Papillary Cystadenoma of Parotid Gland - An Unusual Entity

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Abstract

Papillary cystadenoma is a rare benign salivary gland neoplasm and oncocytic change can be focal or marked. The cytologic features vary. Papillary oncocytic cystadenoma has been reported mainly in the minor salivary glands and occasionally in the parotid glands. Characteristic histological features are diagnostic. This tumor typically presents as a slow-growing, painless mass, and has prediliction for females above 50 years. The purpose of this article is to report a case of papillary cystadenoma in major salivary gland in a 30 year male which is an unusual presentation. The clinical, histopathological features and differential diagnosis of this entity are discussed.

Key words: *Salivary gland tumor, papillary cystadenoma, parotid*

Introduction

Salivary gland tumors account for less than 3% of the head and neck tumors. They are more common in adults than in children. Tumors arising in the salivary glands account for 10% of all salivary gland disorders.[1] Among all salivary gland tumors, pleomorphic adenoma is the most frequently encountered lesion, accounting for approximately 60% of all salivary gland neoplasms. Approximately 8% tumors arise in minor salivary glands, usually presenting as a slowly growing, painless firm swelling that does not cause ulceration of the overlying mucosa.[2] An important point to be noted here is that majority of them are malignant with only 8.5% being benign.[1]

Salivary gland tumors are fundamentally located on palate, lips, cheek mucosa, tongue and floor of the mouth.[2] These tumors represent a heterogeneous group of neoplasms, with a broad range of histological types and growth patterns. They may have a high recurrence rate when surgical removal is incomplete and the possibility of malignant transformation must be taken into consideration.[3]

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Papillary cystadenoma of salivary glands is an uncommon benign neoplasm. In two large reviews, it constituted 2% and 4.7%, respectively, of all minor salivary gland neoplasms.[4] Cystadenoma frequently affects patients in the sixth decade of life, with a female:male ratio of 3:1. The most common locations of papillary cystadenoma are the palate and the buccal mucosa but tumors affecting the lip and the tongue have also been described.[5] The present case was a male patient of 30 yrs age and the lesion was present in the parotid gland, which is an unusual presentation.

Case report

A 30-year-old male reported to the Department of Oral Medicine and Radiology with a painless swelling on left side of his face since past 6 years. There was no history of trauma. His past dental/medical history was not significant. Clinical examination revealed facial asymmetry due to diffuse swelling on the left side of face in the parotid region (Figure 1). The swelling

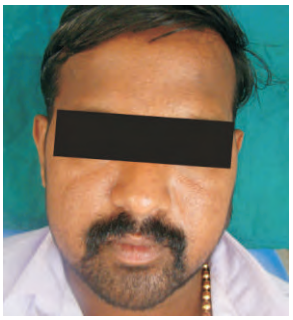

Figure 1 – Extraoral diffuse swelling of left side of face

extended vertically from the left pretragus region to the left angle of the mandible; horizontally 4 cm lateral to ala of the nose to lobule of the left ear, approximately 4×2 cm in size. The ear lobule was not raised. There was no discharge or any other secondary changes (Figure 2). Intraorally there was no evidence of swelling and salivary flow was normal.

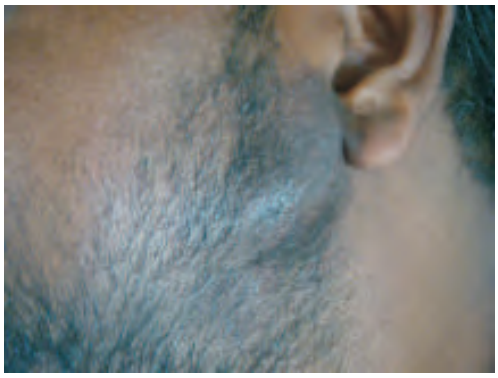

Figure 2 : Swelling of left parotid region

On palpation, the swelling was firm and non-tender. The temperature over the swelling was normal. The swelling was not fixed to the overlying skin. Depending on the history and clinical findings the provisional diagnosis was made as Pleomorphic adenoma of left parotid gland.

Investigations

USG, FNAC and panoramic radiograph were advised. Panoramic radiography revealed no changes. USG report showed heterogenous cystic natured lesion of size 35x22x17 mm with hyperechoeic areas of necrosis seen in superficial lobe of parotid gland suggestive of neoplastic mass lesion. FNAC reports showed cellular smear consisting of small and large monolayered sheets and clusters of oncocytic cells. Many cells showed mild to moderate anisokaryosis. Numerous macrophages, few giant cells and metaplastic squamous cells were seen. The overall features were suggestive of Warthin's tumor and acinic cell tumor. Surgical excision of the lesion was done.

