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Giant Mammary Phyllodes Tumour: A Case Report and Review of The Literature

Meriem SERRAJ ANDALOUSSI[1]*, Soukaina SABIR[1], Mustapha BENHESSOU[1], Mohamed ENNACHIT[1], Mohamed ELKERROUMI[1], Ayoub KHOAJA[2], Mohamed KARKOURI[2]

[1]Mohamed 6 center for cancer treatment1, *Ibn Rochd University Hospital Center*, Casablanca, Morocco
[2]Central Pathological Anatomy Service, Ibn Rochd University Hospital Center, Casablanca, Morocco

***Corresponding author:** Meriem SERRAJ ANDALOUSSI, Mohamed 6 center for cancer treatement, Ibn Rochd University hospital center, Casablanca, Morocco.

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Abstract

Phyllodes tumors are rare fibroepithelial lesions that usually reach 4 cm in size, but tumours> 10 cm in size have been described, known as giant phyllodes tumours. We report the case of a 42-year-old female patient who presented to the Mohamed VI centre for cancer treatment at the university hospital of CASABLANCA with a mass of the left breast. Clinical examination revealed a large, hard mass taking up the whole breast, with shiny skin ulcerated in places and collateral circulation. The trucut biopsy showed a myofibroblastic-like mesenchymal component with a focus of necrosis that may be consistent with a mesenchymal component of a phyllodes tumour or a mesenchymal proliferation. A simple mastectomy was performed and the pathological examination revealed a morphological appearance of a borderline phyllodes tumour according to the 2012 WHO classification.

Keywords: *Phyllodes tumour, giant, borderline, Fibroadenoma.*

Introduction

Phyllodes are rare tumours with an incidence of about 2.1 per million, peaking in women aged 45-49 years. Their name comes from the Greek word "phyllon" meaning a leaf. It is also known as Brodie's disease and accounts for 0.3 to 0.5% of female breast tumours. Phyllodes tumours usually reach a size of 4 cm, with tumours larger than 10 cm being referred to as "Giant Phyllodes Tumours". These neoplasms were first described by Johannes Müller in 1838 and are broadly classified according to WHO guidelines into benign, borderline and malignant types (1). Preoperative pathology diagnosis allows correct surgical planning and also avoids reoperation. There are no convincing data to recommend adjuvant treatment after surgery. A large excision with an adequate margin is necessary to reduce the risk of local recurrence, but if the tumour is giant and involves the whole breast, excision is difficult and a mastectomy should be considered.

Reconstruction of a large defect is difficult and requires careful planning (2). In contrast, phyllodes tumours that are benign on clinical, radiological and cytological examination are often indistinguishable from fibroadenomas and can be cured by excision, which may be either a simple wide excision or a mastectomy provided the margins are respected (1).

Observation

MK, 42 years old, single, without any particular pathological history, presented to the Mohamed VI centre for cancer treatment at the university hospital of CASABLANCA with a mass in the left breast, discovered 2 months before her consultation, increasing in volume progressively, without any signs of inflammation or nipple discharge. The clinical examination notes asymmetrical breasts with a left breast with a voluminous mass taking up the whole breast, hard, with shiny skin ulcerated in places and collateral circulation (figure 1). The rest of the examination was unremarkable.

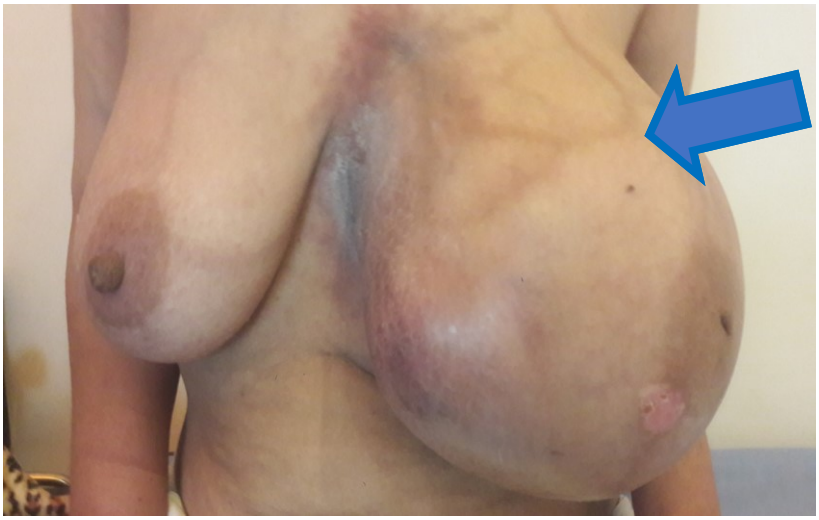

Figure 1: Mass in the left breast with shiny skin ulcerated in places and collateral circulation.

