Case Report

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Report of Two Cases with Simultaneously Detected Tubular Carcinoma and Phyllodes Tumor of the Breast

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ABSTRACT

Tubular carcinoma (TC) is a subtype of invasive breast carcinoma with better prognosis, and phyllodes tumors (PT) are rare fibroepithelial lesions. Accurate preoperative pathological diagnosis allows for correct surgical planning and avoidance of reoperation for these breast neoplasms. A database was created by analyzing the archives of Department of General Surgery of the İstanbul Faculty of Medicine between September 2006 and November 2017, and a total of 105 PTs and 55 TCs were collected. Two cases with concurrence of TC and PT were identified and examined in detail. The first patient was a 33-year-old woman with a 20×12 mm^2 TC and a 65×32 mm^2 malignant PT in the left breast. The second patient was a 28-year-old woman with two masses in the right breast. The first mass was 38×16 mm^2 on the upper outer quadrant, and the second mass was 10×8 mm^2 in size in the lower inner quadrant, accompanied by a 16×10 mm^2 TC and a 33×26 mm^2 borderline PT. Both cases were treated by mastectomies due to patient's decisions or insufficient margin control. This study extrapolated that if two tumors are detected simultaneously, margin control can become more difficult, and breast-conserving surgery should be thoroughly reviewed.

Keywords: Breast, phyllodes tumor, surgery, tubular carcinoma

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Introduction

Tubular carcinoma (TC) is a rare histologic subtype of all breast cancers, which accounts for 1%–4% of all breast carcinomas (1). Pathologically, TC appears like a necklace formed by a string of beads and presents with stellate infiltration. These tumors tend to be of low grade, which means that their cells appear normal, with >90% of tubular formation. Over 90% of tumors with TC are hormone receptor positive and HER2 negative, which indicates favorable oncologic outcomes (2-5).

Phyllodes tumors (PTs) are fibroepithelial breast tumors and account for less than 1% of all breast neoplasms (6). PTs have characteristic epithelial components arranged in clefts, surrounded by a mesenchymal component organized in a leaf-like pattern (7). Simultaneous occurrence of these two tumors is extremely rare.

Case Presentations

Case 1

A 33-year-old woman presented with a 1-year history of a rapidly enlarging left breast lump. Family history was significant for an aunt with breast cancer at age 60 years. Ultrasonography (USG) and mammography (MG) were performed as standard protocol. USG, MG, and additional magnetic resonance imaging detected two masses: a 24×16 mm^2 non-palpable lobulated mass in the lower outer quadrant and a 6×5 cm^2 mass in the upper inner quadrant with sharp margins (Figure 1). Multiple inconspicuous metastasis lymph nodes were detected in the axilla. Core needle biopsies were performed. The breast mass in the lower outer quadrant was diagnosed as TC, and the mass in the upper inner quadrant was diagnosed as mesenchymal neoplasia with core needle biopsy. A USG wire-guided tumor excision and a regular tumor excision were performed for the non-palpable first mass and second mass, respectively, based on the adequate breast volume and sentinel lymph node biopsy (SLNB) to the axilla. Under definitive pathologic examination, the mass found in the lower outer quadrant was a 20×12 mm^2 TC without axillary lymph node metastasis (modified Bloom-Richardson Grade I). The tumor had luminal type A receptor features. No lymphovascular invasion (LVI) was detected. The mass in the upper inner quadrant was diagnosed as a 65×32 mm malignant PT with 10/10 BBA mitosis score, marked cellular atypia,

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pleomorphism, and stromal overgrowth; however, this malignant PT was 5 mm close to the margin. Mastectomy was performed based on the surgeon's suggestion and patient's decision. Non-malignant postoperative changes were detected in the mastectomy specimen. Treatment continued with radiotherapy and anti-estrogen therapy. No further disease was observed in the 10-year follow-up period.

