Kiss Flaps to Repair Large Skin Defect After Excision of Giant Malignant Phyllodes Tumor of the Breast: A Case Report and Literature Review

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Abstract

Background: Malignant phyllodes tumors of the breast are fairly rare and fast-growing tumors. They do not respond to chemotherapy or hormonal therapy except in malignant phyllodes tumors. Therefore, the primary treatment for malignant phyllodes tumors is wide surgical excision.

Case presentation: Herein, we report a case study which featured a 26-year-old woman presented with a giant malignant phyllodes tumor measuring 20 × 17 × 13 cm. In order to reduce the chance of local recurrence, treatment for these types of tumors usually involves extensive excision with at least 1 cm of surgical margins. The patient underwent extended lumpectomy with 1-3 cm surgical margins, which left a large skin defect of 25 × 15 cm. Repair of such a large skin defect is a challenge to breast surgeons. This is the first reported case in which a “kiss” flap was successfully used to repair the skin defect created after resection of a giant malignant phyllodes tumor.

Conclusions: The kiss flap could be considered as an effective method to repair large chest wall defects after resection of giant phyllodes tumors.

Background

Phyllodes tumors are fairly uncommon fibroepithelial neoplasms accounting for 0.3–0.9% of all breast tumors [1]. They are fast-growing tumors with benign, borderline, or malignant behavior, depending on histologic features including stromal cellularity, infiltration at the tumor’s edge, and mitotic activity. Their size ranges from 1–45 cm and they may occupy the entire breast. Giant phyllodes tumors are typically defined as those > 10 cm in diameter [2]. Reports in the literature [1, 3] have been focused on surgical approaches to tumor resection. An adequate surgical margin is preferred with extended lumpectomy or mastectomy to prevent recurrence and metastasis. However, repair of the skin defect created after wide excision with at least 1 cm of surgical margins is a great challenge to breast surgeons. Repair with a pedicled latissimus dorsi kiss flap has not been reported in the literature. We report a case of giant malignant phyllodes tumor treated with extended lumpectomy followed by successful defect repair with a kiss flap.

Case Presentation

A 26-year-old Chinese woman presented with a giant mass on her right breast, which had enlarged in the past year. The patient visited our breast department on September 18, 2016 because of the rapid tumor growth and the occurrence of skin ulceration with bleeding. Her familial and personal history was non-contributory. Except for the breast tumor, laboratory tests showed no significant findings. Physical examination revealed a protruding and hardened palpable mass occupying almost the entire right breast. The mass measured approximately 20 × 17 × 13 cm. The skin had been stretched thin, with areas of ulceration and engorged superficial veins. Bilateral axillary lymph nodes were palpable.
A mammogram was not feasible because of the tumor size. Ultrasound of the breast mass indicated a heterogeneous echo and internal structures containing small cystic components, calcification, and hyperechoic separations, and was classified as BI-RADS 4. Multiple enlarged hypoechoic bilateral axillary lymph nodes were detected and some of them had lost their normal internal architecture and fatty hila. Magnetic resonance imaging (MRI) revealed a giant, lobulated mass with heterogeneous signal intensity, as well as septa and well-circumscribed margins, indicating typical features of phyllodes tumor (Fig. 1).

Image-guided core needle biopsy was performed twice, but a definite diagnosis was not obtained. Subsequently, a right breast excisional biopsy demonstrated a fibroepithelial lesion suspicious for a malignant phyllodes tumor. The results of immunohistochemical (IHC) staining on tumor biomarkers showed: ER(-), CK(-), CD34(+/-), P63(-), SMA(-), and P53(+).

The patient underwent extended lumpectomy and axillary lymph node biopsy. Before surgery, a “kiss” flap was carefully designed to cover the skin defect. First, a paper template of the huge skin defect was created. The template was then split into two small pieces of the same size and shape. These paper templates were strategically oriented onto the kiss donor sites, allowing direct primary donor-site closure, and an exact match of the assembled flaps to the large defect. The skin defect was 25 × 15 cm. The template was used to mark the flaps on the patient's back. The location of the two-lobed flap depended on the vascular pedicle to ensure paddles were nourished by independent myocutaneous perforators from the thoracodorsal artery. An incision was made along the outlined semicircular skin island until the latissimus dorsi muscle was visualized, making sure both flaps were connected with the muscle. Before transposing the flaps to the chest wall through a subcutaneous tunnel, the two narrow skin paddles were arranged side by side and carefully sutured together to create a “kiss” flap. Finally, the donor site was sutured in layers, and the kiss flap was placed according to the design to cover the chest defect and sutured without tension (Fig. 2). The wound recovered well (Fig. 3).

