

Adenomyoepithelioma with malignant transformation and repeated local recurrence: A case report

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Case report

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Abstract

Background Adenomyoepithelioma (AME) is a rare type of benign breast tumor. Many AMEs have demonstrated benign behavior, but very few cases of malignant AME have been reported.

Case presentation A 53-year-old woman visited our hospital with a 16-mm, palpable mass in the right breast. Core needle biopsy was performed. The pathological diagnosis was AME. Partial mastectomy was performed. Two years later, local recurrence developed, and the patient again underwent partial mastectomy. The pathology showed malignant AME, and the margin was negative. Eight months later, local recurrence developed again in the same location, and total mastectomy was performed. The pathological diagnosis was malignant AME. The patient has remained recurrence-free without adjuvant therapy.

Conclusions A very rare case of AME with repeated local recurrences and malignant transformation was reported.

Background

Adenomyoepithelioma (AME) is a very rare type of benign tumor of the breast. Many AMEs have demonstrated benign behavior and are often cured with excision with negative margins, but some have exhibited malignant transformation of the myoepithelium, glandular epithelium, or both [1–6]. In this report, the case of a patient with repeated local recurrences and malignant transformation is presented.

Case Presentation

The course to the first surgery

A 53-year-old woman visited our hospital with a palpable mass in the right breast. The mammogram showed an oval, smooth, and well-defined iso-dense mass in the upper right breast (Fig. 1a). Diagnostic ultrasonography showed a well-defined mass with cystic change, measuring up to 16 mm, at the 2 o'clock position of the right breast (Fig. 1b). Core needle biopsy (CNB) was performed, and the lump was diagnosed as AME. Partial mastectomy was performed. Pathological findings showed benign AME. The gross findings of the tumor were as follows. The tumor was a 20-mm cystic lesion, and the cystic wall had nodules or irregular thickening (Fig. 1c). The pathological findings were as follows. Round or spindle-shaped myoepithelium was proliferating in and around the gland ducts. High mitotic counts were prominent in the myoepithelial component. The border of the tumor was relatively clear (Fig. 1d). Excisional margins were negative. No surrounding invasion or severe cellular atypia was observed, and there was no clear evidence of malignancy. No adjuvant therapy was given.

The course of the first local recurrence

Two years later, the woman visited our hospital with a palpable mass in the same location. Ultrasound showed a well-defined, oval, low-iso echoic mass in the slightly caudal side of the area of postoperative

change (Fig. 2a). The components were mainly cysts, but with a thick wall (substantial part) at the margin. The mass was the same as that at the initial surgery and was suspected to be a recurrence. Vacuum-assisted biopsy was performed, and the diagnosis was recurrent AME. Partial mastectomy was again performed. The gross findings of the tumor were as follows. The tumor was a well-defined, borderline lesion with a thick white coat. Inside the tumor were green to yellowish jelly-like substances that filled it, 45*30*20 mm³ in size. The pathological findings were as follows (Fig. 2b-e). Epithelium with squamous metaplasia grew into the lumen side, and dense growth of spindle-shaped myoepithelium was found on the side of the membrane. They all spread underneath the existing glandular epithelium. Tumor cells showed prominent nuclear atypia and high mitotic counts. Although there was no proliferation of glandular cells, it was diagnosed as recurrent AME with a proliferation pattern similar to that at the initial surgery. In addition, a diagnosis of malignant transformation of AME was made due to the observation of nuclear atypia, a high mitotic count, and invasive growth. No adjuvant therapy was given.