Case 2

A 28-year-old unmarried nulliparous woman presented with a 6-month history of a rapidly enlarging right breast lump. As regards family history, no ovarian or breast malignancy was determined. The masses were located in the right breast: the first mass (38×16 mm^2) was located on the upper outer quadrant (Figure 2) and the second mass (10×8 mm^2) was located in the lower inner quadrant. Core needle biopsies revealed that the first mass was a biphasic tumor and the second mass was a TC. Given the distant localization of these masses, mastectomy, SLNB, and oncoplastic surgery were performed on patient's request. The first mass was defined as a borderline PT (34×20 mm^2) and had 5/10 BBA mitosis score and minimal cellular atypia. The size of the TC was 14×8 mm^2 in definitive pathologic examination, which was a modified Bloom–Richardson grade I tumor. No LVI was detected. The tumor had luminal type A receptor features, and the SLNB result was negative. The patient was on anti-estrogen treatment. No further disease was observed in the 5-year follow-up period.

Informed consent was obtained from each patient for inclusion in this case report.

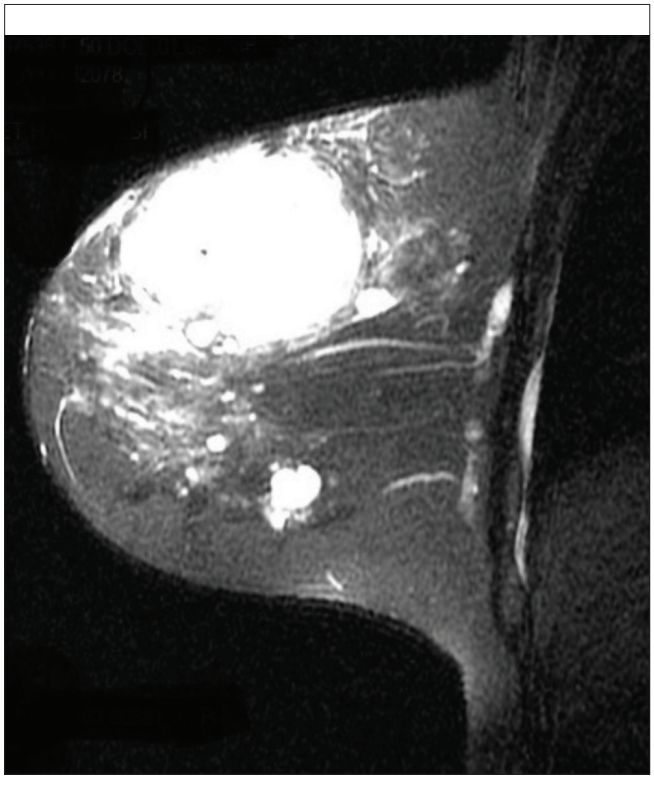

Figure 1. Malignant PT in the upper inner quadrant and TC in the lower outer quadrant

PT: Phyllodes tumor; TC: Tubular carcinoma

Key Points

- Simultaneous and co-detection of invasive breast cancer with PT of the breast is rarely described in the literature; however, the histogenesis has not yet been fully understood.
- If two breast tumors are detected simultaneously, margin control of both tumors can become more difficult.
- In this study, mastectomies were performed in one of the patients given the distance between the tumors and the other patient had positive margin, which supports the above opinion.

Discussion and Conclusion

Because TCs may have a typically favorable prognosis, efforts have been made to reduce unnecessary treatment. Therefore, some investigators have suggested that surgical staging of the axilla may not be necessary for TC <1 cm. In contrast, other researchers have proposed that axillary staging should be considered for all patients with TC, as small tumors <1 cm also showed nodal involvement (8). In our study, no nodal involvement was detected in both TC cases.

PT is one of the fast-growing breast tumors; however, it is generally histologically benign. It might remain latent for many years and then start to grow fast in some patients. MG and USG used in the diagnosis of breast masses are not very reliable in the differential diagnosis of PTs from fibroadenomas. Given the fast growth pattern, there might be suspicious axillary lymphadenopathies, enlargement in the skin and veins, nipple changes, and necrosis. Because of the similarity between PTs and fibroadenomas clinically and radiologically and for avoidance of any axillary procedure due to suspicious lymphadenopathies, preoperative evaluation with core biopsy for PT cases should be performed (8-13).

The underlying etiology for concomitant carcinoma occurring within PTs is unknown. The presence of carcinoma associated with PT is rare, with only anecdotal reports of isolated cases. Table 1 summarizes reports published since 2000, and most of the accompanying cancers were ductal carcinomas in situ (14-28).