Histopathology

Excisional biopsy showed glandular proliferation of parenchymal tissue in the form of finger like projection and these were directed towards cystic spaces. The projections were lined by cuboidal cells and all cells were isomorphic. Fibrous capsule was also noted in periphery. Overall features were suggestive of Papillary Cystadenoma (Figure 3). The patient is currently being followed up and shows no signs of recurrence.

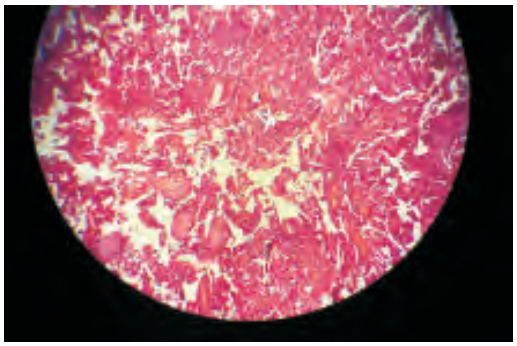

Figure 3- H and E section showing papillary projections (10x

Discussion

Cystadenoma of the salivary glands is a rare benign epithelial tumor, which is predominantly characterized by multicystic growth, exhibiting papillary and, less frequently, mucinous proliferation. The tumor is more frequent in the minor salivary glands, although cases occurring in the major glands, such as the parotid gland, have been reported.[6] The tumor appears to occur more frequently in women; most patients have been older than 50 years of age, with several in their seventies. The most common sites are the palate and buccal mucosa; however, tumors in the lip and tongue also have been described. The usual presentation is an asymptomatic mass.[4] The present case was a male patient of 30 yrs age and the lesion was present in the parotid gland, which is an unusual presentation.

On microscopic examination, the neoplasm is usually well circumscribed and may be surrounded by a rim of fibrous tissue; there are solid areas (usually limited in extent) and cystic areas into which project papillae lined by cuboidal to columnar cells usually two layers thick. The cells usually have eosinophilic cytoplasm, and goblet cells may be present.[4] In the present case same features were noted. In view of the complexity and histomorphological diversity of salivary gland tumors, the differential diagnosis of cystadenoma should include intraductal papilloma, cystadenocarcinoma, low grade mucoepidermoid carcinoma and Warthin tumor. Intraductal papilloma always occurs in a single cystic space and is characterized by numerous and complex papillary projections.These features were not observed in the present case.

Although rare in the oral mucosa, cystadenocarcinomas are morphologically similar to cystadenomas, a fact that tends to make diagnosis difficult. However, some aspects distinguish cystadenocarcinoma from cystadenoma, such

as mode of invasion, pattern 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.[4,5] None of these features was observed in the present case. Similar to cystadenocarcinoma, low-grade mucoepidermoid carcinoma also requires a more in-depth criterion for assessment because it is an infiltrating malignant neoplasm that resembles cystadenoma in terms of the growth pattern and the cell population involved.

In this respect, mucoepidermoid carcinoma presents, in addition to cystic structures, non-cystic epithelial proliferations, a feature that is very important in the distinction between both types of neoplasms. When present, papillary growth is irregular and complex, as observed in intraductal papilloma. Regarding the cell population, low-grade mucoepidermoid carcinoma exhibits a combination of epidermoid, mucosal and, to a lesser extent, intermediate and basaloid cells.On the other hand, Warthin tumor (lymphomatous papillary cystadenoma) is characterized by an epithelium of often oncocytic origin, which shows multiple papillary proliferations projecting into the cystic spaces, supported by a lymphoid stroma, and should not be confused with cystadenoma or other tumors.[5] These lesions are treated by conservative surgical excision, as they are well circumscribed and usually surrounded by fibrous tissue rim. The likely hood of recurrence is low.

Conclusion

The salivary glands may show a diverse range of lesions presenting a challenge to even the most experienced clinician and pathologist. Papillary cystadenoma of major salivary gland is a benign tumor of rare occurrence. It is well-circumscribed tumour, with the cystic cavities containing intraluminal papillary projections and a diagnosis should be made carefully lest a major salivary gland be resected. High index of suspicion and an adequate clearance of the lesion with a cuff of surrounding dispensable normal tissues is the key to successful treatment of such lesions.

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