Despite the use of high constants for left breast mammography the radiographs obtained onthe left breast were not interpretable, the mammogram of the right breast being without abnormalities. Breast ultrasound revealed a large, well-circumscribed mass in the left breast measuring 188 x 152 x 97 mm that was echogenic, heterogeneous and without posterior acoustic attenuation; no detectable tissue mass or cystic formation on the right, nor axillary adenopathy bilaterally (Figure 2).

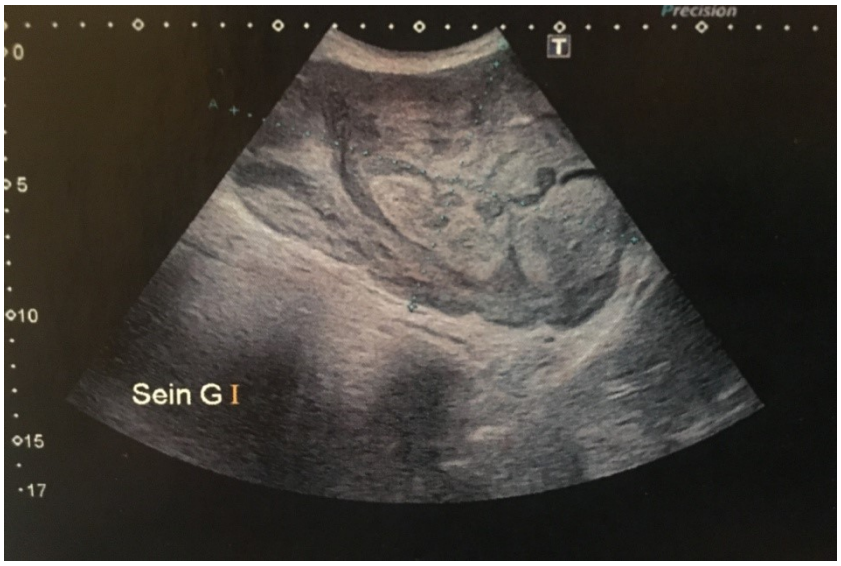

Figure 2: large, well-circumscribed mass in the left breast measuring 188 x 152 x 97 mm.

The trucut biopsy showed a myofibroblastic-like mesenchymal component with a focus of necrosis that may be consistent with a mesenchymal component of a phyllodes tumour or a mesenchymal proliferation whose type and malignancy cannot be established on this specimen.

There was no axillary lymph node involvement on either side. Surgery consisted of wide resection of the large mass (Figure 3 and 4) and the specimen sent for histopathological diagnosis. Pathological examination of the surgical specimen revealed an encapsulated polylobed nodular formation weighing 2411g and measuring 20x17x13cm. The histological examination found a well-limited proliferation with a double fibroepithelial component. The mesenchymal component is made up of a very cellular stroma often occupying a whole field at low magnification. The fibroblasts show moderate nuclear atypia. The mitotic index is estimated at 8 mitoses per 10 fields at high magnification x10 (figure 5) and x20 (figure 6). No stromal necrosis was recognised. Focal points of haemorrhagic suffusion are also seen. The epithelial component is made up of ducts of variable size, with a regular lining. In total: a morphological aspect of a borderline phyllodes tumour according to the 2012 WHO classification.

