



Swelling of the Parotid - A Rare Case Report of Undisclosed Aetiology

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ABSTRACT

Prominent lymphoid component which occur in various lesions of salivary glands represent heterogeneous group of diseases that include both benign reactive lesions and malignant neoplasms. Occasionally, these lesions cause difficulties in the clinical and pathological diagnosis. To assess their prognosis is also difficult. Human immunodeficiency virus infection associated lymphadenitis of the salivary glands (HIVLSG), benign lymphoepithelial lesion and cysts are described as early events in HIV patients. The diagnosis is not usually made by clinical examination as it mimics a salivary gland tumor. We are presenting a rare pathological finding in the salivary gland of an individual who was diagnosed to have HIV infection. She was clinically diagnosed to have a salivary gland tumor which was cytologically diagnosed to be benign lymphoepithelial lesion.

Keywords: Parotid gland, HIV-associated salivary gland disease, Lymphoepithelial sialadenitis, Benign lymphoepithelial lesion and cyst.

Introduction

The lymphocytes can infiltrate the major salivary glands and produce spectrum of diseases that range from reactive to benign and malignant neoplasms. The lymphocytic infiltrate in many cases is a minor inflammatory component that is easily distinguished from the primary disease processes. In some cases, however, the lymphocytic infiltrate is a major component of the

disease and the features that distinguish reactive and benign lesions from malignant lesions are often subtle. Lymphoepithelial sialadenitis (as occurs in Sjögren syndrome), benign lymphoepithelial lesions, HIV-associated sialadenitis, and extranodal marginal zone B-cell lymphoma are good examples of diagnosis that are often problematic to differentiate from one another^[2]. The parotid gland is the only salivary

gland to contain lymphoid tissue within its capsule while the submandibular lymph nodes lie adjacent to, but outside the glandular capsule, so it is difficult to differentiate lymphocytic neoplastic and non-neoplastic lesions of the parotid ^[1, 3, 4]. Benign lymphoepithelial lesions (BLL or BLEL), are usually mixed solid and cystic lesions that cause enlargement of the parotid glands and are usually associated with cervical lymph node enlargement, and nasopharyngeal lymphofollicular hyperplasia. It is also misleadingly known as AIDS-related parotid cysts (ARPC). BLL are seen usually in HIV positive patients without AIDS. ^[5] BLLs are only rarely seen in the submandibular glands or sublingual glands. It may clinically manifest as Sjögren's syndrome. As only a few cases have been published in literature, we are reporting this case to bring this entity and its cytopathological features to the attention of clinicians and pathologists as the treatment of choice is conservative method.

Case history

52 year old female presented to the OPD with swelling in the left parotid region for about one year duration. The swelling was associated with pain and it was increasing in size. It was a firm and well circumscribed swelling measuring 3X 2cm respectively. No other swelling was felt on the contralateral side in the parotid region and in the neck region. The swelling doesn't change in size with the intake of food. The overlying skin was not inflamed and it was free. There was no history of any dryness of the mouth or eyes. Clinically she was diagnosed to have mixed parotid tumor. Ultrasound was done which showed cystic lesion in the lobes of the parotid. The differential diagnosis were granulomatous lesion, auto immune disease and warthin tumor of parotid. Fine needle aspiration was done from the parotid swelling. The smears showed entangled masses of salivary acini with fibrous strands and lymphoid aggregates along with histiocytic collection and occasional epitheloid collection (figure 1). Tingible body macrophages and

occasional cyst macrophages were also seen in a background of dense mixed population of lymphoid cells (figure 2). Seen predominantly were mature small lymphocytes, immunoblasts and plasma cells (figures 3&4). There were no Langhans type giant cells, caseous necrosis, Warthin-Finkeldey giant cells or squamous epithelial cells. A diagnosis of benign lymphoepithelial lesion was initially thought. However in view of the dense population of the lymphocytic infiltrate and clinical history HIV infection was suspected and retroviral assay was suggested. The HIV testing done subsequently was positive and the patient was treated conservatively. The patient has remained free of any AIDS-defining illness in the follow-up period.

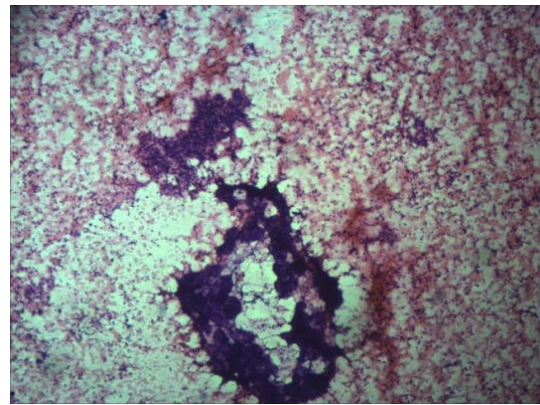


Figure 1: Clusters of acini along with polymorphous population of lymphocytes (4X H&E)

