

CASE REPORT



Papillary cyst adenoma lymphomatosum: A case report and review

Damodhar Arabagatte Mylarappa¹, Vanishri C. Haragannavar², Aparna H. Gopalkrishna², A. Reshma Joy², D. Latha², V. K. Archana²

¹Department of General Pathology, Coorg Institute of Dental Sciences, Virajpet, Karnataka, India, ²Department of Oral Pathology and Microbiology, Coorg Institute of Dental Sciences, Virajpet, Karnataka, India

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Correspondence

Dr. Vanishri C. Haragannavar, Department of Oral Pathology and Microbiology, Coorg Institute of Dental Sciences, Virajpet - 571 218, Karnataka, India. Phone: +91-7204318001, Tel.: +91-(0)20-483825836. Email: vani.haragannavar@gmail.com

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Abstract

Papillary cyst adenoma lymphomatosum is the next common tumor to the pleomorphic adenoma affecting exclusively parotid gland, was first described in 1895 by Hilderbrad. Due to its unique presentation both clinically as well as histopathologically and its unknown origin, this tumor entity is still riveting oral surgeons and pathologist. Here we report a case of parotid swelling clinically diagnosed as benign salivary gland tumor and its review.

Introduction

Papillary cyst adenoma lymphomatosum is a second most benign salivary gland tumor, which was first described by Hilderbrad in 1895 as a form of congenital cyst of the neck. This lesion has long been a point of confusion in terms of nomenclature and histogenesis. In 1929 Warthin's coined the present name as papillary cyst adenoma lymphomatosum, other synonyms for this tumor are adenolymphoma, Warthin's tumor (WT), cystic papillary adenoma, etc. After the initial reports by Hildebrad and Nicholson, this tumor has continued to captivate and enhance the controversies among researchers for understanding of its histogenesis and pathogenesis.^[1]

According to WHO 2005 classification of tumors, WT can be defined as a tumor composed of glandular and often cystic structures, sometimes with a papillary cystic arrangement, lined with characteristic bilayered epithelium, comprising inner columnar eosinophilic or oncocytic cells surrounded by smaller basal cells. The stroma contains a variable amount of lymphoid tissue with germinal centers.^[1,2]

Case Report

We are reporting a case of a 48-year-old male patient, who presented with the swelling in the right parotid region since 4 years, there were no other clinical signs and symptoms associated with the swelling. Provisional diagnosis of benign salivary gland tumor was given, and the patient was subjected to the routine blood investigations and lesional area was excised. On gross pathology, the cut surface showed cystic areas filled with chocolate brown rubbery areas with soft areas in the center [Figure 1]. Microscopically under scanner view H and E stained sections shows cystic space lined by predominantly epithelial component with surrounding scattered lymphoid stroma along with few ductal structures containing eosinophilic material in the lumen. Under higher magnification the cystic space lined by the two cell layers, outer layer nearing the cystic space were columnar cells with centrally placed nucleus with granular cytoplasm interspersed by oncocytes and inner layer cells are of cuboidal shape placed towards the lumen. These epithelial cells showed papillary projections thrown into the cystic space. This epithelial component was demarcated from the surrounding lymphoid stroma by a thin basement membrane [Figures 2-5]. By



Figure 1: Macroscopic appearance of the specimen. Excisional biopsy, the cut surface shows a few cystic spaces containing grayish, yellowish, and gelatinous material

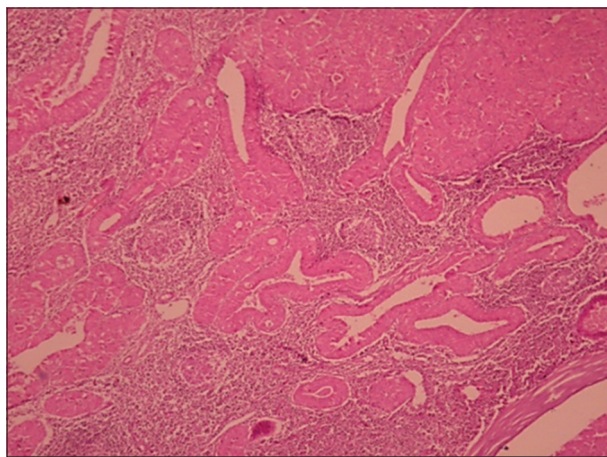


Figure 2: Photomicrograph of H and E stained sections under ×4, ×100 magnification: The tumor consists of cystic spaces lined by bilayered oncocytic epithelium and lymphoid stroma

