Recurrent Fibroadenoma Turns to be Gaint Phyllodes Tumor: A Case Report

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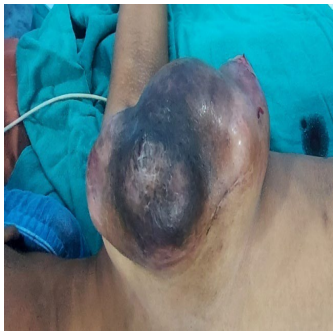
Abstract

Phyllodes tumor of the breast is rare fibroepithelial neoplasm, which accounts for 0.3–1% of all tumors. Few case reports have described the occurrence of giant phyllodes tumor. To our knowledge, about 20% of phyllodes tumors are encounterd as giant. Complete surgical excision is the standard of care for giant benign phyllodes tumors; axillary lymph node metastasis is rare, and dissection should be limited to patients with pathologic evidence of tumor in the lymph nodes. We report a case of 21 years female from remote area of nepal who underwent lumpectomy twice at other centers with diagnosis of recurrent fibroadenoma. She presented at our centre with gaint tumor with Hb % 5.5 gm /dl .Three pint of whole blood was transfused and then simple mastectomy was performed. The pathology reports showed tumor of 4866 gm in weight and 26.2×18.3×7 cm in size and diagnosed as benign phyllodestumor.We do not suggest adjuvant radiation therapy for patients with benign phyllodes tumors that are excised with adequate margin. A review of literature was done for the following patient.

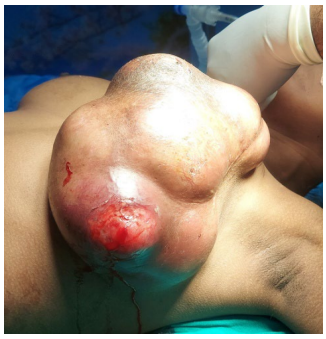
Keywords: Transitional care • Clinical pharmacist • Medication reconciliation • Medication discrepancies

Introduction

Phyllodes tumor of the breast is a rare fibroepithelial neoplasm that accounts for less than 1% (0.3–0.5%) of all female breast neoplasms. The actual incidence of malignant phyllodes tumor is unknown . Metastasis occurs mostly through the haematogenous route, The most common site of distant metastasis for PT appears to be the lungs. Of the 21 studies which mentioned sites of metastasis, 20 documented patients whose PTs had spread were to the lungs. The second most common site for PT metastasis is the bone, with 12 studies containing patients with bone metastases.The term "phyllodes," which means leaf-like, describes the typical papillary projections that are seen on pathologic examination. They were originally called "cystosarcomaphyllodes" by Johannes Müller in 1838 [1-6] . The terminology has since evolved, with over 60 synonyms having been applied to this entity before the term "phyllodes tumors" was adopted by the World Health Organization.Histologically, phyllodes tumors are classified as benign (60%–75%), borderline (15%–20%), or malignant (10%–20%), based upon the assessment of five features: the degree of stromal cellularatypia; the mitotic activity per 10 High-Power Fields (HPFs); infiltrative or circumscribed tumor margins; the presence or absence of stromal overgrowth (i.e., the presence of purestroma devoid of epithelium); and the nature of the tumor borders.Tumor size is variable, ranging from 1 cm to 41 cm (average 4–7 cm). Giant phyllodes tumors are those larger than 10 cm in diameter, and they account for about 20% of all phyllodes tumors . Lymph node metastases are less frequent, although 10.0% – 15.0% of patients may present with clinical lymphadenopathy, they are usually as a result of reactive hyperplasia due to tumour necrosis or infection . In a study conducted by Gullett and colleagues, 9.0% of 1035 cases of patients with PT were subjected to an axillary sampling of ≥10 lymph nodes but nodal involvement was documented only in 9 patients (Figure 1 A-C).



A



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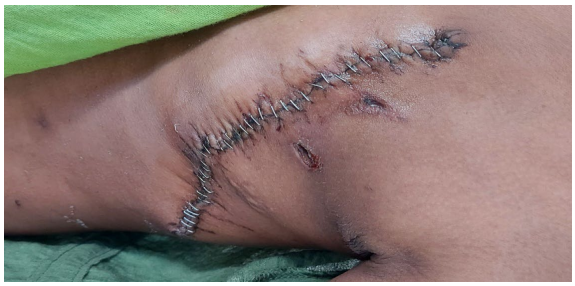

C

Figure 1. A 21 years female present with gaint phyllodes tumor. A and B are preop images, C is post of image.

