

CASE REPORT

Recurrent giant phyllodes tumour in a 17-year-old female: a rare case report

James J. Yahaya^{1,2,*}

¹Department of Pathology, Makerere College of Health Sciences (MakCHS), Makerere University, Kampala, Uganda, ²Department of Biomedical Science, College of Health Sciences (CHS), University of Dodoma, Dodoma, Tanzania

*Correspondence address. Department of Biomedical Science, College of Health Sciences (CHS), University of Dodoma, PO Box 395, Dodoma, Tanzania. Tel: +255 685 820 546; E-mail: jayame76@gmail.com

Abstract

Phyllodes tumours (PTs) are rare fibroepithelial tumours of the breast with incidence accounting for <1% in the general population. Availability of reports on PTs with size of 31 cm or more in diameter in the literature are extremely rare. Herein, the case of a 17-year-old female patient with a giant recurrent right breast PT is reported. Histologically, the tumour showed proliferation of stromal tumour cells consisting of spindle cells with uniform nuclear chromatin, inconspicuous nucleoli, abundant eosinophilic cytoplasm and proliferation labelling index for Ki67 of <10%. PTs require meticulous surgical excision due to the fact that they have a high rate of recurrence and status of surgical margins must be included in the pathology report. This is because recurrent cases of PTs tend to grow faster than the primary ones with a high chance of transforming to malignancy.

INTRODUCTION

Phyllodes tumours (PTs) are rare fibroepithelial tumours of the breast with incidence accounting for <1% of all breast tumours and 2.5% of fibroepithelial tumours, and they are classified as benign, borderline or malignant [1]. These tumours are fast growing tumours and can reach a median size of 4 cm and sometimes can grow up to 50 cm [2]. There are few reported cases of PTs in adolescents. The mean age of PTs at diagnosis is 45 years (range 35–55 years) and commonly occur in third and fourth decades [2, 3]. Until 2015, there were only 20 cases of PTs in adolescents that were already reported in the literature [4]. Approximately 70% of PTs are benign; however, 20% of them undergo malignant transformation whereas 10% are usually borderline at diagnosis [3]. Herein, an adolescent female with a highly recurrent and benign giant PT is being reported.

CASE REPORT

A 17-year-old female was brought as an outpatient in the department of general surgery with a 2-year history of a huge

ulcerating, painful and tender right breast mass. On physical examination, she was ill-looking, febrile and mildly wasted. She reported four different excisions in the past 2 years. Two excisions were done at a health centre and unfortunately the excised tumour masses were not evaluated histologically due to lack of pathology laboratory. The subsequent two excisions after recurrence were done at two different private hospitals. One histological report indicated fibroadenoma and the fourth report revealed benign PT. The right breast was very huge with ulcerating skin surface and was forming nodules (Fig. 1A). Ultrasound revealed a tumour, which measured 31 × 30 × 21 cm, calcified with regular borders and not fixed to the underlying tissue, but the left breast was normal. Chest X-ray showed clear lungs. Both fine-needle aspiration cytology and mammography could not be done because were not available. Modified radical mastectomy was performed by involving a significant amount of pectoralis major muscle. Macroscopically, the tumour was extensively necrotic, haemorrhagic and nodular. Grossly, the surgical margins were 3.4 cm far from the tumour (Fig. 1B). On microscopic examination, the tumour was hypercellular and was composed of uniform and slender spindle cells, and it