Surgery is essential in the treatment of PTs, and wide excision with negative surgical margins (at least 1 cm) is the recommended surgical approach regardless of the histopathological type (16). Surgical approach and margin assessment for TC is similar to invasive ductal carcinoma. In 2016, the Society of Surgical Oncology and American

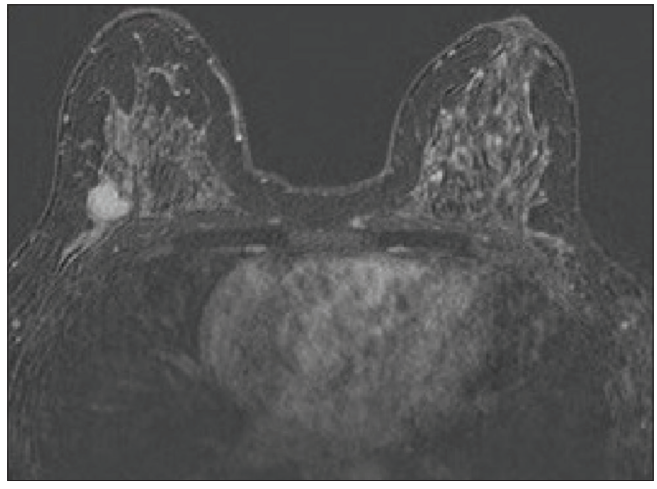

Figure 2. A borderline phyllodes tumor in the right breast

Table 1. Published literature of invasive and *in situ* carcinoma associated with PT

No	Age	Type of PT	Size of PT (cm)	Type of carcinoma	Lymph node involvement	Study
1	39	M	9	DCIS	0	Alo et al. (14)
2	47	B	17	ILC	0	Kodama et al. (15)
3	26	B	3/3	IDC/DCIS	4/13	Parfitt et al. (16)
4	45	M	12	DCIS	0	Lim and Tan (17)
5	59	M	3/5	Undifferentiated	0	Tokudome et al. (18)
6	69	B	NA	SCC	0	Ramdass and Dindyal (19)
7	75	M	3/5	DCIS	0	Nomura et al. (20)
8	65	M	6	IDC	0	Sugie et al. (21)
9	51	M	16	IDC/DCIS	2/12	Korula et al. (22)
10	54	B	15	DCIS	0	Yamaguchi et al. (23)
11	24	Borderline	10	IDC/DCIS	1/2	Kuo et al. (24)
12	70	M	6	IDC	0	Macher-Goepinger et al. (25)
13	49	B	4/8	ILC/LCIS	0	Shirah et al. (26)
14	53	Borderline	6/5	IDC/LCIS	0	Qinlan-Davidson et al. (27)
15	42	B	2/2	DCIS	0	Ghosh and Saha (28)

M: Malignant P; B: Benign; IDC: Invasive ductal carcinoma; ILC: Invasive lobular carcinoma; DCIS: Ductal carcinoma *in situ*; LCIS: Lobular carcinoma *in situ*; SCC: Squamous cell carcinoma

Society for Radiation Oncology announced a margin consensus as "no ink on tumor" for invasive and 2 mm for ductal carcinoma in situ and reported the "no tumor at ink" principle as the standard for an adequate margin with wide excision (29). In this study, both patients underwent mastectomy.

Management steps of TCs and PTs separately are well-known; however, detecting these tumors simultaneously is extremely rare, and the histogenesis has not yet been fully understood. In our case, we could not reveal histomorphologic findings that would definitely support one of the theories suggested in the pathogenesis. However, we think that management can be more complicated in these cases with simultaneously detected different tumors. This study extrapolated that if two tumors are detected simultaneously, margin control can become more difficult and breast-conserving surgery should be thoroughly reviewed. The study has supported this opinion and the performance of mastectomies in both cases.

Informed Consent: Written informed consent was obtained from patients who participated in this case.

Peer-review: Externally peer-reviewed.

Author Contributions

Concept: B.İ.; Design: B.İ.; Supervision: B.İ..; Resources: B.İ., S.E., R.T.; Materials: B.İ.; Data Collection and/or Processing: B.İ., S.E.; Interpretation: B.İ., S.E., R.T.; Literature Search: S.E., R.T.; Writing Manuscript: B.İ.; Critical Review: B.İ., S.E., R.T.

Conflict of Interest: The authors have no conflicts of interest to declare.

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