



Case Report

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## Giant Fungating Borderline Phyllodes Tumor of the Breast

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### Abstract

Phyllodes Tumors are rare fibroepithelial neoplasms of the breast, accounting for less than 1% of all breast neoplasms. Preoperative diagnosis of Phyllodes Tumors is not easy, and a definitive diagnosis depends on a complete excision of the tumor. We present a case of a Giant Fungating Borderline Phyllodes Tumor of the breast. The main objective is to share our experience with the Phyllodes tumor of the breast from Mankweng Academic Hospital, Limpopo, South Africa. *Case Report:* A 50-year-old postmenopausal patient presented to our Breast Oncology clinic with a history of a slow-growing left breast mass for a duration of more than 1 year. It started as a small mass and progressively increased in size. She previously had two core needle biopsies, of which both histological analyses were inconclusive. In the last four months, there were multiple ruptured round masses associated with pain, bleeding, and an offensive discharge. The patient does not have any family history of breast cancer. On examination, the left breast has a fungating mass of 15×15 cm, with areas of necrosis and hemorrhage. There were no associated axillary lymph nodes and no abnormal findings in the right breast. Chest X-ray: normal. A mammogram was not done owing to the fear and risk of bleeding. A mastectomy was performed, and it was a highly vascularized mass. The excised specimen weighed 2.7 kg and was sent for histological assessment, which showed features of fibroepithelial proliferation consistent with Borderline Phyllodes Tumor with areas of stromal atypia, focal increase in cellularity, mitosis 5 per 10 High Power Field (HPF), and clear surgical margins of 0.9 millimeters (mm). The patient was reviewed six weeks after the operation. *Conclusion:* Owing to the rare nature and misdiagnosis of Phyllodes tumors, it is important to obtain a tissue biopsy for histological assessment of all suspicious breast lesions, regardless of age.

*Keywords:* Phyllodes Tumors; Benign; Borderline; Malignant; Breast Cancer; Margins.

### 1. Introduction

Phyllodes Tumors (PT) were first described by Muller in 1838 as cystosarcoma phyllodes. The term Phyllodes is coined from the Latin Phylloidium, which means “leaf-like” based on the gross pathological description of leafy, bulky, cystic, and fleshy tumors of the breast. PTs are rare fibroepithelial neoplasms of the breast, accounting for less than 1% of all breast neoplasms with variable clinical behavior [1]. The World Health Organization (WHO) classified PTs histologically as benign, borderline, and malignant based on stromal cellularity, stromal atypia, stromal overgrowth, mitosis, and tumor margins [2].

Borderline PTs have not been extensively studied as compared to benign and malignant PTs [3]. Benign PTs are more common between 35-64%, Borderline 7-40%, and Malignant can reach up to 30%, and a sharp distinction between benign and malignant is not always possible [4]. Preoperative diagnosis of PTs is not easy, and a definitive diagnosis depends on a complete excision of the tumor [5]. We present a case of a Giant Borderline Phyllodes Tumor of the breast.

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The main objective is to share our experience of Phyllodes tumor of the breast from Mankweng Academic Hospital, Limpopo. To the best of our knowledge, this is the first case of Giant Fungating Borderline Phyllodes Tumor at our institution.

## 2. Research Methodology

A description of rare clinical cases of borderline Phyllodes tumors in the breast of a patient who presented at Mankweng Academic Hospital, Limpopo, South Africa. A written informed consent was obtained from the patient to publish this case report.

### 2.1. Case presentation

A 50-year-old postmenopausal patient with no known comorbidities is married with four children. Presented to our Breast Oncology clinic after being referred from the local hospital with a history of a slow-growing left breast mass for more than 1 year. It started as a small mass and progressively increased in size. She previously had two core needle biopsies (CNB), of which both histology specimens were inconclusive. In the last four months, there were multiple ruptured round masses associated with pain, bleeding, and an offensive discharge. The patient does not have any family history of breast cancer; she is a non-smoker with no history of oral contraceptives. On examination, the left breast fungating mass was 15×15 cm, with areas of necrosis and hemorrhage (Figures 1 and 2). There were no associated axillary lymph nodes and no abnormal findings in the right breast.