The course of the second local recurrence

Eight months later, the patient returned to the hospital because of a palpable mass in the same area. Ultrasound showed a mass of up to 27 mm that was congregated on the right side of the previous surgical wound as a series of masses (Fig. 3a). The largest of the masses showed no echogenicity, with thick walls and internal septal/cystic degeneration. The findings were similar to those of the previously diagnosed AME. Enhanced breast magnetic resonance imaging (MRI) showed multiple masses up to 25 mm in size in the right inner-upper area (Fig. 3b). They were cystic with thick walls, similar to the previous one and diagnosed as recurrent. Two 7-mm nodules were also found within the pectoralis major muscle. Total mastectomy was performed. The tumor was 75*24 mm² in size in the inner-upper area. The center of the lesion was hollow and cystic, and the lesion contained a jelly-like substance (Fig. 3c). The pathological findings were the same as the previous recurrence (Fig. 3c-e). Epithelium with squamous metaplasia and spindle-shaped myoepithelium arranged in a complex or bundled pattern were present, and there was continuity between the two types of epithelium; the epithelial cells showed prominent nuclear atypia and high mitotic counts, especially prominent in the squamous epithelial component. There were also findings of invasion of partially resected pectoralis major muscle and extra-mammary adipose tissue. The diagnosis of recurrent AME with malignant transformation and squamous differentiation was made.

No adjuvant therapy was given. The patient has been disease-free for one year post-treatment.

Discussion And Conclusions

AME of the breast is a rare disorder characterized by the simultaneous proliferation of glandular epithelium and myoepithelium. Characteristically, AMEs tend to exhibit benign clinical behavior, although malignant transformation has been reported in a small number of cases. This transformation is indicated by features such as prominent cytological atypia, an elevated mitotic index, necrosis, and metastasis [7, 8]. Because of the biphasic nature of the tumor, carcinomas may arise from the glandular epithelium, myoepithelium, or both [9–11].

In the present case, the proliferation of myoepithelium was initially more prominent than that of glandular epithelium in the initial surgical specimen. From the time of recurrence, the glandular epithelium was eradicated, and the tumor was mainly myoepithelium and epithelium with squamous metaplasia. Malignant transformation at the time of local recurrence has been reported, but the number of cases is not large [12]. Thus, the proportion of myoepithelium and glandular epithelium proliferation differs from case to case. A case with different proportions between the metastatic site and the primary tumor has also been reported [13].

Mammography findings are a mass without calcification, consistent with previous reports [12]. There were no specific findings on mammography that suggest a diagnosis of AME in previous reports. Macroscopically, AMEs are typically well-circumscribed, unencapsulated, solid tumors that may show focal cystic change. Although there are many reports that do not mention ultrasound examination results, even when they are reported, they are generally hypoechoic, and it seems to be rare for a mass to be a clearly intracystic lesion, as in the present case [13, 14]. Enhanced breast MRI was performed for the first time before the third surgery. In this case, it was useful in diagnosing the spread of the tumor, and it contributed to determining the indication for total mastectomy.

There are no clear treatment guidelines for AME. Partial mastectomy is often performed, but total mastectomy is sometimes performed for large or suspected malignant tumors [6]. Axillary lymph node metastases are rare, and only a few cases of axillary dissection have been reported [15–17]. There may be recurrences even after complete resection with partial mastectomy, as in the present case, and they should be carefully monitored. Ito et al. concluded that, based on previous reports, total mastectomy is often performed for tumors larger than 3 cm [6]. It will become clearer as more cases are reported which patients should undergo total mastectomy, and whether reconstruction is possible after resection. No adjuvant therapy is recommended. Therefore, it was not performed in the present case.

List Of Abbreviations

AME: adenomyoepithelioma, CNB: core needle biopsy, MMG: mammography, US: ultrasonography, MRI: magnetic resonance imaging

Declarations

Ethics approval and consent to participate

The study was approved by the Ethics Committee of Tokyo Medical and Dental University Hospital (Tokyo, Japan). Written, informed consent was obtained from the patient.

Consent for publication

Written, informed consent for publication was obtained from the patient.

Availability of data and materials

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

Competing interests

The authors declare that they have no competing interests.

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No funding was received.

Authors' contributions

GO performed surgery, and wrote the manuscript. TN and HS performed surgery and collected data. MM and TF were responsible for diagnostic imaging and ultrasound-guided needle biopsy. IO was in charge of the pathology. HU designed the present study and wrote the manuscript. The final version was read and approved by all authors.

