Lymphoepithelial Cyst of the Parotid Gland in an HIV-Positive Patient

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ABSTRACT
Lymphoepithelial cysts serve as early detectors of HIV infection. These cysts have been understood to be a localized manifestation of the generalized persistent lymphadenopathy often associated with HIV infection. They can become large and can cause disfiguration. Treatment modalities for benign lymphoepithelial cysts include repeated fine-needle aspiration, radiotherapy, and conservative treatment with highly active antiretroviral therapy, sclerotherapy and surgery. This article presents a case of lymphoepithelial cyst in an HIV positive patient who was diagnosed and treated in a conservative manner.

Keywords: Lymphoepithelial cyst, HIV infection, Ultrasonography, Fine needle aspiration.

INTRODUCTION
Benign lymphoepithelial cysts have been widely recognized as a common cause of parotid gland swelling in patients infected with the human immunodeficiency virus (HIV). These cysts are characteristic of HIV. They vary in size from 0.5 to 5.0 cm, and they are capable of causing facial deformity and discomfort. They are rather painless, soft in consistency and involve mainly the superficial lobes of the parotid glands.

Ultrasound imaging is recognized as a good diagnostic modality. Fine-needle aspiration serves both diagnostic and therapeutic purposes.

CASE REPORT
A 38-year-old male patient came to the dental OP of Ragas Dental College and Hospital, Chennai with the complaint of a swelling in the left side of the face for the past 5 years (Figs 1 to 3). The swelling was slow growing, gradually progressive in size, with no history of pain or pus discharge. Medical history revealed that the patient was HIV-positive and was under HAART for the past 6 years. On extraoral examination, a single swelling measuring around 4 × 3 cm was seen in the preauricular region of the left side of the face. It extended anteriorly 4 cm behind the left alae of the nose, posteriorly in front of the left ear lobe, superiorly at the level of the tragus of the ear, and inferiorly 1 cm below the ear lobe with well-defined borders. The skin over the swelling appeared normal. On palpation, the swelling was soft in consistency, freely mobile, non-tender with no rise in temperature and no pus discharge. A provisional diagnosis of pleomorphic adenoma of the left parotid gland was given.

Ultrasound of the left parotid gland revealed a well circumscribed cystic lesion seen occupying the entire left parotid pretragal region with septations and bright internal echoes (Fig. 4). Normal appearing left parotid parenchyma
was seen around the cyst. No lymph node enlargement was seen in the rest of the cervical region. The ultrasound picture was suggestive of a lymphoepithelial cyst of the left parotid gland (an AIDS related cyst in the parotid).

The cystic fluid was aspirated under local anesthesia using an 18 gauge needle with a 5 cc syringe and the fluid was drained completely (Fig. 5). The cystic fluid was dark reddish brown in color and on analysis showed protein levels of 5.6 gm% (Fig. 6). Histopathological examination of the PAP stained cytological smear showed predominant lymphoid component (Fig. 7). It was finally diagnosed as a lymphoepithelial cyst of the left parotid gland. Patient was under observation for a possible reaccumulation of the cystic fluid, which did not recur even after 6 months (Figs 8 and 9).

**DISCUSSION**

The involvement of salivary intraglandular lymph nodes in HIV infection was first suggested and reported in 1985. \(^4\)

HIV associated salivary gland disease (HIV SGD) is a heterogeneous group of signs and symptoms that has a tendency to vary from person to person coinciding with the progression of the HIV infection in the same individual. A prevalence of 5% has been noted in HIV patients. \(^5\)

Parotid swellings of various types have been reported in patients infected with HIV. \(^2\) These include inflammatory disorders like the sicca syndrome, benign lymphoepithelial lesions, parotitis, intraparotid lymphadenopathy, and neoplasms such as adenoid cystic carcinoma, lymphoma and Kaposi’s sarcoma.

In the last few years, lymphoepithelial cysts have been found in increasing number in acquired immunodeficiency syndrome (AIDS) patients. \(^3\)

Lymphoepithelial cysts can grow to large proportions and can cause disfigurement. It can be rather devastating to the patient and may be a cause of isolation and depression. \(^1\)

The lymphoepithelial cyst has an equal gender distribution, and it may be single in number or multiple, and unilateral.
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The parotid gland is a 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. The parotid gland is a 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.6

These cystic lesions are considered to reflect a localized manifestation of the generalized persistent lymphadenopathy commonly associated with HIV. 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 if they arise as lymphoepithelial lesions from the salivary gland parenchyma, with different authors suggesting different views.7

Lymphoepithelial cysts are considered to be a reactive phenomenon, and are not true neoplasms.8

There are two hypothesized etiologic mechanisms behind the development of benign lymphoepithelial cyst (BLEC). The first hypothesis is suggestive of the ‘obstructive theory’ which states that lymphoid proliferation occurring in the parotid gland leads to ductal obstruction followed by salivary dilation that resembles a true cyst. The second theory is that the reactive lymphoproliferation occurs in the intraglandular lymph nodes. The epithelium of the parotid gland gets trapped in the normal glandular nodes and can result in cystic enlargement.

Diagnosis of lymphoepithelial cysts is made on the basis of a thorough case history, physical examination, and fine-needle aspiration biopsy.1

Ultrasound imaging is an excellent diagnostic modality. Fine-needle aspiration serves both diagnostic and therapeutic purposes.2

There are a large number of treatment options and these include repeated fine-needle aspiration and drainage, surgery, radiotherapy, sclerotherapy, and conservative management, with institution of highly active antiretroviral therapy medication.1

Aspiration of the cystic fluid is a palliative treatment modality which aims at achieving esthetic results, although the cyst recovers their volume after a while. The advantage of this technique is that a specimen for cytological examination can be obtained, which will help in the diagnosis of the condition with minimal discomfort to the patient unlike more invasive techniques such as surgery.

Intracystic injections of doxycycline promote sclerosis and effectively induces regression of these lesions.9

A lot of controversy exists regarding the utility of radiotherapy but overall it has shown to be a rather effective and fairly well tolerated technique.10 Sclerotherapy by alcohol injections is also a proven modality which is simple, and effective in patients who do not qualify for antiretroviral treatment.11

Surgical nucleation of the cyst along with preservation of the gland may not provide 100% resolution because the...
cyst may reoccur in other sites within the gland. Recurrence is related to the tissue alterations promoted by the HIV. Another surgical option is the superficial removal of the parotid gland while preserving the facial nerve, which can be a rather challenging procedure.9

CONCLUSION

Although, BLEC can arise in HIV-negative individuals who have Mikulick’s disease, myoepithelial sialadenitis, Sjögren’s syndrome, infection with HIV should be strongly suspected in patients presenting with bilateral BLEC of the major salivary glands, especially those of the parotid glands.

REFERENCES


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