Recurrent Fibroadenoma and Concomitant Mucinous Cancer with Ductal Carcinoma *In situ* Following Benign Phyllodes Tumor Excision: A Rare Case Report

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Abstract

A 37-year-old woman was diagnosed with a fibroadenoma (FA) with concomitant extensive spreading of ductal carcinoma *in situ* (DCIS) and multifocal mucinous carcinomas in 2022. Previously, she was first diagnosed with a FA and received excision in 2016. The initial clinical presentation was a palpable mass in the left breast. During periodic breast ultrasound examination in 2020, another newly developed tumor was found in the same region. A core biopsy revealed a fibroepithelial lesion. The patient received wide excision surgery. The final pathology report indicated a phyllodes tumor (PT) with a clear margin. All three events occurred in the same region (left breast, at 2 O'clock and 3–5 cm from the nipple). Our patient had a relapsed FA, which progressed into a benign PT and FA concomitant with DCIS and multifocal mucinous carcinomas. We report this case to highlight the need for continuous follow-up for FA cases even after complete resection and the possibility of recurrence with other fibroepithelial tumors.

Keywords: Benign phyllodes tumor, breast cancer, case report, ductal carcinoma *in situ*, recurrent fibroadenoma

Introduction

Fibroepithelial lesions (FELs) are categorized into fibroadenomas (FAs) and phyllodes tumors (PTs), the latter accounting for 0.3%–1% of FELs and usually requiring surgical excision. FAs are usually managed by regular follow-up. An increased risk of breast cancer subsequent to a diagnosis of an FA has been reported,[1] and a special surveillance regimen should be adopted for women with breast cancer risk. The incidence of carcinomas within FAs is estimated at 0.1%–0.3%, and FAs could be a long-term risk factor for invasive breast cancer.[2,3] Distinguishing benign PTs from FAs is difficult in the early stage due to overlapping radiology and microscopic features. Herein, we present a case of recurrent FAs in a patient who underwent two tumor excisions and still developed PTs with recurrent FAs and mucinous cancer with ductal carcinoma *in situ* (DCIS).

Case Report

We present the case of a 31-year-old woman who presented to the breast surgeon outpatient department with a palpable mass lesion in the left breast in 2016. There was no known family history of breast cancer or other gynecological cancer. Her ultrasound showed a 2 cm, partially ill-defined hypoechoic breast nodule over the left breast at 2 O'clock and 4 cm from the nipple (left 2/4) [Figure 1]. The patient underwent tumor excision and the pathology report indicated a FA. In 2020, during a periodic breast ultrasound follow-up, a newly developed, partially ill-defined hypoechoic nodule was detected at left 2/5 [Figure 2]. The result of a core biopsy indicated a FEL. Further wide excision surgery was performed and the final pathology indicated a benign PT with a clear margin (1 cm from the tumor and free of cancer). During regular breast imaging follow-up in August 2022, a new 1.0 cm hypoechoic nodule was found at left 2/5 [Figure 3]. In January 2023, rapid interval growth was noticed in breast sonography [Figure 4]. The size of the left 2/5 hypoechoic

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Abbreviations

CNB	Core needle biopsy
DCIS	Ductal carcinoma in situ
FA	Fibroadenoma
FEL	Fibroepithelial lesion
NCCN	National Comprehensive Cancer Network
PT	Phyllodes tumor
WHO	World Health Organization
WCT	World Health Organization Classification of Tumors

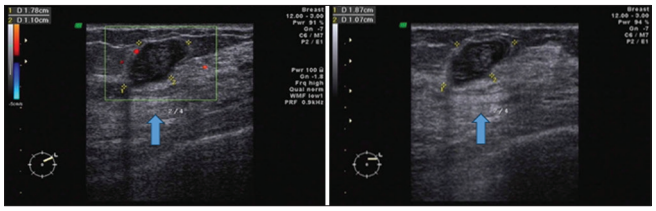

Figure 1: Breast ultrasound showing a partially ill-defined hypoechoic breast nodule in the left breast at 2 o'clock and 4 cm from the nipple (left 2/4)

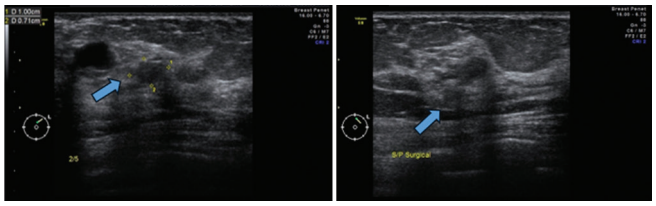

Figure 3: Breast ultrasound showing another new hypoechoic nodule at left 2/5

breast nodule grew to 2.5 cm [Figure 5]. Pathology of the excised tumor revealed an FA concomitant with the extensive spreading of DCIS with multifocal mucinous carcinoma. The patient underwent a partial mastectomy and sentinel lymph node biopsy (0/2) on March 9, 2023. Serial radiotherapy was performed. The recovery was uneventful.

