



CASE REPORT

Pleomorphic adenoma with extensive squamous metaplasia and keratin pearls

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Abstract

Background: Pleomorphic adenoma is the most common salivary gland tumor, accounting for 54 to 65% of all salivary gland neoplasms. 80% of all the benign salivary gland neoplasms is constituted by pleomorphic adenoma. Parotid is the most commonly affected gland followed by submandibular salivary gland and the minor salivary glands. Histo-morphologically a pleomorphic adenoma shows both stromal as well as epithelial components. **Case History:** Here we present the case of a 22 yr. old male who presented to the ENTOPD with complains of swelling in front of the ear on the left side around the parotid region since 2yrs. A biopsy of the swelling was done and sent to the department of Pathology. A diagnosis of pleomorphic adenoma with squamous metaplasia and keratin pearls formation was given. A superficial parotidectomy was performed under general anesthesia. **Discussion:** Pleomorphic adenoma is one of the most common salivary gland tumors the mean age of occurrence being 43.6 yrs. Squamous metaplasia in the pleomorphic adenoma results in formation of an extensive keratin-filled cysts lined by squamous epithelium. Many a times excessive squamous metaplasia is mistaken for malignancy, including mucoepidermoid carcinoma and squamous cell carcinoma. **Conclusion:** The presence of squamous metaplasia in pleomorphic adenoma makes up 25% of the total secondary changes in the tumor. The patient was taken up for surgery & discharged after three days and was on follow-up for one month, after which he recovered completely.

Key Words: Squamous metaplasia; keratin pearls; pleomorphic adenoma

INTRODUCTION

Pleomorphic adenoma is the most common salivary gland tumor, accounting for 54 to 65% of all salivary gland neoplasms. 80% of all the benign salivary gland neoplasms is constituted by pleomorphic adenoma. Parotid is the most commonly affected gland followed by submandibular salivary gland and the minor salivary glands[1]. Histomorphologically a pleomorphic adenoma shows both stromal as well as epithelial components. Many types of metaplasias may at times be associated with a pleomorphic adenoma, for eg. mucous, sebaceous, oncocytic and squamous metaplasia, sometimes with the formation of keratin pearls. Extensive squamous metaplasia can be at times mistaken for malignancies like squamous cell carcinoma & mucoepidermoid carcinoma. Benign adenoma with metaplastic changes may mimic other conditions like keratocystoma, dermoid cyst, squamous cell carcinoma, and mucoepidermoid carcinoma. Hence it's important to differentiate these causes from an adenoma with metaplastic changes. Here we present the case of a 22yr old male who presented with complains of swelling in front of the ear on the left side around the parotid region since 2yrs. A biopsy of the swelling was done and sent to the department of Pathology. A diagnosis of

pleomorphic adenoma with squamous metaplasia and keratin pearls formation was given. A superficial parotidectomy was performed under general anesthesia and the patient was discharged after three days and was on follow-up for one month after that he recovered completely.

CASE HISTORY

A 22 yr old male presented to the ENTOPD with complains of left sided swelling of the face since 2yrs. On examination the swelling was insidious in onset, progressive in nature and painless. There is no history of fever, weakness or ear discharge. His past and personal history is unremarkable except for history of addiction to masala, 4-5 packs per day since 5 yrs. On examination of the swelling, a 2x2 cms firm swelling with irregular borders but smooth surface, 2cms in front of the ear lobule was seen. On examination of the oral cavity grade II tonsillar hypertrophy was also seen.

Investigations

Hematological Investigations: Haemoglobin 16.4gm%, CBC was unremarkable except showing eosinophilia with 12% eosinophils in PBS.

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Radiological Investigations: USG-showed a well-defined round to oval heterogenous, hypoechoic space occupying lesion located in the parotid in the superficial plane measuring (0.7x1.3 cms) s/o Pleomorphic adenoma. CT Head-shows lobular mass with circumscribed borders and smooth surface suggestive of pleomorphic adenoma.

Histopathological Investigation: Grossly a single grayish white soft tissue piece measuring 2x1.5cms received. On microscopic examination lesion showed biphasic appearance with admixture of epithelial and stromal components. Epithelial component being glandular in nature with variable sized ducts and acini lined by cuboidal epithelium with underlying myoepithelial cells. Stroma shows characteristic fibromyxoid areas along with focal areas of squamous metaplasia with formation of keratin pearls. [FIG 1,2] Histomorphologically diagnosis of PA with squamous metaplasia and keratin pearl formation was made. The patient was taken up for surgery and was put on follow-up of 1 month. He was asymptomatic and well post-surgery.

