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The largest and neglected giant phyllodes tumor of the breast—A case report and literature review

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ABSTRACT

INTRODUCTION: Phyllodes tumours are rare fibro-epithelial lesions. The role of the pathologist in the preoperative diagnosis of phyllodes tumours of the breast is critical to appropriate surgical planning. Wide local excision or mastectomy with adequate margin remains the treatment of choice. Local recurrence occurs in approximately 10- 16.1% of patients (Wei et al., 2014) [1] and distant metastases occurs in 6.3–31% of patients with malignant phyllodes tumours (Wei et al. (2014), Chaney et al., 1998) [1,2] but only in 4% of all phyllodes tumours (Salvador et al., 1989) [3]. Emphasis should be given in early diagnosis and intervention to decrease morbidity and mortality.

1. Case presentation

A 44 year old Indo-Trinidadian female was referred to the Breast Clinic of San Fernando General Hospital of Trinidad and Tobago. On examination there was a 24 × 20 cm firm lobulated mobile mass in her right breast with no skin changes, no nipple discharge and her axillary lymph nodes were not palpable. She had no associated risk factors for breast cancer and no family history of breast and ovarian cancer. A bilateral mammogram of breast revealed 20 × 20 cm lobulated mass on right breast with architectural distortion and asymmetry (Fig. 1). Colour Doppler image shows increased vascularity of the mass but no specific features suggestive of malignancy. A needle core biopsy revealed breast tissue with a stromal fragment with occasional mitoses suggestive of phyllodes/fibroadenoma. The patient was counselled for mastectomy but she refused and sought help from alternative remedies despite several attempts at counselling. The patient returned back after one year. At that time, she was emaciated/malnourished and anaemic with a fungating mass (approximately 50 × 50 cm) occupying over the entire right chest wall with enlarged prominent cutaneous blood vessels (Fig. 2a, b). The patient had palpable right axillary lymph nodes and her left breast examination was normal. A repeat needle core biopsy revealed a benign phyllodes tumour with extremes infarction. A CT scan of the chest revealed a heterogeneous enhancing mass in right breast with ill- defined deep margin at the RUOQ with few enlarged axillary lymph nodes with no suspicious features and there were no obvious chest wall or pulmonary metastasis were noted (Fig. 3). CT abdomen and pelvis showed no liver and bony metastasis.

Informed consent was obtained from the patient. Right wide mastectomy with partial excision of pectoralis major with axillary lymph nodes sampling (Figs. 4 and 5) and immediate latissimus dorsi (LD) flap closure (Fig. 6) was performed. The dissection was very tedious as the blood vessels were grossly dilated under the skin. The patient had a successful postop recovery (Fig. 7). Histology of the specimen revealed a low grade (benign) phyllodes tumour with scanty mitoses and minimal nuclear atypia (Fig. 8a, b). Six (6) axillary lymph nodes were identified with reactive changes and no metastasis. All resection margins were otherwise negative. The case was discussed at our MDT meeting and decision was made for long term follow up with no further adjuvant therapy.

After several months, the patient was readmitted with a massive pleural effusion on the contra-lateral side of the tumour. Cytology of the pleural fluid revealed malignant cells. The original histopathology of the specimen was once again reviewed by two senior pathologists, but failed to diagnose any malignancy. Despite further treatment patient demised after 6 months from her initial surgery.

2. Discussion

Phyllodes tumours represent a broad range of fibro-epithelial diseases. They make up 0.3–0.5% of female breast tumours and have an incidence of about 2.1 per million [4]. Most of the tumours arise in women aged between 35 and 55 years, approximately 20 years

