

CARCINOMA EX PLEOMORPHIC ADENOMA: A RARE SIGHT ON CYTOLOGY!

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INTRODUCTION:

Malignancy in pleomorphic adenoma is an uncommon event and has been categorized as carcinoma ex pleomorphic adenoma; carcinosarcoma (true malignant mixed tumor) and metastasizing pleomorphic adenoma.^[1]

Fine needle aspiration cytology (FNAC) has been a widely accepted diagnostic tool for palpable salivary gland lesions. Diagnostic accuracy of FNAC of salivary gland lesions ranges from 80-95% in adequately sampled specimens.^[2, 3] One of the entities rarely diagnosed on cytology is carcinoma ex pleomorphic adenoma.^[4] We present a case report of this uncommon entity diagnosed on cytology.

A 55 year old male presented with swelling in front of the left ear of 10 years duration. It started initially as a small nodule and increased gradually to the present size. Patient also gave history of a rapid increase in size during the last 3 months associated with pricking type of pain. There was no history of skin discoloration or increase in size while chewing. There was no history of fever, ear discharge or pain. General and systemic examinations were within normal limits.

Local examination showed a 3.5x 3cm swelling in front of the left ear. The swelling was firm in consistency, fairly circumscribed and was lifting the ear lobe. There was no warmth, pain or tenderness over the swelling. A clinical diagnosis of pleomorphic adenoma of left parotid gland was made and sent for FNAC.

Fine needle aspiration cytology of the left parotid swelling was performed using a 22 gauge needle and the material aspirated was smeared on to the glass slides. Smears fixed in 95% ethanol were stained with hematoxylin and eosin stain while the air dried smears were stained with

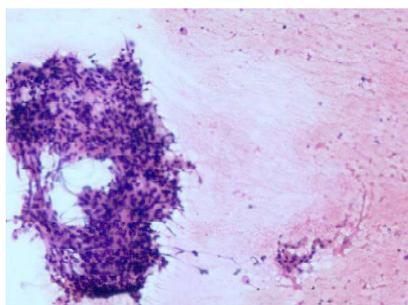


Fig. 1: Scanner view showing cellular cell cluster (Hematoxylin and eosin X 20)

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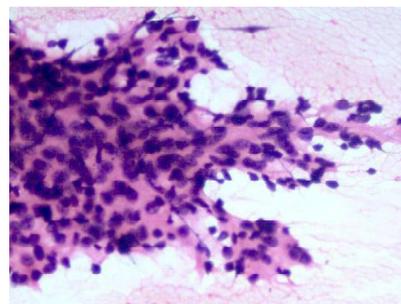


Fig. 2: Closely admixed benign cells with bland nuclei and malignant epithelial cells having coarse nuclear chromatin (Hematoxylin and eosin X 100)

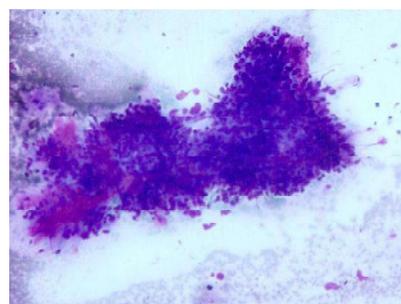


Fig. 3: Cluster showing biphasic pattern with epithelial cells and chondromyxoid background seen as by magenta coloured material (May-Grunwald Giemsa X 40)

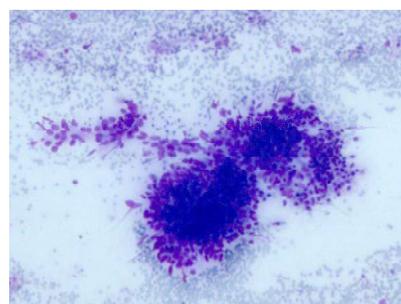


Fig. 4: Benign epithelial cells in the clusters appear small oval with round nuclei having a smooth nuclear membrane. Few atypical cells are also seen (May-Grunwald Giemsa X 40)

May-Grunwald Giemsa stain. Examination of the cytological smears revealed a high cell yield (Fig. 1). Smears showed a mixed population of cells composed of benign and malignant elements (Fig. 2). Some clusters showed a metachromatic stroma in the background (Fig. 3). The benign epithelial component appeared as small round to oval cells with bland oval nuclei (Fig. 4). The malignant population consisted of large cells with large pleomorphic nuclei having coarse chromatin and high nucleo-cytoplasmic ratio (Fig. 2, 4 & 5). Malignant change occurring in a background of pleomorphic adenoma was suggested based on the above mentioned cytological features.

A superficial parotidectomy was performed under general anesthesia and the excised specimen was sent for histopathological examination.

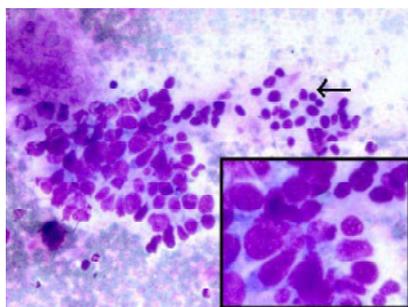


Fig. 5: Loose cluster of highly atypical cells with an occasional cluster of benign epithelial cells (arrow) in the adjacent area (May-Grunwald Giemsa X 100); Inset shows large atypical cells with large pleomorphic nuclei, coarse chromatin and irregular nuclear membrane (May-Grunwald Giemsa X 200)

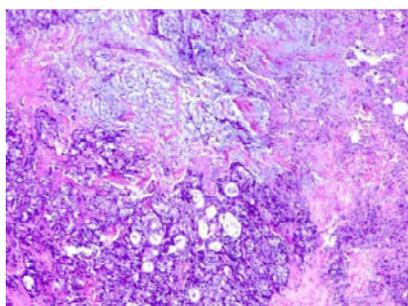


Fig. 6: Microscopic section showing mixed tumor with epithelial cells and mesenchymal chondro-myxoid material (Hematoxylin and eosin X 20)

Gross examination showed a soft tissue mass measuring 5x4x3.5cm. External surface appeared irregular. Cut surface showed a fairly well circumscribed grey tan nodular tumour measuring 3cm in diameter having focal myxoid areas compressing the adjacent salivary gland tissue to the periphery (Fig. 6). The margins were uninvolved.

