Coexistent adenomyoepithelioma and invasive ductal carcinoma of the breast: presentation as separate tumors

Abstract Adenomyoepitheliomas are rare breast tumors. We report an unusual case of adenomyoepithelioma associated with invasive ductal carcinoma here. Histologically, the lesion consisted of two separate tumors. One nodule corresponded to invasive ductal carcinoma consisting of tubular and trabecular arrangements of columnar or cuboidal neoplastic cells. The other tumor corresponded to adenomyoepithelioma consisting of an inner layer of neoplastic cells with basophilic cytoplasm and the outer layer of neoplastic cells with clear cytoplasm. Immunohistochemically, some myofibroblasts were observed in the stroma of both adenomyoepithelioma and invasive ductal carcinoma, but no CD34-positive stromal cells were seen in the stroma of either lesion. The stromal reaction of adenomyoepithelioma resembles that of intraductal papilloma in the previous study. To the best of our knowledge, this is the first case of coexistent adenomyoepithelioma and invasive ductal carcinoma of the breast that were discovered as separate nodules. Clinicians and pathologists should be aware of such an association because they need to distinguish such a case from malignant neoplasms arising in adenomyoepithelioma. Additionally, our preliminary report suggests that the stromal response of adenomyoepithelioma may resemble that of intraductal papilloma.

Key words Adenomyoepithelioma · Invasive ductal carcinoma · Breast

Introduction Adenomyoepitheliomas are rare breast tumors, and various types of malignant neoplasms arising in adenomyoepithelioma or a malignant counterpart of adenomyoepithelioma have been reported to date. However, there have been no descriptions of coexistent carcinoma and adenomyoepithelioma as separate tumors in the same breast of the same patient. We report here a case with invasive ductal carcinoma of the breast in the adjacent area of adenomyoepithelioma and discuss the clinical and pathological significance of such a case. Additionally, we studied the stromal response of adenomyoepithelioma and invasive ductal carcinoma in the present case.

Case report A 66-year-old Japanese woman noticed a breast tumor and consulted the surgeon at our hospital. Ultrasound sonography and mammography disclosed two different tumors (Fig. 1). Aspiration cytology extracted from one of the two breast tumors showed suspected malignancy, and subsequent core needle biopsy of the two breast tumors disclosed the invasive carcinoma and a benign neoplasm. Subsequently, the patient underwent partial mastectomy. Her clinical course was uneventful at 5 months after the operation.

Breast tissue obtained by mastectomy was fixed in 10% formalin and embedded in paraffin. Sections 3 μm thick were stained with hematoxylin and eosin. Additionally, immunohistochemical stain was performed using a Histofine Simple stain-PO (multi) kit (Nichirei, Tokyo, Japan). Antibodies against p63 (A4A, dilution 1:200; Lab Vision,
Fremont, CA, USA), alpha-smooth muscle actin (ASMA) (1A4, dilution 1:50; DAKO, Glostrup, Denmark), h-caldesmon (h-CD, dilution 1:50; DAKO, Santa Barbara, CA, USA), CD31 (JC/70A, dilution 1:20, DAKO, Glostrup, Denmark), and CD34 (MY10, dilution 1:20, Becton-Dickinson, San Jose, CA, USA) were employed in the present study. We considered ASMA-positive and h-caldesmon-negative stromal cells as myofibroblasts, and CD34-positive and CD31-negative stromal cells as CD34-positive stromal cells, namely, fibroblasts, as shown in the previous study.15–17

**Pathological findings**

**Macroscopic findings**

The cut surface of the tumor disclosed a tan-brown and well-demarcated tumor measuring 0.8 cm in maximum diameter in the area adjacent to a whitish tumor measuring 3.5 cm in maximum diameter and showing an irregular margin (Fig. 2).

**Microscopic findings**

Histologically, the two lesions were recognized as separate tumors (Fig. 3a). The whitish tumor consisted of tubular and trabecular arrangements of columnar or cuboidal neoplastic cells, resulting in the diagnosis of invasive ductal carcinoma (Fig. 3b). The tan-brown tumor was composed of two layers of neoplastic cells. The inner layer formed a tubular pattern, with columnar cells with basophilic cytoplasm and neoplastic cells with clear cytoplasm regularly proliferated along the outer layer of tubular cells (Fig. 3c). These histological features corresponded to the diagnosis of adenomyoepithelioma.

**Immunohistochemical findings**

In the adenomyoepithelioma area, p63 (Fig. 4a) and ASMA were expressed in neoplastic cells of the outer layer, namely, myoepithelial cells (Fig. 4b). In the stroma of adenomyoepithelioma, some myofibroblasts, which were positive for ASMA and negative for h-caldesmon, were observed (Fig. 4b), but no CD34-positive stromal cells were seen (Fig. 4c). In contrast, no myoepithelial cells positive for p63 and ASMA were detected in the invasive ductal carcinoma area. In the stroma of invasive ductal carcinoma, some myofibroblasts positive for ASMA and negative for h-caldesmon were identified (Fig. 4d), but no CD34-positive stromal cells were observed (Fig. 4e).

**Discussion**

Tavassoli et al. classified myoepithelial lesions into three categories, namely, myoepitheliosis, adenomyoepithelioma, and myoepithelial carcinoma in 1991.2 Myoepithelial lesions are divided into four major categories, namely, myoepitheliosis, adenomyoepithelial adenosis, adenomyoepithelioma, and malignant myoepithelioma (myoepithelial carcinoma) according to the recent WHO classification.18 Malignant neoplasms arising in adenomyoepithelioma or the malignant counterpart of adenomyoepithelioma have been described in many patterns1–14 and have been subclassified as myoepithelial carcinoma, epithelial carcinoma, malignant epithelial and myoepithelial components, sarcoma, and carcinosarcoma, according to the recent WHO classification.18 However, to the best of our knowledge, there have been no reports of cases of coexistent invasive ductal carci-