Case Report

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Breast Adenomyoepithelioma with Predominance of Morules, A Cytological Dilemma

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ABSTRACT

Adenomyoepithelioma is a rare, benign proliferative tumour that can involve the breast. It is usually present as a solitary, unilateral, painless mass at the periphery of the breast in women range in age from 26 to 82 years (average 63 years). Tumour sizes range from 0.5 to 8 cm. (average size 2.5 cm).We report a case of adenomyoepithelioma of right breast in a 58 years old female since two months with diagnostic difficulty on cytology, especially with morules predominance that merits documentation due to its rarity.On physical examination, it was a single well defined, lobulated, non mobile, firm mass of 10 x 8 x 5 cm in upper outer quadrant of right breast without associated axillary lymphadenopathy. Sonomammography showed well defined lobulated right breast mass with macrolobulations and cystic changes suggestive of phyllodes tumour. Wide local excision was performed and histopathological study revealed adenomyoepithelioma which is confirmed by P63 immunostain.

Keywords: *Adenomyoepithelioma, Breast, Morules.*

Introduction

We are presenting a case of adenomyoepithelioma breast with diagnostic difficulty on cytology, especially with morules predominance. FNAC diagnosis of adenomyoepithelioma of breast can be very challenging problem for pathologist. Appropriate special stain masson trichrome, immunostain and histopathology may play a vital role in such situation for arrival at diagnosis.

Adenomyoepithelioma of breast was first reported by Hamprel in 1970,more than 60 cases had been reported since then[1].It is a rare tumour characterized by biphasic proliferation of an inner layer of epithelial cells and a prominent peripheral layer of myoepithelial cells. Usually, it is benign but occasionally may have malignant potential. Incidence of adenomyoepithelioma in breast could not be ascertained due to rarity of lesion and paucity of literature with few case reports [2] Only few cases of adenomyoepithelioma with detailed fine needle aspiration biopsy findings have been described in the literature. Adenomyoepithelioma may be detected by sonomammography but it is in apparent on mammography. A case described herein represents a rare case of adenomyoepithelioma breast with diagnostic difficulty on cytology, especially with morules predominance.

Case Report

A 58 year old, postmenopausal female clinically presented with a rapidly increasing painless lump in right breast since two months. On local examination 10 x 8 x 5 cm lobulated, non mobile, firm, mild tender lump noted in upper outer quadrant occupying the substantial part of right breast, overlying skin and nipple-areola appear normal with few superficial dilated veins (Figure 1A). Left breast was normal with no palpable bilateral axillary lymph nodes. Sonomammography showed well defined lobulated 10 x 7 x 5 cm mass, occupying entire right breast with macrolobulations and cystic changes suggestive of Phyllodes tumour. Routine haematological parameters, chest x-ray and USG abdomen were normal.

Fine needle aspiration cytology (FNAC) of right breast lump was done and stained with Papanicolaou stain showed a cellular smear with ductal epithelial cell hyperplasia. Epithelial cells were in cohesive flat sheets, cribriform pattern and globi surrounded by epimyoepithelial cells. The epithelial cells have round to oval monotonous nuclei, bland chromatin, inconspicuous nucleoli and smooth nuclear membrane with scanty cytoplasm. Fibrillary myxoid stromal fragments and myoepithelial cells were present.(Figure 1B&1C).No necrosis or mitotic figures were seen. Cytological diagnosis was given as? Phyllodes ? Low grade adenoid cystic carcinoma and tissue diagnosis was advised.

Grossly, a wide local excision specimen of the patient showed 10 x 8 x 5 cms skin covered mass with lobulated greyish white, firm cut surface showing leaf like and cystic areas (Figure 2A). The formalin-fixed tissue sections were

stained with Hematoxylin and eosin stain showed, well demarcated tumour arranged in tubular architecture with biphasic proliferation of glandular epithelial cells and surrounding myoepithelial cells with pale clear cytoplasm along with the intervening fibro vascular stroma. Some glandular lumens contain secretion. Histomorphological features suggestive of Adenomyoepithelioma (Figure 2B). After review of FNAC smears again a massontrichrome stain showed collagen present in the cribriform spaces (Figure 3A). Massontrichrome stain on tissue section also showed collagen present in the cribriform spaces (Figure 3B). Immunostain for P63 showed strongly positive nuclear reactivity in the proliferating myoepithelial cells, while the epithelial cells fail to react confirming a diagnosis of adenomyoepithelioma(Figure 4). The resection margins were free. No additional treatment was performed.

