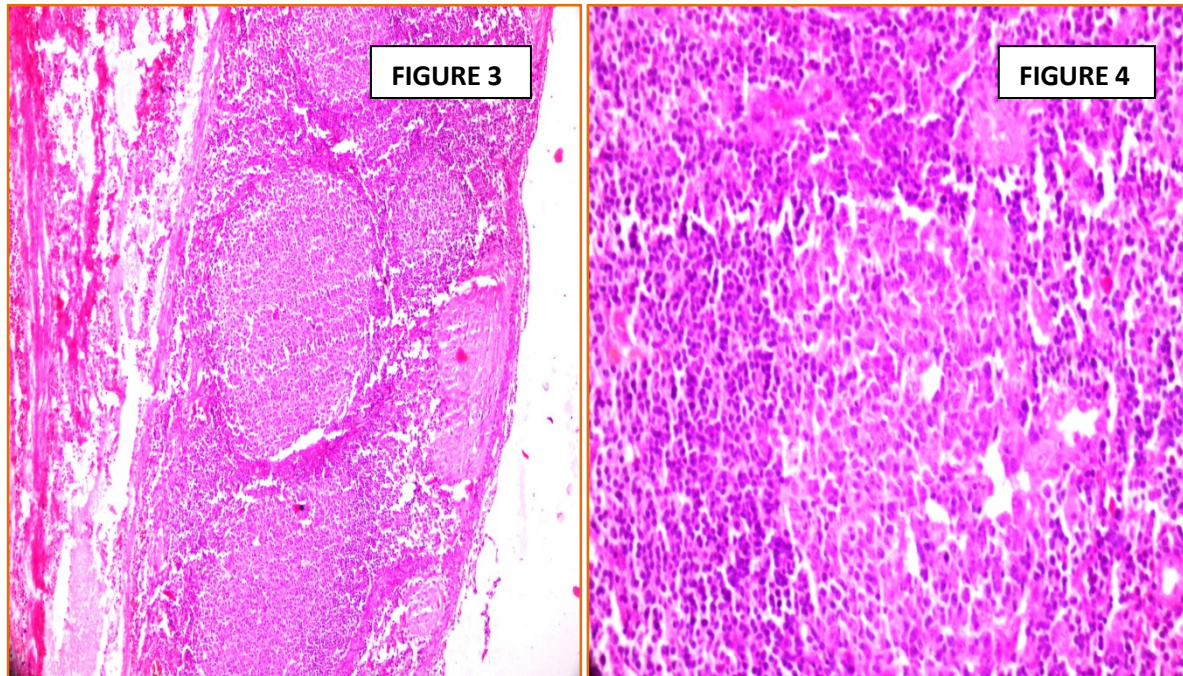


Fig. 1- Gross image of the Eucleated cyst showing gray white in appearance and inner surface smooth.

Fig. 2 & 3- Microphotograph showing cyst wall lined by flattened epithelium and subepithelially lymphoid follicles with prominent germinal centres and congested blood vessels.

Fig. 4- Microphotograph showing one Lymphoid follicle with prominent germinal centre and lymphoid cells.

DISCUSSION

The definitive diagnosis of the cystic lesion of the parotid gland depends solely on histological examination (HPE). In this present case the diagnosis of parotid lymphoepithelial cyst was made after the HPE. Lymphoepithelial cysts within the parotid gland are rare. The first reported case of lymphoepithelial cyst in the parotid gland was in 1895 by Hildebrandt. Since then about 70 cases of this type of cysts have been reported (Camilleri and Lloyd, 1990). It is considered that the epithelial remnant of the parotid gland can give rise to Lymphoepithelial cyst inside the parotid gland and cervical lymph nodes. Ductal obstruction phenomenon is considered as one of the

mechanism of parotid lymphoepithelial cyst associated with the HIV. This obstruction goes along with a follicular hyperplasia in the periductal parotid lymph nodes, induced by the virus observed in patients manifesting AIDS⁽³⁾. The parotid gland is the preferential site for these cysts to occur and this is probably due to the presence of intraparotid lymph nodes in the glands which are absent in all the other salivary glands⁽⁴⁾. Lymphoepithelial cyst can grow to large proportions and may lead to disfigurements. It can be rather devastating to the patient and may be the cause of isolation and depression⁽⁵⁾. The lymphoepithelial cysts have equal gender distribution, and they may be single or multiple,

unilateral or bilateral. They are usually painless, soft in consistency, and involve the superficial lobe of the parotid gland ⁽⁶⁾. Lymphoepithelial cyst is considered to be a reactive phenomenon, and they are not true neoplasm ⁽⁷⁾. However, it is not clearly understood whether these lesions develop as a result of pre existing salivary gland inclusions in the intraparotid lymph nodes or they arise as lymphoepithelial lesions from the salivary gland parenchyma. Diagnosis of the lymphoepithelial cysts is made on basis of a thorough case history, general physical examination and fine needle aspiration cytology. Ultrasound imaging is an excellent diagnostic modality. Fine needle aspiration cytology serves both diagnostic and therapeutic purposes ⁽⁸⁾. In this case report, both ultrasound and fine needle aspiration cytology gave a wrong diagnosis.

There is large number of treatment options available and these include repeated fine needle aspirations and drainage of the cyst fluid, conservative management, radiotherapy, sclerotherapy, surgery, highly active retro-viral therapy or combinations of therapies.

Surgical nucleation of the cyst along with the preservation of the gland may not provide total resolution because the cyst may recur in other site within the gland. Recurrences are mainly due to the

tissue alterations promoted by the Human Immuno Deficiency (HIV) virus. Best treatment is surgical removal of the parotid gland with preserving the facial nerve; however it is the most challenging procedure.

CONCLUSION

The lymphoepithelial cysts are considered to be the indicators of the HIV infection as they are more common in the HIV infected individuals. In the HIV era Lymphoepithelial cysts are becoming an increasingly common cause of parotid gland enlargement. Although intraparotid cystic masses are uncommon, we should be able to distinguish it from the benign tumors of the parotid gland as it may greatly affect the treatment plan. Thorough clinical examination, appropriate investigations and diagnosis will assist in planning the treatment. Patients who are on conservative therapy complete superficial parotidectomy is the best treatment as it has negligible chance of recurrences. Clinicians should watch for the possible transformation into B-cell lymphoma. A six monthly check-up is recommended, supplemented by a fine needle aspiration biopsy when indicated by the clinical behavior of the lesion in the patients who are surgically unfit or who are on medical/conservative management.

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