Discussion

The majority of PTs occur in women, with a median presentation age of 45 years. In Asian women, the incidence is higher and the age younger. PTs usually present clinically as well-defined, firm nodules or masses, usually located in the upper outer quadrant of the breast. They are unilateral and grow rapidly, with an average size of 5–7.2 cm. PTs of the breast are infrequent fibroepithelial neoplasms, accounting for 0.3%–1% of all breast neoplasms; the remaining 97.5% are FAs.[4-6] Triple assessment by clinical, radiological, and histological examination is the initial assessment regimen to evaluate PTs. Large PT tumors tend to grow faster than FA do. They can stretch and ulcerate the skin, and they are associated with blue discoloration, dilated skin veins, skin ulcers, nipple retraction, and in rare cases, and palpable axillary lymph nodes.

An increased risk of breast cancer subsequent to a diagnosis of FA has been reported, and a special surveillance regimen should be adopted for women at risk of breast cancer.[1] Organ

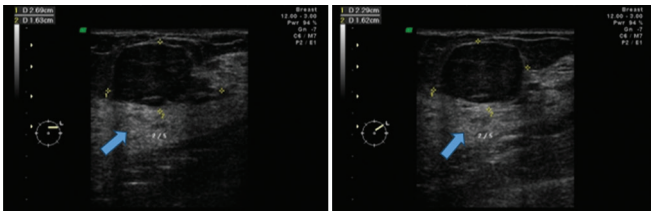

Figure 2: Breast ultrasound showing a newly developed, partially ill-defined hypoechoic nodule detected in the left breast at 2 O'clock and 5 cm from the nipple (left 2/5)

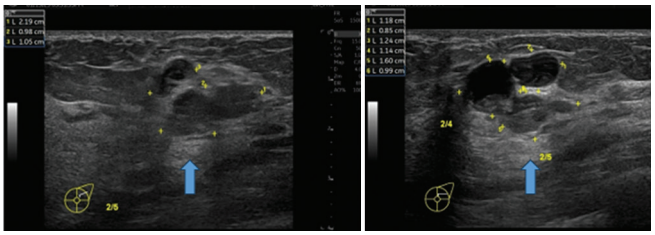

Figure 4: Breast ultrasound showing rapidly enlarged hypoechoic nodule at left 2/5. The size progressed with peripheral cystic change

et al. reported a recurrence rate of 17% for surgically excised FAs.[7] This rate may reflect the multifocality of tumors or the presence of associated "fibroadenomatoid change" in the surrounding breast tissue.[6,8,9] DCIS, arising within an FA, is a specific pathological type of tumor that is rarely encountered in practice. Its incidence ranges from 0.02% to 0.125%, and it is usually discovered incidentally during pathological examination of resected FA tissue.[10] Lobular carcinomas *in situ* can also arise within a breast FA. Overall, the incidence of carcinomas within FAs is estimated at 0.1%–0.3%. Hence, FAs could be a long-term risk factor for invasive breast cancer.[2,3]

The differences between FAs and PTs in ultrasound and mammogram imaging include their size, shape, margin, and posterior acoustic enhancement. FAs typically present as smaller in size, usually <3 cm, with a round or oval shape. The tumors appear as homogeneous hypoechoic masses with clear margins. PTs, on the other hand, often exceed 3 cm in size and have irregular shapes, lobulations, a heterogeneous internal texture, a cystic component, horizontal linear clefts, and the absence of microcalcifications. They may exhibit leaf-like protrusions at the edges and are more likely to show posterior acoustic enhancement on ultrasound imaging.[11-13] Most FAs grow to 2–3 cm at most, after which their size tends to stabilize. The average PT tumor size ranges from 4 to 5 cm.[4] FELs larger than 3 cm increase the risk of malignancy by 3.87 times, and FELs larger than 8 cm almost invariably develop into malignant PTs.[14] The degree of FEL deterioration is positively correlated with size.[14] There is no clear indicator for PTs in ultrasound. MRI has limited usefulness in distinguishing PTs from FAs.[11-13,15]

FAs and PTs have overlapping characteristics in core needle biopsy (CNB) and thus are not easy to distinguish. Surgical excision and pathological examination are needed for a more