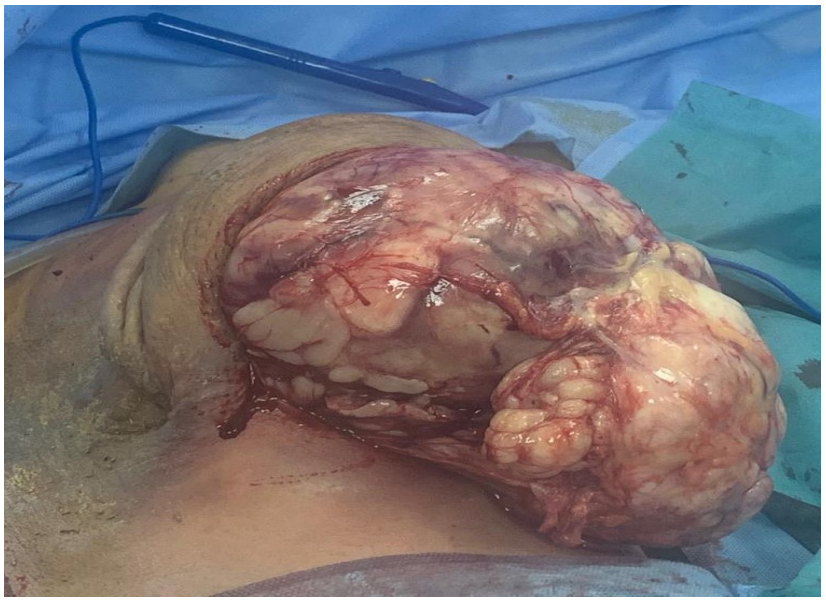

Figure 3: Intraoperative photograph of the large mass removal.

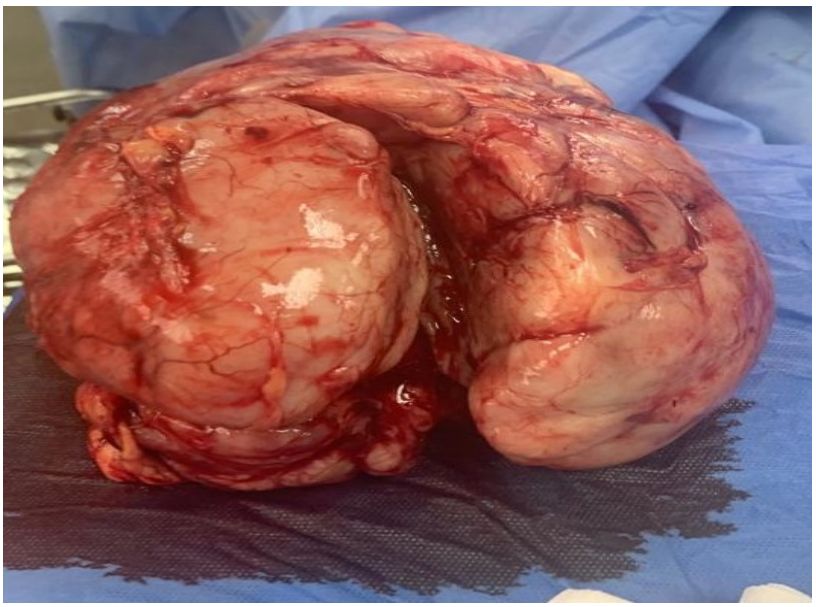

Figure 4: Surgical specimen as a large, firm, lobulated, well-circumscribed mass with a beige-brown appearance.

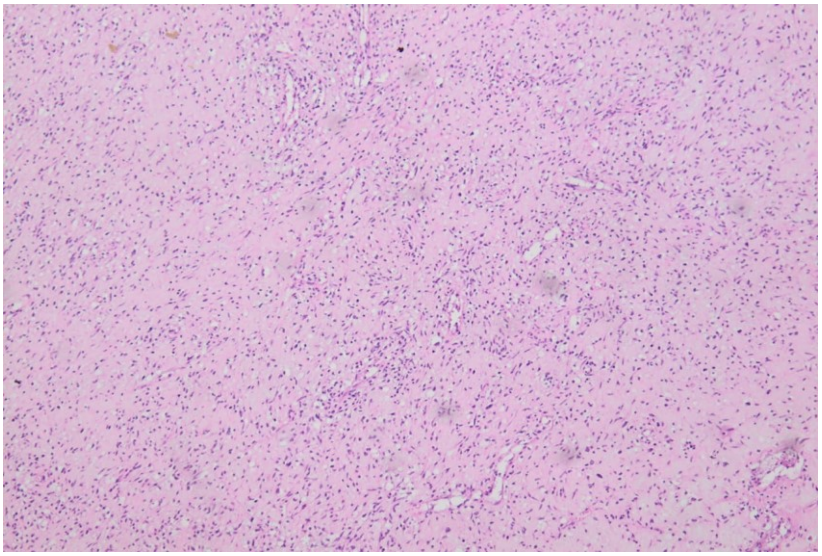

Figure 5: Histological aspect of a phyllodes tumourGrossingx10 (Laboratory of anatomopathology CHU IBN ROCHD).