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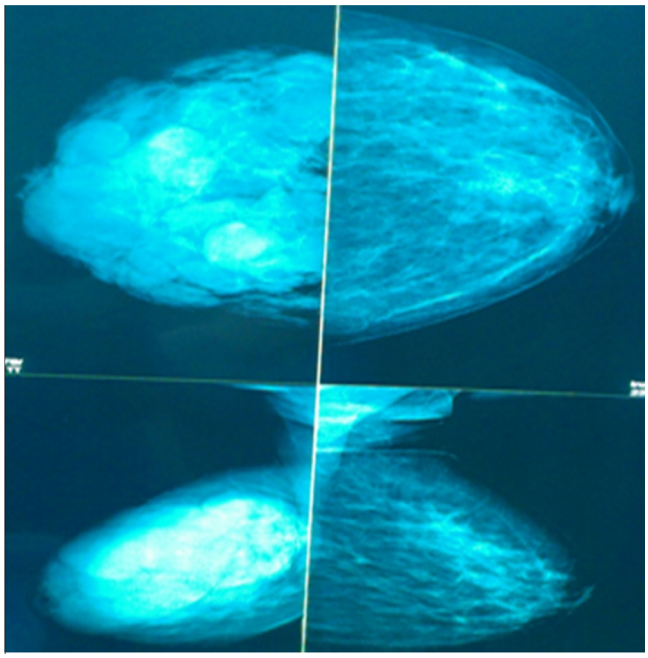

Fig. 1. Initial mammogram shows lobulated mass on right breast with architectural distortion.

later than fibro-adenoma. The tumour is rarely found in adolescents and the elderly [3,5]. It is more prevalent in the Latin American, White and Asian populations.

Chelius in 1827 first described this tumour [6]. Johannes Muller was the first person to use the term cysto-sarcoma phyllodes. It was believed to be benign until 1943, when Cooper and Ackerman reported on the malignant biological potential of this tumour. In 1981 the World Health Organization adopted the term phyllodes tumour and as described by Rosen sub-classified them histologically as benign, borderline, or malignant according to the features such as tumour margins, stromal overgrowth, tumour necrosis, cellular atypia, and number of mitosis per high power field [7]. Because of limited data, the percentage of benign versus malignant phyllodes tumours is not well defined. Reports suggest that approximately 85–90% of phyllodes tumours are benign and 10–15% were malignant [8].

At present time, the exact aetiology of phyllodes tumour and its relationship with fibro-adenoma are unclear. It has been suggested that, in a proportion of fibro-adenomas, a somatic mutation can result in a monoclonal proliferation, histologically indistinguishable from the polyclonal element, but with a propensity to local recurrence and progression to a phyllodes tumor, which has also been supported by clonal analysis [9].

Classically, patients present with a firm, mobile, well defined, round, macro-lobulated, and painless mass [10,11]. The median size of phyllodes tumours is around 4 cm; however, <10% of tumours grows larger than 10 cm which are rare and these have been defined as giant phyllodes tumour and very rarely they can be up to 48 cm in diameter. In 10–15% cases axillary lymph nodes are palpable but only <1% had pathological positive nodes. In recent years several giant phyllodes tumour have been reported (Hsu et al., 30 × 25 cm; Tarun et al., 50 × 25.2 cm; Juliana Alves De Suza et al., 14 × 10 cm; Ramesh sarvanandan et al., 40 × 35 cm; Junaid Nabi et al., 9 × 8 cm; Mohammed A. Sbeih et al., 25 × 20 cm; Tapanutt Likhitmaskul et al., 20 × 20m; Dong Xia et al., 47.5 × 37 cm; Rumi Khajotia et al., 24 × 22 cm; Banno A. et al., 30 cm) [12–21].

