

Life threatening bleeding from untreated phyllodes tumor following earthquake induced follow up delay

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Abstract

Phyllodes tumors, representing 0.3%–0.5% of breast masses, are infrequent and sophisticated neoplasms of the breast. Based on their histological features, they are classified into three categories: benign, borderline, or malignant; preoperative diagnosis is challenging, and surgical removal with clear margins is the primary treatment option. This report describes the case of a 35-year-old woman whose longstanding breast mass underwent rapid transformation. The patient missed her follow-up appointment due to the February 6, 2023, earthquake in Hatay and presented with hemorrhagic shock from bleeding caused by the tumor eroding the internal thoracic artery.

Keywords: Phyllodes tumor, breast tumor, hemorrhage, emergency surgery

INTRODUCTION

Phyllodes tumors are rare, but complex breast tumors. They comprise approximately 0.3%–0.5% of breast masses (1). Unlike other types of breast cancers, distinguishing benign phyllodes tumors from fibroadenomas and malignant phyllodes tumors can be challenging. The World Health Organization divides these tumors into three distinct groups: benign, borderline, and malignant. This classification is determined by examining various characteristics, including the edges of the tumor, cellular abnormalities, and rate of cell division (2). Patients may notice a lump or have symptoms such as skin ulceration and bleeding. Phyllodes tumors cannot be easily distinguished from fibroadenomas by imaging tests. They are often difficult to diagnose preoperatively and are commonly diagnosed later in life. Surgical removal with clear margins is the primary treatment, but borderline and malignant subtypes can still recur locally in up to 20% of cases (3). This case report describes a patient with a phyllodes tumor who developed life-threatening bleeding due to the tumor eroding an artery after missing a follow-up appointment because of the February 6, 2023 earthquakes in Hatay.

CASE

A 35-year-old woman sought medical attention at our facility in January 2023, reporting a breast mass that had experienced sudden growth. Based on the anamnesis, it was learned that the mass in that area had been followed up for two years. Three tru-cut biopsies were performed, fibroadenomas were evaluated, and follow-up was recommended. Physical examination revealed a regular, mobile, painless mass with regular borders in the upper outer quadrant of the right breast. Thereupon, the requested breast ultrasound was reported as ‘a lesion with central vascularity, regularly and occasionally angular contours was observed in the Doppler examination performed at the 1 o'clock position of the right breast, adjacent to the areola, measuring approximately 24*46 mm’. Therefore, a tru-cut biopsy was performed. The biopsy result was reported as ‘ductus and gland structures were observed in the stroma of spindle cells showing myxoid degeneration in sections. Excisional biopsy was recommended for definitive diagnosis of the mass. Preoperative frozen examination was interpreted as compatible with phyllodes tumors. The operation was terminated when surgical margins were negative. Following the surgical procedure, the patient

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was sent home on the third day. A follow-up appointment at the outpatient medical oncology department was advised. In May 2023, the patient was admitted to the emergency room with dizziness and bleeding in the right breast (Figure 1).



Figure 1. Bleeding lesion in the right breast

In the anamnesis, it was learned that she was buried under rubble and lost her relatives in the earthquakes on February 6, 2023, that she had no one to care for her, and that she could not go for a postoperative check-up due to transportation problems. The vital signs indicated a blood pressure of 60/40 mmHg and a heart rate of 150 beats per minute for the patient. Physical examination revealed an ulcerated bleeding lesion measuring approximately 10 cm×10 cm in the upper inner quadrant of the right breast. Laboratory tests revealed Hb:7.3 g/dl, Htc:23 %. Following the application of a compressive dressing to the mass, the patient was relocated to the intensive care unit for ongoing management. After vital signs were stable with massive fluid and fresh whole blood resuscitation, breast magnetic resonance imaging (MRI) was performed (Figure 2).