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Figures

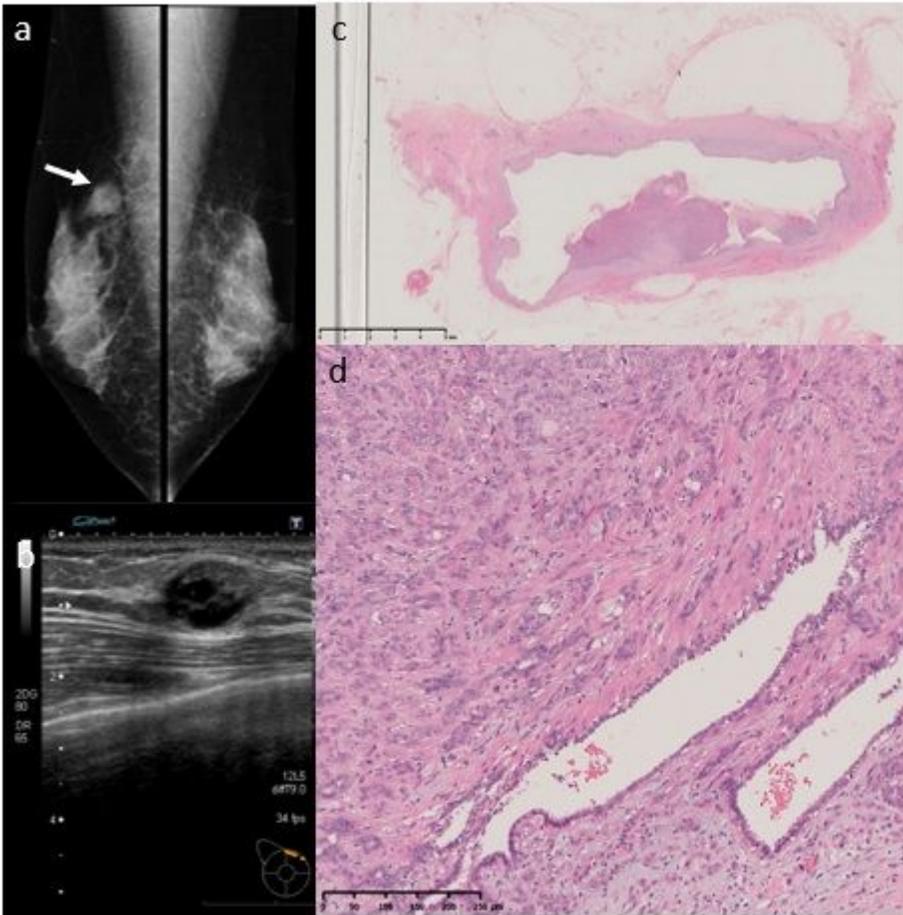


Figure 1

Imaging and pathology findings at the time of initial surgery a: The mammogram shows an oval, smooth, well-defined, iso-dense mass in the upper right breast (white arrow). b: Ultrasonography shows a well-defined mass with cystic change, measuring up to 16 mm, at the 2 o'clock position of the right breast. c: (x65 magnification) The tumor is a cystic lesion, and the cystic wall has nodules or irregular thickening. d: (x100 magnification) Round or spindle-shaped myoepithelium is proliferating in and around the gland ducts. High mitotic counts are prominent in the myoepithelial component.