Our patient presented with one of the largest giant phyllodes tumour in the English literature, measuring 50 × 50 cm. It was a fungating mass, fixed to the skin, but mobile over the underlying

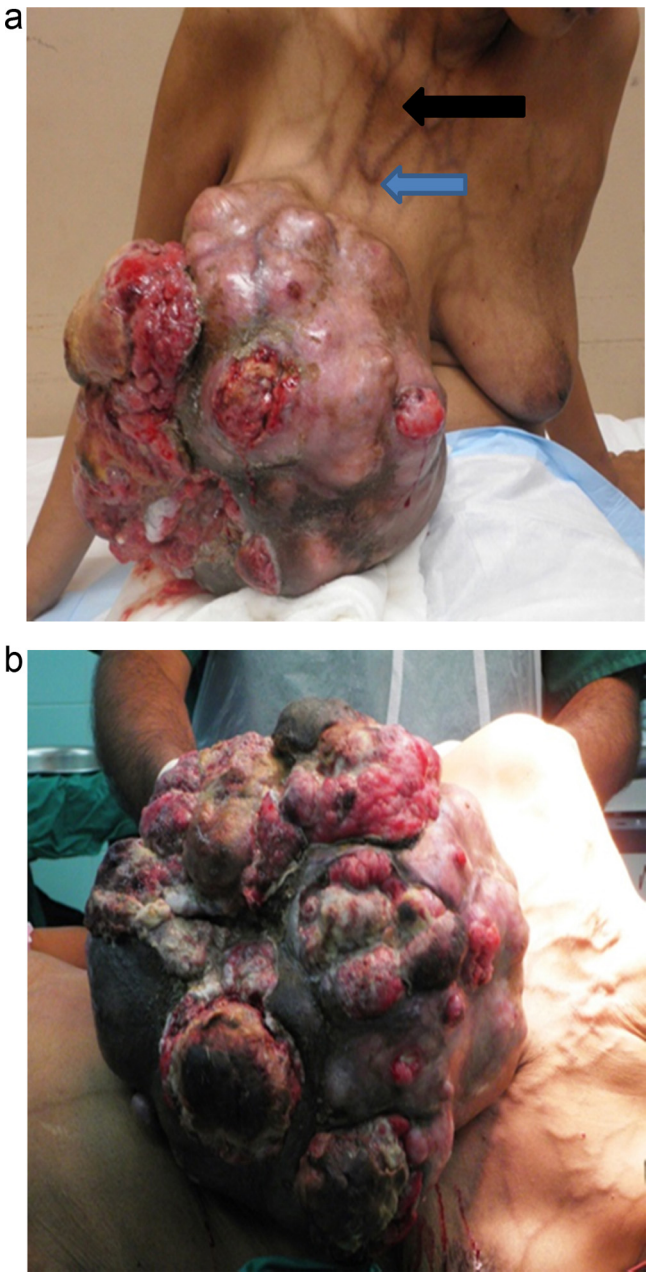

Fig. 2. (a) Giant phyllodes tumour with prominent dilated subcutaneous vessels.(b) The fungating giant phyllodes tumour. (a, b): The fungating giant phyllodes tumour with prominent dilated subcutaneous vessels.

muscle and her axillary lymph nodes were palpable. There were multiple hugely dilated feeding blood vessels to the tumour, which can be a challenge surgically and have to be looked for especially subcuatenous veins and perforating intercostal vessels.

A variety of techniques have been utilized to improve the pre-operative diagnosis of phyllodes tumours, such as ultrasound, mammogram, MRI and core needle biopsy. Ultrasound and mammography are the most commonly used first line investigation for this disease. There are no pathognomonic mammographic or ultrasound features of phyllodes tumour [22].

Core tissue biopsy represents the preferred means of pre-operative diagnosis for giant breast tumours with a sensitivity of 99% and negative predictive value and positive predictive value 93% and 83%, respectively [23].

