

CASE REPORT

Breast Necrosis: A Rare Case of Neglected Phyllodes Tumour

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ABSTRACT

Phyllodes tumour (PT) of the breast is firstly described in literature as early as 1838. This article reports the youngest recorded case of giant complicated PT and the role of toilet mastectomy as a salvage surgery. A 19-year-old lady presented with a gigantic lump of the left breast for 6 months which was rapidly growing (20 x 20 cm) and complicated with a foul-smelling blackish discoloration of the breast skin and tissues. Toilet mastectomy was performed as a result. The specimen weighed 4.0 kg with histopathological report of the tumor being borderline phyllodes. Herein, we describe a case of PT, who presented with breast necrosis and we discuss its medico-social aspect of it.

Keywords: Phyllodes tumour, necrosis, mastectomy

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INTRODUCTION

Phyllodes tumor (PT) was firstly described by Johannes Muller in 1838 and has been a quagmire in both diagnosis and treatment (1). Being a disease of middle-aged women, it is also found to develop at earlier age in the Asian populations¹. The Malay race is reckoned as the highest number of cases according to a local study (2). PT has a distinct biphasic characteristic of epithelial and stromal components, similar cytologically to fibroadenoma. Unlike breast carcinoma, PT usually originates from the stromal layer which consists of the connective tissue, fatty tissue and ligaments. Histologically, PT is classified as benign, borderline or malignant based on its distinct features as described by Rosen (3).

Majority of the cases presented as painless breast lumps. They have a tendency to rapidly enlarge to a giant mass until causing pressure necrosis, ulceration, secondary infection, bleeding or foul-smelling tissues. As far as we concerned, this is the first case of a neglected PT complicated with breast necrosis, who was subjected for a toilet mastectomy for a salvage surgery. We discuss the clinico-social aspect of her problem with literature review of PT.

CASE REPORT

A 19-year-old non-local girl presented to the emergency department with septic shock requiring fluid resuscitation. She elicited a history of left breast mass that was progressively increasing over the past 6 months. She tried to hide her illness by wearing a baggy cloth. However, due to the increasing size of the mass, she opted to stay with her ailing grandmother back in the village 474 km from her hometown in Kota Kinabalu. The condition had aggravated in which the breast skin became blackish and culminated with an offensive odor. Thus, this debilitating issue finally prompted her to seek medical therapy. Otherwise, she denied any risk factors for breast or ovarian cancer in the family. Clinically, there was a firm, mobile, lobulated mass of 20 x 20 cm arising from her left breast resulting in a distortion and asymmetry as compared to the right breast. There was an extensive area of skin and tissue necrosis associated with foul smelling odor (Figure 1). There was no palpable axillary node.

Owing to the diagnosis of breast necrosis, she was subjected for a toilet mastectomy. The dissection was relatively uneventful. The tumor was removed en bloc with the specimen weighted 4 kg (Figure 2). The pectoralis major was relatively preserved, however a wide area had to be removed due to the necrosis and infection. The resulting mastectomy wound was left open for secondary healing and planned for split skin graft later (Figure 3). She had an uneventful post-



Figure 1 : Patient’s condition during her first presentation with necrotic left breast tissue.



Figure 2: The specimen showing a bosselated mass with discoloration of the skin and disruption of the nipple areolar complex, weighting 4 kg

operative recovery and was discharged home. The final histology of the specimen was consistent with borderline PT (Figure 4). Unfortunately, the margin was involved which warrants a redo surgery and radiotherapy. Due to her non-local citizenship, she was denied from underdoing further treatment. Instead, she is currently under our care of three monthly follow up.



Figure 3 : Remaining area is left exposed for granulation

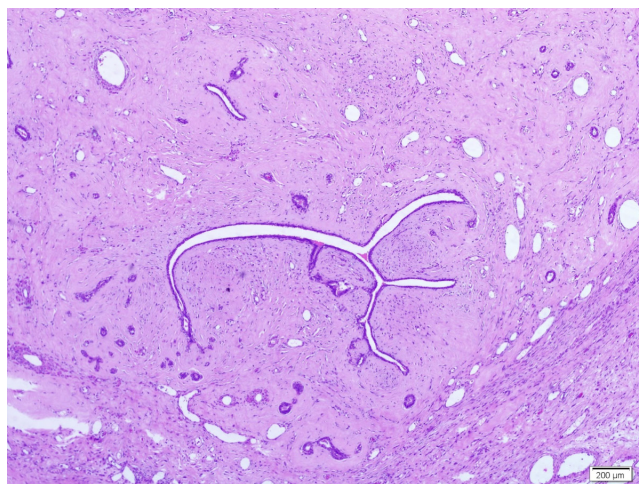


Figure 4 : Proliferation of breast duct and stroma pushing the duct into slit-like spaces with infiltrative border (H&E x4)