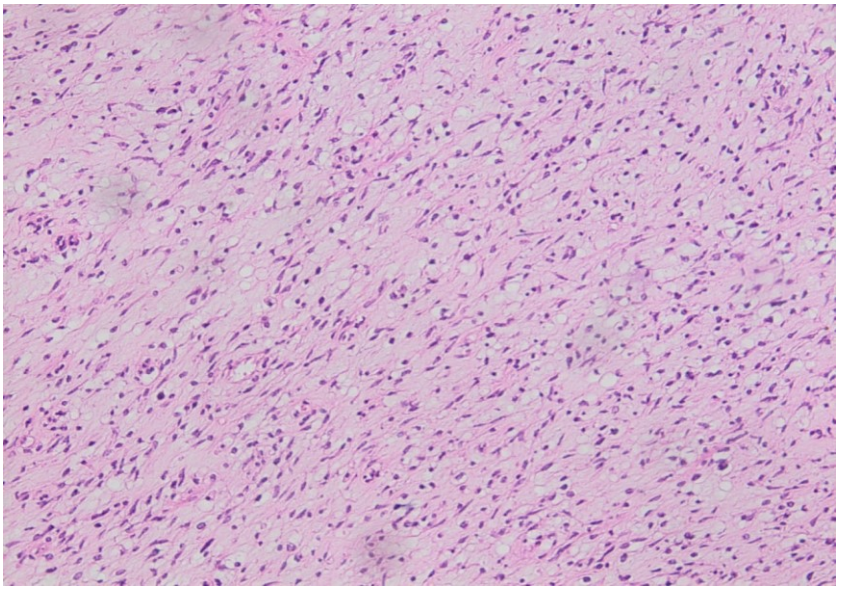

Figure 6: Histological aspect of a phyllodes tumour (Laboratory of anatomopathology CHU IBN ROCHD).

Discussion

Phyllodes tumours of the breast are rare fibroepithelial lesions containing both stromal (connective) and glandular (lobules and ducts) tissue (3).

It is a rare neoplasm, accounting for 0.3% to 1% of all breast tumours (4) and typically presents as a large, painless breast mass stretching the overlying skin. The average size is 4-5 cm and 20% of tumours exceed 10 cm (5).

Tumours larger than 10 cm are called giant tumours and are found in 20% of phyllodes (6) and take up the whole breast as in our patient's case.

The lesion mimics the benign fibroadenoma from which it is important to differentiate. Phyllodes tumours contain a wide spectrum ranging from benign to malignant via borderline according to characteristics such as tumour margins, stromal proliferation, tumour necrosis, cellular atypia and the number of mitoses per high power field (7). In our patient, this was a borderline phyllodes tumour.

Women between 35 and 55 years of age are most often affected, the median age of presentation being 45 years (8) which is exactly the age of our patient.

Chelius in 1827 first described this tumour (9). Johannes Muller was the first person to use the phyllodes term cystosarcoma. It was thought to be benign until 1943, when Cooper and Ackerman reported the malignant potential of this tumour. In 1981, the World Health Organisation adopted the term phyllodes tumour and, as described by Rosen, sub-classified them histologically as benign, borderline or malignant according to previously cited characteristics (10). Due to limited data, the percentage of benign versus malignant phyllodes is not well defined. Reports suggest that approximately 85-90% of phyllodes tumours are benign and 10-15% are malignant (11).

At present, the exact etiology of the phyllodes tumour and its relationship to the fibroadenoma is unclear. It has been suggested that in a proportion of fibroadenomas, a somatic mutation may result in monoclonal proliferation, histologically indistinguishable from the polyclonal element, but with a tendency for local recurrence and progression to a phyllodes tumour, which has also been supported by clonal analysis (12).

Macroscopically, benign phyllodes tumours show a well circumscribed solid lobulated mass with a beige-brown appearance on the cut surface, which is similar to a fibroadenoma.On the section of our patient's surgical specimen, the pathologist also found a firm beige-white appearance with haemorrhagic, myxoid and calcific changes.

