Case Report

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Synchronous phyllodes tumor and invasive breast carcinoma in contralateral breasts- a rare case report

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ABSTRACT

Phyllodes tumours are rare breast tumours, accounting for 0.3-1% of all breast cancers and 2.5% of fibroepithelial tumours. They occur in younger age women in Asian countries. These can be classified into benign, borderline and malignant based on stromal hypercellularity, mitotic figures, cellular pleomorphism, stromal overgrowth and nature of the tumour borders. Synchronous coexistence of phyllodes tumour and invasive breast carcinoma in the contralateral breast is very rare, and probably the second one reported in the literature. Hereby, we report a case of a middle-aged woman presenting with a lump in the bilateral breast, diagnosed to have benign phyllodes of one breast and Invasive carcinoma of the contralateral breast.

Keywords: Phyllodes tumor, Carcinoma breast, Synchronous, Carcinoma thyroid

INTRODUCTION

Phyllodes tumours are rare breast tumours, accounting for 0.3-1% of all breast cancers. They are grouped under fibroepithelial tumours according to World Health Organization classification of breast tumors and constitute up to 2.5% of them.¹ They occur in a younger age group (25-30 years) in Asian women. They are classified into benign, borderline and malignant based on stromal hypercellularity, mitotic figures, cellular pleomorphism, stromal overgrowth and nature of the tumour borders.^{2,3} Coexistence of phyllodes tumour in one breast with invasive ductal carcinoma in the other is extremely rare. We feel this is the second case of synchronous coexistence of these tumours reported in the literature after Neto et al in 2012.⁴

CASE REPORT

A 46-year-old female presented with complaints of a lump in the left breast since three years and a lump in the right breast for seven months. The left breast swelling was firm to hard in consistency and progressed in size gradually. Seven months back, she also noticed a lump in the right breast with white discharge from the nipple. The swelling was hard in consistency. The patient gave a history of fine needle aspiration of left breast suggestive of fibrocystic disease and right breast suggestive of invasive ductal carcinoma. She also had a history of total thyroidectomy three years back and was diagnosed to have papillary carcinoma of the thyroid. There was no history suggestive of ovarian/ breast malignancy in the family. On examination, Right breast: Lump of size 3×3cm was palpable in the upper inner quadrant, no skin changes, nipple and areolar complex appeared normal, no dilated veins or ulceration or puckering was present. No palpable axillary lymph nodes were present. Left breast: lump of size 3×2cm was felt in the upper outer quadrant. Nipple and areolar complex appeared normal. No dilated veins, ulceration or puckering was present. No palpable axillary lymph nodes. Trucut biopsy of a lump in the right breast was performed, which showed invasive tumour cells in nests and tubules with high nucleus to cytoplasmic ratio, pleomorphic hyperchromatic nucleus and scant cytoplasm. Peritumoral desmoplasia, sparse lymphocytic infiltrate and occasional mitotic figures were noted. Therefore, a diagnosis of invasive carcinoma breast, Nottingham Bloom Richardson (NBR) grade 1 was given. The patient underwent right modified radical mastectomy and left breast lumpectomy (Figure 1).



Figure 1: Right (R) modified radical mastectomy specimen showing a grey white tumour in upper inner quadrant and left (L) lumpectomy specimen showing a homogenous pale lobulated tumour.

The right modified radical mastectomy specimen measured 20×19.5×7cm with elliptical skin measuring 15×12.3cm. The overlying skin and nipple areola complex appeared normal. On cutting through, a hard grey white tumour was seen in the upper inner quadrant measuring 3.8×2×2 cm, located at a distance of 1cm from the medial resected margin, 2 cm from the superior resected margin, 10.2 cm from the lateral resected margin, 8.2 cm from the inferior resected margin, 0.5cm from the deep resected margin and 1.6 cm from the overlying skin. Microscopically, the tumour cells were arranged predominantly in tubules and focal nests, displaying moderate nuclear pleomorphism, and some showing a prominent nucleolus. The tumour cells were seen invading the underlying muscle. Focus of ductal carcinoma in situ (DCIS) was noted (Figure 2).

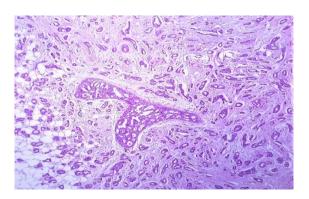


Figure 2: Microscopic image showing pleomorphic tumour cells in tubules with a focus of DCIS.

All the resected margins were free of tumour. Thirty-six lymph nodes dissected from the axillary pad of fat

showed reactive changes. The left breast lumpectomy specimen measured 8×8×4.5cm, with overlying skin measuring 4x3 cm. On serially slicing, a homogeneous pale white lobulated mass was noted, with slit-like spaces. Areas of haemorrhage and mucoid areas were also identified. Microscopically, a tumour with hypercellular stroma was identified (Figure 3).