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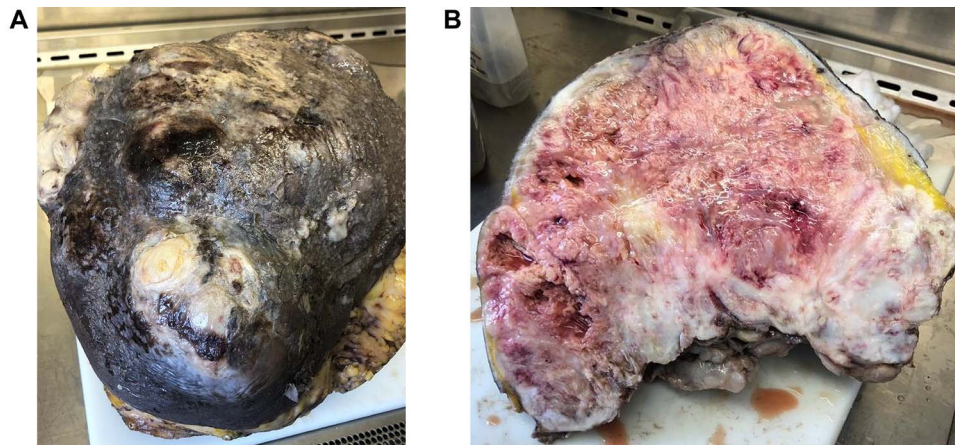


Figure 1: (A) Clinical appearance of the lesion; (B) cut surface of the tumour is showing haemorrhage and necrosis.

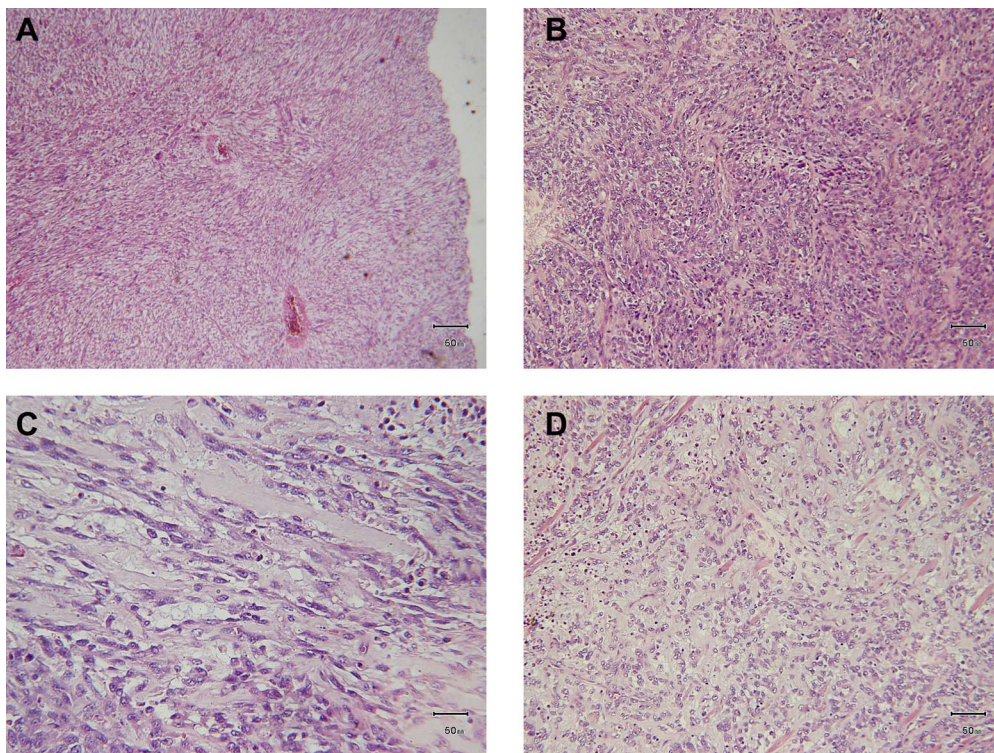


Figure 2: (A) Stromal hypercellularity of the lesion. The proliferating spindle cells are uniform and slender with vascularization in some areas (haematoxylin and eosin staining, $\times 100$); (B) the packed hyperchromatic tumour cells are showing streaming of the cytoplasm in some areas with few mature lymphocytic infiltrate (haematoxylin and eosin staining, $\times 200$); (C) the cells have spindle nuclei and some with prominent nucleoli and marked hyalinization (haematoxylin and eosin staining, $\times 400$); (D) the plasma cells are proliferating in a stroma with marked oedema and vascularization (haematoxylin and eosin staining, $\times 200$).

