

Cystic Pleomorphic Adenoma: A Diagnostic Challenge On Cytology

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Dear Sir,

Although fine needle aspiration cytology (FNAC) is a highly accurate tool for the diagnosis of pleomorphic adenomas, even this common salivary gland neoplasm can be diagnostically challenging and cause pitfalls in cytodiagnosis. In particular, the presence of cystic degeneration and squamous metaplasia may cause diagnostic confusion and stresses the need for guarded approach while interpreting such lesions.

A 36-year-old male presented with a painless, left preauricular swelling since 3 months. Ultrasonography of the lesion revealed a lobulated hypoechoic mass lesion measuring approximately 27x18 mm in upper lobe of left parotid gland. The lesion showed few cystic areas. Diagnosis of Pleomorphic Adenoma with Cystic Degeneration was given. Routine FNAC yielded a mucoid fluid mixed with blood. Cytologic smears revealed predominantly foamy macrophages in a mucoid and hemorrhagic background. Only one smear revealed fibrillary chondromyxoid material. An ultrasound-guided FNAC was then performed from the solid part of the lesion. The smears revealed oval to spindle myoepithelial cells entrapped within fibrillary chondromyxoid matrix. Multiple hyaline stromal globules were evident. Squamous metaplastic cells were seen in fair numbers. Few oncocyctic cells were also noted. Background was mucoid and hemorrhagic and showed fair number of foamy macrophages. A cytologic diagnosis of Pleomorphic Adenoma with Cystic Change was rendered and histopathology was advised. The patient then underwent left superficial parotidectomy. Grossly, the specimen had 4 pieces measuring 3.5x1.7x1.7 cm to 1.4x1x0.2 cm and weighed 15 gm. On cutting a capsulated growth of 2x1.5 cm with tan fleshy appearance was seen. It was solid with partial cystic area. Microscopically, biphasic

appearance was seen resulting from intimate admixture of epithelium with stroma. Epithelial component was forming tubules within chondromyxoid matrix. There were cystic spaces filled with mucinous material. Squamous and oncocyctic metaplasia was seen. No necrosis or nuclear atypia was appreciated, thus the histopathologic diagnosis was consistent with Pleomorphic Adenoma with Cystic Change.

A wide variety of neoplastic and non-neoplastic lesions of the salivary glands may present as either partial or completely cystic masses. The non-neoplastic cystic lesions include developmental anomalies such as polycystic disease of the parotid glands, benign lymphoepithelial cysts and degenerative changes of the salivary gland duct system, such as mucus retention cyst and mucus extravasation reaction. The neoplasms that may present with a cystic component include pleomorphic adenoma, Warthin's tumor, low-grade mucoepidermoid carcinoma, intraductal papilloma, acinic cell carcinoma and mucinous adenocarcinoma.^[1] Other than cystic changes, pleomorphic adenomas may often be associated with changes such as squamous,^[2-6] mucinous and sebaceous cell metaplasia.^[4,7] Diagnosing pleomorphic adenoma accurately in such a situation may be challenging for cytologists.^[7] There have been rare studies^[1] and a few case reports^[4,6] in the literature highlighting the diagnostic problems encountered with the cystic lesions of the salivary glands on FNAC. Such interpretive problems may result in false positive or false negative diagnosis. Most often, the false positive diagnosis occurs as a result of metaplastic epithelial components in the aspirates, in particular the squamous metaplastic cells.^[8,9] and false negative diagnosis occurs as a result of dilution of diagnostic tumor cells by the cyst fluid. In cytology, aspirates from cystic pleomorphic adenomas showing epithelial metaplastic changes, especially in

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the absence of characteristic chondromyxoid stroma, may be misinterpreted as malignancy in general, and mucoepidermoid carcinoma in particular.^[7] There have been cases in the literature in which such metaplastic changes have misled cytologists, resulting in either suspicion or misinterpretation of pleomorphic adenoma cases as mucoepidermoid carcinomas.^[4,7] However, the presence of characteristic fibrillary chondromyxoid stroma as in our case excludes this possibility. It is also worth remembering that conditions such as mucus retention cysts, mucus escape reactions and papillary cystadenocarcinoma may also have a mucoid or mucoid/myxoid background in the cytologic smears. However, in general, an epithelial component is absent in cases of mucus retention cysts and the mucus escape reactions, with a cellular component consisting of lymphocytes and histiocytes only; papillary cystadenocarcinomas with mucoid aspirates generally contain at least a few papillary fragments or other features of glandular differentiation. Due to the fact that the mucoid aspirates are seen also in nonneoplastic lesions, salivary gland tumors with mucoid aspirates may often be misinterpreted as non-neoplastic cysts. Layfield and Gopez,^[1] in their series of cystic lesions of the salivary glands, encountered a case of pleomorphic adenoma that yielded predominantly mucoid extracellular material, initially suggesting a mucus retention cyst. However, additional aspirates in that case showed characteristic myxoid and chondroid fragments, leading to an accurate diagnosis. This fact emphasizes the importance of a repeat FNAC in such situations, to obtain a more representative sample as in our case.

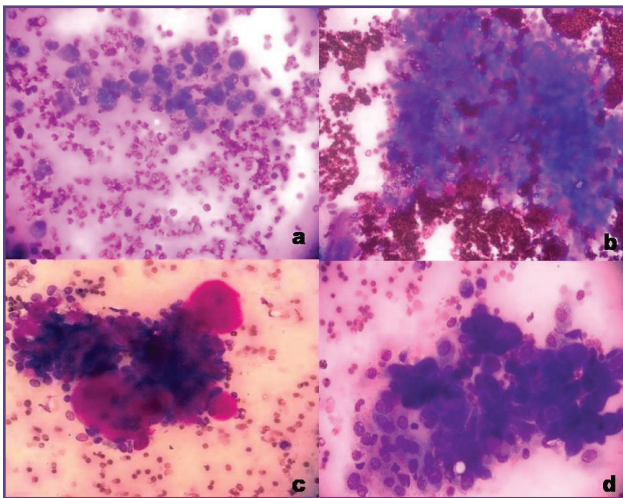


Fig. 1: Cytologic smears showing (a) foamy macrophages (MGG, x400), (b) myoepithelial cells entrapped within fibrillary chondromyxoid matrix (MGG, x400), (c) hyaline stromal globule (MGG, x400), (d) squamous metaplastic cells (MGG, x400)

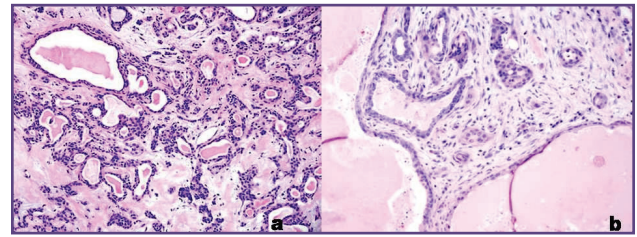


Fig. 2: Histologic sections showing (a) epithelial component forming tubules within chondromyxoid stroma (H & E, x100), (b) cystic spaces filled with mucinous material (H & E, x100)

A cystic pleomorphic adenoma with squamous metaplasia is a diagnostic dilemma for cytopathologists. A cautious and systematic approach in such a situation helps in its accurate diagnosis.

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Competing Interest

None declared

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