Microscopic sections showed tumor with features of biphasic pattern consisting of epithelial and mesenchymal components. Tumor displayed islands of bland cells arranged in glandular pattern and cords with a chondro-myxoid background.

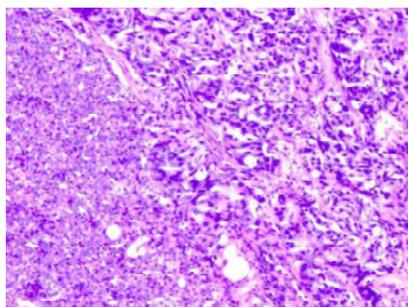


Fig. 7: Histology illustrating benign epithelial cells arranged in glandular and trabecular pattern along with a focus of highly atypical cells showing hyperchromatic pleomorphic nuclei (Hematoxylin and eosin X 40)

In addition, one large focus showed groups of highly atypical epithelial cells with pleomorphic nuclei and

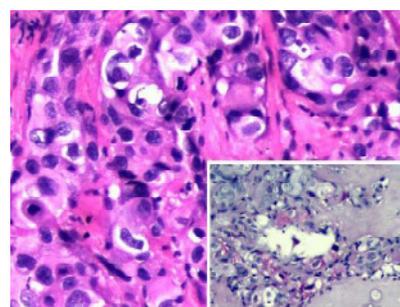


Fig. 8: Malignant cells showing cytoplasmic mucin and desmoplastic stroma (Hematoxylin and eosin X 100); inset shows magenta colour in cytoplasm highlighting mucin (Periodic Schiff stain X 100)

increased nucleo-cytoplasmic ratio (Fig. 7 and 8). Pleomorphic cells were arranged in squamoid nests, glandular pattern and sheets. Few of the tumor cells showed presence of cytoplasmic mucin demonstrated by periodic Schiff (PAS) (Fig. 8). Surgical margins were negative for tumour infiltration. A final diagnosis of mucoepidermoid carcinoma occurring in a background of pleomorphic adenoma was made.

DISCUSSION

Fine needle aspiration cytology is a well accepted tool for the pre-operative diagnosis of salivary gland tumours as they are easily accessible. However, cytology of many of the salivary gland lesions may not always display classical features leading to diagnostic dilemma to the cytopathologist.

The wide spectrum of morphological patterns and frequent overlap of cytomorphological features in a salivary gland lesion leads to difficulty in cytological diagnosis. The other possible reason for inaccurate diagnosis is needle miss in a heterogenous tumor leading to a sampling error.^[5] Carcinoma ex pleomorphic adenoma accounts for 3.6% of salivary gland tumours.^[6] It is difficult to diagnose this rare entity on cytology.^[7] In a series studied by Verma et al on pleomorphic adenoma, all the cases of carcinoma ex pleomorphic adenoma were missed on cytology resulting in a false negative pre-operative cyto-diagnosis.^[4]

Quite often the cytologist is satisfied with a single needle pass in FNAC, provided the aspirated material has adequate cellularity for deriving an opinion. In our case, since we made multiple needle passes from different areas of the swelling, we were able to sample the areas of malignant transformation. There are no prescribed criteria for the number of aspiration attempts to be made on salivary gland lesions. However, it has been recommended to make at least 3 passes to completely sample the lesion for a presumptive diagnosis.^[8]

The common clinical feature suggestive of a malignant change includes sudden increase in size of a longstanding lesion, pain and facial nerve palsy. In our case, there was a sudden increase in size for the past 3 months in a swelling present for 10 years. The possibility of malignant transformation in a longstanding pleomorphic adenoma

increases from 1.6% in < 5 years to 9.5% in a tumor present for 15 years.^[9] The common malignancies occurring in a background of pleomorphic adenoma are adenocarcinoma not otherwise specified (42.4%) and salivary duct carcinoma(32.8%).^[8] The other uncommon malignancies that can arise in a setting of pleomorphic adenoma are adenosquamous carcinoma, adenoid cystic carcinoma, undifferentiated carcinoma, myoepithelial carcinoma, epithelial-myoepithelial carcinoma and sarcomatoid carcinoma. Mucoepidermoid carcinoma arising in a pleomorphic is a rare event.^[9] On cytology, it may show features of non-specific high grade malignancy as noted in our case. Presence of mucin producing cells in addition can support a diagnosis of mucoepidermoid carcinoma. Although mucin producing cells were appreciated on histopathological sections of our case, it was not evident on cytological smears and hence due to lack of specific findings only a diagnosis of malignancy owing in a background of pleomorphic adenoma was offered in cytology. Our inability to provide a precise cyto-diagnosis as mucoepidermoid carcinoma was due to lack of specific cytomorphological features on cytosmears.

A careful scrutiny of all smears helped us in making a cytodiagnosis of malignant change in a background of pleomorphic adenoma. This vital information is necessary for the surgeon to plan out the therapeutic modalities for a better outcome.

Considering the polymorphic nature of some salivary gland tumours, it is advisable to aspirate from more areas to render the correct cytodiagnosis. Aspiration from multiple sites is mandatory in salivary gland lesions clinically suspicious of malignancy.

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