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Malignant Phyllodes tumor in a young female: A Case Report

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Abstract

Phyllodes tumor is a rare fibroepithelial neoplasm of the breast and constitutes 0.3–0.5% of all breast tumors. They are usually benign and only few of them undergo malignant transformation. Benign or borderline phyllodes tumor typically never recur however, malignant phyllodes tumors do have a recurrence and a metastatic potential. Differentiating these two groups is of prime importance for proper management of the patient. We report a rare case of Malignant Phyllodes tumor in an 18 year old female.

On cytology, difficulty can occur in differentiating between low grade phyllodes tumor and fibroadenoma as they share many overlapping features. These tumors vary greatly in size and the diagnosis and sub-classification is based upon histological characteristics. Based on the established histological criteria Phyllodes tumors (PT) are classified into three categories—benign, borderline and malignant. In addition to the stromal overgrowth and hypercellularity, malignant phyllodes tumor has infiltrative margins, atypia, and mitotic activity of at least 10/10 HPF.

The extent of the surgery is usually determined by the size of the tumor in relation to the breast. In large multifocal PT, breast—conserving surgery is not recommended because of the risk of inadequate local excision and associated high recurrence.

Benign or borderline phyllodes tumor typically never recur. On the other hand, Malignant phyllodes tumors do have a recurrence and a metastatic possibility. Differentiating these two groups is of prime importance for proper management of the patient. We report a rare case of Malignant Phyllodes tumor in a young female who presented with recurrence within two months of surgery.

Case Presentation

An 18–yr old female presented with a left breast lump since 2 months. The mass gradually increased in size. The patient complained of intermittent pain in the left breast. There was no significant past medical or surgical history. She had no family history of breast or ovarian cancer. Physical examination revealed a 10x10 cm mass which involved the entire left breast. The mass was nodular, firm and was not fixed to the chest wall.

Within two months of surgery patient had a recurrence of a huge breast lump with overlying skin ulcerations and underwent a left simple mastectomy. Patient is currently under follow up period and free of disease. A definitive preoperative diagnosis is of utmost importance in correct surgical management of the patient and in order to avoid local recurrences.

Keywords: Phyllodes tumor, Malignant, recurrent.
Multiple engorged veins were noted on the overlying skin surface. Right breast was normal. There was no axillary or supraclavicular lymphadenopathy. Ultrasound revealed single rounded confluent mass of well defined and regular contours with heterogeneous echotexture and cystic areas were also noted. Fine needle aspiration cytology (FNAC) was performed which revealed many clusters of cells showing moderate nuclear pleomorphism, irregular nuclear membrane and inconspicuous to prominent nuclei. Many singly lying cells were also noted showing pleomorphism, binucleation and multinucleation in a proteinaceous background and a diagnosis suggestive of an atypical cytology (C3) was given. Patient was advised to undergo urgent biopsy. After this a core needle biopsy was performed and a possibility of malignant phyllodes tumor was rendered. Microscopically, the spindle cells showed moderate anisonucleosis (Figure 1-b) and increased proliferative activity with 8–10 mitosis/HPF, Ki-67 showed 20–25% positivity (Figure 1-c). Following this, lumpectomy was performed and intraoperative

Fig. 1.A (4x, H & E stain) and 1.B (40x, H & E stain) – Photomicrograph of core biopsy showing stromal overgrowth, atypical spindle cells and mitotic figures.

Fig. 1.C Immunohistochemical study with Ki–67 to assess the index of proliferation and malignant potential – Photomicrograph showing ki–67 positivity of about 40% of the tumor cell nuclei. (10x, IHC stain). 1.D

Fig. 1.D Photomicrograph showing vimentin positivity. (10x, Vimentin)
touch and crush smears were also prepared. Specimen was sent for histopathological examination. We received a grey–white to grey–brown lobulated mass measuring 12x10x4 cms. On cut section of the mass, multiple cysts were identified largest measuring 7x5 cm and a variegated appearance with greyish white and hemorrhagic areas were also noted. On microscopic examination, hypercellular stroma with marked cellular pleomorphism and atypical mitotic figures were noted (Figure 3–e) and a diagnosis of Malignant Phyllodes tumor was given with involvement of the resected margins by the tumor. Within two months of surgery patient had a recurrence of a huge breast lump with overlying skin ulcerations and underwent a left simple mastectomy. The specimen was sent for histopathological examination, a diagnosis of malignant phyllodes was rendered with deep resected margins and nipple–areola complex free of tumor. Patient is currently under follow up period and free of disease.

