

# Adenomyoepithelioma of the Breast — A Case Report

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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. Accurate diagnosis and differentiation from more aggressive tumors are important. Although the tumor bears a potential for recurrence, local excision with safe margins usually leads to a relatively benign course. We describe a 76-year-old woman who presented with a right breast lump for several months. Physical examination revealed a single well-defined mass with firm consistency and no evidence of associated axillary lymphadenopathy was found. Mammography revealed an opaque mass with linear microcalcifications and focal blurred margins. Excisional biopsy was performed and pathologic study revealed adenomyoepithelioma. (*Tzu Chi Med J* 2006; **18**:65-67)

*Key words:* adenomyoepithelioma, breast, myoepithelial cell

## INTRODUCTION

Adenomyoepithelioma is a rare benign breast tumor with characteristic dual proliferation of glandular and myoepithelial cells, typically presented by a single unilateral painless nodule. After the first fully described case report by Hamperl in 1970 [1], only a few case reports and three studies have been presented in the English literature [2-4]. Tavassoli [4] divided myoepithelial lesions into three different clinicopathologic categories: (1) myoepitheliosis, (2) adenomyoepithelioma (3) myoepithelial carcinoma. All the reported cases were female except for two male patients [5,6]. Although it is a benign neoplasm, failure to achieve an adequate resection margin may lead to recurrence [2]. Therefore, local resection with free margins is the treatment of choice for these patients. Differential diagnosis of this tumor includes sclerosing adenosis, fibroadenoma, and tubular adenoma [7]. The case described herein represents a rare case of adenomyoepithelioma arising from the breast in a postmenopausal woman with clinical presentation of a breast lump. A correct diagnosis was not

achieved until surgical excision of the tumor.

## CASE REPORT

A 76-year-old woman was referred due to a right breast lump for several months. The family history was unremarkable. Physical examination revealed a single well-defined mass in the lower inner zone of the right breast with no evidence of associated axillary lymphadenopathy. The tumor had a firm consistency. Routine laboratory test results were all within reference ranges. Breast sonography revealed a hypoechoic tumor with microcalcification (Fig. 1). Mammography showed an opaque mass with linear microcalcifications and focal blurred margin found in the right breast (Fig. 2). Excisional biopsy was performed due to the suggestion of malignancy.

Gross examination of the surgical specimen disclosed a well-delineated rounded nodule that measured 2.0 × 2.0 cm. All of the tissue was formalin fixed and processed for paraffin sections. Sections were stained with hematoxylin-eosin and immunohistochemically

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with smooth muscle actin, S-100, and cytokeratin.

On light microscopic examination, the tumor was well-demarcated and composed of biphasic proliferation of glandular epithelial cells and surrounding myoepithelial cells. Proliferative epithelial cells displayed tubular growth patterns (Fig. 3). Prominent myoepithelial cells with clear cytoplasm around epithelial cells were noted (Fig. 4a). Both epithelial and myoepithelial cells were blended looking without cytological atypia. Less than 1 mitosis /10 high power field (HPF) was noted in the mitotic activity. Central infarction was presented in the focal areas. Immunohistochemical staining of myoepithelial cells was strongly positive for smooth muscle

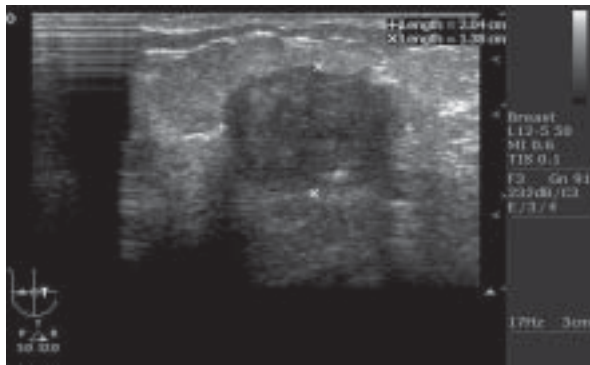


Fig.1. Breast sonography reveals a hypochoic tumor with microcalcification.

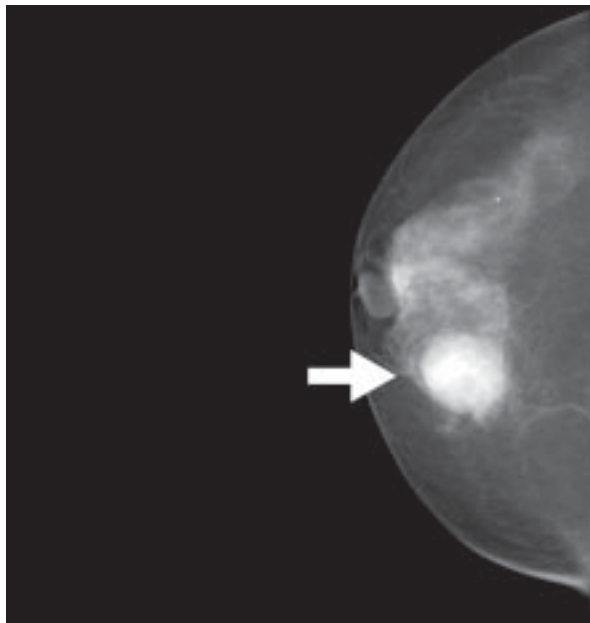


Fig.2. Mammography shows an opaque mass with linear microcalcifications and focal blurred margin (arrow).

actin (Fig. 4b), weak positive for S-100 (Fig. 4c), and negative for cytokeratin (Fig. 4d).