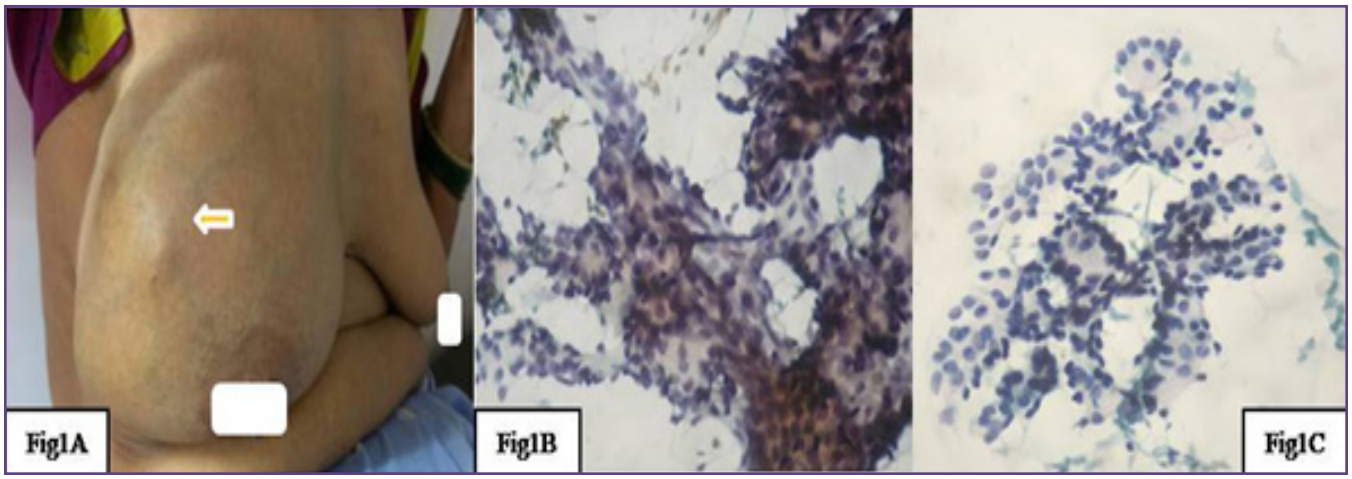

Fig. 1A: Large 10 x 8 x 5 cm lobulated, non mobile, firm, mild tender lump [arrow] noted in upper outer quadrant occupying the substantial part of right breast, overlying skin and nipple-areola appear normal with dilated veins. Left breast appears normal. Figure 1 B: Right breast FNAC reveals, cellular smears with ductal epithelial cells hyperplasia, cribriform pattern, fibromyxoid stromal fragments and myoepithelial cells are present in background. (Pap stain: 100 X) Figure 1C: Cribriform pattern and globi surrounded with epimyoepithelial cells in sheets (Pap stain: 100 X).