Discussion

Phyllodes tumor (PT) is a rare breast tumor comprising of about 0.3–0.9% of all breast tumors. Incidence of PT is 2.1 per one million and it is usually benign occurring over a wide age range with a median age of 45 years.\(^{2,8}\) Its occurrence in a younger age group is quite rare and in our case the patient was only 18 years old. It usually presents as a large, mobile, non–tender mass with a tendency to grow rapidly. It usually involves the left breast more commonly than the right and spares the skin and nipple–areola complex as was seen in our case.\(^{9}\) However in case of large tumors it erodes the overlying skin with ulcerations and in our case also patient had multiple bleeding ulcers on the overlying skin.

According to World Health Organisation PTs are classified into benign, borderline and malignant depending upon the degree of stromal overgrowth, nuclear atypia, mitotic activity and tumor margin status.\(^{10}\) This classification helps in predicting the likelihood of developing local recurrence or metastasis or both. But not all the malignant tumors recur or metastasize and some of the borderline tumors do. Various studies have reported the local recurrence rate of 20% despite the histological classification.\(^{11,12}\) Other studies report recurrence rates of malignant phyllodes tumor in the range of 20–65%.\(^{7,13}\)
In our case, the patient presented with a recurrence of breast lump within two months of surgery.

Important factors which determine the local recurrence rate are tumor size, positive surgical margins, stromal overgrowth, high mitotic count and necrosis. In a study by Oktar et al, stromal overgrowth increased the possibility of local recurrence by seven times whereas surgical margin less than 1cm increases the risk by five times and if the tumor size is larger than 10 cm then the probability of local recurrence is four times greater than the smaller tumors. The median size of PT is 4 cm and around 21% of them grow larger than 10 cm as in our case size of the tumour was about 12cm.

A correlation between MIB1 (Ki–67) positivity and histologically high– grade tumors was demonstrated by Daciec et al. In our case Ki–67 positivity was about 40%.

Diagnostic accuracy of phyllodes tumor preoperatively allows correct surgical management of the patient, avoiding reoperation because of inadequate excision or surgical overtreatment. In our case, the patient was advised a lumpectomy before and on histopathological

Fig. 3.A and B (4x, H & E stain) – Microphotograph showing increased stromal cellularity.

Fig. 3.C (10x, H & E stain) and 3.D (40x, H & E stain) – Microphotograph showing highly atypical spindle cells with bizarre nuclei and frequent mitosis is noted.

Fig. 3E (40x, H & E stain) – Microphotograph showing numerous mitotic figures.
examination there was involvement of all the resected margins by the tumor and therefore within two months of the surgery she presented with a recurrence of a huge breast lump and then she had to undergo a simple mastectomy.

The Paddington Clinicopathologic Suspicion Score outlines the criteria to assist in the selection of patients for core biopsy. Core biopsy is preferred over fine needle aspiration cytology (FNAC) for a correct preoperative diagnosis because of higher false negative findings associated with FNAC and similar cytological features between fibroadenomas and phyllodes tumors. Scolyer et al proposed that the most useful feature in distinguishing phyllodes tumor from fibroadenoma, is the presence of hypercellular stromal fragments and the presence of nuclear atypia helps in differentiating malignant type of phyllodes tumor from the benign ones. Imaging techniques like Ultrasonography and Mammography are also not reliable in distinguishing between benign and malignant phyllodes tumor.

Standard primary treatment for malignant PT is surgical. Preferred therapy is a wide excision with clear surgical margins which is the only proven protective factor. Contraindications for breast conservation surgery include large size of the tumors, multifocality and malignancy because of a higher risk of recurrence. Mastectomy is indicated in cases of giant tumors or if free margins cannot be achieved or in cases of local tumor recurrence. The risk of local recurrence increases in incompletely excised tumors as was seen in our case. Mean time of local recurrence is around 2 yrs according to reported studies, but in our case patient had a recurrence within a short duration of only two months.

Since PT spread through hematogenous route, lymph node metastasis is very low less than 5% thus axillary dissection is not required during the initial surgical intervention. Chemotherapy, hormonal therapy and radiotherapy have no proven benefits in the treatment of phyllodes tumor in regards to its recurrence and metastasis.

Due to its rarity in younger age groups, a subjective case based approach and a regular follow—up is advisable.

**Conclusion**

Malignant phyllodes tumor (MPT) is a very rare malignancy of breast with its tendency to locally recur and metastasize. A rapidly growing mass of the breast may be one of the earliest signs of MPT in a young female and it should be considered so that it can help in prompt management of the patient. It can be easily diagnosed on core biopsy, a definitive preoperative diagnosis is of utmost importance in correct surgical management of the patient and in order to avoid local recurrences.

Declaration of Consent for publication: Written informed consent was obtained from the patient and her parents for publication of this case report and accompanying images.

**References**