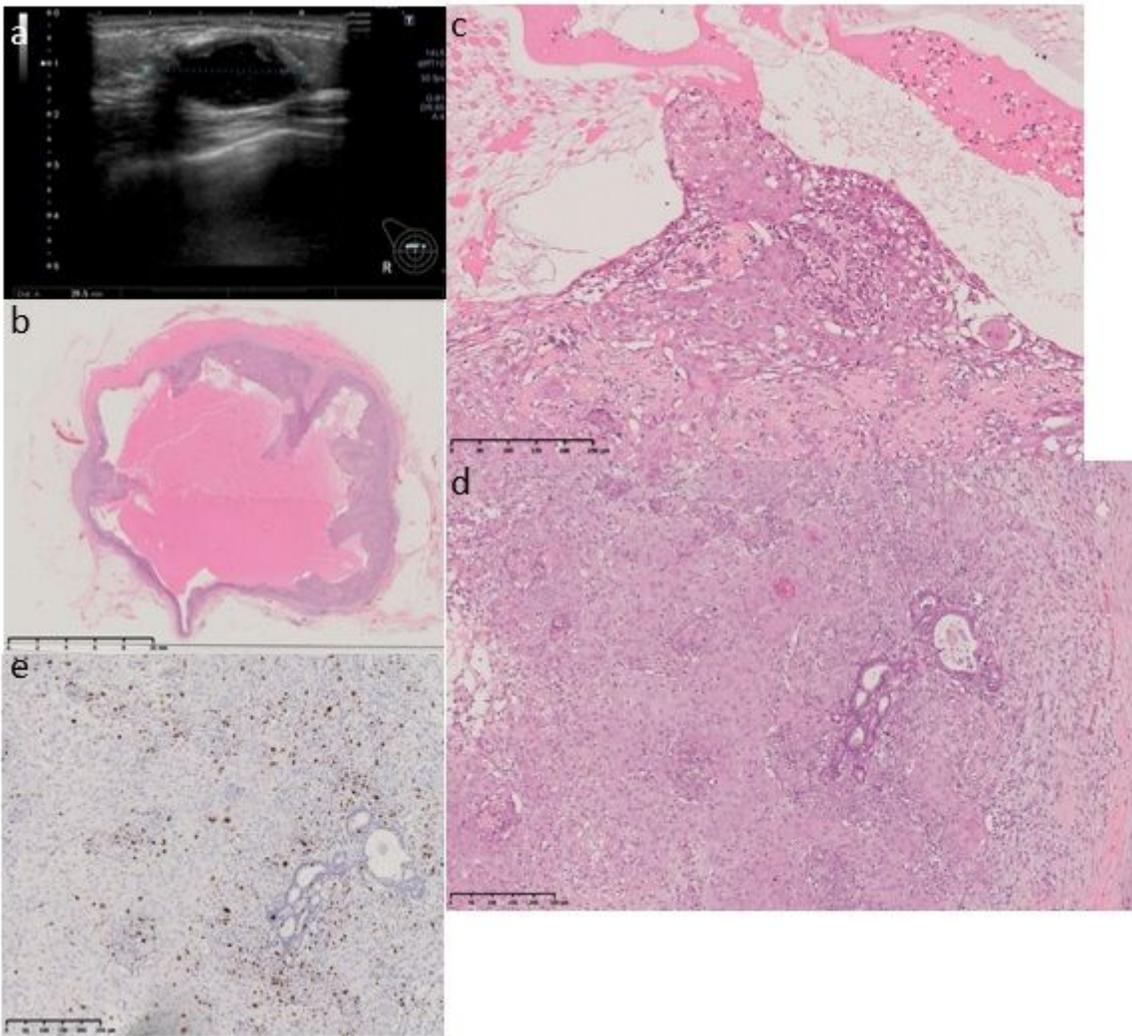


Figure 2

Imaging and pathology findings at first recurrence a: Ultrasonography shows a well-defined, oval, low-isoechoic mass. b: (x32 magnification) The tumor is a cystic lesion, and the cystic wall has nodules or irregular thickening. The cysts are full of mucus. c: (x100 magnification, lumen side) and d: (x100, membrane side) Epithelium with squamous metaplasia grows into the lumen side. Dense growth of spindle-shaped myoepithelium is found on the side of the membrane. They all spread underneath the existing glandular epithelium. Tumor cells show prominent nuclear atypia and high mitotic counts. e: (x100 magnification, Ki67) The Ki 67 hot spot is 57.1%.