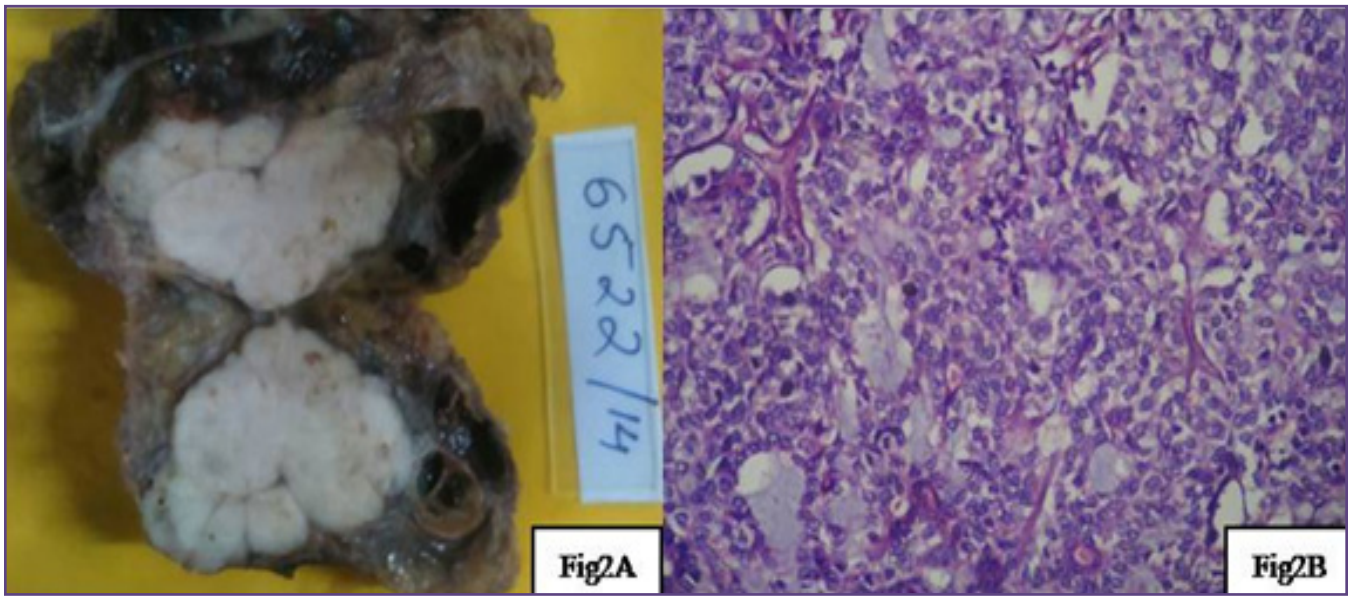

Fig. 2A: Wide local excision specimen of the right breast lump measuring 10 x 8 x 5 cms with lobulated greyish white, firm cut surface showing leaf like and cystic areas. Figure 2B: Histologically, tumour arranged in tubular architecture with biphasic proliferation of glandular epithelial cells and surrounding myoepithelial cells along with the intervening fibro vascular stroma. Some glandular lumens contain secretion.(H&E stain:100 X).

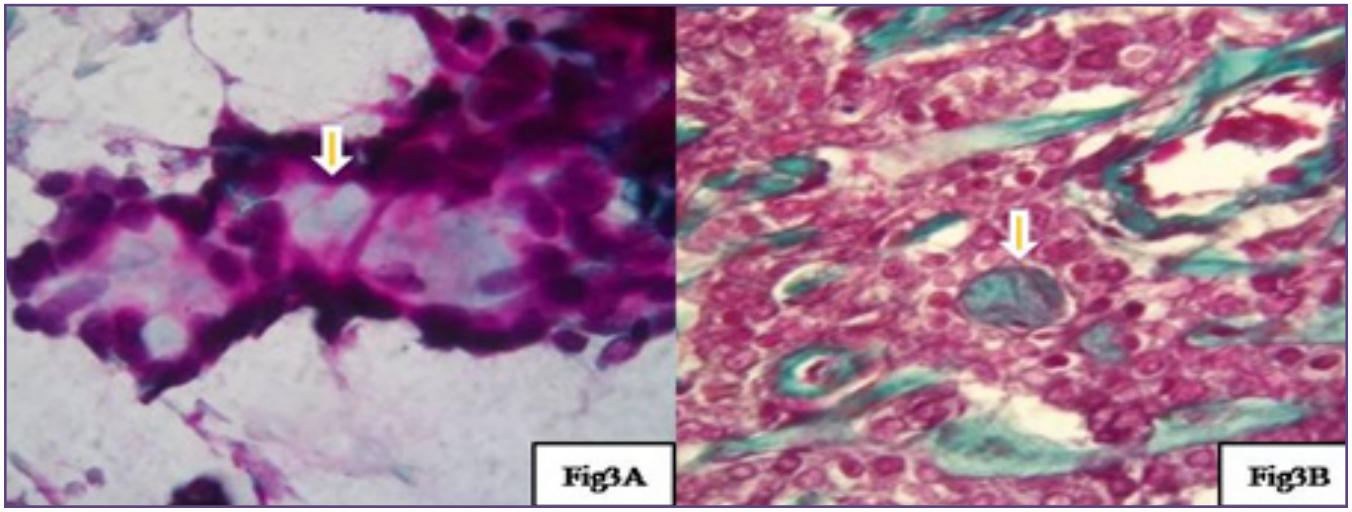

Fig. 3A: Masson trichrome stain on FNAC smear showed collagen[arrow] in the cribriform spaces. (400 X)Figure 3B: Masson trichrome stain on tissue section showed collagen[arrow] in the cribriform spaces. (400 X).

