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A giant malignant phyllodes tumor with lymph node metastasis: A case report

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Abstract

Phyllodes tumors are fibro-epithelial neoplasms that account for 0.3% to 0.9% of all breast neoplasms. They are mostly benign, but can be malignant having a hematogenous metastasis. Usually they are small but can reach huge sizes. Axillary lymph nodes metastasis is rare and routine axillary lymph node dissection is not usually recommended. Within this content we present a case of a 32 years old woman with an 18 months history of a huge left breast mass that was found to have a 29 cm phyllodes tumor with 10 lymph node metastasis.

Keywords

Phyllodes; giant; lymph node; metastasis.

Introduction

Phyllodes tumor was first described by German physician Johannes Müller in 1838 as cystosarcoma phyllodes despite being rarely malignant and having uncommon cystic component [1]. The term phyllodes tumors was first adopted by the WHO in 1981 and currently these tumors are divided into as benign, borderline and malignant based on the stromal component [2].

Phyllodes tumors are defined as a fibro-epithelial neoplasm composed by a double layered epithelial component forming clefts with a hypercellular stroma elaborating leaf-like structures. They are mostly benign but recurrences are common. Following a diagnoses of malignant phyllodes tumor, the hematogenous metastasis are rare [3]. Phyllodes tumor account for 0.3% to 0.9% of all breast neoplasms [4,5] . The mean age of presentation is 40 to 50 years with malignant tumor developing 2 to 5 years later than benign tumors [3]. The average tumor size is around 4 cm with less than 10% of tumors growing more than 10 cm. These are defined as giant phyllodes tumors and very rarely they can reach 48 cm [6].

Axillary lymph nodes metastasis is rare and routine axillary lymph node dissection is not usually recommended [7-9]. The first method of management of malignant phyllodes tumor is surgery [10] .

Here we present the case of a giant malignant phyllodes tumor (The postoperative specimen measured 30 cm) with 10/35 metastatic lymph nodes.

Case Presentation

A 32 years old woman presented with an 18 months history of a huge left breast mass. The patient had neglected her rapid growing mass due to religious concerns. There was no personal history of breast cancer. She had no significant medical history. On physical exam the mass was huge, mobile, well-circumscribed, cystic solid in consistency and non-tender and occupied almost the whole breast. The overlying skin was erythematous with normal nipple areola complex. The right breast was normal (Figure 1A, 1B, 1C, 1D).

PET scan was done as out and showed locally invasive large tumor with no distant metastasis. A left radical mastectomy and axillary node dissection was performed.

Grossly the specimen weighed 6200 grs and measured 30 X 25 X 16 cm. The nipple was fixed. Slicing revealed a tumor which occupied almost the whole breast, measuring 29 X 22.5 X 14.5 cm (Figure 2). The tumor was heterogeneous composed of myxoid and cystic necrotic pattern invading all margins except the deep one. This tumor ulcerated the skin. Several hard-white nodules, well limited at 1cm from the tumour, were observed.

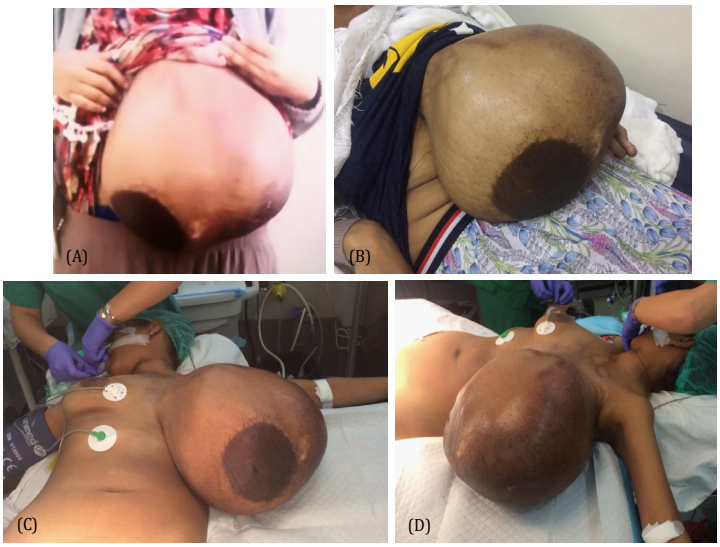

Figure 1 (A,B,C,D): Preoperative clinical photographs showing a marked asymmetric enlargement of the left breast due to a huge, mobile, well-circumscribed mass with erythematous overlying skin occupying almost the whole breast.