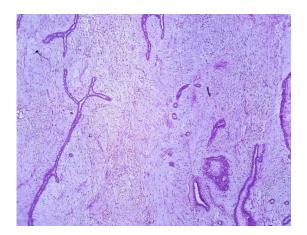


Figure 3: Microscopic image showing hypercellular stroma with epithelial and myoepithelial cells stretched into clefts.

The epithelial component consisted of luminal epithelial and myoepithelial cells stretched into clefts. The stromal cells were relatively uniform and showed<5 mitosis/ 10 high power field. Areas of hyalinization and myxoid change were also seen. Thus, the final diagnosis of invasive ductal carcinoma of right breast, not otherwise specified with a focus of DCIS, NBR grade 1, TNM stage pT2N0 and benign phyllodes tumour of the left breast was made. The post-operative period was uneventful. Immunohistochemistry was performed which showed positivity for oestrogen receptor (ER), progesterone receptor (PR); negativity of Her2/neu and Ki-67 index of 40%. Thus, the tumor categorised into luminal B-like (Her2-negative). The patient is now on regular follow for chemotherapy (fluracil+epirin+endoxan: FEC regimen). The patient is tolerating the chemotherapy well.

DISCUSSION

Breast tumours are broadly classified as epithelial tumours, mesenchymal tumours, fibroepithelial tumours, tumours of the nipple, malignant lymphoma, metastatic tumours, tumours of male breasts and clinical patterns according to WHO classification of the tumors of breast. Fibroepithelial tumours are biphasic neoplasms consisting of proliferating epithelial as well as stromal components. They include fibroadenoma, phyllodes tumour and hamartoma. Phyllodes tumour is much rarer than fibroadenoma and resembles intracanalicular fibroadenomas forming a leaf life pattern. They are thought to arise from the intralobular or periductal stroma. It is characterized by bilayered epithelium arranged in clefts surrounded by hypercellular stroma.

Most of the phyllodes tumours are benign but may often recur. Malignant phyllodes may metastasize via hematogenous route, around 3-12%. The most sites for metastasis are lung and bone. Lymph node metastasis is rare. ^{5,6} Grossly, they may range from a few centimetres to large size (>10cm) causing the overlying skin to stretch out with dilation of superficial veins. Due to their well-demarcated margins, they can be easily shelled out. On cut surface, whorled pattern with curved clefts may be seen giving it a leaf-like appearance. Smaller lesions have a more of homogenous appearance.

Microscopically, phyllodes tumour consists of an enhanced intracanalicular pattern with leaf-like projections into dilated elongated lumina. They can be classified into benign, borderline and malignant based on stromal hypercellularity, cellular pleomorphism, mitotic figures, stromal overgrowth and nature of the tumour borders. ^{2,3} The differential diagnosis for benign phyllodes tumour is fibroadenoma and for malignant phyllodes can be spindle cell sarcoma.

Surgical resection is the mainstay of treatment, with a 10mm clearance margin.⁷ Axillary lymph node clearance is usually not performed. Clinical and radiological follow up is then recommended every year, as local recurrence is common even in mild cases, more than 8% of subjects in 10 years. Rate of recurrence is higher in borderline and malignant phyllodes.⁸ Invasive carcinomas arising within or adjacent to phyllodes have been reported quite often.^{9,10} But, the coexistence of benign phyllodes in one breast with invasive carcinoma in the contralateral breast is very rare. Only one such case is reported previously by Neto et al. in 2012.4 Also, similar coexistence of phyllodes and invasive lobular carcinoma is reported To et al in 2018.¹¹ Ours is the second case reporting coexisting benign phyllodes tumour and invasive carcinoma in contralateral breasts.

Also, another diagnosis to be considered in such patients is Cowden syndrome, showing germline mutation in PTEN gene. Cowden syndrome or Cowden-like syndrome show an increased risk of various cancers including breast and thyroid cancers.¹²

Also, germline mutation in PARP4 gene shows susceptibility to primary breast and thyroid cancers. Similarly, radiotherapy can increase the risk of second primary cancers. However, this possibility was not considered in our patient as she had not taken any radiation therapy post-thyroidectomy.

CONCLUSION

The association of phyllodes tumour with invasive carcinoma of the breast is extremely rare, especially independent and synchronous in contralateral breasts. We have reported the second case of the coexisting benign phyllodes tumour and invasive ductal carcinoma in

contralateral breasts. Additionally, our patient also had a history of papillary carcinoma of the thyroid.

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