CASE REPORT

Ductal carcinoma in situ of the breast arising in encapsulated mammary hamartoma; A case report

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Abstract: Mammary hamartoma is benign lesion and relatively rare. 17 cases of breast cancer associated with a hamartoma had been previously documented in the literature. We describe herein a case of noninvasive ductal carcinoma of the breast arising in hamartoma in a woman of 60's. The discordance of images of the mass between mammogram and ultrasonogram can lead us to detect the carcinoma within the hamartoma in our case. J. Med. Invest. 67 : 368-371, August, 2020

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INTRODUCTION

Mammary hamartoma is commonly shown an encapsulated mass containing fat and parenchymal densities in mammogram or ultrasonogram. Hamartoma is a distinct subtype of benign tumor, forming circumscribed or lobulated mass consisting of a mixture of fat tissue and normal parenchyma with fibrous stroma. Hamartomas are essentially disorganized overgrowth of normal mammary tissue, and they are named fibroadenolipoma or adenolipofibroma in other word. Stromal element of hamartoma uncommonly shows myoid change or muscle differentiation (1). Coexistence of breast carcinoma with a mammary hamartoma is extremely rare. Cases with hamartoma associated carcinoma had been reported in the literature (2-6). We describe a case of hamartoma from which ductal carcinoma in situ had arisen. In our case, ductal carcinoma in situ had developed within hamartoma and had limited to the region of hamartoma. The discordance between mammogram or ultrasonogram and CT image of the lesion had led us to do core needle biopsy of the lesion, and we were finally able to detect breast cancer in hamartoma. The report emphasizes the importance of adequate imaging analyses or tissue sampling of mammary hamartoma.

CASE REPORT

A woman of 60's had jointed to mass screening for breast cancer every other year. She is healthy except undergoing medication for hypertension. Her family history of breast pathology or hereditary disease is absent. As she was pointed out an abnormal shadow in her left mammography, she visited to the outpatient clinic for further examination. Ultrasonography and mammography were routinely done. Mammogram revealed an irregular and lobulated dense shadow in the upper-outer quadrant of left breast (Fig. 1a). A physical finding revealed a firm, ill-defined, oval tumor with a smooth surface measuring 3 × 2 cm at the upper-outer quadrant of the left breast and no swelling of axillary lymph nodes. As the images of the lesion are quiet different between ultrasonogram and mammogram, the patient was examined MRI.

A T2-weighted MRI image revealed no emphasized mass. A T1-weighted dynamic MRI revealed irregular and lobular mass, measuring 15 mm and being shaped like mammographic image (Fig. 1c). Dynamic curve of MRI imaging showed steep and constant pattern. From MRI image the lesion was thought to be malignancy, such as scirrhous type of ductal carcinoma or noninvasive carcinoma.

Core needle biopsy using ultrasonography was performed. Two specimens were collected. Microscopically, cancer cells which were medium-sized to large nuclei filled many ducts and proliferated in flat, solid and comedo pattern. Diagnosis of the lesion was ductal carcinoma in situ of the breast. Preoperative CT study showed the lesion in her left breast, be emphasized lobular shadow within an oval mass (Fig. 1d). It

Fig 1a. Mammogram revealed an irregular and lobulated dense shadow (arrow) in the upper-outer quadrant of left breast. Fig 1b. Ultrasonogram revealed an oval tumor with heterogeneous internal echoes, representing the hamartoma. A physical finding revealed a firm, ill-defined, oval tumor with a smooth surface measuring 3 × 2 cm at the upper-outer quadrant of the left breast and no swelling of axillary lymph nodes. As the images of the lesion are quiet different between ultrasonogram and mammogram, the patient was examined MRI.

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The patient underwent a partial glandectomy and also sentinel nodes biopsy. No metastasis to a sentinel lymph node was proven by OSNA (One-step nucleic acid amplification) assay for intra-operative detection of lymph node metastasis. The lesion was resected with surrounding normal tissue. The specimen of the breast was fixed in buffered formalin, and it cut into eleven slices having 6 mm width. In the 7th slice, capsuled yellowish mass in which grey and slightly firm area was observed (Fig. 3). Postoperatively, the patient has been doing well for several months.

Pathology

Microscopically, Hamartoma composed of mature fat tissue with scattered mammary ducts and lobules with clear encapsulation by delicate fibrous tissue included ductal carcinoma in situ inside (Fig. 4a). The cancer cells have high grade nuclei with central necrosis (Fig. 4b). The spreading of cancer cell was 13 × 4 mm, and hamartoma was 22 × 7 mm in size.
**Immunohistochemistry**

Immunohistochemically, the cancer cells were both negative for estrogen and progesterone receptors, and human epidermal growth factor receptor type 2 (HER2/neu) protein expression was also negative. Some parts of cancer cells show immunoreactivity for epidermal growth factor receptor (EGFR) with cell membrane staining pattern, and they were negative for CK5/6. The carcinoma was considered as basal cell type carcinoma.

**DISCUSSION**

The first description of mammary hamartoma had been written by Hogeman and Ostberg in their report in 1968 (7). Mammary hamartoma is a circumscribed lesion composed of disorganized overgrowth of normal breast tissues, and showing various composition of ductal and lobular epithelium and stromal elements. Hamartoma is often named as fibroadenolipoma or adenolipobroma. The pathogenesis of the development of mammary hamartoma is not fully clear. Sevim and Kocanay et al. reported a clinicopathologic analysis of 27 cases of hamartoma including one myoid hamartom, and mentioned the mean age and tumor size of them was 41.8 years and 3.9 cm, respectively, and described that breast hamartoma approximately accounts for 5% of all benign breast lesion (8). Mammary hamartomas have a typical area of radioolucent and smooth edges in mammography where fat and various amounts of fibrous and adenomatous tissues were shown. Although radiologic feature is characteristic, Helvie et al. reported that 29% of the tumors in their study could not be detected on mammography (9). Typical ultrasonogram of hamartoma showed sonolucent fat and a heterogeneous internal echoes. Images of several diagnostic procedure of hamartoma are reflected their pathologic structures. Although pathologic diagnosis and treatment are completely able to make by excisional biopsy of the lesion, pathologic diagnosis cannot be always done by ultrasonography-guided core needle biopsy because of above mentioned internal texture of hamartoma. Herbert et al. reported immunohistochemical studies of 24 breast hamartoma cases, in which all cases showed estrogen and progesterone receptors positivity in epithelial cells as well as in the stromal cells and no HER2/neu positivity were noted in these cases (10). Breast cancer associated with hamartoma is rare. Mendiolaa et al. had described the first case report of a malignant lesion associated with hamartoma in 1982 (2). Nami et al. had reported 15 cases with coexistence of breast cancer and hamartoma (11), and after the report Kai and Fukai were described a new case of carcinoma arising in mammary hamartoma. Kajo K, Zubor P, Danko J : Myoid (Muscular) hamartoma of the breast: case report and review of the literature. Breast Care 5(5) : 331-4, 2010

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