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Staging resection and reconstruction with temporary wound VAC coverage in a case of giant cystosarcoma phyllodes of the breast

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ABSTRACT

INTRODUCTION: Cystosarcoma phyllodes (CP) is a rare breast tumor occurring most often in females in their fifth decade. While usually benign, some CP tumors exhibit aggressive growth patterns and extensively invade chest wall structures; resecting these tumors to negative surgical margins can be challenging. We present a case of malignant CP involving the chest wall where using a negative pressure vacuum-assisted closure (VAC) system after resection enabled complete histopathologic margin assessment prior to reconstruction. This is the first known report of staged breast tumor resection and reconstruction with interim VAC coverage.

CASE PRESENTATION: A 48 year-old woman presented with rapidly increasing left breast size, fevers, and fatigue. On examination, the left breast was massively enlarged with engorged vessels and skin necrosis. Lab analyses revealed unusual metabolic abnormalities requiring preoperative hospitalization. We performed a left modified radical mastectomy with partial resection of pectoralis major and minor muscles, temporarily sealing the wound with a VAC due to concern for deeper tumor extension that could require further resection. Pathology revealed malignant CP with a negative deep margin. The 38 cm defect was then repaired with latissimus myocutaneous flap plus skin graft. At three-year follow up the patient remains free of disease.

CONCLUSION: In cases of malignant CP involving the chest wall, minimizing the extent of chest wall resection is critical for reducing morbidity, while completely clearing tumor margins is essential for reducing recurrence risk. Using temporary wound VAC coverage enables cautious debulking followed by histopathologic margin assessment prior to definitively reconstructing the breast.

1. Introduction

Cystosarcoma phyllodes (CP) represents less than 0.5% of all breast neoplasms. It occurs almost exclusively in women and incidence peaks in the 4th and 5th decades of life. Phyllodes tumors oddly occur more frequently in the left breast, and though the average size is quite large for a breast tumor – about 5 cm – lesions exceeding 10 cm, termed "giant," are common. Generally, tumors are well demarcated, freely mobile, and are often discovered clinically by patients. Both clinically and on imaging studies, the lesions appear similar to benign fibroadenomas; biopsy is thus critical for capturing the diagnosis [1,2].

Histopathologically, phyllodes tumors appear as diffuse cystic spaces among overgrown stromal connective tissue. In general, proliferation of stromal fibroblasts characterizes benign lesions, while cellular atypia and increased mitoses characterize malignant lesions. Up to 90% of the tumors are histologically benign. However, in many cases, histopathologic appearance does not predict tumor behavior. Furthermore, benign tumors that recur may do so at a higher-grade. Malignant tumors can demonstrate sarcomatous behavior and metastasize hematogenously to the lungs, skeleton, heart, and liver. Because of these aggressive growth patterns, both benign and malignant lesions should be excised completely with no tissue left behind in order to prevent recurrence [3–5].

While studies have shown surgical margin status to be a principal determinant of local recurrence and disease metastasis, there are no official guidelines on how much to excise around lesions. Many practitioners advocate for wide local excision with >1 cm margins, citing higher recurrence rates with margins below this [1,4–6]. In cases where disease extends to the chest wall, however,

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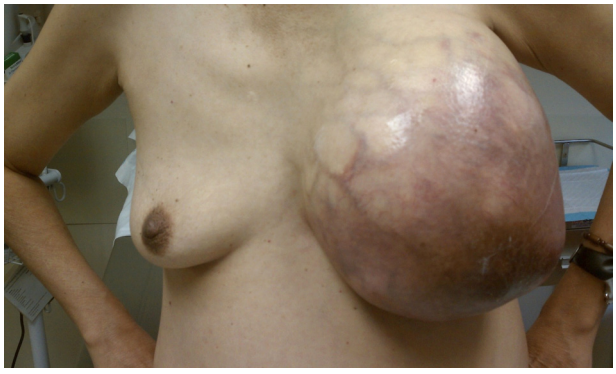

Fig. 1. Left breast mass, anterior view at presentation.