DISCUSSION

PT is a rare disease of breast pathology. It has biphasic features namely epithelial and stromal components. The existence of stromal overgrowth in such tissues is pathognomonic of PT. Its pathogenesis is perplexing either from existing fibroadenoma or de novo (1). Our

case differs from the conventional norm of PT as the individual involved is in her late adolescents. She is more prone to be diagnosed as huge fibroadenoma if she comes earlier. Fibroadenoma is labeled as giant if the tumor weighs more than 500 grams. Clinically, PT and fibroadenoma share much in common especially during initial presentation. Both are relatively mobile and rounded. They usually do not present with palpable lymph nodes. Preoperative diagnosis poses a diagnostic difficulty as the fine needle aspiration cytology may not be able to distinguish a PT from a fibroadenoma. However, with thorough history and complete clinical assessment, one may be differentiated from the others. Rapidly growing breast tissues with skin necrosis are diagnostic for PT regardless of her age. Giant phyllodes tumour is considered if the mass size is more than 10 cm. The largest reported case of giant phyllodes tumour was 50 x 50 cm (4).

Since she presented in sepsis, resuscitation is the paramount mode of initial treatment. She was managed according to surviving sepsis campaign bundles. Nevertheless, the conventional protocol for investigation of breast lump was not able to be implemented. Rather, an en bloc resection and reduction of septic load were implemented via toilet mastectomy. Thus, it greatly improved patient's morbidity and avoided the mortality of patient. Though an R0 or R1 resection was not achieved, the final histopathological report was consistent with a borderline PT, unfortunately with involved margin. Therefore, re-excision is warranted by resecting the superficial surface of the pectoralis major muscle with inclusion of radiotherapy to the chest wall.

There are no differences in term of disease free survival and overall survival between breast conserving surgery versus mastectomy (5). However, conservation shows higher rate of local recurrence (5). Thus, in malignant subtype, mastectomy is the gold standard of therapy especially when dealing with giant PT (4). Getting an R0 is mandatory to improve the survival. Most experts have advocated the operating surgeons to achieve at least 1 cm margins on primary excision or else, a surgical resection of the pectoralis major with a reconstruction or radiotherapy can be attempted (4). As malignant PT usually spreads via hematogenous route and the proportion of patient with lymph node metastasis is extremely minimal, routine axillary clearance is not advisable. In the case of systemic metastasis, therapy is according to soft tissue sarcoma protocol.

Neglect is the fundamental issue of this reported case. The patient should ideally seek a medical attention earlier so that a complete and thorough management

of breast lesion can be undertaken smoothly without a delay. Even in PT, triple assessment is mandatory. Dealing with smaller and simpler breast pathology is easier and safer rather than the complicated lesion as reported in our case. Emphasis on proper health education and self-examination is essential especially by the health care providers and authority concerned. The negative consequences of alternative or traditional treatment should be highlighted as unnecessary delay can lead to diseases progression, increased morbidity and mortality.

CONCLUSIONS

The diagnosis of PT is warranted in all patients presenting with rapid enlargement of breast lump. Excision should be done as soon as possible especially in septic patient. Emphasis on proper health education and the negative consequences of alternative treatment or neglect are utmost important as unnecessary delay can lead to diseases progression, increased morbidity and mortality.

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REFERENCES

1. Hayati F, Lian HH, Azizan N, Ali AA, Abidin ZAZ, Suhaili MA. Approaches to phyllodes tumour of the breast: a review article. *Int Surg J* 2017; 4(3): 841-5
2. Munawer NH, Md Ali SA, Md Zin R, Muhammad R. The co-expression of estrogen receptor beta (ER β) and smooth muscle actin in phyllodes tumour of breast: a tissue microarray study. *Sains Malaysiana* 2014; 43: 233-40
3. Rosen PP. *Rosen's Breast Pathology*. 2nd edition. New York, NY, USA: Lippincott William Wikins; 2001
4. Islam S, Shah J, Harnarayan P, Naraynsingh V. The largest and neglected giant phyllodes tumor of the breast—A case report and literature review. *Int J Surg Case Rep* 2016; 26: 96-100
5. Cohn-Cedermark G, Rutqvist LE, Rosendahl I, Silfversward C. Prognostic factors in cystosarcoma phyllodes: a clinicopathologic study of 77 patients. *Cancer*. 1991; 68(9): 2017–22.