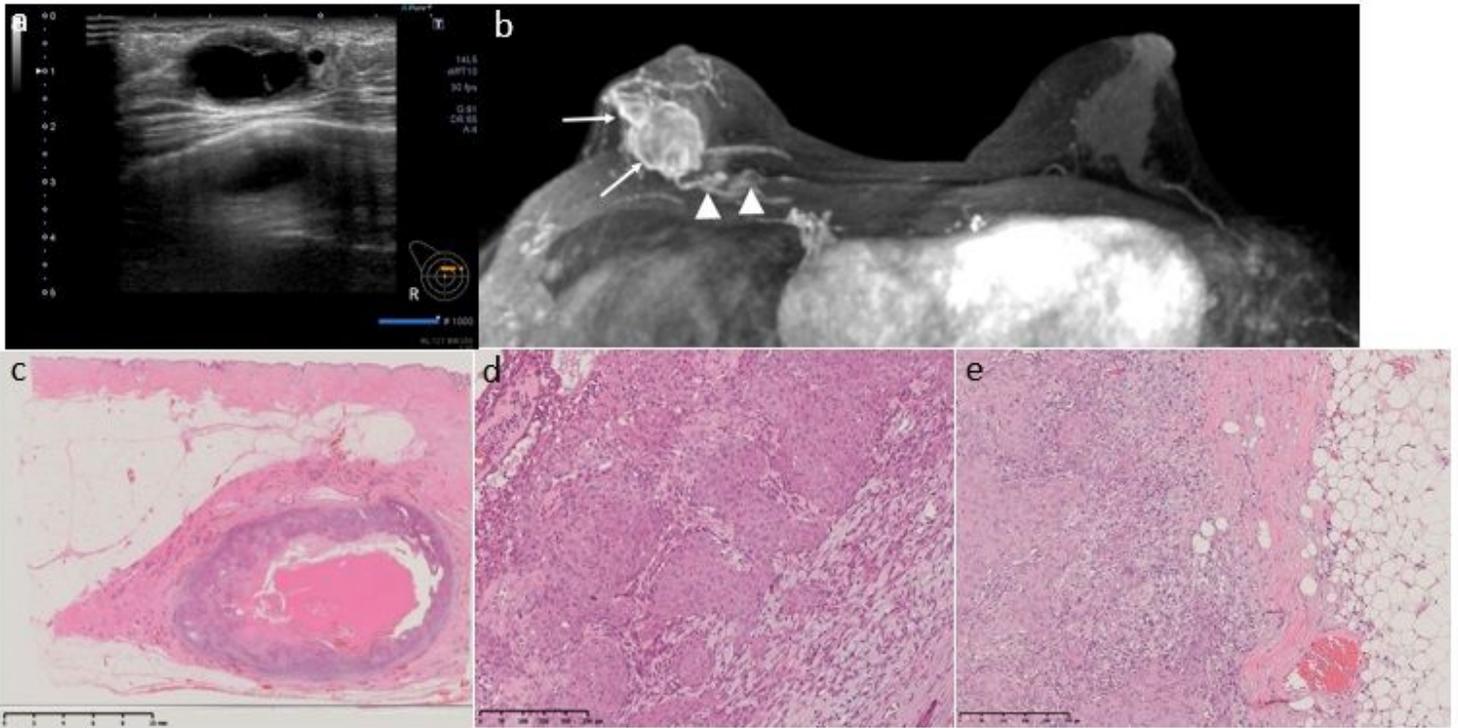


Figure 3

Imaging and pathology findings at the 2nd recurrence a: Ultrasonography shows a mass of up to 27 mm that is congregated on the right side of the previous surgical wound as a series of masses. b: Enhanced breast MRI shows multiple masses up to 25 mm in the right inner-upper area (white arrow). They are cystic with thick walls, similar to the previous one and diagnosed as recurrent. Two 7-mm nodules are also found within the pectoralis major muscle (arrow head). c: (x3 magnification) The tumor is a cystic lesion, and the cystic wall has nodules or irregular thickening. Mucus fills the cyst. d: (x100 magnification) Biphasic proliferation of both inner epithelium with squamous metaplasia and outer spindle-shaped myoepithelium is seen. Both cell types show prominent nuclear atypia and high mitotic counts, especially prominent in the epithelial component. The histological findings are the same as at the previous surgery. e: Findings of tumor cell invasion into extra-mammary adipose tissue.