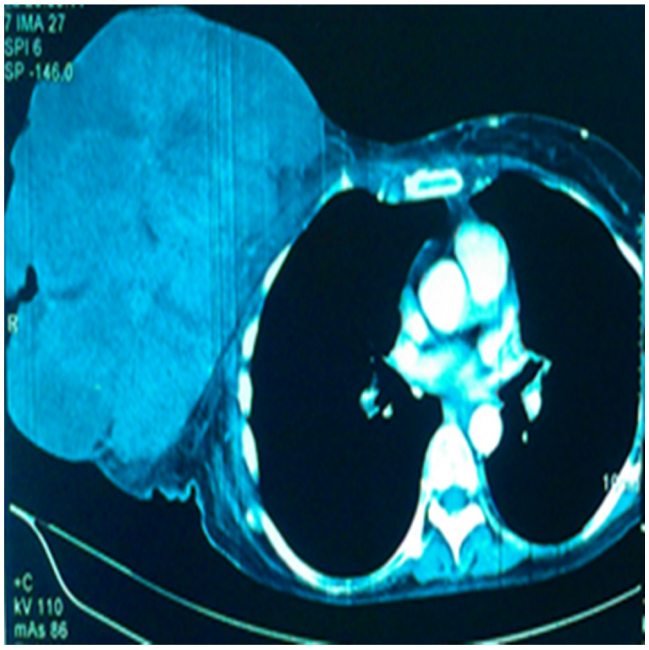

Fig. 3. CT chest showing heterogeneous enhancing mass in right breast with ill-defined deep margin.

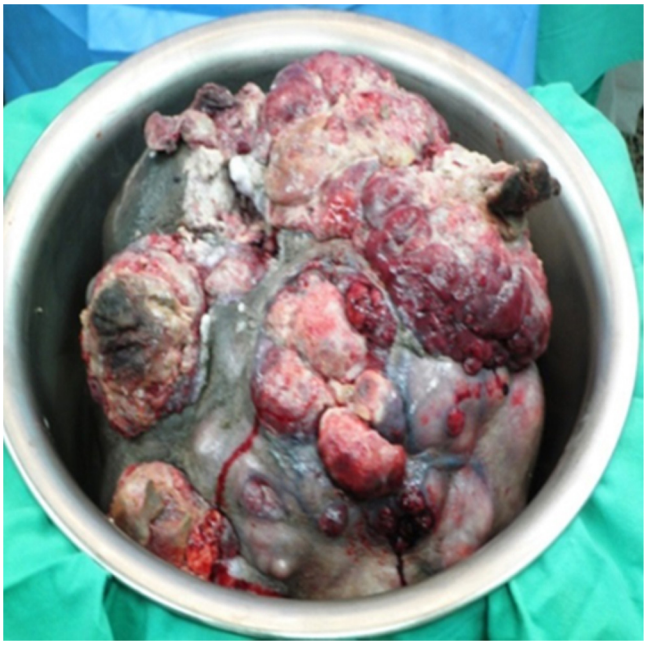

Fig. 5. Phyllodes tumor after excision.

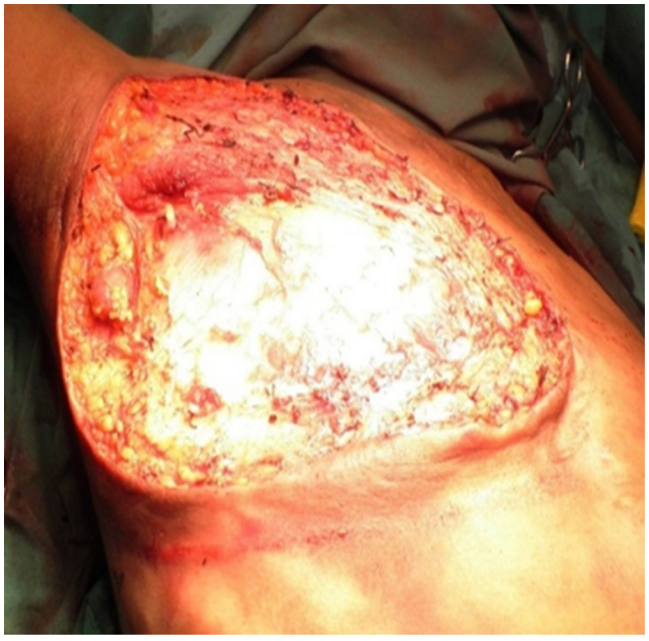

Fig. 4. Chest wall after excision of phyllodes tumor.