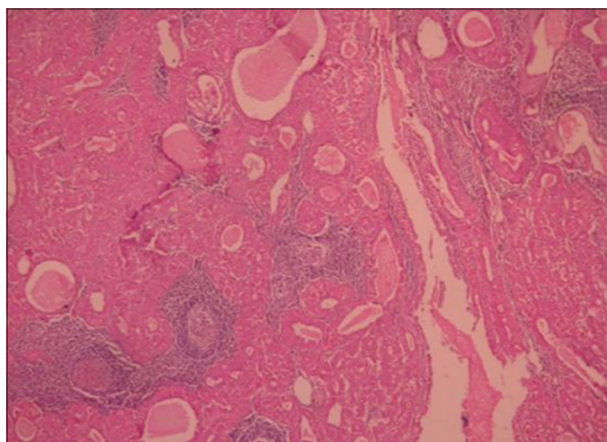


Figure 3: Photomicrograph of H and E stained sections under ×4, ×100 magnification: The tumor consists of cystic spaces lined by bilayered oncocytic epithelium surrounded by the areas of lymphoid aggregates

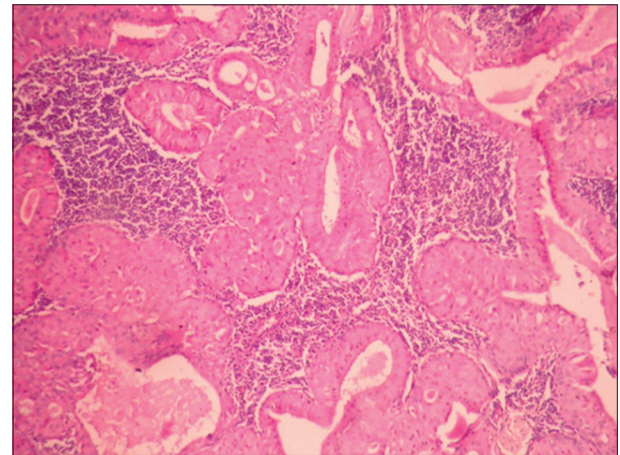


Figure 4: Photomicrograph of H and E stained sections under ×10, ×100 magnification: The tumor consists of cystic spaces lined by cuboidal and columnar cells with granular cytoplasm with eosinophilic areas in the center of the duct

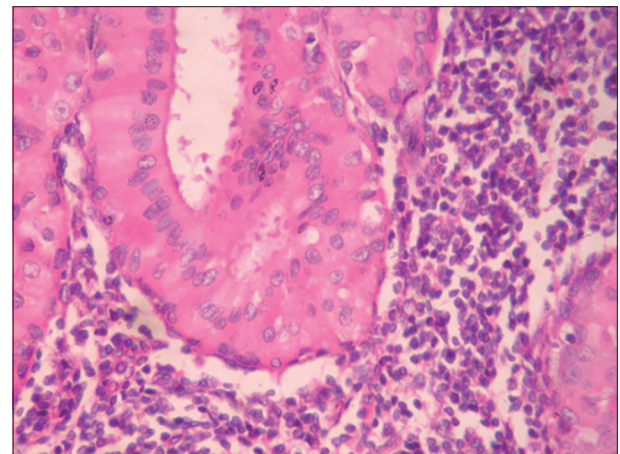


Figure 5: Photomicrograph of H and E stained sections under ×40, ×100 magnification: Both cuboidal cells and columnar cells with lymphoid aggregation surrounding the epithelium

considering the clinical and histopathological features, we gave a diagnosis of WT.