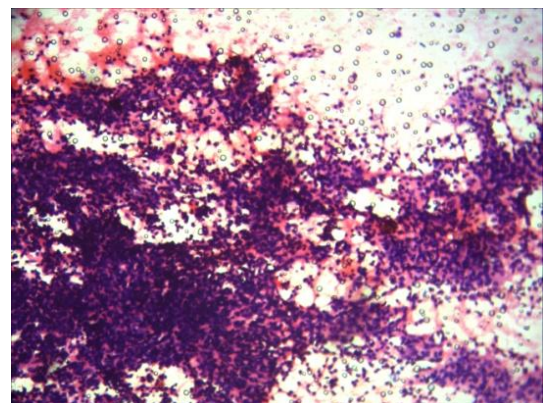


Figure 2- Intermingling of lymphocytes with ductal epithelial cell clusters (10X H&E)

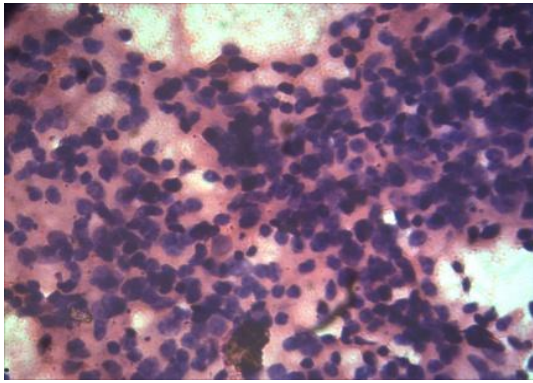


Figure 3- Sheets of salivary gland epithelial cells with lymphocytes (20X H&E)

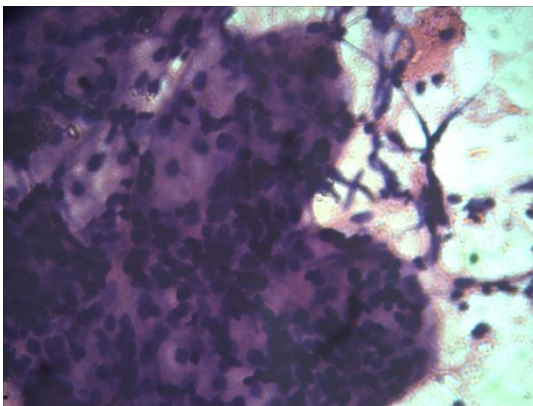


Figure 4- Clusters of acini with lymphocytes (20X H&E)

Discussion

Salivary gland gets involved exclusively in HIV infection and it was first reported in 1985. [1,6] Salivary gland disease usually develops before acquired immunodeficiency syndrome, and is sometimes the first manifestation of human immunodeficiency virus infection^[2]. Many patients with human immunodeficiency virus present with atypical features and the salivary swellings are frequently diagnosed as salivary gland tumors. Patients with HIV infection have been reported to have parotid swellings of various types including salivary gland inflammatory disorders such as sick syndrome, diffuse infiltrative lymphocytosis syndrome, parotitis, intraparotid lymphadenopathy and benign lymphoepithelial lesions, as well as salivary gland neoplasms, such as adenoid cystic carcinoma, Kaposi sarcoma and lymphoma^[7]. Many of the reported lesions are diagnosed on routine examination. Benign lymphoepithelial lesions are

a well-known entity associated with the AIDS-related complex but they occur rarely in patients on highly active antiretroviral therapy (HAART)^[9]. These lesions are seen in 3-6 per cent of patients^[8]. Benign lympho-epithelial lesion (BLEL) and cystic BLEL seen in this condition occur within the gland parenchyma. The pathophysiology of this disease is thought that HIV-infected cells migrate into the parotid glands, which triggers lymphoid proliferation, inducing metaplastic changes in the salivary ducts^[9]. Cysts form due to ductal obstruction secondary to cellular proliferation^[10]. Cysts are not encapsulated since they arise from intralobar ducts^[11]. A dramatic reduction in HIV-associated BLEL occurs with initiation of HAART^[9]. HIV patients present with unilateral or bilateral tumor-like masses, especially in the parotid gland, without any other symptoms.^[3] Differential diagnosis for unilateral lesions containing cysts includes sialolithiasis, chronic sialadenitis, HIV-associated BLEL, adenoid cystic carcinoma, and benign mixed tumor.^[12] With bilateral disease, considerations include sialosis, sarcoid, systemic lupus erythematosus, and BLEL associated with either HIV or Sjogren's syndrome can be considered.^[12] Warthin's tumor and lymphoma can be either unilateral or bilateral; however, lymphoma is usually solid. Lymphoepithelial cysts associated with HIV infection are distinguished from Sjogren's syndrome by lymphoid infiltrates in the parotid glands and can cause both solid and cystic parotid lesions but associated with diffuse cervical lymphadenopathy.^[9, 12] Sialadenitis presents with more inflammatory cells and not associated with HIV infections. sarcoid infection show many non caseating granulomas commonly not associated with HIV infections. The most important differential diagnosis is lymphoma, mainly MALT lymphoma. This often requires immunological studies, most conveniently by flow cytometry of aspirated material. Branchial cyst in which only the lymphoid component has been sampled should also be considered.

Conclusion

HIV predisposes infected individuals to a broad spectrum of head and neck diseases but salivary gland diseases are important diagnostic and prognostic indicators in HIV infection. We have aimed to highlight the association between benign lymphoepithelial lesion of parotid gland and human immunodeficiency virus infection, in order to aid early diagnosis and management of the disease. This benign condition can be diagnosed on cytology and the treatment is usually conservative so that surgery can be avoided.

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