

Adenomyoepithelioma of breast: Report of a rare case

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ABSTRACT

Breast adenomyoepithelioma is a rare benign proliferative tumor. It usually presents as a solitary unilateral painless mass at the periphery of the breast. It is characterized by proliferation of epithelial and myoepithelial cells. This benign tumor is known for its recurrence and metastatic potential. Hence accurate diagnosis with close follow up is mandatory. We report a rare case of adenomyoepithelioma in a 38-year-old female who presented with a painless lump of 3 × 2 cm size in the left breast for 3 months.

Key words: Adenomyoepithelioma, breast, myoepithelial cells

INTRODUCTION

Myoepithelial cells are a normal component of breast tissue. Adenomyoepithelioma is a rare benign breast tissue tumor with proliferation of glandular and myoepithelial cells. It was first described by Hamperl in 1970.^[1] The myoepithelial lesions are divided into three different categories (1) myoepitheliosis, (2) adenomyoepithelioma, and (3) myoepithelial carcinoma.^[2] Majority of adenomyoepitheliomas are benign. Few cases of adenomyoepitheliomas are reported to have local recurrences, metastasis, and malignant transformation.^[2,3] Failure to recognize this tumor may lead to inappropriate diagnosis and management. Here we report a case of adenomyoepithelioma in a 38-year-old female.

CASE REPORT

A 38-year-old female presented with lump in left breast since 3 months. Lump was mobile, painless, size of 3 × 2 cm in upper outer quadrant of left breast with firm

consistency. Neither axillary nor supraclavicular lymph nodes were palpable. Craniocaudal mammography showed a 2.3 cm well defined nodule with calcification in left breast. Clinical impression was fibroadenoma. Lumpectomy was performed. Gross examination showed a globular specimen of size 3 cm in diameter. Cut surface was well circumscribed, lobulated, and grayish white in color. Microscopy revealed well circumscribed lesion [Figure 1]. On high power view showed small lobules formed by biphasic proliferation of an inner layer of epithelial cells and a prominent peripheral layer of clear myoepithelial cells. Epithelial cells were cuboidal and monotonous with vesicular nucleus and prominent nucleolus. Both components were cytologically bland showing minimal pleomorphism and low mitotic rate.

Immunohistochemical study

Inner layer of epithelial cells show positivity for epithelial membrane antigen (EMA) [Figure 2] and cytokeratin [Figure 3].

Outer layer of myoepithelial cells show positivity for smooth muscle actin (SMA) [Figure 4] and S-100 [Figure 5].

DISCUSSION

Myoepithelial cells are normally present in the breast and may appear spindle shaped or as large ovoid cells, sometimes with a clear cytoplasm. Neoplasms of pure myoepithelial or mixed epithelial and myoepithelial origin are described in the salivary glands but are very rare in the breast. The myoepithelial lesions are divided into three different

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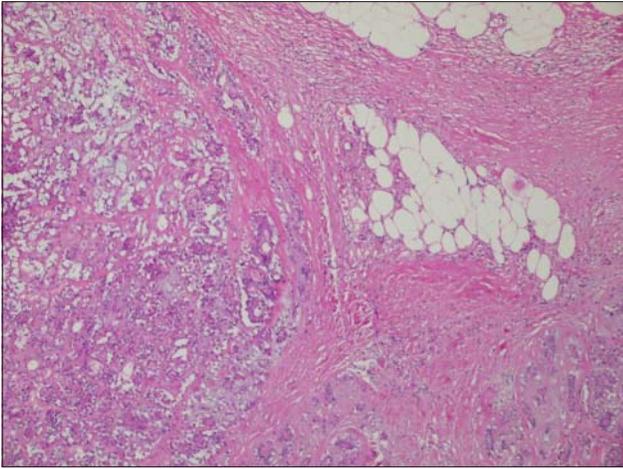


Figure 1: Microscopy revealed well circumscribed lesion

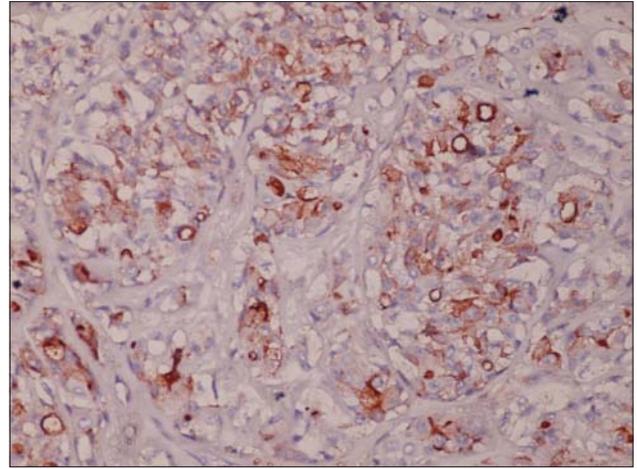


Figure 2: Immunohistochemical study: Inner layer of epithelial cells show positivity for epithelial membrane antigen

