Recurrent Malignant Phyllodes Tumor of the Breast in a Young Nulliparous Female

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DOI: https://doi.org/10.33545/surgery.2020.v4.i1f.359

Abstract
Phyllodes tumor are fibroepithelial lesions accounting for less than 0.5% of female breast tumors. They are known to recur locally in up to 19% of patients. Surgery is the mainstay of the treatment and inadequate local excision has been associated with local recurrence. The aim of this study is to prevent local recurrence in a post-operative case of recurrent phyllodes tumor of the breast, with histopathology report suggestive of atypical ductal hyperplasia. We present a 23-year-old nulliparous female with recurrence of malignant phyllodes tumor who underwent mastectomy, following which there are no signs of recurrence till date.

Keywords: Phyllodes, recurrence, atypical ductal hyperplasia

Introduction
Phyllodes tumor derived from the Greek word “phyllon” or leaf are fibroepithelial lesions accounting for less than 0.5% of female breast tumors and have potential for recurrence [1]. They are also known as serocystic disease of Brodie. They are known to recur locally in up to 19% of patients. These tumors are more common in 3rd to 4th decade [2]. Role of FNAC in diagnosis of phyllodes tumor remains controversial, however core needle biopsy has a high sensitivity and negative predictive value. They account for less than 1% of all breast neoplasms. The common treatment for phyllodes is wide local excision [3]. Surgery is the mainstay of the treatment and inadequate local excision has been associated with local recurrence. The tumor is poorly responsive to chemotherapy and radiotherapy. Accurate preoperative pathological diagnosis allows correct surgical planning and avoidance of reoperation. We present a young nulliparous female with recurrent malignant phyllodes tumor who underwent wide local excision of benign breast lump in the same breast twice in the past, with an aim to understand the transformation from benign to malignant.

Methods
A 23-year-old nulliparous female presented with lump in left breast since 1 month, that was gradually increasing in size. On local examination there was a 8x7 cm bosselated lump in the lower inner quadrant, freely mobile. There were no palpable axillary lymphadenopathy. Patient reported having undergone 2 previous surgeries of the left breast in the past. Wide local excision was done and the histopathology reports suggested benign phyllodes in the first and borderline phyllodes with atypical ductal hyperplasia and sclerosing adenosis in the second surgery. Immunohistochemistry for ki67 was positive for malignant phyllodes. Patient was advised left simple mastectomy but did not follow up. She presented 6 months after the second surgery. Left simple mastectomy was done with primary closure. Histopathology was consistent with features of malignant phyllodes, margins uninvolved by the tumor. Patient was discharged on the seventh post-operative day after removal of the drain. She is on regular follow up and there has been no signs of recurrence till date.

Discussion
Phyllodes tumors are rare fibroepithelial tumors characterized by a combination of hypercellular stroma and cleft-like or cystic spaces lined by epithelium, into which the stroma classically project in a leaf-like fashion and have a potential to recur and metastasize. They were originally described as “cystosarcoma phyllodes” by Johannes Muller in 1838 [4]. The incidence of
Phyllodes tumor is about 2.1/million, the peak is seen in women aged 45-49 years \[5\]. The tumor is rarely found in adolescents and elderly \[6\]. Clinical presentation is usually a painless rapid growing mass. Triple assessment by clinical, radiological and histological examination forms the fundamental basis for the evaluation of Phyllodes tumor. Images from mammography and ultrasonography should indicate phyllodes tumor if the tumor is a well-circumscribed oval or lobulated mass with rounded borders \[7\]. The clinical course of phyllodes tumor is unpredictable and it is difficult to differentiate between phyllodes tumor and fibroadenoma by imaging and even cytology. In 1981, the World Health Organization (WHO) adopted the term phyllodes tumors and subclassified them into benign, borderline, or malignant tumors according to histopathological characteristics such as stromal cellularity, cellular atypia, mitotic activity, stromal overgrowth, and tumor border \[8, 9\].

Age, tumor size, surgical approach, mitotic activity, stromal overgrowth and surgical margin have been reported as prognosis-predictive factors related to local recurrence \[10\]. Kaprisi et al. \[11\] concluded that tumor size and surgical margins were found to be the principal determinant of local recurrence. Given the high rate of local recurrence and their large tumor size, mastectomy has been the preferred surgical option for malignant phyllodes tumor of breast \[12-15\]. However, breast conserving surgeries have become increasingly common in management of PT for cosmetic reasons. In the past, several literatures recommended that the standard treatment for phyllodes tumor is surgery to remove tumor with a free margin of at least 1 cm to reduce local recurrence \[3, 7\]. Radiotherapy in cases of phyllodes is still controversial \[16\]. There is no consensus on the role of chemotherapy in malignant phyllodes tumor. Some authors suggest adjuvant RT to decrease the possibility of local recurrence in both borderline and malignant phyllodes tumor patients who are treated with breast-conserving surgery \[17\]. Increased p53 protein and Ki-67 antigen expression has been detected in malignant phyllodes tumors and they may be valuable in differentiating fibroadenomas from phyllodes tumors. Furthermore, in phyllodes tumors, p53 and Ki-67 expression has been shown to correlate with negative prognostic factors. Therefore in our case, mastectomy was done due to recurrence, large tumor size, ki 67 positivity and non compliance of the patient.

**Conclusion**

Phyllodes tumor should be accurately diagnosed in the first visit, as these tumors have a high risk of recurrence and malignant potential. This case has brought to attention that there is a need for intraoperative frozen section, close follow up and the importance of ensuring clear surgical margins.

**References**


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**Fig 1:** Pre-operative preparation of the breast lump

**Fig 2:** size of the resected specimen: 15x20cm

**Fig 3:** Left breast tissue with lump
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