Phyllodes tumors should be completely excised; axillary lymph node dissection is not necessary. Adjuvant Radiation Therapy (RT) may benefit borderline or malignant, but not benign, tumors. However role of chemotherapy is not advocated.

Here, we report a case of giant benign phyllodes tumor seen at our hospital. After standard mastectomy with adequate free margins, we encounter no complications. The patient recovered well.

Case Report

A 21 years unmarried female presented to our hospital with a giant tumor in her left breast. The patient had noticed the small lump 2 years earlier; she was diagnosed as a case of fibroadenoma and underwent excison twice. One year later, blisters had appeared on the nipple. In the last year, the mass had presented with erythema, a venous network, a foul smell, and skin ulceration with bleeding, and it had also increased considerably in size. Due to the rapid growth, the patient consulted us. The family and personal history did not provide any information relevant to the case. Physical examination revealed an enlarged left breast, approximately 26.2×18.3×7 cm , with irregular margins, erythematous and hyperemic skin, ulceration of the nipple with hemorrhagic discharge, pain on palpation, and the axillary region without nodes. Laboratory tests only showed leukocytosis (12,700) with neutrophilia (87), hb of 5.5gm% ,without any other significant finding. Sonography of the breast showed a huge mass with multiple areas of cystic degeneration alternating with solid tissue, poorly defined margins, irregular

vascularity, and thick hyperechogenic septa. A tru-cut biopsy was performed, and the histopathology results showed a fibroepithelial neoplasm with hyalinized, myxoid and hypercellular stromal areas, compatible with fibroadenoma/ benign disease. The patient underwent blood transfusion and optimization up 7 gm% followed by left total mastectomy. The postoperative period was uneventful; the patient recovered well and she went home after 7 days of hospitalization [7-9]. The pathology results showed a solid, multilobed, and heterogeneous tumor 2866 gm in weight and 26.2×18.3×7 cm in size. Moreover there was a skin ulceration of 8 cm in the nipple. Microscopically, the sections showed a benign mixed fibroepithelial neoplasm with areas of fibroadenoma and benign phyllodes tumor, as well as tumor-free surgical margins. During follow-up, there has been no evidence of local relapse or distant metastases to date.

Discussion

Phyllodes tumors can vary in size but are frequently large, with a median size of 4–5 cm. Few case reports have described the occurrence of giant phyllodes tumors, which are phyllodes tumors of a size greater than 10 cm. Sizes described range from 15 to 50 cm. The tumor described here is one of the largest reported in the literature. 73% of benign phyllodes tumors are smaller than 5 cm, and those that are larger than 7 cm are associated with malignancy. About 20% of phyllodes tumors would be considered giant benign. Phyllodes tumor occurs mainly in women, although there are reports of some cases in men. They can occur in women of a median age at presentation of 42–45 years (range 10–82), about 15–20 years later than fibroadenomas. In men, phyllodes tumors usually occur in association with gynecomastia. Higher-grade tumors are more common in older patients. Genetic risk factors for phyllodes tumors are largely unknown, but the literature describes phyllodes tumors in Li-Fraumeni syndrome patients and a mother-daughter pair. Stromal induction of phyllodes tumors can occur due to growth factors produced by the breast epithelium and stromal expression of endothelin-1, insulin-like growth factors (IGF-I and II), and epithelial overexpression of Wnt5a in benign/borderline phyllodes tumors.

Trauma, pregnancy, increased estrogen activity, and lactation occasionally have been implicated as factors stimulating tumor growth . Phyllodes tumors may grow slowly or rapidly or exhibit a biphasic growth pattern. As they grow larger, phyllodes tumors can form a visible mass that distorts the contour of the breast or even cause pressure necrosis of the overlying skin. Unlike breast carcinomas, phyllodes tumors start outside of the lobules and ducts, in the breast's connective tissue, called the stroma, which includes the ligaments and fatty tissue that surround the lobules, ducts, and lymph and blood vessels in the breast. Phyllodes tumors can also contain stromal cells. They most likely develop de novo, although there have been reports of progression of fibroadenoma to phyllodes tumor.