Investigations performed included a full blood count (FBC) and Urea & Electrolytes (U&E). The patient had a low HB of 8.9 g/dl and a low MCV (mean corpuscular volume). U&E -Normal. Chest X-ray: normal. A mammogram was not done owing to the fear and risk of bleeding. The patient was transfused with red blood cells and consented to a mastectomy after being optimized. A mastectomy was done, and it was a highly vascularized mass with an estimated blood loss of 1 litre. The excised specimen (Figure 3) weighed 2.7 kg and was sent for histological assessment. Her post-operative condition was uneventful, and she was subsequently discharged on day 4.

Histology was reviewed and showed features of fibroepithelial proliferation consistent with Borderline Phyllodes Tumor with areas of stromal atypia, focal increase in cellularity, mitosis 5 per 10 High Power Field (HPF), and clear surgical margins of 0.9 millimeters (mm). The patient was reviewed six weeks after the mastectomy (Figure 4).



Figure 1. Preoperative period



Figure 2. Preoperative period



Figure 3. Excised Specimen



Figure 4. Post-Operative Follow-up (6 weeks)

### 3. Discussion

The peak incidence of Phyllodes Tumors (PT) is between the ages of 35-50 years in women across many cases [5-8]. Our patient had a mass at the age of 49 and was diagnosed at the age of 50 after an excision. PTs have been diagnosed in patients as young as 11 years [9], and therefore it is advisable to always have a high index of suspicion.

Benign PTs have consistent nuclear morphometry of the interstitial nucleus; mitosis is uncommon when present, usually less than 5 mitoses per 10 HPFs, and comprise 60-75% of all PTs [3]. It can be challenging to differentiate benign PTs from Fibroadenoma (FA) because increased stromal cellularity is a prominent feature in both; however, the leaflike pattern is the typical feature in PTs and is not seen in FAs [3]. Malignant PTs are characterized by pronounced stromal cellularity and nuclear pleomorphism, stromal overgrowth, and more than 10 mitoses per 10 HPF. The presence of heterologous sarcomatous elements alone is diagnostic of malignant PTs. The differential diagnosis of malignant PTs includes sarcomas and metastatic (sarcomatoid) carcinoma, the distinction of which is based on morphology [3].

Borderline PTs are the histology and topic of interest because of our patient's histology. Our patient had features of fibroepithelial proliferation, areas of stromal atypia, a focal increase in cellularity, and mitosis 5 per 10 High Power Field (HPF). According to the WHO definition, PTs that do not possess all the features for malignancy are classified as Borderline and they have focally invasive borders and frequent mitoses (5-9) per 10 HPF. In our case, the patient had mitosis at 5 per 10 HPF. Borderline PTs have not been extensively studied as compared to benign and malignant PTs [3]. The development of PTs is reported to be higher in single, infertile, postmenopausal women, and those using oral contraceptives at childbearing age [5]. Our patient was also postmenopausal at presentation; however, she had never used oral contraceptives before.

The diagnosis of PT can be difficult preoperatively [5, 10], as this was in our case. The patient had two inconclusive core needle biopsies (CNB), which led to a delay in the patient's referral. Previously, some studies found that CNB rarely produced a definitive preoperative diagnosis [11, 12]. Due to the lack of distinctive clinical symptoms that distinguish PTs from FAs, the rate of misdiagnosis is high [5, 10]. It is important to note that even on radiographic imaging, the features of PT are similar to those of Fibroadenoma, and these features include a smooth, large, round or oval, well-circumscribed appearance [1]. However, histologically, PTs have leaf-like cellular lobulations and a biphasic fibroepithelial constitution, which does not feature in FAs [8]. PTs presenting without rupture of the skin are difficult to distinguish from FAs [8]. However, a large size of > 3 cm and rapid growth should raise suspicion. On average, PTs can range between 4-7 cm, and those with a diameter of more than 10cm are referred to as Giant PTs [13], hence we refer to ours as Giant PT.

Wang et al. (2017) [7] found PTs of variable histology in 2 sisters who had masses of 3 cm; one sister had a Borderline PT and the other had Malignant PT. Tumor size has generally been of prognostic significance, but this has not been proven [4]. However, the size may influence surgical management. Benign tumors may grow to a large size, similar to our case, and small lesions may be malignant. In a case reported by Rathore [14], a patient had a mass that measured 30x25 cm, of which the histology was benign, and therefore concluded that size does not directly correlate with the histology.