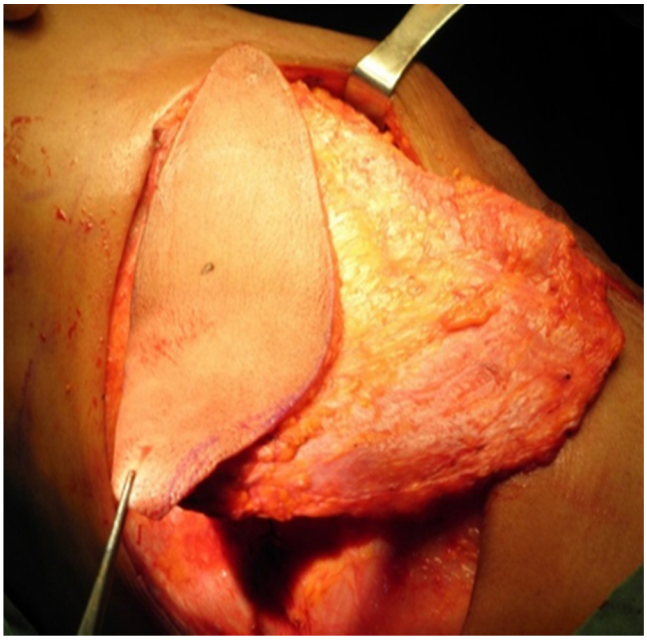

Fig. 6. LD flap to cover the anterior chest wall.

Surgical management of the phyllodes tumour includes either breast conserving surgery or mastectomy with or without reconstruction [24–26]. Most experts currently advocate that surgeons obtain at least 1 cm margins on primary excision or re-excision of a tumour removed with close margins, as long as the tumour to breast size will permit [2,3,10,27,28]. Mastectomy should be reserved for larger tumours between 5 and 10 cm in diameter [11–21,24,25] and should be considered in recurrent tumours, and especially of the malignant tumours [3].

Closure of wound after excision of a giant phyllodes tumor remains a great challenge for the surgeons. There is no contraindication to immediate reconstruction after mastectomy in cases of giant phyllodes tumour, and this decision can be made solely based upon patient preference. Most often immediate reconstruction of the chest wall was performed with prosthetic implant or chest wall are covered with splits skin graft, marlex mesh or latissimus dorsi muscular/myo-cutaneous flap; if the fascia or muscle is infiltrated [10–21,25,29–31].

The role of adjuvant radiotherapy and chemotherapy remains uncertain, but consideration can be given for their use in cases of malignant phyllodes tumours [2,32,33].

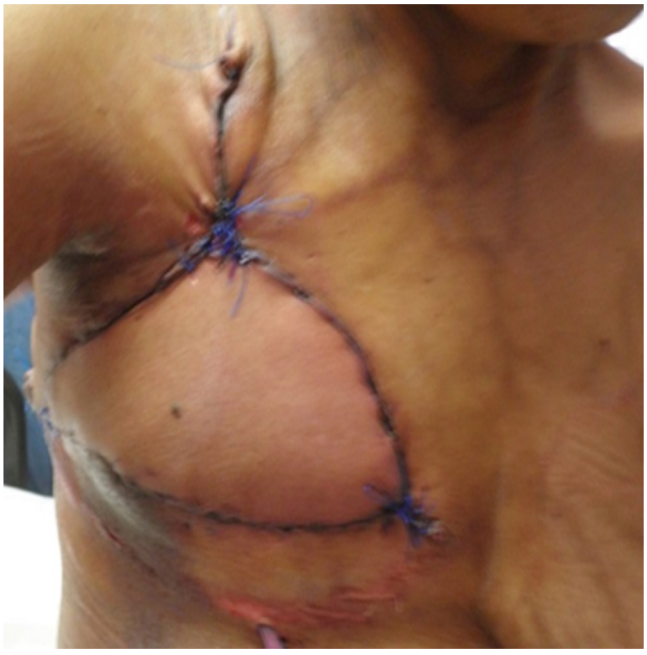

Fig. 7. Photograph taken on postop follow up.

