

Giant Borderline Phyllodes Tumor Mimicking Inflammatory Breast Carcinoma: A Case Report and Brief Literature Review

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Phyllodes tumor (PT) is a rare breast fibroepithelial neoplasm that account for 1% of all breast tumors. The giant PT (GPT) is defined as tumors larger than 5 cm in its maximum dimension. The differential diagnosis of a benign and malignant PTs based on imaging findings is difficult. They often may be confused as malignant breast carcinoma for the larger size and irregular shape on sonographic imaging. If they are associated with skin ulceration, It can mimics inflammatory breast carcinoma. Here, we report a case of giant borderline phyllodes tumor of 43-year-old woman who had rapidly growing breast mass and skin ulceration mimicking inflammatory breast carcinoma with literature review.

Index words: Breast; Neoplasm; Phyllodes tumor; Inflammatory breast carcinoma

Introduction

Phyllodes tumor (PT) is a rare fibroepithelial neoplasm that accounts for 1% of all breast tumors and 2% to 3% of fibroepithelial breast tumors (1). PTs has both epithelial and stromal components, and it is somewhat similar histologically to a fibroadenoma. However, it is characterized by a leaf-like structure with more abundant and cellular stroma than a fibroadenoma (2). Histologically, PTs are classified into categories of benign, borderline and malignant. However, most of these tumors tend to behave in a benign fashion clinically. But local recurrences and distant metastases can occur regardless of the histologic type (3). In some cases with rapidly growing mass, it could be associated with skin ulceration, which mimicking inflammatory breast carcinoma.

Here, we report a case of 43-year-old woman who had borderline GPT with skin ulceration mimicking inflammatory breast carcinoma

Case Report

A 43-year-old woman presented with a huge right breast mass that had necrosis and a foul odor. She has no remarkable past medical history and no significant family history. She noticed the small mass in her right breast one year ago, but she did not undergo any clinical work-up at that time. Then, approximately 4 or 5 days prior to presentation of this case, she had itching sensation with induration of right breast skin. And she noticed a mass extrusion of the outside skin with rapid growth. On physical examination, a huge necrotic mass 18 cm in diameter was seen in the entire right breast. The mass was extruded skin outside, and skin overlying the mass was thickened with ulceration, due to rapid growing breast mass. The axilla showed no remarkable finding. Initial chest X-ray reveals a huge high density

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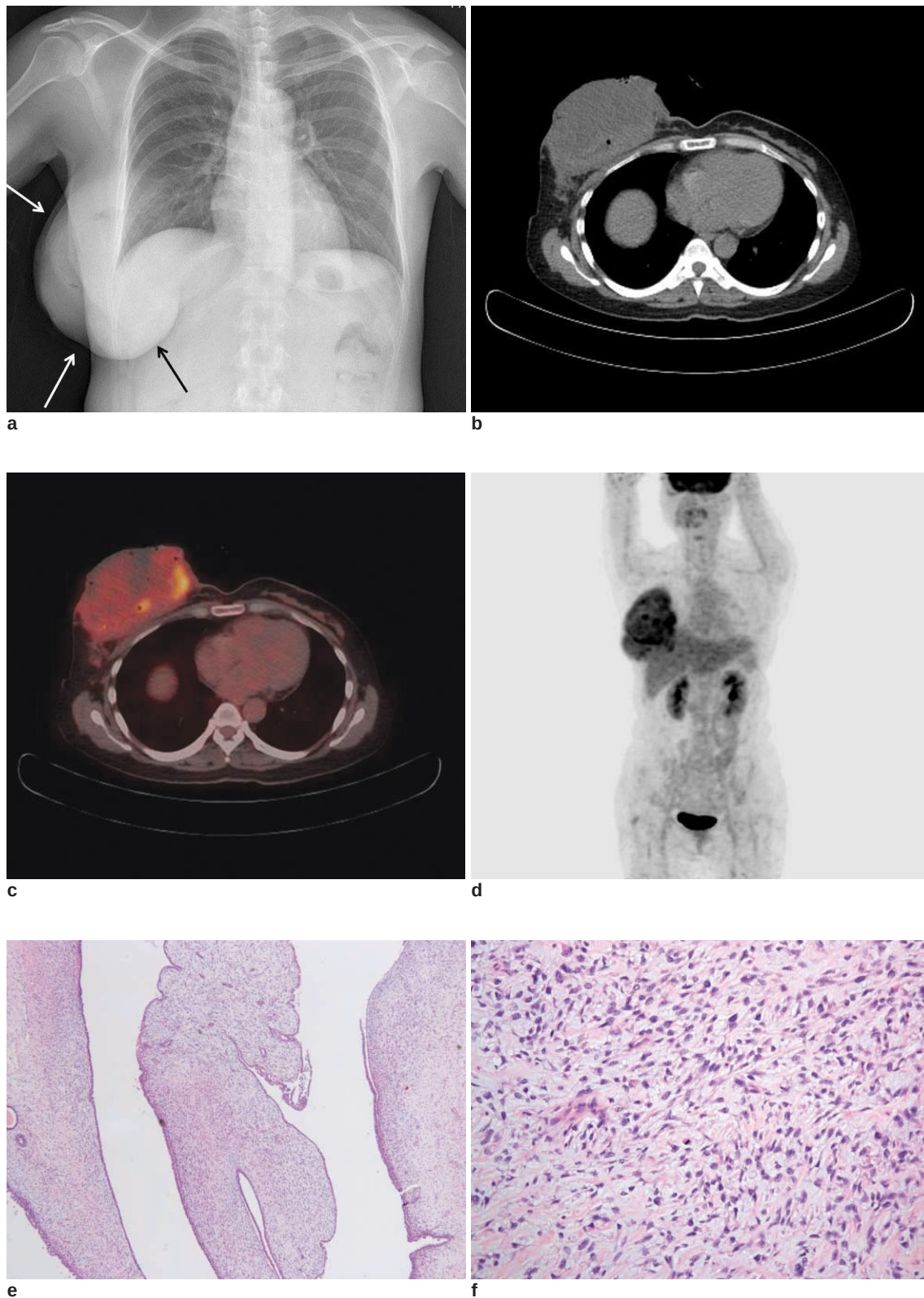