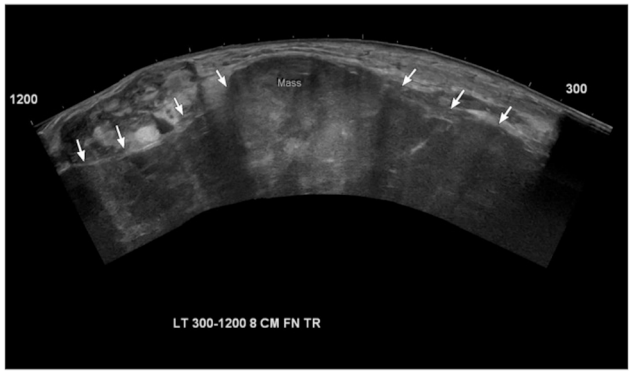

Fig. 2. Transverse ultrasound, 3:00–12:00 axis, demonstrating a large hypoechoic mass (arrows) that extends beyond the field of view and compresses the normal breast parenchyma seen superficially.

resecting to wide margins would violate chest wall integrity and increase operative morbidity.

We present a case of malignant CP so invasive that wide excision was impossible and tumor resection left a significant possibility of positive surgical margins. To address this possibility of reoperation following the final pathology report, we delayed chest wall reconstruction and temporarily covered the surgical defect with vacuum-assisted closure (VAC, Kinetic Concept Inc. – San Antonio, TX, USA), a novel practice in breast surgery according to our review of the literature.

2. Case presentation

A 48 year-old woman from the Philippines with no prior mammographic screening was referred by her primary care physician for a painful left breast mass that had rapidly enlarged over a five-month period. The patient originally noticed a small lump in her left breast about 1 cm in diameter, which in several months' time grew to the size of what she described as a "watermelon". The patient also reported a recent history of daily fevers to 39.4 °C, fatigue, dizziness upon standing, and shortness of breath.

On physical examination, she was tachycardic to 140 and febrile to 38.9 °F. Her left breast was massively enlarged and tender, with a flattened nipple, engorged superficial vessels, and deeply erythematous, partially necrotic skin (Fig. 1).

Labs drawn at that time revealed widespread electrolyte and hematologic disarray. Notably, hematocrit was low at 22.8, white cell count elevated at 16,000, and platelets elevated at 690,000. Liver function tests were abnormal and the patient was both coagulopathic and hemolytic. Iron studies suggested concomitant anemia of chronic disease. The patient was immediately hospitalized for electrolyte correction and multiple blood and plasma transfusions prior to surgery.

Imaging of the left breast was limited due to its massive size. Though mammography could not be performed, ultrasound revealed an enormous heterogeneous mass underlying the left breast and compressing the normal breast tissue anteriorly (Fig. 2). A CT-chest scout image clearly showed the large extruding left breast mass (Fig. 3), but abdominal and pelvic CTs showed no evidence of distant disease. Incisional biopsy of the mass suggested a malignant phyllodes tumor versus breast tissue sarcoma, and the patient was scheduled for immediate surgery.

A left modified radical mastectomy (MRM) was performed in which all gross disease was removed, including large regions of the pectoralis major and minor muscles involved by tumor along with the entire mass and breast. Tumor tissue was scraped off the sternum and costochondral junction at ribs 3–5 and bone shavings were taken for biopsy. After a 4 h resection requiring 4 units of packed red blood cells, 4.5 L of crystalloid, and 500 cc of albumin, intraoperative consultation was held with plastic surgery. The large soft tissue defect on the chest wall measured 38 × 28 cm and exposed ribs and intercostal muscles. Because of concern for microscopic tumor extension through bone and muscle requiring further resection, it was decided that reconstruction be delayed until final margin status was known. The defect was left open and temporarily

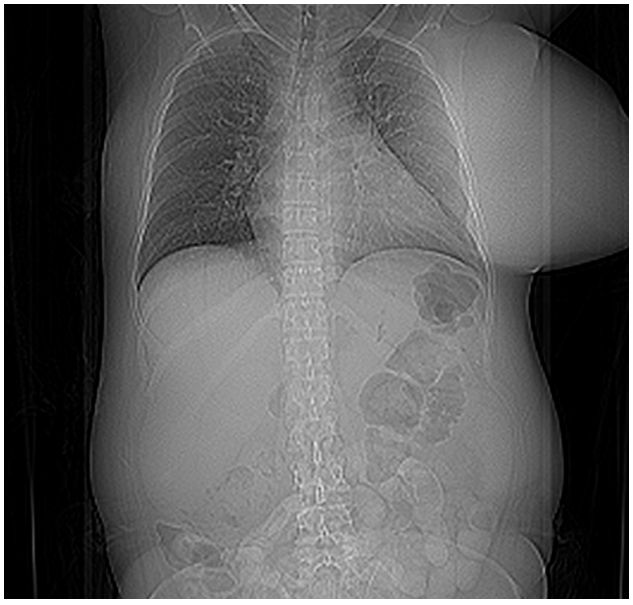

Fig. 3. Scout image from CT of the chest demonstrates large left breast mass.