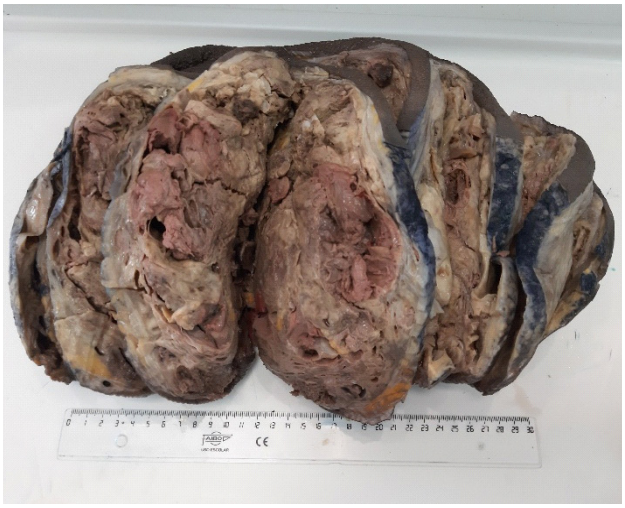

Figure 2: Macroscopic appearance showing a 30 X 25 X 16 cm tumor, occupying almost the whole breast.

Histological examination showed a poorly limited and infiltrating neoplastic proliferation, formed of a double component. The first was epithelial made up of stretched lactiferous ducts coated with a double epithelial layer. The second component was stromal, showing spindle cells with atypical nuclei (Figure 3A, 3B) and many mitotic figures (> 10/10HPF) (Figure 4). Large areas of necrosis were seen throughout the tumour (Figure 5). No heterologous component was found. Tumor implants were found elsewhere in the

breast parenchyma and in the adipose tissue at the level of axillary dissection, in addition to neoplastic vascular emboli near and at a distance from the tumor. This tumor infiltrated the skin which was largely ulcerated and necrotic but the nipple was free. The surgical limits were in pathological zone. An adjacent fibroadenoma was infiltrated by the tumour. The three dissected lymph nodes were reactive.

The immunohistochemical study showed that the epithelial component expressed AE1/AE3. Desmin was negative. The Ki67 proliferation index was estimated at around 60% (Figure 6).

The final diagnosis was made as malignant phyllodes tumour, and the patient received radiotherapy.