Fig. 1. Imaging findings of 43-year-old woman with giant borderline phyllodes tumor.

(a) Initial chest PA reveals a huge high density mass in the right breast (arrows). (b) Nonenhanced axial CT image of ^{18}F -FDG PET/CT showing a huge breast mass with free air and skin defect in the right breast. (c, d) Axial CT image (c) and MIP image (d) of ^{18}F -FDG PET/CT showing a huge uneven hypermetabolic mass ($\text{SUV}_{\text{max}}=4.2 \text{ g/mL}$) in the right breast without the evidence of distant organ metastasis. (e) Photomicrographs of tumor showing the characteristic leafy architecture with epithelium-lined clefts (H & E stain, $\times 40$). (f) Higher magnification of stromal cells demonstrate moderate cellularity and nuclear atypia with mitotic activity (H & E stain, $\times 200$).

mass in the right breast (Fig. 1a). ^{18}F -fluorodeoxyglucose (^{18}F -FDG) positron emission tomography (PET)/CT scan was performed. Nonenhanced axial CT image of ^{18}F -FDG PET/CT showed a huge soft tissue mass in the right breast with focal free air and skin defect (Fig. 1b). MIP and axial CT images of ^{18}F -FDG PET/CT showed a huge uneven hypermetabolic mass (maximum standardized uptake value (SUVmax)=4.2 g/mL) in the right breast, and there was no evidence of distant organ metastasis (Fig. 1c, d). Under strong clinical suspicion of inflammatory breast carcinoma, we performed a core needle biopsy on the tumor itself. Core needle biopsy revealed a fibroepithelial tumor with increased stromal cellularity and rare mitosis, favoring benign phyllodes tumor. We discussed the discrepancy about the clinical diagnosis and core biopsy results at the preoperative meeting. The patient underwent a total mastectomy and reconstruction with TRAM. Macroscopic examination of the resection specimen revealed a firm and well-demarcated mass with hemorrhage, measuring approximately 18 cm in the largest dimension. Histologically, the tumor was confirmed as a borderline phyllodes tumor, which composed of focal hypercellularity, focal nuclear pleomorphism of stromal cells and increased mitotic activity (5–9 mitotic figures per 10 high-power fields) (Fig. 1e, f). The skin was thickened and showed ulceration and necrosis. On immunohistochemistry, both epithelial and stromal components of the PT showed no immunoreactivity for estrogen and progesterone receptors and HER2, except for CD34. Finally, the patient was diagnosed with borderline phyllodes tumor.

