



A Rare Case of Benign Breast Tumor – Adenomyoepithelioma

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Introduction

Benign adenomyoepithelioma of the breast is a rare tumor characterized by biphasic proliferation of both an inner layer of epithelial cells and a prominent peripheral layer of myoepithelial cells. It was first described by Hamperl^[1] in 1970. Benign adenomyoepitheliomas outnumber their malignant counterparts by far. Malignancy may arise either through malignant transformation of 1 of the 2 cellular components or through malignant transformation of both. The difficult differential diagnosis, potential for recurrence and malignant evolution of this lesion merit a careful approach

Clinical Presentation

A 43-year-old woman presented with a swelling in left breast for the past 8 months. On examination, a 3 × 2.5 x 2 cm firm, non-tender, and mobile mass was palpated. Fine needle aspiration cytology of the mass showed clusters and sheets of ductal epithelial cells with apocrine metaplasia and hyperplasia along with clusters of myoepithelial/ stromal cells in background of eosinophilic fluid and cyst macrophages (figure 1,2). Hence a diagnosis of a Fibrocystic disease was made on cytological evaluation. Subsequently the mass was excised in toto and sent for histopathological examination.

Gross Examination

Single, grey-white, globular, soft tissue mass weighing 36 grams, measuring 3 x 2.5 x 2cm was received. On sectioning, the cut surface showed multiple cysts like areas of varying sizes along with solid areas. The cystic areas were filled with gelatinous material. All the tissue was embedded.

Microscopy

Histopathology showed multiple cysts lined by flattened epithelium (figure 3). The cyst wall showed foam cells and chronic inflammatory cells predominantly composed of lymphocytes. The lumen showed many cyst macrophages and cholesterol clefts. (Figure 4) Few foci showed compressed ducts lined by epithelium and stromal hyperplasia. The solid areas showed proliferation of myoepithelial cells in whorls and fascicles (figure 5). Few foci showed clear cell change (figure 6) and epitheloid differentiation.

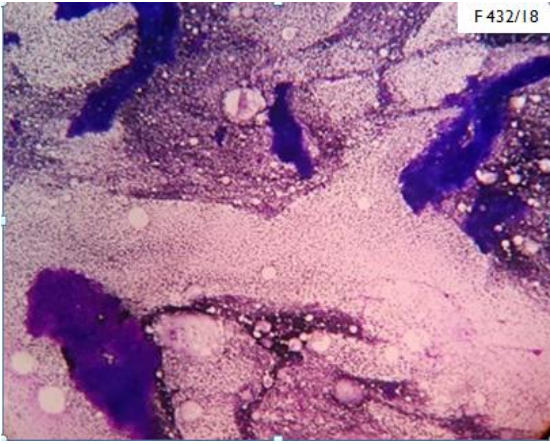


Figure 1: Photomicrograph (4X) showing both epithelial and myoepithelial clusters.

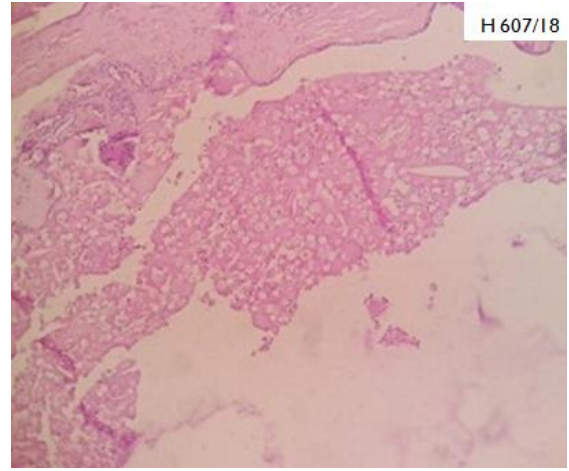


Figure 4: Photomicrograph (10X) cyst wall and lumen showing cyst macrophage, inflammatory and cholesterol clefts

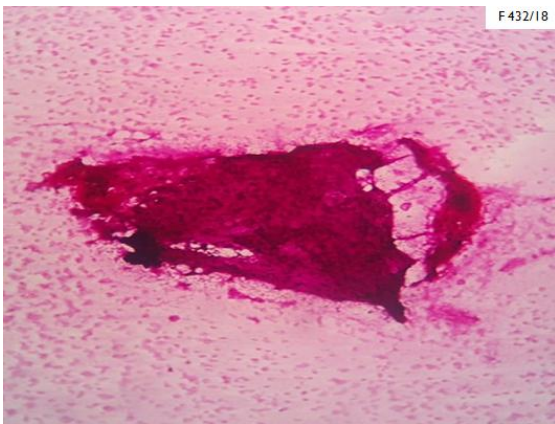


Figure 2: Photomicrograph (10X) showing myoepithelial clusters.

