

Malignant adenomyoepithelioma of the breast

A case report

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Abstract

Rationale: Adenomyoepithelioma (AME) is a rare biphasic tumor consisting of epithelial and Myoepithelial cell. Most of the AME is benign, and only a few will progress to malignancy, Here, we report a case of low-grade malignant adenomyoepithelioma, and review the related literature, in a bid to investigate its clinical and pathological features and thus, enhance our understanding of this tumor.

Patient concerns: A 64-year-old woman visited our hospital with a 1-year history of a painless mass in her left breast. Physical examination revealed a palpable painless mass, measuring approximately 4.5 cm, in the left breast.

Diagnosis: Histological examination confirmed the diagnosis of malignant adenomyoepithelioma

Interventions: The patient underwent local excision of the mass, with frozen section analysis revealing ductal carcinoma in situ. Mastectomy and sentinel lymph node biopsy were then performed.

Outcomes: We conducted a one-year follow-up, and relapse was not observed.

Lessons: Treatment of AME remains controversial owing to the lack of high volume data and absence of prospective studies. Simple mastectomy is an acceptable treatment of this tumor.

Abbreviations: AME = adenomyoepithelioma, DCIS = ductal carcinoma in situ, MAME = malignant adenomyoepithelioma, SLNB = sentinel lymph node biopsy.

Keywords: adenomyoepithelioma, breast, malignant, myoepithelium

1. Introduction

Adenomyoepithelioma (AME) was first reported by Hamperl^[1] in 1970. In 2012, the World Health Organization (WHO) classified malignant adenomyoepithelioma (MAME) as an “adenomyoepitheliomatous variant,” including carcinomas of epithelial, myogenic, and epithelial-myogenic origin.^[2] Because of the heterogeneity of AME, the diagnosis is challenging and a range of histological patterns can be observed. These lesions are particularly common in patients undergoing core needle biopsy. While most of AME is benign, there have been reports of distant metastasis. All the reported malignant AMEs with metastases

have shown significant cytological atypia and brisk mitotic rates. Therefore, a complete resection of the tissue is necessary for pathological diagnosis.

2. Case report

A 64-year-old woman visited our hospital with a 1-year history of a painless mass in her left breast. The patient reported rapid mass growth over the previous 6 months. Physical examination detected a palpable painless mass, measuring approximately 4.5 cm, in the left breast. Ultrasonography revealed a hypoechoic lump measuring 39.4 mm × 22.6 mm at 6 o'clock in the left breast with poorly defined borders. Mammography revealed a nodule of approximately 40 mm × 40 mm in size with poorly defined borders and no calcification visible in the left breast (Fig. 1). The patient underwent local excision of the mass, and frozen section analysis revealed ductal carcinoma in situ. Subsequently, mastectomy and sentinel lymph node biopsy were performed. Definitive histological examination diagnosed low-grade MAME, with local micro-invasion (0.1 cm), malignant degeneration of the myoepithelium in the tumor, and easily visible mitotic figures (local foci >10/10 HPF). Further, intraoperative examination (left sentinel lymph node) did not detect any cancer metastasis by serial sections (0/3). Immunohistochemical results were as follows: ER (–), PR (–), Her-2 (0), S-100 (+), Calponin (+), CK5/6 (+), and P63 (+) (Fig. 2A–C). No further treatment was administered. We conducted a 1-year follow-up, and the patient showed no signs of relapse.

3. Discussion

AME is a rare tumor composed of myoepithelial and epithelial cells, whereas MAME is even more uncommon. There are less than 20 reports on MAME and most literature reports on female cases.^[3] The onset age of MAME is between 26 and 81 years, and

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The patient has provided informed consent for publication of the case.

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

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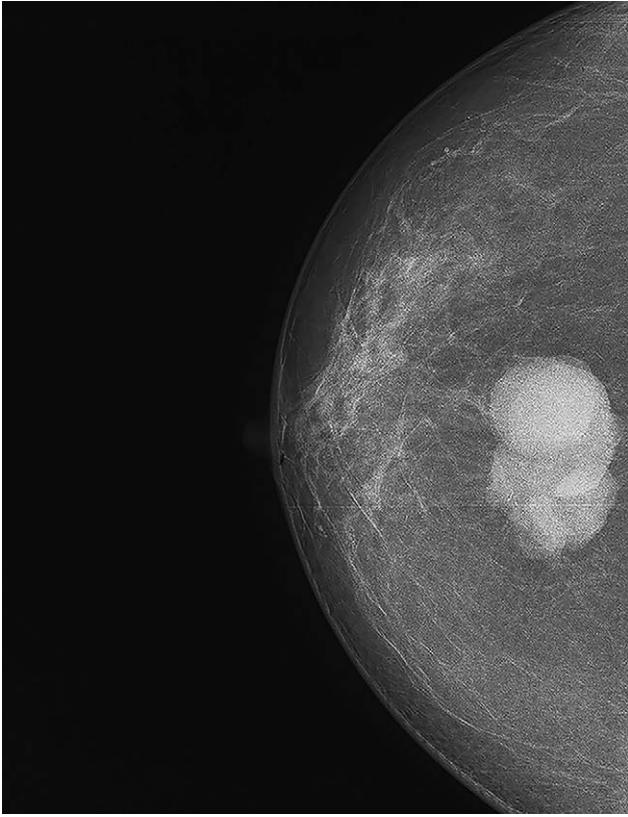