Discussion

Benign salivary gland tumors account for approximately 60-80% of parotid neoplasms and among these, WT is the second most common benign neoplasm accounting for approximately 15% of all parotid epithelial tumours. The understanding of etiopathogenesis of WT still remains as controversial.

Earlier theories suggest that the WT arises from the proliferation of aberrant salivary glands in the lymph nodes or lymphatic tissues. But this theory does not explain the following issues such as an increase in the incidence of WT in males who

have a habit of smoking and occurrence of WT in middle age or old age. Soon after this hypothesis other theories of histogenesis came into existence stating that the enclaved parotid epithelium in the lymph node can be a reason for the development of this lesion, which was suggested by the Azzopardi and Smith. Along with this Allergro has proposed that hypersensitivity as the main cause resulting in the oxyphilic metaplasia of striated ducts followed by papillary formations with secretion and cyst formation causing infiltration of the basement membrane by chronic inflammatory cells, thus indicating delayed type of hypersensitivity reaction, which shows the formation of lymphoid stroma. Few studies have also shown the correlation between Epstein berr virus (EBV) in the pathogenesis of WT, especially in multiple and bilateral cases. This virus might infect ductal epithelial cells, and the release of EBV gene products or cytokine by infected cells might activate lymphoid tissue and result in a polyclonal B-cell response. Many of the studies suggest that there is increase in the incidence rate of WT in smokers and there is equal predilection for males and females due to change in the socioeconomic status, according to this benzopyrene, arsenic, N-nitrosoguanidine etc. present in the tobacco these irritants can affect the aberrant salivary gland tissue in the lymph nodes and result in metaplasia of the gland tissues and secondary tumor change.^[1,3-5] Most commonly WT occurs unilaterally affecting major salivary glands, they can occur synchronously or metachronously. It is mainly found in males although its incidence among women is on the higher range due to the increase in the use of tobacco. Racial predominance is more in whites. Clinically it presents as a solitary, nodular, slow growing, and painless swelling with varying size. Superficial lesions can cause facial asymmetry, most commonly these lesions are located in the inferior pole of the parotid gland and these lesions can also occur in deeper part and intra parotid lymph nodes, on palpation they are firm to fluctuant in nature.^[1,2]

WT has got good correlation between cytological and histopathological findings. Classically WT shows mixture of apocrine type of cells, lymphocytes, macrophages along with oncocytic epithelium and mast cells, which does not create any diagnostic dilemma until and unless single type of cells shows predominance. On gross examination lesion present as round to oval, well-circumscribed masses that are typically encapsulated. Their cut surfaces may be brown to tan-white, depending on the relative proportions of epithelium and lymphoid stroma. They contain a variable number of cysts, ranging from small slits to spaces up to several centimeters, which contain clear, yellowish, mucoid, creamy white, or brown fluid and, rarely, semisolid caseous material.^[1,2]

Microscopically this lesion is made up of epithelial and lymphoid components, which can vary within the same lesion or can differ from lesion to lesion. Depending upon the distribution

of the stromal component it can be sub classified into three groups typical-both components approximately equal, stroma poor - epithelial component >70%, and stroma rich - epithelial component of <30%. Classically lesion presents with the epithelial cells which are arranged in two cell layers in uniform rows called as inner layer cells and outer layer cells. The outer layer is composed of columnar cells containing darkly stained, pyknotic nuclei which are situated centrally near the luminal space and inner layer is made up of cuboidal and polygonal cells with nuclei having prominent nucleoli, cytoplasm of both the type of cells is finely granular and distinctly eosinophilic, with variation in the staining intensity due to the distribution of mitochondria within the cells and this can be demonstrated by the use of special stain phosphotungstic acid - hematoxylin stain. The epithelial lining shows papillary projections into the cystic spaces which contain homogenous eosinophilic granules, and these can be demonstrated with periodic acid Schiff's reagent. This epithelial component is separated from the lymphoid stroma by a thin layer of basement membrane. The lymphoid component present in this appears as reactive with germinal centers, sinusoids and subcapsular spaces. The amount of lymphoid stroma can vary from case to case.^[1,6-7]

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