Discussion

PT is a rare neoplasm of the breast that was first described as cystosarcoma phyllodes by Johannes Muller in 1838. It was finally renamed as a phyllodes tumor by the World Health Organization in 1982 (4). It is difficult to differentiate PTs from fibroadenomas, clinically and histologically. Comparing with fibroadenomas, the mean age of the patients in PTs is between 40 and 50 years, which higher than fibroadenomas and the PTs have higher stromal cellularity, cytologic atypia, and increased mitoses (2). The PT was histologically classified

as either the benign, the borderline or the malignant, according to the criteria proposed by Azzopardi and Salvadori and adopted by the World Health Organization, which include the followings: number of mitoses per 10 high-power fields in the most mitotically active area of the slides (0–4, 5–9, or 10 or more), tumor margins (pushing or infiltrating), stromal cellularity (slight or severe), stromal overgrowth (absent, slight, or severe), tumor necrosis (present or absent), and cellular atypia (absent, slight, or severe) (4). Sometimes PTs are presented with large tumors associated with skin ulceration for its rapid growing (5). The GPT is defined as tumors larger than 5 cm in its maximum dimension.

The differential diagnosis of a benign and a malignant PTs based on imaging findings is difficult. The mammographic findings in PTs are variable according to the size of mass. Some lesions may be missed because of small size. If the GPT is too large to take mammography, chest X-ray or CT could be used for evaluation of the whole contour of mass. Hence, the value of mammograms is very limited for PTs. The ultrasound findings are also notorious for their inability to distinguish the benign or malignant PTs. Chung et al. reviewed sonographic findings of 14 GPTs with circumscribed, lobulated, and heterogeneous masses in US (6). They reported that the larger the size of GPTs, the more malignant pathology was found. Tan et al reported that borderline PTs were likely to have irregular shapes on sonographic imaging in their series and appeared to have a multinodular-like fusion shape, which may correlated with an asynchronous growth rate within different components (7). But in their series, 50% of the tumors were initially misdiagnosed as fibroadenomas.

MRI could provide a clue to differentiate a GPT from other breast diseases. GPTs showed initial rapid and delayed persistent enhancement pattern on dynamic breast MRI, while other breast cancer revealed delayed washout pattern (8). In addition, internal non-enhanced septations, slit-like patterns in enhanced images and internal signal voids were also differential points of GPTs from other breast masses. A preoperative MRI could have elevated the diagnostic accuracy in present case, but we would have been unable to study an MRI because of the huge mass.

PET could be helpful to differentiate malignant from

benign PTs (9). The SUV of more than 2.5 is frequently used as the criteria for malignancy. In our case, PET was performed with SUV of 4.2 g/mL but it could not be pathognomonic for differentiate the borderline PT from inflammatory breast cancer. We could not evaluate the enhancement pattern of tumor because of the nonenhanced study of PET/CT.

PTs, especially GPT, required core needle biopsy to rule out a rapid growing breast cancer. Komenaka et al reported the positive and negative predictive values of core biopsy to diagnose the PT were 83% and 93% respectively (10).

Choice of surgical treatment depends on the size of mass and simple mastectomy is the best surgical procedure for the breast affected by large borderline or malignant PT (3). Conservative surgery is justified only if the breast volume is large enough to obtain clear margins.

In conclusion, most PTs are very difficult to distinguish from other benign or malignant tumors on the basis of radiologic features, especially from fibroadenomas. PT can present with a huge palpable mass which can grow rapidly associated with skin ulceration mimicking inflammatory breast carcinoma clinically. MRI is useful tool for the diagnosis of breast PTs and ¹⁸FDG-PET/CT could be helpful to differentiate between a be-

nign and a malignant PTs.

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염증성 유방암과 유사한 소견을 보이는 거대 경계성 엽상육종: 증례보고 및 문헌고찰

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유방 엽상종양은 전체 유방종양의 1% 미만을 차지하는 매우 드문 섬유상피성 종양으로, 최대 직경이 5 cm 이 넘을 경우 거대엽상종양으로 정의한다. 영상소견만으로 양성 엽상종양과 악성 엽상종양을 구별하기는 어렵다. 간혹 크기가 크며 초음파소견에서 불규칙한 모양을 보이는 경우 악성유방암으로 오인하기도 한다. 특히 피부궤양과 동반된 경우에는 염증성 유방암과 유사한 소견을 보일 수 있다. 저자들은 43세 여성에서 급격히 커진 종괴와 피부궤양으로 염증성 유방암과 유사한 소견을 보였던 거대 경계성 엽상종양의 예를 문헌고찰과 함께 보고하고자 한다.

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