Figure 2. Right breast mass on preoperative magnetic resonance imaging

The patient underwent emergency surgery because of ongoing bleeding, and the angiography unit was inactive in our clinic after the earthquake. During exploration, it was

observed that the mass lesion eroded the internal thoracic artery from the second intercostal space and that there was active pulsatile bleeding from this area. Rib resection was performed to reach the artery (Figure 3). After the artery was clipped and ligated, nipple-areola preserving mastectomy was performed (Figure 4).

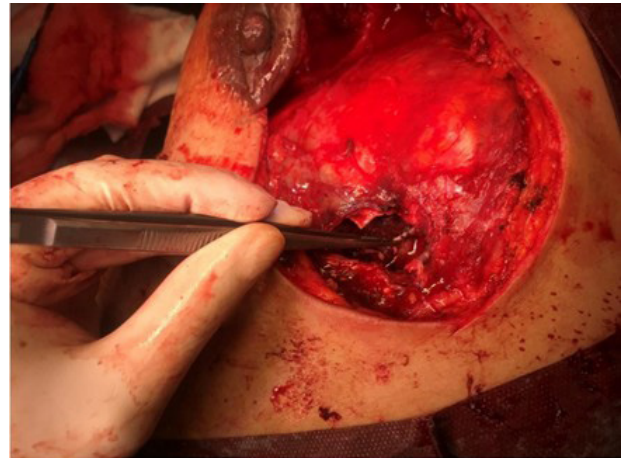


Figure 3. After a rib resection and reach the artery

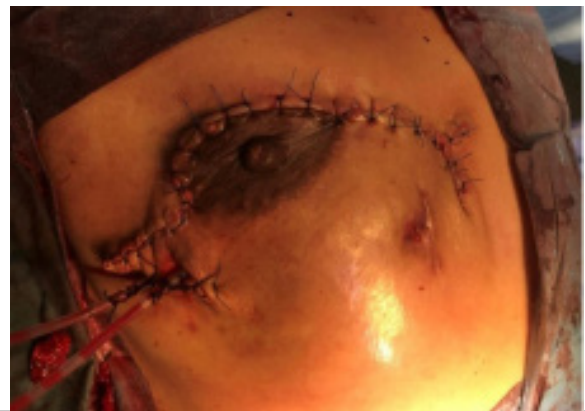


Figure 4. The nipple-areola preserving mastectomy was performed

After a successful recovery, the patient was sent home on the 7th day following the surgical procedure. The final pathology report was reported as follows: The excised lesion was diagnosed as a fibroepithelial tumor consistent with a malignant phyllodes tumor. The tumor measured 15×10×8 cm. Histologically, there was marked stromal atypia, with more than 50 mitoses per 10 high-power fields. No heterologous elements were identified. The tumor margins could not be clearly delineated, but no tumor involvement was observed at the surgical margins. Additionally, no evidence of lymphovascular invasion was detected (Figure 5). The patient was referred to the oncology department for adjuvant treatment. Written informed consent was obtained from the patient for the publication of this case report.

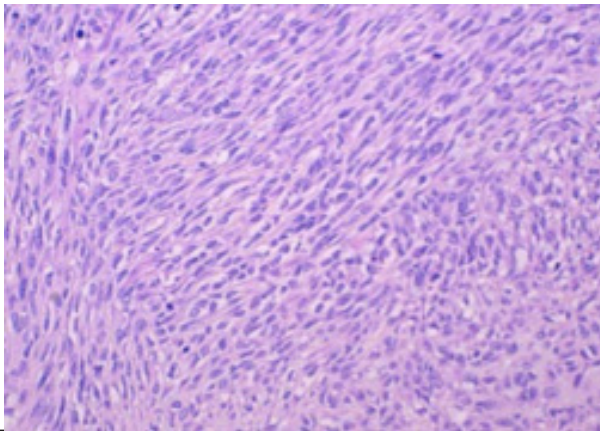


Figure 5. Stromal atypia in malignant phyllodes tumor (20x magnification)