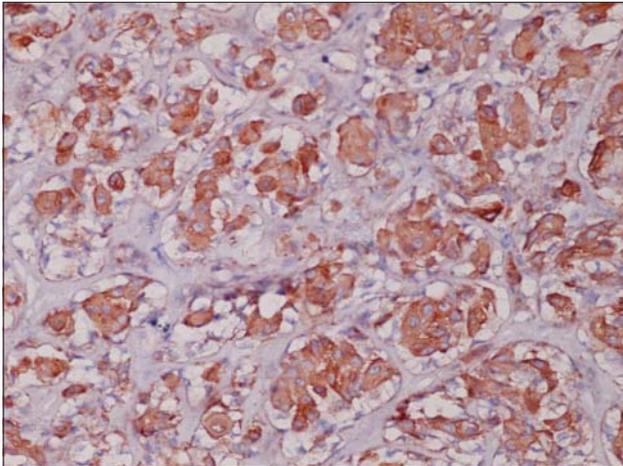


Figure 3: Immunohistochemical study: Inner layer of epithelial cells show positivity for cytokeratin

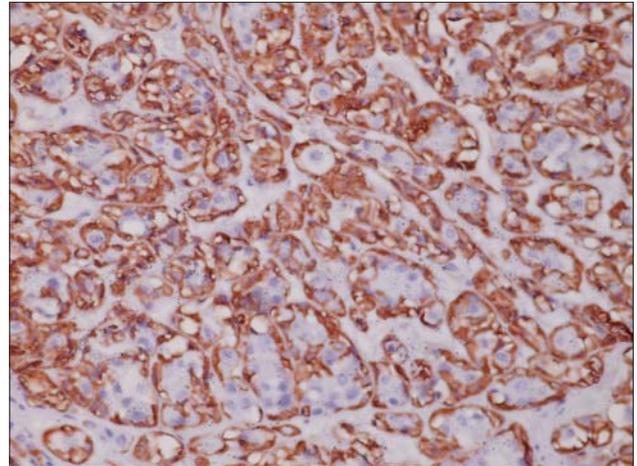


Figure 4: Outer layer of myoepithelial cells show positivity for smooth muscle actin

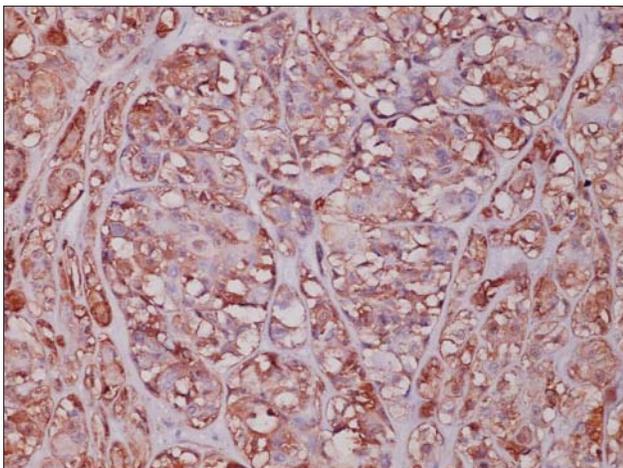


Figure 5: Outer layer of myoepithelial cells show positivity for S-100

categories (1) myoepitheliosis, (2) adenomyoepithelioma, and (3) myoepithelial carcinoma.^[2] Adenomyoepithelioma are rare lesions and only 27 cases have been described

in the literature.^[2,4-6] Most of the adenomyoepitheliomas are characterized by biphasic proliferation of an inner layer of epithelial cells and a prominent peripheral layer of myoepithelial cells.^[7] Majority of the cases of adenomyoepithelioma are reported in the fifth to sixth decade of life. The exact etiology of adenomyoepithelioma is still obscure. The adenomyoepithelioma presents as solid well-delineated rounded nodules varying from 0.3 to 7 cm, with an average size of 2.5 cm, firm, whitish yellowish nodules. Adenomyoepithelioma are classified as tubular, lobulated, or spindle subtypes.^[2] The most common pattern is tubular type with features characterized by proliferation of glandular cells and surrounding myoepithelial cells with abundant clear cytoplasm. Both the components are cytologically bland with minimal pleomorphism and low mitotic rate generally <2/10 High Power Field. Malignant changes from epithelial, myoepithelial or both components have been described.^[8] Immunohistochemistry reveal positivity for AE1/AE3 and epithelial membrane antigen

in the epithelial component, whereas the myoepithelial cells express several specific markers including muscle specific actin and myosin, calponin, P63, CD10, and S-100 protein.^[9] On Immunohistochemistry study our case shows positivity for epithelial membrane antigen (EMA) and cytokeratin (CK) in inner epithelial cells and positivity of smooth muscle actin (SMA) and S-100 in outer myoepithelial cells.

Considering the histological features and immunohistochemical findings, a diagnosis of adenomyoepithelioma of breast was made. Differential diagnosis of adenomyoepithelioma includes tubular adenoma, sclerosing adenosis, fibroadenoma, and pleomorphic adenoma. Tubular adenoma, sclerosing adenosis, and fibroadenoma have less prominent proliferative features compared with adenomyoepithelioma. Pleomorphic adenoma usually has prominent areas of chondroid and osseous differentiation. The prognosis of patients with adenomyoepithelioma of breast is usually good. Failure to achieve a free resection margin may result in local recurrence or rarely malignant transformation. Therefore it is important to make an accurate diagnosis and arrange proper management for this kind of rare breast tumor. Malignant predictors include a high mitotic rate, cytologic atypia, and infiltrative peripheral border.^[10] Further clinical and pathological investigations of breast adenomyoepithelioma may help to elucidate the true nature of this tumor.

CONCLUSION

Adenomyoepithelioma is a benign tumor of breast with potential for recurrence, metastasis, and malignant transformation. It mimicks other benign and malignant lesions of breast and hence poses diagnostic challenge.

Failure to recognize this tumor may lead to inappropriate diagnosis and management. Hence accurate diagnosis with close follow-up is mandatory.

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