The resected primary tumor measured 18 × 14 × 14 cm (Fig. 4A). The cut surface of the tumor was white to gray with hemorrhagic foci (Fig. 4B). There was no involvement of the cutting edges, skin, tissue beneath the nipple, or superficial fascia. The sentinel lymph nodes were negative (0/4) with IHC biomarkers ER(-), CK(-), CD34(+/-), P63(-), SMA(-), and P53(+). Postoperative pathology revealed a high-grade malignant phyllodes tumor with multifocal necrosis (Fig. 4CD).

After surgery, the patient was given six cycles of chemotherapy with docetaxel (160 mg), epirubicin (100 mg), and cyclophosphamide (800 mg). The patient also received twenty-five fractions of adjuvant chest irradiation treatment (Total 50 GY). The patient was followed up for more than 4 years postoperatively, and there was no local recurrence or distant metastasis. Currently the timing of further reconstructive surgery is appropriate for her to plan a delayed implant reconstruction of the right breast.

**Discussion**

Phyllodes tumor is a disease of the epithelial and stroma tissue in the breast. It is classified as benign, borderline, and malignant. Malignant tumors have high stroma cellularity and tend to be permeative
whereas benign tumors have low stroma cellularity and are circumscribed[4]. Malignant phyllodes tumors is distinguished only pathologically by identification of marked stromal cellularity, more than 5 mitoses per 10 high-powered fields, invasive margins, and marked stromal overgrowth[5]. Giant phyllodes tumors are rare fibroepithelial breast neoplasms typically >10 cm in diameter by definition [2]. In general, it is difficult to differentiate phyllodes tumors from benign fibroadenoma by clinical presentation, radiology, or even core needle biopsy [6]. The most accurate diagnosis of breast phyllodes tumor is postoperative pathology [7]. Unlike breast cancer which can be downsized by neoadjuvant therapy, phyllodes tumor is not sensitive to chemotherapy or radiotherapy or endocrine therapy [8–9]. Surgery is regarded as the primary treatment method of phyllodes tumors. Negative margins rather than surgery type, such as extended lumpectomy or total mastectomy, determine the recurrence rate [10]. The National Comprehensive Cancer Network guidelines[11] advocate a wide excision with surgical margins of 1 cm or more. A negative margin is an independent prognostic factor for disease-free survival and local recurrence [5, 12–13]. Patients with a positive margin and malignant histology should undergo further surgery to obtain clear margins [3].

Although extended lumpectomy or mastectomy with adequate surgical margins is the best choice for large malignant phyllodes tumors, the resulting large skin defect always requires a skin graft or transplanted flap. To the best of our knowledge, this case is the first documented use of a kiss flap to repair the large skin defect resulting from removal of a giant malignant phyllodes tumor of the breast. There are some other options to repair the defect such as a transverse rectus abdominis myocutaneous (TRAM) flap or a deep inferior epigastric artery perforator ( DIEP) flap. TRAM and DIEP can immediately reconstruct a new breast after mastectomy. Although DIEP and TRAM have little influence on pregnancy, the young girl has not been married or pregnant, she worried about a long scar on the abdominal wall would affect beauty and future pregnancy. Moreover, DIEP and TRAM need a long operative time and are highly traumatic, and DIEP requires microsurgical techniques. The young girl also worried about a quick recurrence soon after surgery, so she refused complicated immediately breast reconstruction such as DIEP and TRAM and wanted to choose a simple procedure. The latissimus dorsi flap is close to the postoperative chest wound and usually be used to repair the chest wall defect after breast dissection. However, the skin of the back lacks elasticity, the donor area cannot be directly sutured and generally requires a skin graft if the width of the flap exceeds 8 cm. This prolongs postoperative recovery and limits the wide application of the latissimus dorsi flap. The kiss flap involves the excision of double skin paddles, which has an independent blood supply from the donor stem. These paddles are spliced in the recipient area, so that they “kiss” each other side-by-side, to create a much larger flap, accurately matching the size of the defect. This technique allows flexible design of the flap shape, while increasing the surface area of skin flap coverage and minimizing incision dehiscence and non-healing complications [14]. A careful presurgical flap design is necessary to make maximum use of the limited human tissue available and ensure minimum damage while performing the autologous tissue transfer. The functional and aesthetic outcome of the donor site should also be considered. In this case, the postoperative flap had a good appearance with no hyperplastic scar and the activities of shoulder joint were not affected. The outcome of this case suggests that the kiss flap is a simple and feasible
technique for repair of large skin defect following giant phyllodes tumor resection. If there is no recurrence or metastasis more than one year after surgery, the patient may plan a delayed breast reconstruction. After communication with the patient, she planned a delayed implant breast reconstruction plus fat grafting.