The results further support the existence of myoepithelial cells around the glandular cells. The final diagnosis was adenomyoepithelioma. The resection margins were free. Post operation course was smooth and uneventful. No additional treatment was performed.

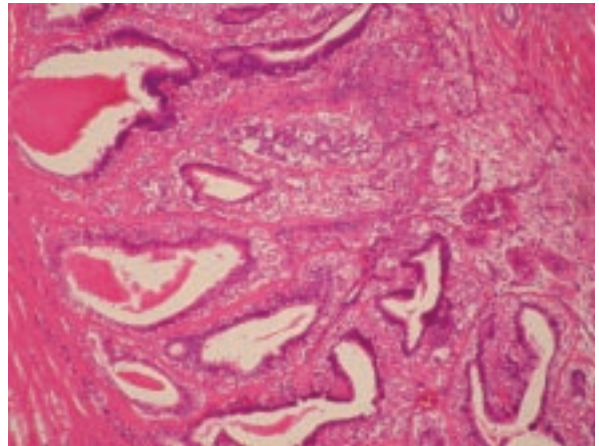


Fig.3. Medium power photograph shows tubular architecture of tumor with biphasic proliferation of glandular epithelial cells and surrounding myoepithelial cells (H&E  $\times$  100).

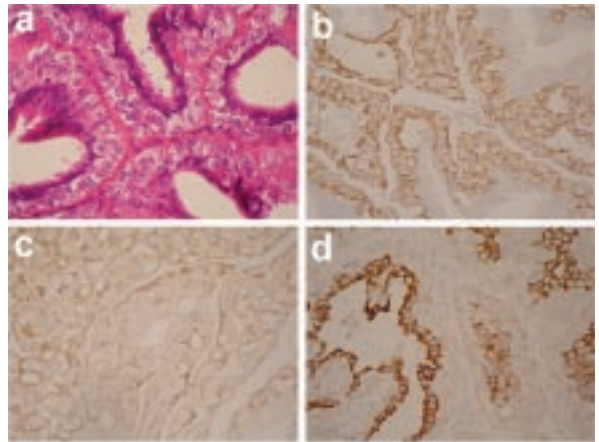


Fig. 4. High power micrograph reveals inner layer of epithelial cells and outer layer of prominent myoepithelial cells with clear cytoplasm. Both epithelial and myoepithelial cells are blended looking without cytological atypia (a). Immunohistochemical staining of myoepithelial cells was strongly positive for smooth muscle actin (b), weak positive for S-100 (c), and negative for cytokeratin (d) (Original magnification  $\times$  400).

## DISCUSSION

Breast myoepithelial lesion is a rare tumor. Three forms have been described by Tavassoli, including myoepitheliosis, adenomyoepithelioma, and myoepithelial carcinoma. Most adenomyoepitheliomas are characterized by biphasic proliferation of an inner layer of epithelial cells and a prominent peripheral layer of myoepithelial cells.

Adenomyoepitheliomas have been further classified as tubular, lobulated, or spindled subtype [4]. The most common pattern is the tubular type with features characterized by proliferation of glandular cells and surrounding myoepithelial cells of abundant clear cytoplasm. Malignant changes from epithelial, myoepithelial, or both components have been described [8].

The exact etiology of breast adenomyoepithelioma is still obscure. All cases have been sporadic and no familial aggregation has been observed. Kiaer et al [9] 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 [7] proposed that adenomyoepithelioma was derived from a myoepithelial long standing underlying breast disease, such as adenosis and fibroadenoma.

Accurate diagnosis can be difficult based solely on radiological observation; therefore, histological examination results are required to make the precise diagnosis. 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. Proliferative myoepithelial cells with clear cytoplasm may mimic malignancy in an intraoperative frozen section; therefore, it is difficult to make an accurate diagnosis.

Immunostaining for smooth muscle actin and calponin can support the myoepithelial differentiation and no reactivity is noted in epithelial cells. S-100 protein have been used as a marker of myoepithelial cells in the breast, however, its immunoreactivity is not consistent and may be expressed in both myoepithelial cells and epithelial cells. Therefore, it is not a reliable marker

to myoepithelial cells in adenomyoepitheliomas [10].

The prognosis of patients with adenomyoepitheliomas of the 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 pathologic diagnosis and arrange proper management for this kind of rare breast tumor. Three malignancy predictors including high mitotic rate, cytologic atypia, and infiltrative peripheral border have been proposed by Loose et al [2] in their series of case studies. Further clinical and pathological investigations of breast adenomyoepitheliomas may help to elucidate the true nature of this rare tumor.

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