3. Conclusion

The diagnosis of phyllodes tumor should be entertained in all patients presenting with progressive enlargement of breast lump. Emphasis should be given on proper health education on breast cancer and the negative consequences of alternative or delayed treatment. Excision should be done as soon as possible, as unnecessary delay can leads to diseases progression and increase morbidity and mortality.

Conflicts of interest

The authors declare no conflicts of interest.

Author contribution

All authors have contributed significantly in designing and organizing to write manuscript, collecting data as well help in critical analysing the manuscript. All authors have approved the final version of this manuscript

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Consent

Patient consent was obtained.

Ethical approval

No ethical approval is required to publish this case report.

Guarantor

The corresponding author will accept the full responsibility for the work.

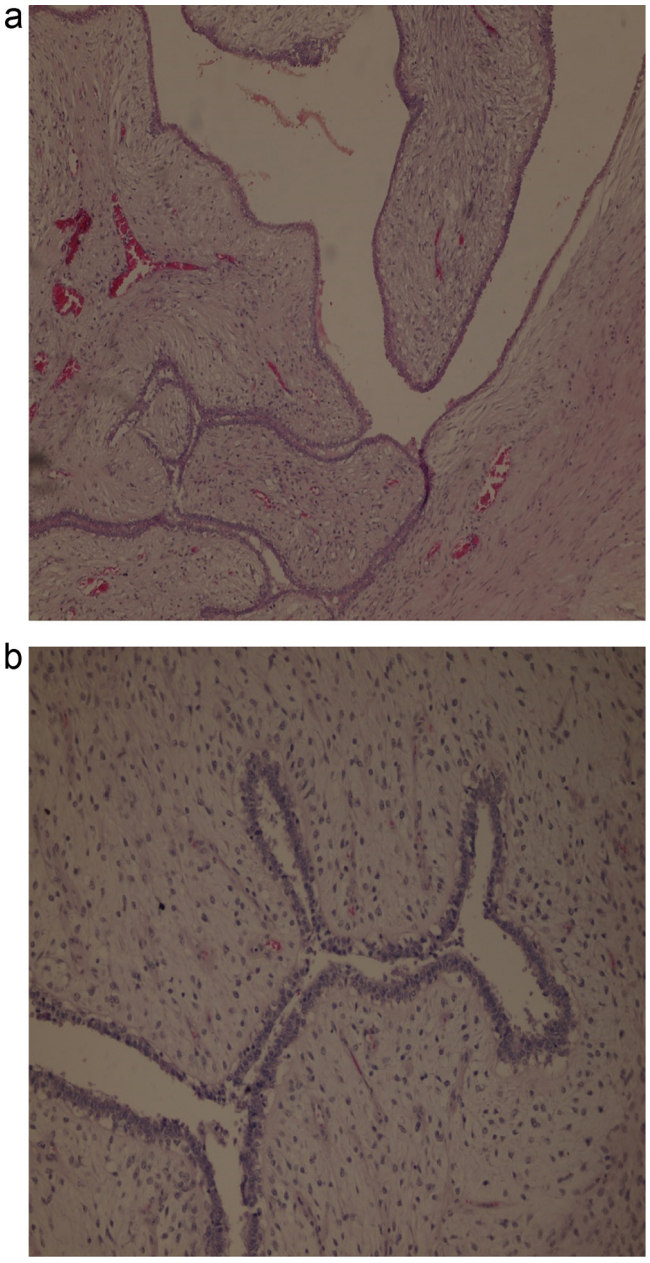

Fig. 8. (a) Leaf like growth pattern with hypercellular stoma characteristic of benign phyllodes. (b) Stromal cellularity with scanty mitoses and minimal nuclear atypia.

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