Figure 1. Mammography scan showing a 40 × 40 mm nodule with unclear margins and no calcification observed within the mass.

the incidence increases with age.^[4] Painless breast lumps are common in clinical practice, with no lymph node involvement. In mammography, tumors tend to appear well defined, dense, and calcified. Ultrasonography often reveals well-defined, homogeneous solid nodules or cystic solid lesions.^[5,6]

Although AMEs are commonly benign, they must be considered malignant when they present with cytological atypia, an infiltrative growth pattern, and a high mitotic index (local >3/10 HPF).^[7] The cellular composition of MAMEs was confirmed by immunolabeling, with epithelial elements typically showing positive staining for low molecular weight cytokeratin, including CAM 5.2, CK7, and EMA. In contrast, positive staining for high molecular weight cytokeratin, such as CK5/6, was characteristic of myoepithelial components. In addition, myoepithelial markers also included S100, SMA, and p63.^[3,5,8,9]

Treatment of AME remains controversial owing to the lack of high volume data and absence of prospective studies. Based on the available evidence, benign AMEs may only require complete surgical resection, whereas malignant tumors involving the breast and/or regional lymphatics could respond to radiotherapy.^[7,9,10] Whether chemotherapy is effective for MAME is unclear owing to the paucity of literature. However, rare instances of metastatic disease would presumably require chemotherapy, and studies of targeted anti-cancer therapy have also been presented in case reports.^[11] Routine use of sentinel lymph node biopsy may not be necessary compared to invasive breast cancer.^[12] Therefore, there is currently no standardized treatment for MAME.

It is challenging for us to assess the prognosis of MAME owing to the low volume of data. Nevertheless, a retrospective study of 110 patients evaluated the prognosis of MAME. The study indicated that the expected 5-year OS for all patients of MAME was 74.4%.^[12]

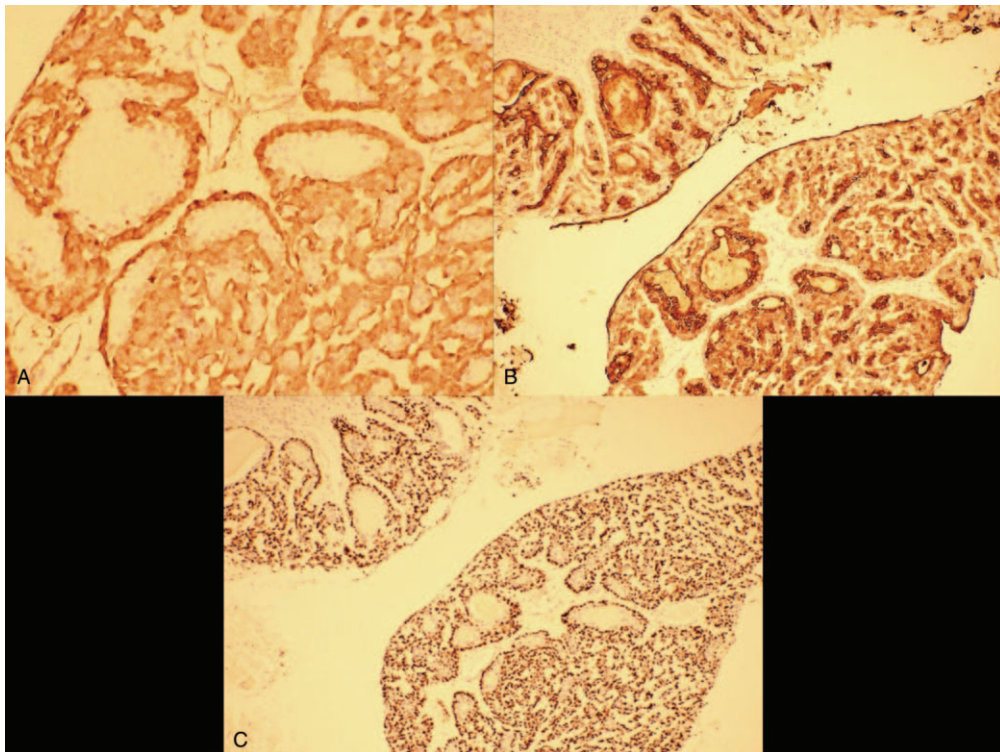


Figure 2. A, Immunohistochemical staining of the resected tissue. Positive calponin staining in myoepithelial cells (200 × magnification). B, Immunohistochemical staining of the resected tissue. Myoepithelial cells were very strongly positive for cytokeratin-5 (CK5) (100 × magnification). C, Immunohistochemical staining of the resected tissue. Nuclear P63 staining in the myoepithelial component confirmed differentiation of the myoepithelium (100 × magnification).

4. Conclusion

In conclusion, treatment of AME remains controversial owing to the lack of high volume data and absence of prospective studies. Simple mastectomy is increasingly accepted by clinicians. In addition, MAME should be distinguished from other breast diseases, such as myoepithelial carcinoma, mammary gland sarcoma, invasive breast cancer, tubular adenoma, and intraductal papilloma, among others.^[4,9] MAME has the possibility of distant metastasis and recurrence. Therefore, it is imperative for clinical pathologists to monitor the progress of MAME.

Author contributions

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