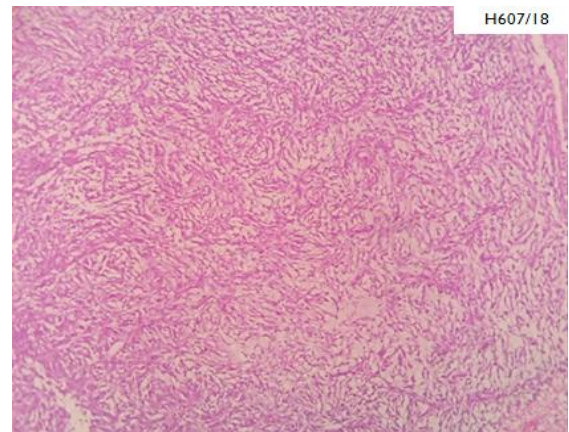


Figure 5: Photomicrograph (10X) showing myoepithelial cells

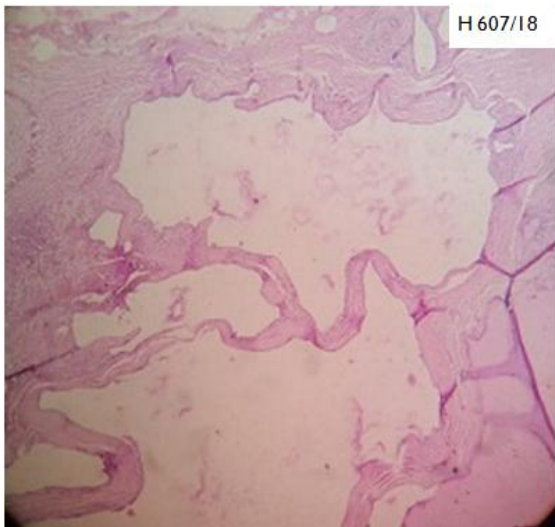


Figure 3: Photomicrograph (10X) showing multiple cyst lined by flattened epithelium

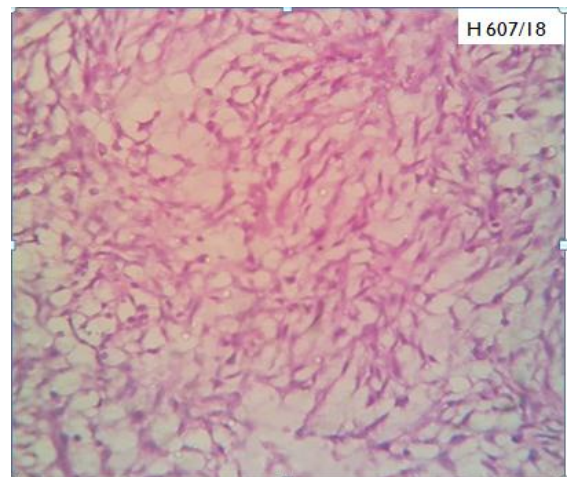


Figure 6: Photomicrograph (40X) showing myoepithelial cells with clear cell change.

Discussion

Myoepithelial cells are a normal component of breast tissue and are located between luminal cells and the basal lamina, of the mammary duct and lobular system. The most common lesion affecting these cells is sclerosing adenosis. Presence of myoepithelial cells in neoplastic lesions has been considered as the hallmark of benignity. Recently, breast neoplasms have been described, ones that are entirely or partially composed of myoepithelial cells. Neoplasms of purely myoepithelial origin have been called myoepitheliomas and may be benign or malignant in approximately equal proportions. Tumors derived from myoepithelial cells have been reported in skin, breast, salivary glands, and lungs; mammary tumors containing myoepithelial elements are not frequent. Tumors with bicellular proliferation of both epithelial and myoepithelial cells are called adenomyoepitheliomas. Tavassoli classification divides adenomyoepitheliomas into spindle cell, lobulated, and tubular (or adenosis) types.

It is closely related to adenomyoepithelial (apocrine) adenosis and sometimes also considered an uncommon variant of intraductal papilloma. The exact etiology of breast adenomyoepithelioma is still obscure. All cases have been sporadic and no familial aggregation has been observed. Kiaer et al.^[2] reported a case of sequential changes from adenomyoepithelial adenosis into adenomyoepithelioma which eventually became low grade malignant adenomyoepithelioma during the course of 18 years. From this observation, Choi et al.^[3] proposed that adenomyoepithelioma was derived from a myoepithelial, long-standing, underlying breast disease such as adenosis and fibroadenoma. Malignant adenomyoepithelioma of the breast is a rare lesion characterized by malignant proliferation of epithelial and myoepithelial cells that show characteristic histologic and immunohistochemical features. Most of the cases demonstrate malignant transformation of only one

cellular component, either epithelial or myoepithelial, though more often epithelial. The epithelial elements stain with antibodies to cytokeratins and carcinoembryonic antigen. The myoepithelial cells stain with S100 and actin. More recently, C10 and p63 have been used as reliable markers of myoepithelial phenotype. Our case shows equivocal staining for both ck-7 and p63, possibly due to extensive cystic change /sclerosis. Other reasons for negative or equivocal stains are partial atrophy which can mimic a malignant transformation in staining characteristics and the quality of immunomarker used. Presence of isoforms will give equivocal staining.

In our case, the clinical, gross and microscopic features did not have any feature of malignancy in the multiple serial sections studied. There was no feature predictive of malignant transformation. The resection was complete with adequate margin clearance.

Conclusion

Prognosis of benign adenomyoepitheliomas of breast is usually good, but it has the potential for local recurrence, especially in the tubular and lobulated variants. Total surgical excision with an adequate margin of uninvolved breast tissue is therefore recommended. Failure to achieve a free resection margin may result in local recurrence.

Hence, the take home message is that, Adenomyoepithelioma, an indolent breast neoplasm has local recurrence and metastasis, especially if malignant transformation occurs focally within the original mass. Therefore, a meticulous approach, detailed study and proper follow up are warranted.

References

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