Surgical excision is the gold standard of treatment; wide local excision, breast conserving surgery, simple mastectomy, and modified radical mastectomy [8]; in cases of large tumors, a mastectomy may be necessary [8, 15]. Our patient was offered a simple mastectomy due to the size of the mass, which measured 15×15 cm (Giant). It was fungating and had a foul-smelling discharge with areas of hemorrhage. It is recommended to obtain a surgical margin of > 1cm to reduce the risk of local recurrence [7]. However, Yom et al. (2015) found that a clear margin of 0.1 mm is not inferior to a margin of 1cm in the benign and borderline subgroups [16]. Tremblay-Lemay et al. also found that margins <1 mm are sufficient if tumor-free [17]. Toussaint et al. conclude that margins ≥10mm were associated with low LR, regardless of the histology of the PT [18]. In our case, we achieved a clear margin of 0.9 mm. In the malignant group, a margin >1cm is recommended.

Moutte et al. [19] also did not find any increase in local recurrence when compared to recurrence rates reported in the literature. A South African study by Spinks (2019) [20] found that no clinical or histological factors, including margin status, were found to significantly predict local recurrence (LR). Choi et al. [21] found that margin status was the only independent risk factor for LR in Borderline PTs. Moutte et al. [19] found that a second procedure to increase margins in patients with positive surgical margins in the benign and borderline subgroups is not necessary [19]. Ben et al. [22] also found that a second procedure to increase margins in patients with positive margins in the same group as Moutte et al. [19] was unjustified. In all the subgroups, axillary lymph node dissection is not necessarily due to the common metastasis through the hematogenous pathway to the lungs, pleura, and bone [5]. Currently, there are no studies supporting the use of radiotherapy and post-operative chemotherapy; however, a previous study by Barth et al. showed that post-operative radiotherapy could lower the rate of Local Recurrence and disease-free survival in the borderline and malignant subgroups [23].

#### **4. Conclusion**

Owing to the rare nature and misdiagnosis of Phyllodes Tumors, which have overlapping clinical and radiological features with other breast pathologies, it is important to obtain a tissue biopsy for histological assessment of all suspicious breast lesions, regardless of age. Preoperative diagnosis of Phyllodes Tumors is not easy, and a definitive diagnosis depends on a complete excision of the tumor with negative surgical margins. When a core needle biopsy is inconclusive, the decision to re-excite positive margins should be subject to clinical assessment. Therefore, we strongly recommend a triple-stage assessment of all breast lesions, obtaining a tissue biopsy, as we do not want to conservatively manage a malignant lesion that will be diagnosed at an advanced stage. Due to the rarity and small number of cases reported across the literature, there are no randomized control trials recommending treatment protocols, and as a result, we do need prospective studies looking particularly at this subject to help better manage patients with this kind of pathology with its variable clinical behavior, as timely diagnosis will ensure timely management.

#### **5. Declarations**

##### **5.1. Author Contributions**

Conceptualization, F.C.M. and M.M.Z.U.B.; methodology, F.C.M. and M.M.Z.U.B.; validation, M.M.Z.U.B.; formal analysis, F.C.M. and M.M.Z.U.B.; investigation, F.C.M.; data curation, F.C.M. and M.M.Z.U.B.; writing—original draft preparation, F.C.M.; writing—review and editing, F.C.M. and M.M.Z.U.B.; visualization, F.C.M. and M.M.Z.U.B.; supervision, M.M.Z.U.B.; project administration, M.M.Z.U.B. All authors have read and agreed to the published version of the manuscript

##### **5.2. Data Availability Statement**

Data sharing is not applicable to this article.

##### **5.3. Funding**

The authors received no financial support for the research, authorship, and/or publication of this article.

##### **5.4. Ethical Approval**

Permission was obtained from the clinical Executive Director of Mankweng Academic Hospital and informed consent was given by the patient.

##### **5.5. Institutional Review Board Statement**

Approval was obtained from the clinical Executive Director of Mankweng Academic Hospital.

##### **5.6. Informed Consent Statement**

Informed consent was obtained from patient.



### 5.7. Declaration of Competing Interest

The authors declare that there is no conflict of interests regarding the publication of this manuscript. In addition, the ethical issues, including plagiarism, informed consent, misconduct, data fabrication and/or falsification, double publication and/or submission, and redundancies have been completely observed by the authors.

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