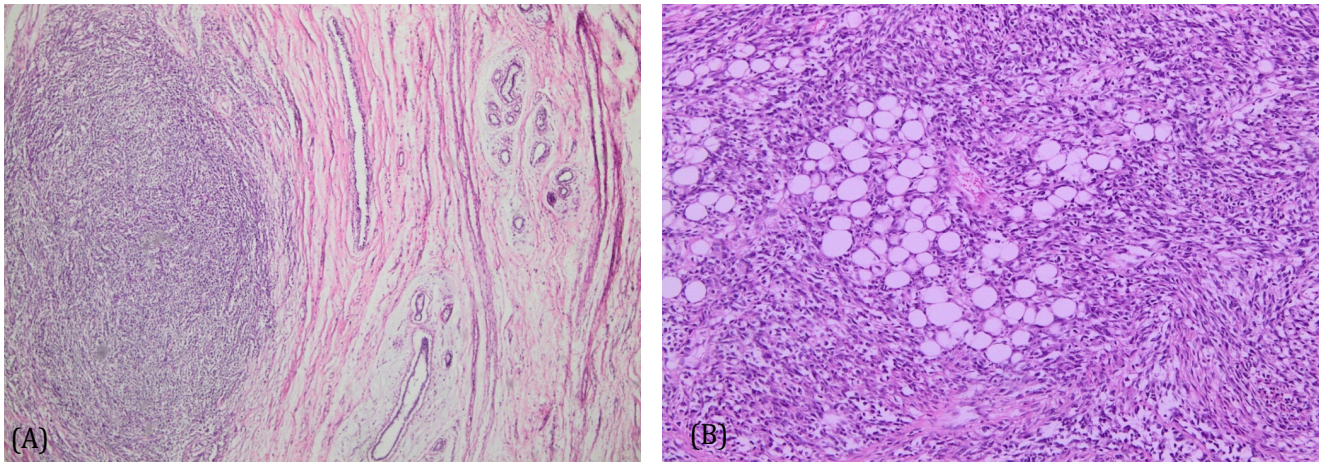

Figure 3: (A) Low power, magnification showing a poorly limited and infiltrating neoplastic proliferation. **(B)** High power magnification highlighting the double component of the tumour, epithelial and stromal with spindle cells and atypical nuclei.

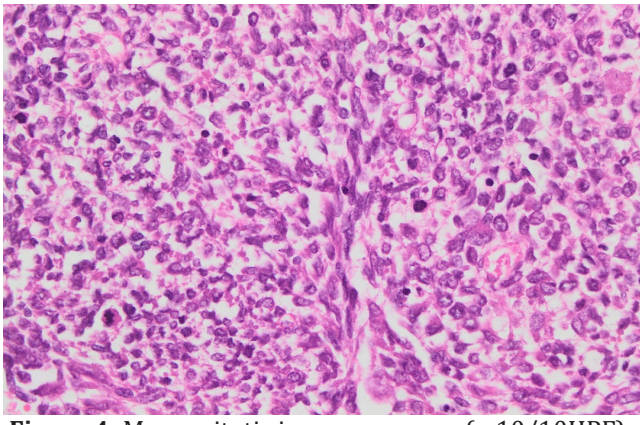

Figure 4: Many mitotic images are seen (> 10/10HPF).

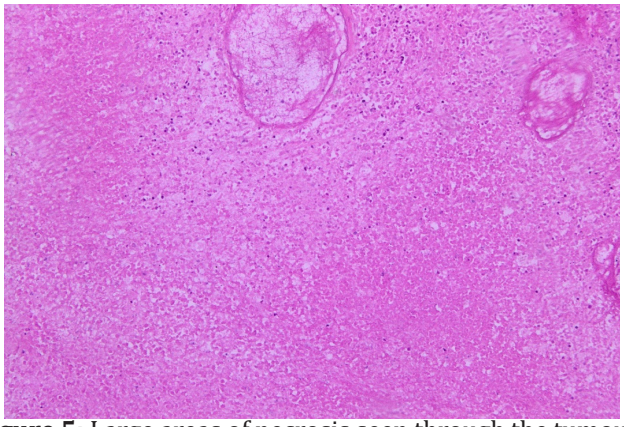

Figure 5: Large areas of necrosis seen through the tumour.