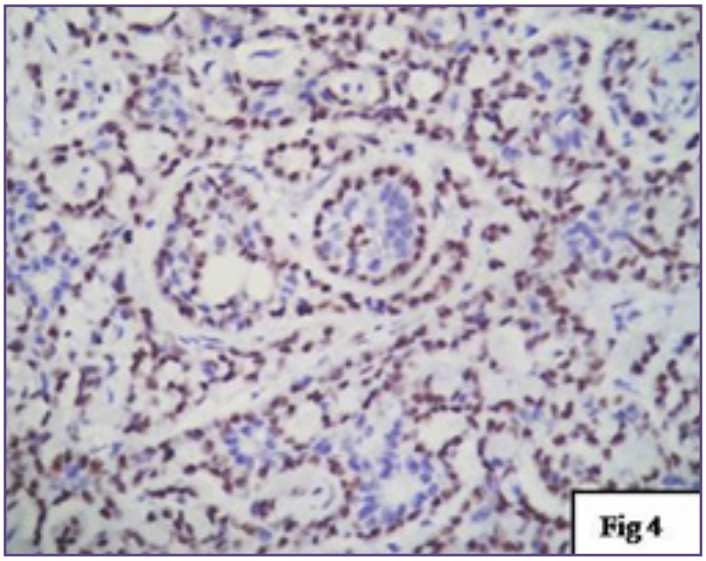

Fig. 4: Immunostain for P63 showing nuclear reactivity in the proliferating myoepithelial cells, while the epithelial cells fail to react (100 X).

Discussion

Myoepitheliomas or even myomas are purely myoepithelial benign tumours and the presence of the epithelial component in these tumours make them adenomyoepitheliomas[3] .There may be predominance of any of these components. Tumours derived from myoepithelial cells had been reported in skin, salivary glands, breast and lungs. Myoepithelial cells derived from ectoderm are widely present in breast where they comprise part of microanatomy of lobules and ducts. This cell layer is present between the basement membrane and the epithelial cells and it is referred to as the basal layer. The myoepithelial cells containing myofilaments in their cytoplasm show contractility, they support the parenchyma and contribute to the production of laminin, collagen type IV and fibronectin to maintain the basal lamina. Myoepithelial cells in FNAC can appear epitheloid, plasmacytoid, spindled or mixed cell morphologies; they may have clear cytoplasm with intracytoplasmic vacuoles and intranuclear inclusions. [4] Their presence in FNAC smears is an indicator of benignity.

Adenomyoepithelioma is difficult to diagnose on cytology exclusively and may mimic other myoepithelial stromal rich lesions like phyllodes tumour and adenoid cystic carcinoma.

Cellular stroma with an increase in cellular pleomorphism and mitotic activity of the plump spindle cells suggest malignant phyllodes tumour. Adenoid cystic carcinoma of breast, another biphasic tumour yields characteristic three dimensional tubular or cribriform structures associated with numerous hyaline globules which are periodic acid Schiff positive and were surrounded by a monotonous and cellular population of neoplastic cells with hyperchromatic nuclei and high nuclear to cytoplasmic ratio.[4]

In this case, the presence of typical cribriform arrangement of cells with spaces surrounded by neoplastic cells on FNAC lead to the misdiagnosis of low grade adenoid cystic carcinoma as a differential diagnosis. It was ruled out by using masson's trichome staining on FNAC, as well as histological tissue section that showed cribriform spaces filled with collagen.

Adenomyoepithelioma shows strong positivity for keratins CAM5.2 ,EMA in the epithelial component and myoepithelial cells show positivity with smooth muscle actin, S-100, P63, CD10, CK5, Myosin, and Calponin[5]. However, p63 produces the best results with consistent intense nuclear staining.

Malignant transformation was rarely documented and may be limited to either epithelial or myoepithelial component or both elements may be involved. In addition to nuclear atypia, mitoses, areas of necrosis in FNAC material along with evidence of definitive invasion and spindle cell overgrowth on histological sections should raise suspicion of a malignant process.

Prognosis of patients with benign adenomyoepithelioma of the breast was usually good, but it had a potential for local reoccurrence, especially in the tubular and lobulated variants. Total surgical excision with an adequate margin of uninvolved breast tissue was therefore recommended. [5,6,7]

Conclusion

In summary, the cytological features diagnostic of AME were difficult to define because of varied histomorphology. No features alone and no features in combination should be regarded as specific or characteristic of AME. Hence awareness of this entity and accurate identification of the myoepithelial cells was crucial. On cytology smears, a simple special stain like massontrichrome can be useful for suggesting the diagnosis of AME by confirming collagen present in cribriform spaces surrounded by epimyoepithelial cells. In uncertain cases, a cell block preparation and immunocytochemical staining for myoepithelial markers may be used. Therefore the difficult differential diagnosis, potential for recurrence and malignant evolution of this lesion merit a careful approach.

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