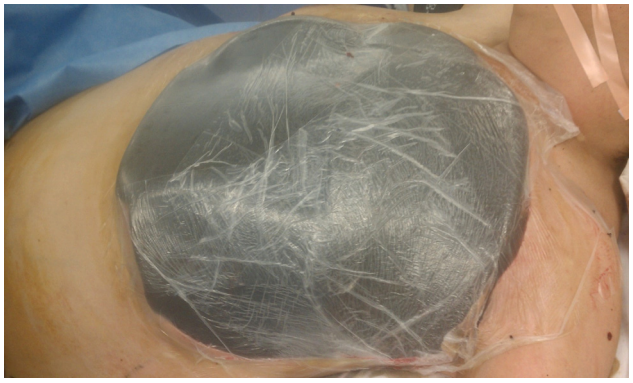

Fig. 4. VAC sponge covering left breast/chest wall defect measuring 38 × 28 cm.

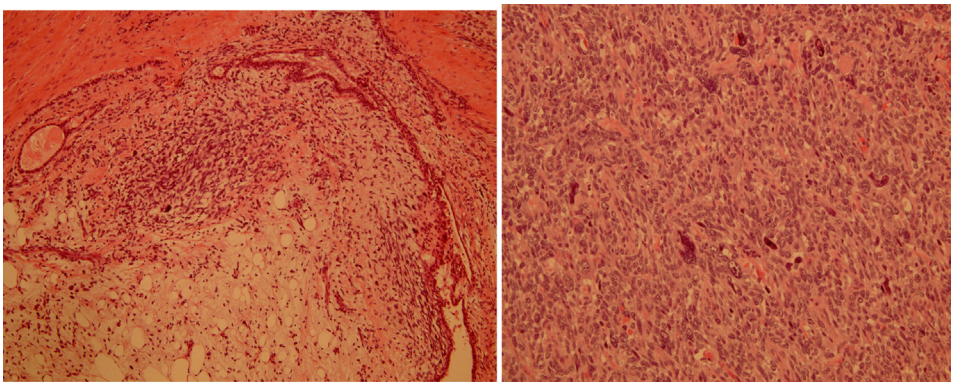

Fig. 5. Left pathology slide shows an intra-canalicular (leaf-like) pattern of growth with periductal variable cellular stroma in CP (100X mag). Right slide shows a high-grade sarcomatous component composed of spindle and giant cells and scattered mitoses (200X mag).

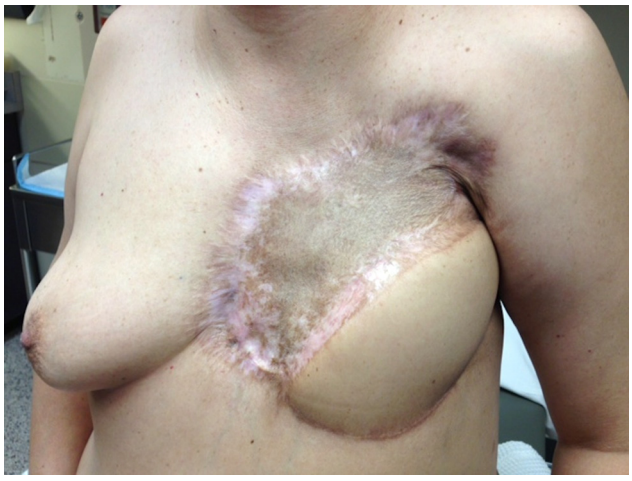

Fig. 6. Left latissimus myocutaneous flap/left thigh split-thickness skin graft reconstruction of the left breast, 2 years after surgical reconstruction, anterior view.

sealed with a VAC sponge (Fig. 4). The excised mass weighed 5.5 kg and measured 26.2 × 23.9 × 15.4 cm.

The final pathology of the mass was consistent with CP with malignant features. Surgical margins were negative for tumor cells but no margin exceeded 1 mm; the smallest was the posterior at 0.3 mm. Histopathologic viewing showed sheets of spindle cells interspersed with malignant-appearing multinucleated giant cells, cystic degeneration, and necrosis. There was a high nuclear grade and a mitotic index of more than 20 mitoses per 10 HPF (Fig. 5). Cytokeratin staining as well as ER, PR, and HER2-Neu were negative, illustrating the complete stromal overgrowth and lack of epithelial layers typical of sarcomatous tumors. Axillary lymph nodes contained reactive lymphocytes only with no invading tumor cells.