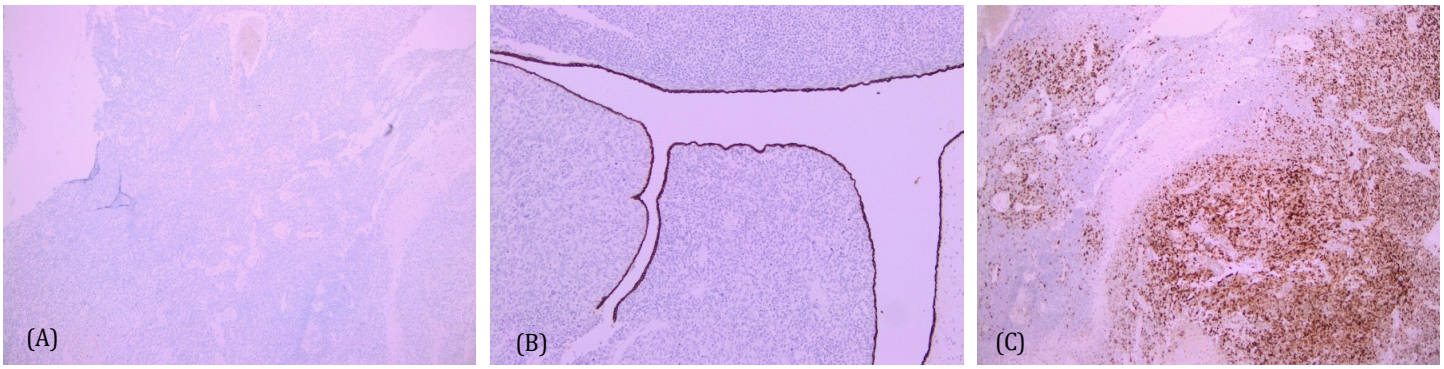

Figure 6: The immunohistochemical study done: **(A)** Desmin was negative, **(B)** Epithelial component expressing AE1/AE3. **(C)** The Ki67 proliferation index was estimated at around 60%.

One month later, she was reoperated due to the positive surgical margins of the first operation. Histological examination showed the presence of several residual nodules in the breast parenchyma. The tumor was at less than 0.1 cm of the superior margin, while the others margins were clear. 35 lymph nodes were dissected, 10 of which were infiltrated by the tumor (Figure 7A, 7B).

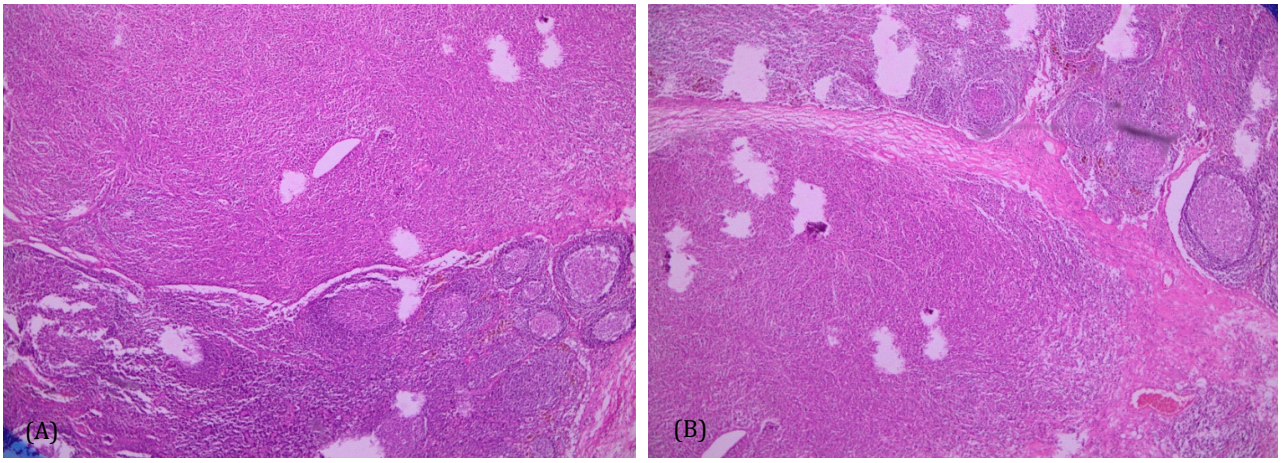

Figure 7 (A-B): Lymph nodes invasion by the tumor.

Discussion

Phyllodes tumors are defined as well circumscribed fibro-epithelial neoplasm. They are divided into benign, borderline and malignant based on the degree of stromal hypercellularity, mitoses and cytological atypia, stromal overgrowth and nature of the tumour margins. They are mostly benign but recurrences are common [3]. Despite being benign tumors, they have always been a great deal of controversy among fibroepithelial tumors of the breast because they might acquire malignant features [11]. These tumors account for 0.3%-0.9% of all breast neoplasms of which 10%-30% are malignant. The median age of presentation for malignant phyllodes tumors is 50 years. Among malignant phyllodes tumors 80% present with localized disease, 8.2% with regional disease or lymph nodes involvement and 1.5% with metastasis [11].