On examination, most patients have a smooth, multinodular, well-defined, firm mass that is mobile and painless. Shiny, stretched, and attenuated skin may be seen overlying a large tumor. Nipple retraction, ulceration, chest wall fixation, and bilateral diseases are rare (33%), but have been described for phyllodes tumors. The most frequent location is in the right breast, being multicentric in a third of cases; 35% are in the upper external quadrant, 15% in the upper internal quadrant, 10–25% in the lower external quadrant, and fewer than 10% in the lower internal quadrant. Although palpable axillary lymphadenopathy can be identified in up to 20% of patients, most cases are reactive; metastatic involvement of lymph nodes with phyllodes tumor is rare. Phyllodes tumors should be suspected when a patient presents with a large (>3 cm), rapidly growing breast mass that is usually palpable. Although imaging features of a phyllodes tumor can be suggestive of fibroadenoma, the large size and history of rapid growth indicate otherwise [10-12].

Regarding treatment, complete surgical excision is the standard of care for phyllodes tumors, and with greater than 1cm margins is often curative and reduces the risk of local recurrence . Mastectomy is generally not indicated for benign phyllodes tumor, unless negative margins cannot be achieved and/or if a tumor is so large that breast-conserving surgery would result in suboptimal cosmetic outcomes. A 2019 meta-analysis of 54 observational studies also found that a positive margin only correlated with a higher local recurrence risk of malignant, but not of benign and borderline, phyllodes tumors . Surgical margins of greater than or equal to 1 cm have been associated with a lower local recurrence rate in borderline and malignant phyllodes . When adequate surgical margins cannot be achieved because of tumor location, adjuvant RT should be administered, even after mastectomy. However, if adequate surgical margins can be achieved, there is less agreement about the need for adjuvant RT. We base our decision about adjuvant RT on tumor grade; thus, we do not suggest adjuvant RT for patients with benign phyllodes tumors that are widely excised, whereas we suggest adjuvant RT for patients with borderline or malignant phyllodes tumors following surgical excision. Axillary lymph node involvement by phyllodes tumors is rarely reported, even when tumors are malignant. In the SEER database study, only 8 of 498 women with known lymph node status had involved nodes . Thus, axillary surgery is rarely indicated in patients diagnosed with phyllodes tumors. Due to scarce data, the role of systemic chemotherapy in phyllodes tumors is limited. Patients with benign or borderline phyllodes tumors are usually cured with surgery and should not be offered chemotherapy unless they develop unresectable metastases. Based on experience and limited data, we recommend adjuvant chemotherapy only to a small minority of patients with high-risk (>10-cm) or recurrent malignant phyllodes tumors who have excellent functional status and minimal comorbidities, and only after a thorough discussion about the risks, benefits, and controversial nature of such treatment. When systemic chemotherapy is indicated, malignant phyllodes tumors should be treated according to protocols designed for soft tissue sarcoma rather than breast cancers. Hormone therapy is not effective against phyllodes tumors.

When phyllodes tumors recur, they typically recur locally within 2 years of the initial excision. Some series have found that the time to local recurrence was shorter for malignant than for benign or borderline tumors. Although recurrences typically have the same grade as the original tumors, there have been several case reports of benign tumors transforming into malignant ones upon recurrence . Despite the best surgical efforts, phyllodes tumors are known to recur locally at rates that vary with tumor grade. As an example, a 2019 meta-analysis of 54 retrospective studies reported an overall local recurrence rate of 12% (95% CI 10–14), as well as pooled local recurrence rates of 8, 13, and 18% for benign, borderline, and malignant tumors, respectively. Local recurrences generally develop within 2–3 years.

The impact of histology on survival was explored in the Sarcoma and Phyllodes retrospective (SAPHYR) study. The overall 3-year survival rate for combined benign and borderline tumors was 100% . The overall 3-year survival rate for malignant phyllodes tumors was 54%, similar to that for non-angiosarcoma primary breast sarcomas (60%). Also, the 5-year overall survival rate for patients with benign/borderline and those with malignant tumors was 91 and 82%, respectively.

Conclusion

Phyllodes tumors are uncommon fibroepithelial breast tumors that are capable of a diverse range of biologic behaviors. Giant phyllodes tumors account for about 20% of all phyllodes tumors. Given the rarity of the disease, treatment principles are based mainly on retrospective series and case reports. Mastectomy is the standard of care for giant benign phyllodes tumors.

Acknowledgments

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Ethics Statement

The patient gave informed written consent to publish her case (including the publication of images).

Conflict of Interest Statement:

The authors have no conflicts of interest to declare.

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