was forming fascicles (Fig. 2A). The cells had uniform vesicular nuclear chromatin and abundant eosinophilic cytoplasm (Fig. 2B). A few mitotic figures < 5 per 10 high-power fields were also seen (Fig. 2C and D). All ER, PR, HER2, SMA, S-100, vimentin and P53 antibodies were negative except Ki67 that showed proliferation labelling index of $< 10\%$. After a postoperative period of 8 months, she was found to be free from local recurrence or metastasis following evaluation with chest and abdominal computed tomography scan.

DISCUSSION

Patients with PT usually present with round, mobile and painless breast lumps, which are more commonly in the upper outer

quadrant with equal chance of the two breasts to be involved [5]. About 20% of the patients have palpable axillary lymphadenopathy and a significant proportion of the patients have history of fibroadenoma with up to 20% of them being with high local recurrence rate [2].

Recently, a systematic review and meta-analysis study done by Lu et al. in China reported the pooled local recurrence rates for benign, borderline and malignant PTs to be 8, 13 and 18%, respectively. This shows that the tendency of PTs to recur locally increases with increase in aggressiveness of the tumour. The study also reported that mitoses, tumour border, stromal cellularity, stromal atypia, stromal overgrowth and tumour necrosis can independently predict development of local recurrence [5]. The categorization of PTs into

benign, borderline and malignant was described in the study by Salvadori et al. [6].

Confirmation of diagnosis of PTs is done by histological examination [7]. Mammographically, PTs are clearly defined masses with a smooth and occasionally lobulated border, whereas on ultrasound they often show smooth contours with low-level homogenous internal echoes, intramural cysts without posterior acoustic enhancement [6]. Mammographically, both PTs and fibroadenoma show coarse microcalcification, but 'malignant' microcalcification in PTs is rare [6]. It has been found that the presence of both epithelial and stromal elements within the cytological smear supports the diagnosis of PTs and not fibroadenoma [8].

Surgery is the initial treatment for PTs with preferred wide excision for clear surgical margins, particularly for malignant ones [6]. The mean time for local recurrence has been reported to be around 2 years. This is different from the present case in which there were three consecutive local recurrences within a period of 2 years [8]. Options for failure to obtain negative surgical margins include re-excision, radical resection of the chest wall and reconstruction using a composite mesh (inner polytetrafluoroethylene and outer vypro), pedicle latissimus dorsi flap and a split skin graft for the recurrent [5].

Chemotherapy, hormonal therapy and radiotherapy have no proven benefits in the treatment of PT in regards to its recurrence and metastasis [9]. In a study by Kario et al. [10] reported that stromal overgrowth increases the possibility of local recurrence by seven times, whereas surgical margin <1 cm increases the risk by five times.

The case described in this report sheds light on the importance of evaluating histologically all excised breast lumps in order to confirm the diagnosis and not generalizing that every benign looking breast lump is fibroadenomas. This may help to avoid missing of the proper diagnosis as it was in the present case. This report also explains the importance of wide local excision for giant PTs (>10 cm in diameter), which helps to prevent local recurrence.

In summary, PTs are extremely rare in adolescents and they grow rapidly with a high risk of local recurrence. Ensuring wide excision during surgery is of utmost importance for obtaining negative surgical margins in order to prevent possibility of local recurrence.

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CONFLICT OF INTEREST STATEMENT

None declared.

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None declared.

ETHICAL APPROVAL

No ethical approval was needed.

INFORMED CONSENT

The author confirms that he had obtained a written informed consent from the patient's parents for publication of the case details and any accompanying images. A copy of this consent is available upon request by the Editor-in-Chief of this journal for review purposes.

GUARANTOR

Dr J.J.Y. is the guarantor of this paper.

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