Local recurrence of phyllodes tumors has been associated with positive margins, younger age, larger tumor size, and malignant pathologic diagnosis [1, 15–16]. The tumors rarely spread via the lymphatic system and axillary lymph node metastasis rate is < 5%. Therefore, axillary lymph node dissection is unnecessary, yet the removal of suspicious axillary lymph nodes is recommended [17, 18]. In this case, axillary lymph node biopsy was carried out because some lymph nodes were found adjacent to the tumor during surgery. However, postoperative pathology proved that all the lymph nodes were negative. Studies have shown that adjuvant radiotherapy can lower the rate of local reoccurrence, particularly for patients with positive margins for borderline and malignant tumors [19–21]. Margin-negative resection combined with adjuvant radiotherapy is very effective for local control and prevention of recurrence [6]. Adjuvant chemotherapy is not the standard care since it is of controversial value for malignant phyllodes tumors, yet some institutions support doxorubicin-based adjuvant chemotherapy for first-line treatment of breast sarcomas, especially with > 5.0 cm large high-risk tumors [22–23]. During postoperative follow-up, no local recurrence or distant metastasis were found.

**Conclusion**

The kiss flap could be considered as an effective method to repair large chest wall defects after resection of giant phyllodes tumors.

**Declarations**

**Ethics Statement**

We obtained written informed consent from the patient presented in this case report in accordance with the Helsinki Declaration of 1975. The patient consented to the treatment and the use of her data, including photos, for research, and publication. This study was approved by the medical ethics committee of Shenzhen People's Hospital.

**Consent**

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

**Availability of Data and Materials**

This was not applicable to this manuscript.
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Conflict of Interest Statement

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Author Contributions Statement

YL and WZ wrote the original draft of the manuscript. YL, CZ, PZ, YZ, DZ and WZ were involved in data acquisition. CZ and PZ acquired the funding. JH and JL provided pathology figures and review. WZ revised the manuscript for critical intellectual content. All authors read and approved the final manuscript and were involved in the review and editing of the manuscript.

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Figures

Figure 1

Imaging data of the patient. (A) Ultrasound of the right breast mass shows small cystic components, calcification, and hyperechoic separations. (B) Magnetic resonance imaging of the breast reveals a giant, lobulated mass with heterogeneous signal intensity, as well as septa and well-circumscribed margins, typical of a phyllodes tumor.

Figure 2
(A) Preoperative view. (B) The skin defect measuring 25×15 cm. (C) A double-paddle latissimus dorsi kiss flap is marked on the patient's back, with each flap measuring 15 × 6 cm. The two flaps have an angle of about 90 degrees. (D) Harvesting of parts A and B of the two-lobed flap. (E) Flaps A and B are sutured together before being transposed to the chest wound. (F) Immediate postoperative view.

**Figure 3**

Postoperative view of the chest at 3 months.

![Figure 3](image)

Figure 4

Macroscopic findings and pathology images of the right breast tumor. (A) The tumor measured 18 × 14 × 14 cm with massive hemorrhagic necrosis. (B) The cut surface of the tumor. (C) Tumor cells are mainly

![Figure 4](image)
spindle cells, which are bidirectionally differentiated. (D) Tumor cells proliferate actively, mitotic figures are easy to see, and the cells are moderately and severely atypia.