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Case Report

Papillary mucous cystadenoma of parotid gland

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ABSTRACT

Cystadenomas are rare benign salivary gland tumours characterised by prominent epithelium-lined papillary projections into the cystic spaces. Cystadenoma accounts for 2% of all salivary gland tumors, occurring most commonly in major salivary glands. Histopathologically there are two variants, papillary and mucinous variant. However very few cases are being reported in literature. We report a case of papillary cystadenoma occuring in parotid gland in a 28 year old male patient. Clinically, it presents as a soft, fluctuant, sessile and non-tender swelling measuring about 4×3.5 cm in diameter. Excisional biopsy was performed. The histopathological features were characteristic of papillary cystadenoma. This article highlights its clinical and histopathological features along with review of literature.

Keywords: Cystadenoma, Mucinous cystadenoma, Papillary cystadenoma, Oncocytic cystadenoma

INTRODUCTION

Cystadenoma of the salivary glands is a rare benign neoplasm in which the epithelial proliferation is characterized by the formation of multiple cystic cavities containing intraluminal papillary projections occurring mostly in major salivary glands.[1]

The Armed Forces Institute of Pathology (AFIP) files contained 96 cases of cystadenomas which constituted 0.7–8.1% of all benign salivary gland tumours, 7% of all minor salivary gland tumours and 3.1% of major salivary gland benign tumours. Among the major salivary glands, 58% of these tumours occur in the parotid gland, submandibular gland and sublingual gland. Among the minor salivary glands majority of cases occur in the lips followed by cheeks, palate and other intraoral sites.

Cystadenoma occurs in 70-80 years of life. A female predilection with F:M ratio of 2:1 or 3:1 is elicited.[1] Cystadenoma typically presents as a slow growing, asymptomatic mass with a clinical picture similar to mucocele. Surgical excision is the indicated treatment.

CASE REPORT

A 28 year old male patient presented with an enlarging lump in the right parotid region present since 3 months. Extraoral examination revealed a solitary, localized roughly round swelling in the right pre-auricular region measuring about 2 cms supero inferiorly and 2 cms anteroposteriorly. Superioinferiorly it extended from the inferior border of zygomatic arch to the angle of the mandible. Posteriorly the swelling was bounded by the tragus of the ear extending anteriorly to the anterior border of ramus of mandible. No lymphadenopathy was evident. The skin over the swelling appeared normal with no evidence of scars, sinuses and discolorations. The computerized topography scan revealed a cystic lesion of size 4×3.5 cm.

Based on these features a clinical differential diagnosis of pleomorphic adenoma, warthin's tumor, basal cell adenoma, mucoepidermoid carcinoma was given. Surgical excision of tumor mass was carried on. Gross appearance of the lesion revealed a cyst measuring 4cm x 3cm. The tissue was fixed in 10% formalin, processed,

embedded in paraffin wax, 4µm sections were cut and stained with hematoxylin and eosin.

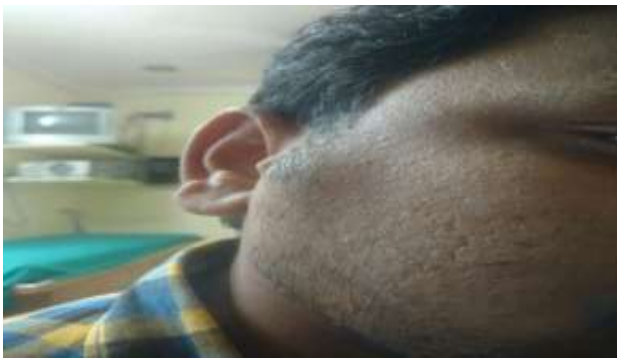

Figure 1: The extra-oral swelling on the right parotid gland region.

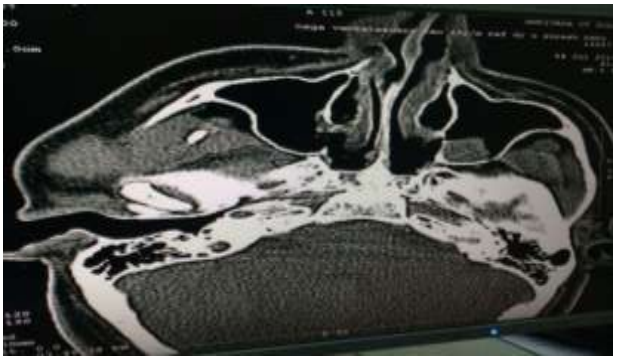

Figure 2: The CT scan revealing a cystic lesion of size 4 cm × 3.5 cm.