The MRM was complicated by a left hydropneumothorax, found incidentally on chest X-ray and attributed to the depth of resection necessitated by tumor invasion. A chest tube was placed on the open chest wound and removed after five days with full resolution.

Reconstruction of the breast and chest wall defect took place 1 week after the mastectomy with a left latissimus myocutaneous flap and left thigh split-thickness skin graft for additional skin coverage (Fig. 6). A latissimus myocutaneous flap was specifically chosen in order to provide coverage for the soft tissue defect and not for cosmetic reconstruction of the breast mound. It also served as barrier protection for the chest wall since the patient was planned for post-operative radiation. Reconstruction took 6 h and there were no complications.

As early as 1 week after surgery, the patient's systemic symptoms and abnormal lab values began to trend down, with complete resolution evident within the month. After hospital discharge, she initiated a 6-month course of experimental valproate as histone deacetylase inhibitor (HDACi) therapy, which has been postulated to have both a cytotoxic and cell-differentiating effect (from high grade to low grade tumor cells) on myeloid malignancies and tissue sarcomas [7,8]. She also completed 6 weeks of external beam radiation therapy. Three years after treatment, the patient continues to do well with no evidence of disease recurrence.

3. Discussion

This report illustrates a unique case of CP, notable for its unusual clinical presentation and therapeutic interventions.

Our patient's malignant giant tumor grew rapidly, extended through the chest wall, and caused widespread systemic symptoms and metabolic abnormalities. According to our review of the literature, profound systemic effects are extremely rare in cases of malignant CP. Based on the size of our patient's tumor and its histopathological appearance, the prognosis was guarded and the disease managed aggressively.

We chose to perform a modified radical mastectomy with takedown of all chest wall regions involved or adjacent to tumor, leaving the defect temporarily sealed with a wound VAC until the final pathology report determined that all margins were negative; only then was definitive reconstructive surgery performed. The VAC system involves applying a foam sponge to a wound space and adding continuous controlled suction, which has been shown to drain excess fluid, increase blood flow, and stimulate growth of granulation tissue [9]. VAC devices are commonly used at the bedside to cover wounds that will ultimately self-appose; less commonly, they are used in the primary coverage of surgical wounds.

Our review of the literature revealed cases of VACs used in general surgery to temporize the coverage of surgical wounds after colorectal abscess debridements, perforated bowel resections, and scalp malignancy excisions [10–12]. In plastic surgery, VACs may be applied directly to tissue flaps and skin grafts at the time of reconstruction to add seal and enhance fusion [13]. In cases of breast tumor resection and reconstruction, however, the use of temporizing VAC has not been reported to our knowledge, making this treatment approach novel in our case.

Additional unusual treatment modalities used in this case were adjuvant radiation and systemic chemotherapy. While there is much debate over the use of radiation in CP, radiotherapy was recommended for our patient since we could only attain just-negative surgical margins [4–6]. Likewise, given the malignant tumor's potential for metastasis and its dramatic systemic impact

prior to surgery, our patient underwent experimental HDACi medical therapy thought to provide a cytotoxic and downgrading effect on tumor cells left behind [7,8].

We propose that in cases of malignant CP extending to chest wall, staging the resection and reconstruction with the aid of a temporary closure device allows for definitive determination of pathologic margins. If the margins on the chest wall are positive, further chest wall resection can be performed at the time of reconstruction.

4. Conclusion

In the surgical management of cystosarcoma phyllodes, 1 cm tumor margins are usually attempted to prevent local recurrence; however, a 1 cm margin is not always feasible on the deep (chest wall) margin. In cases where the extent of disease precludes obtaining wide margins without introducing increased morbidity, a goal of complete tumor excision with negative margins may suffice in eliminating existing disease and preventing recurrence. Staging tumor resection and reconstruction with temporary wound VAC coverage allows for definitive assessment of margins in a situation where margin clearance is essential for minimizing local recurrence risk and chest wall resection carries significant morbidity.

Conflicts of interest

There are no conflicts of interest to disclose.

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Consent

Written, informed consent to publish this paper was obtained from the patient on August 22, 2014. A copy of the signed consent form may be requested and faxed to you for your reference.

Author contribution

All of the authors—Danielle R. Heller, B.A., Christine Rohde, M.D., and Preya Ananthakrishnan, M.D., contributed to all aspects of this case report, including but not limited to: management of the specified case, data collection for the case report, and manuscript writing and editing.

Other contributors to the paper include Hanina Hibshoosh, M.D., and Allyson Parnes, M.D., who respectively contributed pathologic slide images/ captions and radiographic images/ captions.

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