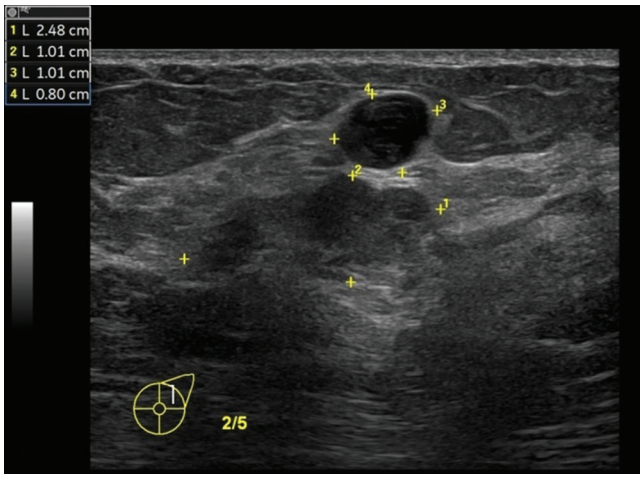

Figure 5: Breast ultrasound showing rapid enlargement of hypoechoic nodule at left 2/5. The size progressed from 1.0 cm (2022/8) to a maximum size of 2.5 cm (2023/1)

complete tissue analysis. FAs and PTs are categorized in the FEL lesion group. Once an FEL tumor is suspected, surgical excision is the recommended and safer option.[16] In 2020, the National Comprehensive Cancer Network modified the recommendation to surgical excision for FELs larger than 3 cm. For FELs, surgery recommendations should not be solely based on tumor size (3 cm) but rather on the initial pathological diagnosis classification from CNB and the rapid growth of the tumor. FAs with rapid growth in a short period are more likely to progress to PTs.[17]

The fifth edition of the World Health Organization Classification of Tumors classifies PTs into three types: benign, borderline, and malignant. The 5-year survival rates are approximately 96% for the benign type, 74% for the borderline type, and 66% for the malignant type.[5] This classification helps determine whether additional treatments (such as radiotherapy) should be provided postoperatively and predicts recurrence rates and survival rates. Surgical resection remains the mainstream for PT treatment. Local recurrence can still occur even with surgical resection, and the recurrence rate increases with tumor grades.[4,18] One-quarter of PTs have the potential for metastasis, which implies a poor prognosis.[14] PT metastasis occurs through blood circulation, with the most common sites being the lungs (66%), bones (28%), brain (9%), liver, and heart. In addition, because of the hematogenous spread, sentinel lymph node excision is not necessary even for malignant PTs.[14]

Clear grading of PTs is essential for surgeons to establish management strategies and provide comprehensive clinical prognostic predictions. Tumor surgical excision is the cornerstone of PT tumor management, which includes (1) local excision (cancer-free margin: 5 mm), (2) wide excision (cancer-free margin: 10 mm), and (3) mastectomy. Among these, wide excision is the more widely practiced approach because it can address up to 20% of the high recurrence characteristics of PTs. Only benign PTs can be considered for local or wide excision; borderline or malignant PTs require wide excision or mastectomy based on breast preservation (breast size).[19]

Adjuvant radiation therapy following surgery for sarcoma lesions may offer a better disease prognosis. Adjuvant radiation therapy is often recommended for borderline PTs and malignant PTs, especially in cases of larger tumors and younger ages, which may provide better survival rates and reduce the chance of ipsilateral breast cancer recurrence, leading to overall better disease prognosis.[14] Prophylactic excision of sentinel lymph nodes before surgery is generally not recommended because PTs typically spread through the hematogenous route.[6,14]

Conclusion

FAs typically exhibit benign characteristics in ultrasound imaging, namely regular borders, slow growth, and rarely, expansion beyond approximately 3 cm. PTs often exceed 3 cm in size, and they have an irregular shape, lobulations, a heterogeneous internal texture, a cystic component, horizontal linear clefts, and the absence of microcalcifications. PTs may exhibit leaf-like protrusions at the edge and are more likely to show posterior acoustic enhancement on ultrasound imaging. This case highlights the importance of clinicians remaining vigilant even if imaging suggests a benign FA, particularly if rapid growth is observed, to rule out the possibility of a phyllodes tumor. It is recommended that patients with benign FAs and phyllodes tumors undergo regular follow-up breast imaging examinations for a period of 3–5 years following tumor excision.

Ethics statement

This study was conducted in accordance with the ethical principles outlined in the Declaration of Helsinki and its amendments. The authors certify that they have obtained all appropriate patient consent form. On the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that neither her name nor initials will be published and that due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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