Larger tumours of the malignant type tend to develop haemorrhagic and necrotic areas with curved projections in the breast parenchyma; unlike phyllodes, fibroadenomas have a true capsule.

A benign phyllodes tumour is characterised by slightly increased stromal cellularity and irregular borders. A fibroadenoma, on the other hand, may have increased stromal cellularity and a circumscribed border that cannot be assessed by biopsy (1).

In our patient, the trucut biopsy revealed a myofibroblastic mesenchymal component with a focus of necrosis that may be consistent with a mesenchymal component of a phyllodes tumour or a mesenchymal proliferation whose type and malignancy could not be established on the specimen.

Therefore, it can be difficult to distinguish between a fibroadenoma and a phyllodes tumour at biopsy. Furthermore, the distinction between benign and borderline phyllodes using microscopic criteria (7) can be very subjective and therefore the precise diagnosis is usually made only on the surgical specimen. A malignant phyllodes tumour can be distinguished from a benign phyllodes tumour by the presence of marked cellularity, cellular atypia, permeative margins and a mitotic activity of

at least 10/10 HPF. In our patient the mitotic index is estimated at 8 mitoses per 10 fields at high magnification.

Most fibroadenomas have polyclonal elements and are considered hyperplastic rather than neoplastic lesions (13). Phyllodes tumours have a similar polyclonal element, but a somatic mutation is thought to occur, resulting in monoclonal proliferation (13). Stromal proliferation can also occur as a result of induction of growth factors in the mammary epithelium. Increased endothelin-1 levels have also been shown in phyllodes tumours (14,15). Trauma, lactation, pregnancy and increased oestrogenic activity have sometimes been implicated as factors stimulating tumour growth. Genetic predilection, namely Li-Fraumeni syndrome, is the most commonly cited genetic alteration in phyllodes tumours (16).

Although imaging can help in the diagnosis in some cases, mammography often does not contribute to the diagnosis. Reports using ultrasound characterise a phyllodes tumour as a solid lobulated nodule with sharp contours and heterogeneous echostructure, which may be associated with cystic components (17). Biopsy remains the cornerstone for establishing the diagnosis.

It is the preferred means of preoperative diagnosis for giant breast tumours with a sensitivity of 99%, a negative predictive value of 93% and a positive predictive value of 83%, respectively (18).

Surgery is the mainstay of the management of phyllodes tumours. Generally, either breast conserving surgery is performed, as was done in our patient despite the size of the mass, with skin reduction, or mastectomy without axillary curage. In case of a large excision, the tumour must be resected with a margin of more than 1 cm. Routine axillary dissection is not recommended because phyllodes spread only by the haematogenous route, whereas spread to the axillary lymph nodes occurs in less than 1% of patients (19).

In our patient's case, a mastectomy was required due to the large size of the tumour. The treatment of choice for borderline and malignant phyllodes is simple mastectomy. Malignant phyllodes are more likely to recur after breast-conserving surgery than benign types (20). In some cases, immediate breast reconstruction, particularly a rotation flap, may be performed at the time of mastectomy for skin closure or cosmetic purposes (19).

The role of adjuvant radiotherapy is still uncertain and under investigation. It is generally recommended for the positive postoperative surgical margin and for local control of borderline and malignant phyllodes (21). The results of adjuvant radiotherapy are encouraging in patients with high-risk features such as large tumours, hypercellular stroma, high nuclear pleomorphism, high mitotic rate, presence of necrosis and increased vascularity within the tumour (21). Therefore, administering adjuvant radiotherapy in a healthy margin resection may provide an effective means of local control of borderline and malignant phyllodes tumours. The role of chemotherapy remains unclear but its use may be considered in malignant phyllodes. Adjuvant chemotherapy using doxorubicin and ifosfamide with 6 cycles and a 28-day interval between each cycle has been performed with promising results (22). The lung is the most common metastatic site, followed by bone and visceral locations (23).

Conclusion

The diagnosis of a phyllodes tumour should be made in all patients presenting with progressive hypertrophy of a breast mass. Preoperative anatomopathological diagnosis allows a correct surgical management based on a large exeresis and also avoids a re-operation. Radiotherapy has precise indications or it allows a local control of these tumours avoiding metastatic evolution. Delayed management can lead to a progression of the disease and increase morbidity and mortality.

Conflicts of interest

The authors declare no conflicts of interest.

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