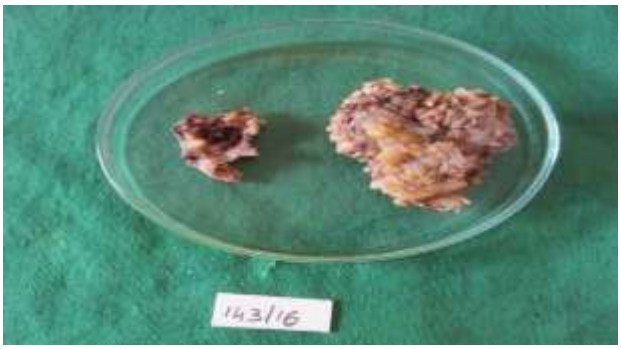

Figure 3: The gross appearance of the lesion revealing a cyst measuring 4 cm × 3 cm.

Figure 4: The specimen was grossed and the two bits were processed separately.

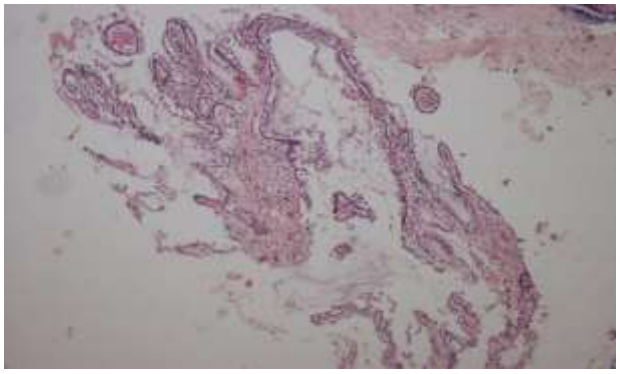

Figure 5: Histopathological examination revealing salivary glandular tissue with numerous duct like structures and clusters of acinar cells with few showing oncocytic transformation, suspended in the lipomatous background. Connective tissue showing multiple cystic cavities lined by low columnar cells admixed with number of mucous cells which are thrown into papillary projections. Numerous ducts exhibited tubular pattern and acinar cells showed degenerative changes.

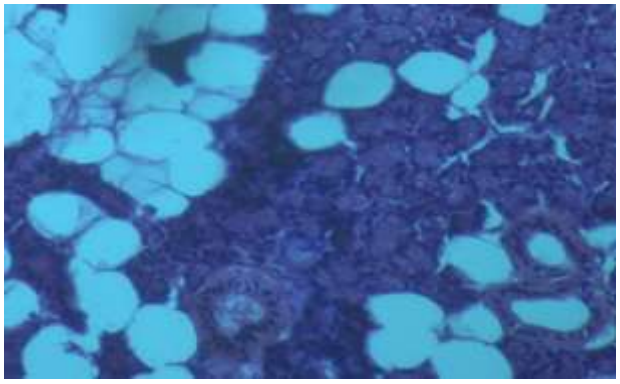

Figure 6: PTAH staining showed cells with centrally placed nuclei and abundant dense granular eosinophilic cytoplasm indicating oncocytic cells. These cells were present in normal gland tissue than at the site of tumor cells.

Histopathological examination revealed a well circumscribed thick fibrous connective tissue capsule within which mixed salivary glandular tissue consisting of few large cells with granular eosinophilic cytoplasm were present. Adipose tissue was distributed evenly. With few ducts and acinar cells exhibited degenerative changes. Numerous ducts exhibited tubular pattern and acinar cells showed degenerative changes. Multiple cystic cavities were lined by double layered low columnar cells admixed with mucous cells. In some areas the thickness of epithelium increased abruptly and is thrown into papillary projections with central core of connective tissue. These features are suggestive of papillary variant of cystadenoma. To confirm the presence of oncocytes, Phosphotungstenic acid haematoxylin staining was done and it showed, cells with centrally placed nuclei and abundant dense granular eosinophilic cytoplasm

indicating oncocytic cells. These cells were present in normal gland tissue than at the site of tumor cells.

DISCUSSION

Cystadenoma is a rare benign salivary gland tumor, characterized by prominent cysts and papillary endophytic projections. Most of these occur in the major salivary glands (65%) followed by minor salivary glands (35%). Among the major salivary glands, parotid is commonly affected. The average age of occurence is about 59 years and over 70 percent of patients are over 50 years of age. Cystadenoma was once considered as a reactive cystic hyperplasia rather than a true neoplasm and was designated as 'duct ectasia', 'salivary duct cyst' and 'intraductal papillary hyperplasia'.[1,2] Now, cyst-adenoma is believed to be neoplastic because of its proliferative properties.[3] Bauer and Bauer suggested that the cystadenomas arise principally from the undifferentiated epithelium of the intercalated ducts of the salivary gland, characterized by cywstic growth within a fibrous connective tissue stroma. The papillary variant of cystadenoma is more common and exhibits papillary proliferations that project into the cystic lumen whereas mucinous variety is rare with mucous cells in the epithelial lining of the cystic lumen. In the World Health Organization's Histological Classification of Salivary Gland Tumors published in 1991, cystadenoma was more clearly defined as a distinct histopathological entity that is further subdivided into papillary and mucinous types.