Clinically it usually presents as a rapidly growing unilateral painless breast mass. The skin overlying the tumor may have a blue discoloration with dilated veins but ulceration is very rare. A high number of patients have a history of fibroadenoma [5] . In fact some suggest that fibroadenoma may be the possible precursor of phyllodes tumor [12]. The mean size of phyllodes tumors is around 4 cm. These tumors can reach sizes up to 40 cm and are therefore called giant phyllodes tumors [5,6]. Recently studies reported many giant phyllodes tumour (Hsu et al., 30 X 25 cm [13]; Tarun et al., 50 X 25.2 cm [14]; Ramesh sarvanandan et al., 40 X 35 cm [15]; Mohammed A. Sbeih et al., 25 X 20 cm [16]; Tapanutt Likhitmaskul et al., 20 X 20m [17]; Dong Xia et al., 47.5 X 37 cm [18]; Rumi Khajotia et al., 24 X 22 cm [19]; Banno A. et al., 30 cm [20].). Our patient presented with one of the largest giant phyllodes tumours measuring 29 X 22.5 X 14.5 cm. Furthermore malignant phyllodes tumor present with larger tumor size as compared to benign [21].

Radiologically malignant phyllodes tumors often mimic fibroadenomas. On mammography, a phyllodes tumor typically appears as a round or lobulated mass, with well-defined margins, heterogeneous internal structures. The presence of internal septations are more in favour of phyllodes tumors than fibroadenomas [22]. On ultrasound, the presence of fluid-filled spaces, lobulations, and marked posterior

acoustic enhancements are used to differentiate phyllodes tumors from fibroadenoma. Additional features of malignant phyllodes tumors are the irregular shape, larger size, and the presence of cystic spaces [23]. Imaging play an important role as a first diagnostic tool, however only biopsy can give a final diagnosis [24].

Histopathologically, in benign phyllodes tumor the stroma is more cellular than in fibroadenomas with spindle cell shape and no nuclear atypia. The mitosis are rare (<5 per 10 high-power fields). Stromal heterogeneity can be found (hyalinization or myxoid changes). Malignant phyllodes tumors are a combination of nuclear pleiomorphism and stromal overgrowth with increased mitosis (>10 mitoses per 10 high-power field), stromal hypercellularity and infiltrative tumor borders [3,4]. When malignant heterologous component is present the tumor is classified as malignant regardless of other histological features [3,25].

Metastasis in a phyllodes tumor is almost always haematogenous [3,4]. Lymph node metastasis is rare and axillary lymph node dissection should be restricted only for rare patients showing proven metastatic lymph nodes [5,8,9,21,25,26]. In fact, a retrospective review of 172 Cases done by WEI-HONG CHEN et al in 2005, showed no axillary lymph node metastasis in the 44 patients with lymph node dissection [9]. Moreover, a retrospective analysis of 150 patients with phyllodes tumor done by Ramakant P et al in 2013, axillary lymph node dissection showed only one positive nodal metastases out of 26 patients that was from infiltrating duct carcinoma while all the rest were negative only showing reactive hyperplasia [21]. Furthermore in a clinic-pathologic study of 77 cases of phyllodes tumor in a Hispanic cohort in 2015 done by Carlos Andres Ossa et al all axillary lymph nodes were negative [27]. In this case the patient had 10/35 metastatic lymph nodes.

The main treatment of phyllodes tumor is surgery with complete excision, an adequate margin, and mastectomy is required only if a malignant phyllodes cannot be adequately excised [24,27].

Local recurrence occurs in patients with positive surgical margins, and in patients with breast conserving therapy [24]. Also, local recurrence was higher in giant phyllodes tumors and in malignant ones [21]. Therefore, when a malignant histology and a positive margin are present patients should undergo further surgery to obtain negative margins [28]. In our case the patient was diagnosed as a giant malignant phyllodes tumor positive margins and was later reoperated to achieve adequate surgical margins.

Radiotherapy is used to minimize local recurrences mostly in recurrent tumors, tumors with close margins, malignant histology and in case of conservative surgery [21,27].

The role of adjuvant therapies such as radiotherapy and chemotherapy is controversial and depends on the patient's status [26].

Large tumour size, malignant histotype and breast conserving surgery are associated with increased risk of distant metastasis [24,29].

Conclusion

In summary, we report an extremely rare presentation of a neglected giant malignant phyllodes tumor with 10 positive lymph nodes. Giant phyllodes tumor should not be neglected but treated early and further studies on axillary lymph node dissection should be done for better management.

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