DISCUSSION

This case highlights several fundamental elements in the management of phyllodes tumors. Notwithstanding the results from multiple core-needle biopsies indicating a benign fibroadenoma, subsequent excisional biopsy and frozen section analysis disclosed a malignant phyllodes tumor, illustrating the complexities associated with the diagnosis of these neoplasms. Phyllodes tumors may exhibit histological characteristics that are indistinguishable from those of fibroadenomas during core needle biopsy, primarily due to the overlapping stromal and epithelial components, especially in the context of early-stage or diminutive lesions. Insufficient sampling might not adequately capture critical diagnostic features such as stromal overgrowth, augmented mitotic activity, and cellular atypia. The inherent heterogeneity present within phyllodes tumors further complicates the diagnostic process, as malignant foci may go undetected (4). In this case, the patient presented with a similar condition, which resulted in a delayed diagnosis.

Although phyllodes tumors are recognized for their rapid proliferation and propensity for local recurrence, instances of vascular invasion resulting in hemorrhagic shock are indeed exceptional (5). In this particular case, the delay in follow-up attributable to the February 6 2023 earthquakes likely exacerbated the tumor's uncontrolled advancement and the eventual erosion of the internal thoracic artery. A total of seven occurrences of ruptured phyllodes tumors have been documented in the literature (6-12). Yang and their team detailed a clinical instance where swift surgical intervention was essential to tackle ongoing hemorrhage resulting from a ruptured phyllodes tumor. In that instance, the mass exhibited rapid expansion over a two-month period and was postulated to have ruptured due to accelerated growth and invasion of the skin. Ruptured phyllodes tumors generally manifest as substantial masses, measuring approximately 9 to 21 cm in

diameter (11). With the exception of the case reported by Nabi et al., all other documented cases involved tumors categorized as borderline or malignant. Nabi et al., who encountered a benign tumor, executed a local excision, whereas all other patients underwent simple mastectomy (7). In this specific case, a nipple-areola-preserving mastectomy was performed following the confirmation of negative margins, in accordance with current guidelines for the management of malignant phyllodes tumors, thus emphasizing the significance of complete resection in reducing the risk of recurrence. This represents the inaugural documented case of a malignant phyllodes tumor being addressed with nipple-areola-sparing surgical intervention.

The strategy of emergency rib resection for the management of hemorrhage underscores the necessity for flexibility in critical surgical contexts, particularly in case where interventional radiology services are lacking. This represents the inaugural documented instance in which hemostasis was achieved through costal resection. Phyllodes tumors predominantly disseminate through hematogenous routes and typically do not necessitate axillary lymph node dissection unless there is the presence of enlarged nodes or evident metastases, owing to the infrequent occurrence of lymph node involvement. Instances of axillary lymph node metastasis have not been documented in cases involving ruptured phyllodes tumors. Regardless of the tumor's considerable dimensions, there were no signs of distant spread. Should there have been no urgent requirement for surgery, the patient could have derived benefits from receiving radiotherapy ahead of the operation (13). Various research works in the available corpus imply that preoperative radiotherapy may lower the likelihood of local recurrence; still, in this unique situation, the patient lacked follow-up care or radiotherapy after the first surgical operation. This case elucidates critical public health and ethical dilemmas pertaining to patient follow-up during natural disasters. The disruption of standard healthcare services occasioned by such events can lead to postponed diagnoses and treatments, thereby exacerbating clinical outcomes. This circumstance highlights the critical need for developing comprehensive emergency protocols that safeguard the continuity of healthcare services for susceptible patient demographics.

CONCLUSION

Breast phyllodes tumors are uncommon fibroepithelial neoplasms that may exhibit rapid enlargement and hemorrhage, particularly in malignant cases. Clinicians should be aware of the potential for these rare tumors to cause significant morbidity and mortality if left untreated or if the follow-up is delayed. Urgent surgical management is

critical for preventing complications such as hemorrhage, as demonstrated in this case report.

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