Clinical differential diagnosis of this case included pleomorphic adenoma, warthin's tumor, basal cell adenoma, mucoepidermoid carcinoma and plemorphic adenoma.

Warthin's tumor and pleomorphic adenoma primarily affect the parotid gland and present as a slow growing, asymptomatic swelling in the parotid gland. Pleomorphic adenoma occurs in young and middle aged adults between 30- 60 years of age whereas warthin's tumor is seen in older individuals (age 60–70 years). Warthin's tumor is seen more commonly in whites and occurs bilaterally and usually presents as multifocal lesion. Basal cell adenoma also presents as slow growing, painless, freely movable tumor in elderly people, mostly in 6-8th decades and is seen more in whites with female prelidiction. Mucoepidermoid carcinoma clinically presents as painless, solitary, well circumscribed swelling initially with a slight female prelidiction. Slowly the lesion is fixed to underlying skin and facial nerve palsy is present in most cases. The present case being a 28 year male, with the painless, slow growing, firm swelling involving the superficial lobe of parotid showed propensity for a provisional diagnosis of pleomorphic adenoma.

But histopathology picture came as a bolt from the blue with much variation in cell morphology and in growth patterns and was diagnosed as papillary mucous cystadenoma. Cystadenomas are generally well circumscribed and may have a thick, encapsulating band of fibrous connective tissue. However, cystic structures are haphazardly arranged over a background of fibrous connective tissue or salivary gland parenchyma. Cystadenoma usually lacks an extra luminal, solid epithelial component. Foci of lymphocytic cell aggregations are sometimes evident in the fibrous stroma.[2] Although papillary type is commonly seen in this tumour, the term Papillary Cystadenoma is applied only when the lesion has multilocular cyst formations with conspicuous multiple papillary projections into the cystic spaces.[4,5] It is an uncommon benign, well-circumscribed or encapsulated tumour.[6] These cystic spaces may contain eosinophilic, protinaecious material, few epithelial and inflammatory cells in some cases. Psammoma bodies or crystalloids (tyrosine rich crystals) are rarely present within the luminal secretions.

The lining of cystic structures varies from flattened to tall columnar epithelium and cuboidal cells. Mucous, oncocytic, squamous, and apocrine cells are also present in the epithelium focally or occasionally extensively; a mixture of several cell types may commonly be seen.[6] The lining may be one to three epithelial cells thick and may then abruptly become focally thickened or form ramifying papillary projections with central cores of connective tissue.[1,7] In the present case multiple cystic spaces were predominantly lined by mucous columnar epithelium also with the presence of papillary projections, so we termed the lesion as papillary mucous Cystadenoma.

Papillary oncocytic cystadenoma is composed of papillary-cystic proliferation of a single or double layered oncocytic epithelium, superficially resembling a Warthin's tumour without a lymphoid stroma.[1] It lacks the dense lymphoid stroma characteristic of Warthin's tumour. The lining epithelium of oncocytic cystadenoma may focally be admixed with cuboidal or columnar cells.[2] One case of oncocytic cystadenoma having prominent signet ring cell component has been reported in literature.[8] Some cases of papillary oncocytic cyst-adenoma in minor salivary glands have been reported in literature.[8,9] But in the present case PTAH positivity was restricted to normal salivary gland, ruling out the presence of oncocytes in the tumor proper. The presence of oncocytes in the salivary gland could be explained by metaplastic transformation of cells in response to adverse changes, probably the adjacent tumor component with the normal cells losing their original specialization.

The histopathological differential diagnosis of cystadenoma is Warthin tumor, low grade mucoepidermoid carcinoma and cystadenocarcinoma. Warthin tumor (lymphomatous papillary cystadenoma) is characterized by an epithelium of oncocytic origin which shows multiple papillary proliferations projecting into the cystic spaces.[10,11] But because of the absence of lymphoid component we ruled out its possibility. Mucoepidermoid

carcinoma shows cystic structures and also non-cystic epithelial proliferations, a feature that is very important in distinction between both types of neoplasms. When present, papillary growth is irregular and complex, as observed in intraductal papilloma. Because of the absence of irregular, complex papillary growth pattern, epidermoid cells and intermediate cells we ruled out mucoepidermoid carcinoma. Cystadenocarcinomas are morphologically similar to cystadenomas but occur very rarely in the oral cavity.[12] Absence of solid growth in focal areas, cellular atypia in some cases, permeation or destruction of the glandular parenchyma, and breakdown of the glandular lobe architecture, as well as infiltration of adipose, muscle or bone tissues led us to the diagnosis of cystadenoma.

Conservative but complete surgical excision that is superficial parotidectomy is the treatment of choice. Recurrence is very rarely seen in cystadenoma of parotid gland. The present case was reviewed for one year with no recurrence.

CONCLUSION

The knowledge of salivary gland tumour pathology is increasing day by day. Due to the rarity of this tumor, we aimed to document this lesion for easy understanding and better knowledge to the readers.

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