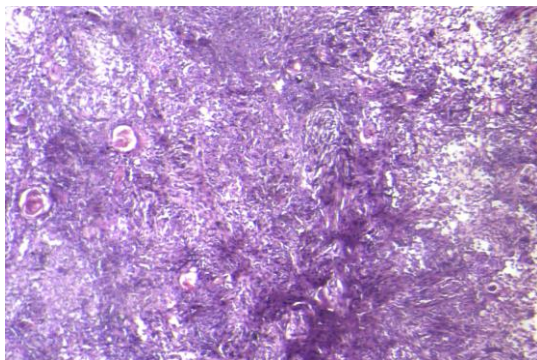


Fig.1:

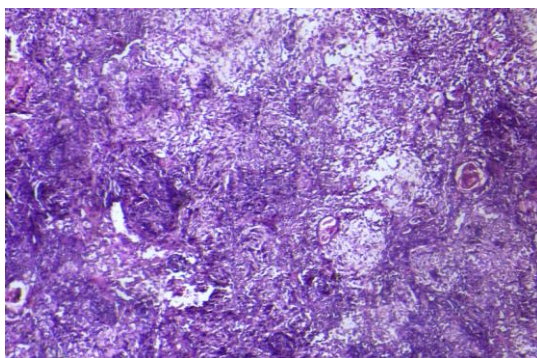


Fig. 2:

DISCUSSION

Pleomorphic adenoma is one of the most common salivary gland tumors, accounting for

54 to 65% of all salivary gland neoplasm's and 80% of the benign salivary gland tumors. Mean age of occurrence is 43.6yrs[2]. The first gland to be affected is the parotid gland followed by submandibular gland, the minor salivary glands being least commonly affected. Microscopically various changes are seen in this tumor like, mucous, sebaceous, oncocytic and squamous metaplasia, sometimes with the formation of keratin pearls. 25% of pleomorphic adenomas show squamous metaplasias[2]. At times squamous metaplasia in the pleomorphic adenoma results in formation of an extensive keratin-filled cysts lined by squamous epithelium. Many a times excessive squamous metaplasia is mistaken for malignancy, including mucoepidermoid carcinoma and squamous cell carcinoma. The IHC markers commonly seen positive in Pleomorphic adenoma are CK 7,8,19, EMA, CEA, GFAP, S-100, SMA, MSA, p63[2]

Squamous metaplasia is also seen in non-neoplastic entities like chronic sialadenitis, necrotizing sialometaplasia, lymphoepithelial cysts, and salivary duct cyst and in neoplastic lesion like Warthin's tumor. Tumor with extensive squamous metaplasia and marked nuclear atypia can be misdiagnosed as SCC. Tricho-epitheliomatous differentiation due to adenexal differentiation in the form of extensive keratin-filled cysts has also been reported. [3,4] Variable stromal changes like chondroid, osseous, myxoid or mucoid may also be seen. Scanty or absence of stroma is an unusual finding. Benign conditions like keratocystoma might be confused with cystic changes in squamous metaplasia [5,6,7,8]. Cystic changes might be due to secretions from the duct of salivary glands, necrosis and haemorrhage. [9,10,11] Important differential diagnosis of this common benign entity with unusual presentation are keratocystoma, dermoid cyst, squamous cell carcinoma, and mucoepidermoid carcinoma [12,13]. Keratocystoma, previously known as choristoma, is a benign salivary gland tumor resembling a trichoadenoma. The unique feature of a keratocystoma is presence of keratinized squamous cells without granular layers [11]. Goulart MC *et al.*, believes that the PA and keratocystoma may constitute related lesions, representing different stages in the evolution of a specific type of salivary gland tumor. Anucleated squamous cells and skin appendages will lead to a diagnosis of dermoid, if present, but not so in this case. The

features favoring squamous cell carcinoma in this case are the presence of atypical keratinized squamous cells and keratin. Cytologic atypia in metaplastic squamous cells are the main culprit for the misdiagnosis. However, presence of fibrous capsule and absence of invasion are against the squamous cell carcinoma on histology. [9,10] Mucous and intermediate squamoid cells are seen in mucoepidermoid tumor not seen in present case. Chondromyxoid stroma of pleomorphic adenoma is a very typical stromal feature, which helps to differentiate from other tumors although not diagnostic. Chances of recurrence is 5-30% and treatment of choice being wide local excision [2]. The problem of satellitism, when being operated by the surgeons is common to occur.

In the present case patient showed features of pleomorphic adenoma with keratin pearls but no cystic changes. The patient was operated and is well. This case presented with an excellent opportunity of presenting a variation present in only 25% of the cases only.

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