Phyllodes tumor of the breast with nipple discharge
A case report

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Abstract
Rationale: Phyllodes tumors (PTs) of the breast are rare biphasic fibroepithelial neoplasms which usually present with unilateral palpable breast lumps. Here we reported a case of borderline PTs with multifocal intraductal lesions whose primary clinical manifestation was nipple discharge.

Patient concerns: A rare case of PT with nipple discharge in a 26-year-old young lady accepted a wide local excision on her left breast, and the pathological examination turned out to be a malignant phyllodes tumor. After a 12-month follow-up, no local or distant recurrence was observed.

Diagnosis: Borderline PTs with multifocal intraductal lesions

Interventions: The patient received wide local excision with clear margin. After the operation, the patient is undergoing surveillance through ultrasonography and physical examination every 3 months.

Outcomes: After a follow-up of 12 months, no local or distant recurrence was observed.

Lessons: The lesions of the PTs are often manifested with unilateral, palpable masses in the breast. There are very few case reports of PTs manifested with nipple discharge in the literature. We should improve the understanding of the multiple clinical manifestations of PTs.

Abbreviations: PTs = Phyllodes tumors, WHO = World Health Organization.

Keywords: breast, case report, nipple discharge, phyllodes tumors

1. Introduction

Phyllodes tumors (PTs) of the breast are uncommon neoplasms originating from fibroepithelial component that account for less than 1% of all breast neoplasms.\textsuperscript{1,2} PTs occur in women of all ages, including adolescents and elderly. The majority of PTs arise in women between 35 and 55.\textsuperscript{3} World Health Organization (WHO) has graded PTs to benign, borderline and malignant categories on the basis of stromal characteristics.\textsuperscript{4} The lesions of the PTs are often manifested with unilateral, palpable masses in the breast. Here we reported a rare case of a young lady with nipple discharge, whose ultrasonography and ductoscopy highly suggested an intraductal tumor. However, histological examination after surgery confirmed a borderline PT.

2. Case presentation

A 26-year-old young woman was admitted to the West China Hospital, Sichuan University, China, complaining of spontaneous nipple discharge in her left breast for one month. Her past history showed that she had accepted a lumpectomy on her left breast because of a palpable breast mass. The postoperative pathological report showed a complex fibroadenoma, with stromal cells proliferation, local lobulated structures were observed. She denied any family history of breast cancer or ovarian cancer.

On physical examination, a firm mass with a clear margin was palpated in lower inner quadrant near the nipple of the left breast, which measured approximately 2.5 cm in diameter. Single duct non-bloody discharge from the left nipple was evident after squeeze. There was no nipple retraction and skin dimpling evident.

An ultrasonography detected mammary duct ectasia and a solid-cystic mass measured $28 \times 20 \times 20$ mm near nipple-areola complex (NAC) of the left breast (Fig. 1). Left breast galactography showed mammary duct ectasia and multiple irregular filling defects in the dilated duct (Fig. 2).

The mass and the related mammary duct were excised through a circumareolar incision. Two intraduct neoplastic lesions measured $20 \times 20 \times 20$ mm and $28 \times 20 \times 20$ mm respectively were found in the dilated duct system. The cytology of the nipple discharge showed no tumor cells. The histological examination
showed a biphasic tumor, with mesenchymal and epithelial components (Fig. 3A). The mesenchymal cells highlighted moderate stromal hypercellularity (Fig. 3B), marked nuclear atypia and occasional mitoses (Fig. 3C). Ki67 immunohistochemistry to assess the index of proliferation showed 20% to 30% positive (Fig. 3D). The histologic diagnosis was a borderline PT. The margin of resected tissue showed to be free of tumors.

After operation, the patient is undergoing surveillance through ultrasonography and physical examination every 3 months. Till now, she has been followed up for 12 months, and no local or distant recurrence was observed.

3. Discussion

PTs are unusual neoplasms of the breast, accounting for less than 1% of breast tumors. Most of PTs lesions are detected as unilateral, single, nodular, macro-lobulated and painless mass.[5,6] Bloody nipple discharge caused by spontaneous infarction of the tumors has been reported.[4,7,8] However, PTs with the clinical manifestations of primary nipple discharge are very rare. Only 3 case reports were found through searching of the literature.[9–11] The Pt of breast is thought to arise from the periductal rather than intralobular stroma.[8] Our case represents an unusual macroscopic variant of Pts mimicks intraductal tumor. Its origin is from the periductal stroma of large subareolar dilated duct, and its growth within it explains its gross appearance.[8]

The past history of this patient showed a complex breast fibroadenoma with local lobulated structure on her left breast 4 years ago. Although it is still unclear the exact relationship between PTs and breast fibroadenoma, it is clear that these fibroepithelial tumors share molecular similarities.[12]

WHO has graded PTs to benign, borderline and malignant categories on the basis of stromal characteristics, including the
degree of stromal cellularity and atypia, mitotic count, stromal overgrowth, and the nature of their tumor borders. However, criteria for degrees of stromal hypercellularity and atypia is objective. Moreover, PTs usually show intratumoral heterogeneity, with features of benign lesions in some areas, and borderline and malignant lesions in other areas. A practical approach is to grade a phyllodes tumor as malignant when it shows all of the histological changes of malignancy, and as borderline when not all malignant characteristics are present. In our case, the Phyllodes tumor manifested with stromal hypercellularity and atypia of moderate degree (Fig. 3B), hyperchromatic nuclei, occasional mitoses (Fig. 3C), and Ki67 20% to 30% positivity (Fig. 3D). All of the above suggested a borderline PT.

With regard to treatment of PTs of the breast, surgical excision including local excision, wide local excision, and mastectomy, plays a critical role in the management of PTs. It is recommended that wide local excision with clear margin was the primary approach, because of the insufficient clearance of resection margins related to local recurrence. It is generally agreed that PTs of the breast did not need axillary dissection, which can be explained by the low lymph node metastatic rate. Moreover, the effect of the adjuvant radiotherapy and chemotherapy remains unclear. Based on above statements, in our report, the patient received wide local excision with clear margin. She received neither adjuvant radiotherapy nor chemotherapy after operation. After a follow-up of 12 months, no local or distant recurrence was observed.

4. Conclusion

We reported a case of borderline PTs with multifocal intraductal lesions whose primary clinical manifestation was nipple discharge. There are very few case reports of PTs manifested with nipple discharge in the literature. Most of them are benign PTs. To our knowledge, this is the first report of a borderline PT with primary manifestation of spontaneous nipple discharge.

Author contributions

Xiaoqin Yang designed the research. Pengfei Jing and Bing Wei developed the literature search and collected the photographs. Pengfei Jing drafted the manuscript. Conceptualization: Xiaoqin Yang. Resources: Bing Wei. Supervision: Xiaoqin Yang. Writing – original draft: Pengfei Jing. Writing – review & editing: Pengfei Jing.

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