

Case report

Resection and reconstruction following recurrent malignant phyllodes—Case report and review of literature



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HIGHLIGHTS

- Aggressive surgical approach is advocated for locally advanced malignant tumors.
- Pathological progression of histology may occur in phyllodes tumors.
- Phyllodes tumors do not respond well to radiotherapy.

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ABSTRACT

Introduction: Phyllodes tumors are uncommon biphasic fibroepithelial neoplasms of the breast of varying malignant potential occurring in middle aged women. They exhibit diverse biological behavior. Margin free excision is the mainstay of treatment.

Case presentation: A 27 year-old lady was referred with a painless ulceroproliferative right breast lesion which had rapidly progressed over six months. Three years back, she had been diagnosed with a borderline phyllodes tumor and underwent a wide local excision followed by a right mastectomy for recurrence. The resection margins were positive hence she underwent postoperative radiation. We performed a radical resection of the chest wall and reconstruction using a composite mesh (inner PTFE and outer vypro), pedicled latissimus dorsi flap and a split skin graft for the recurrent malignant tumor. She recovered uneventfully thereafter.

Discussion: Malignant phyllodes tumor is uncommon and treatment principles are from case reports and retrospective studies. Aggressive resection of the lesion and reconstruction of the chest wall with bone cement and two meshes—a composite mesh (inner layer -polytetrafluoroethylene and outer layer of polypropylene) and a Vypro mesh is a possibility. This case highlights the challenges encountered in managing these patients and presents a radical solution.

Conclusion: Treatment of phyllodes tumor necessitates adequate excision of the tumor and adjacent tissues to ensure tumor free margins. Pathological evolution from intermediate to malignant histology may be exhibited. A full-thickness chest wall resection and reconstruction although radical is a feasible option as these tumors rarely respond to other modalities of cancer management.

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1. Introduction

Phyllodes tumors are biphasic fibroepithelial neoplasms of the breast in which the stroma may show diverse biological behavior. First described by Johannes Muller, they represent roughly 0.3–0.9% of all breast cancers in women [1]. Benign, borderline and

malignant histotypes are determined based on histology. Malignant phyllodes tumors can both recur and metastasize.

We present a patient with recurrent phyllodes tumor who was managed in a tertiary hospital. The current standard of care is a mastectomy. Our management of this recurrence involved a radical resection of the chest wall with a subsequent reconstruction using a double mesh.

1.1. Patient information

A 27 years-old lady was referred to our institution with a history

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of painless ulceroproliferative right breast lesion, rapidly progressive over the last six months (Fig. 1).

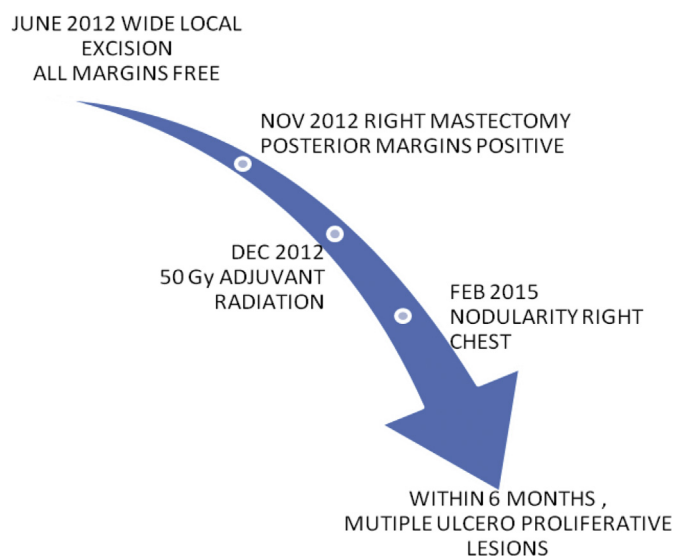
1.2. Clinical findings

On examination, an ulceroproliferative lesion of 18 × 15cm with an isolated mass of nodular growth was seen on the right breast. The surrounding skin was erythematous. The growth was firm in consistency and fixed to the underlying chest wall. The left breast was normal and there were no palpable lymph nodes bilaterally.

1.3. Timeline

Three years back, this patient was diagnosed with a borderline phyllodes tumor on core needle biopsy and underwent wide local excision. Postoperative histopathology confirmed the diagnosis and the resected margins were tumor free. However the lump recurred in 4 months and she underwent a right mastectomy. The resected margins were positive and hence postoperative radiation of 50Gy in 18 fractionated doses was administered. She was asymptomatic on follow up but defaulted follow up after one year.

Six months back she developed a small nodularity over the right chest which rapidly progressed into several ulceroproliferative lesions. She has no family history of breast cancer. There was no other medical or surgical history. She attained menarche at 13 years of age and had never been pregnant.



1.4. Diagnostic assessment

MRI chest revealed a heterogeneously enhancing multilobulated lesion with solid and cystic components suggestive of malignant phyllodes tumor invading the chest wall (4th/5th ribs and serratus anterior). Core needle biopsy confirmed the diagnosis. CT chest, brain and ultrasound of abdomen showed no evidence of distant metastasis. A high grade recurrent non-metastatic malignant phyllodes tumor was diagnosed. A multidisciplinary approach for radical resection of the chest wall and reconstruction was proposed by the tumor board.

1.5. Therapeutic intervention

The surgery was performed by a senior surgical oncologist through an elliptical incision with an aim to achieve a skin clearance of 2 cm on all sides. The tumor was excised in-toto with the overlying skin, pectoral muscles, anterior half of 3,4,5,6 ribs, right sternal border, intercostals and the parietal pleura. The thoracodorsal neurovascular bundle was preserved. The residual defect measured 13.5 cm × 18.5 cm. A 32F intercostal drainage tube was inserted in the right pleural cavity. Reconstruction was performed using a composite mesh (inner layer -polytetrafluoroethylene and outer layer of polypropylene) of size 15.9 × 21cm fixed all around with prolene. The PTFE layer prevents the mesh adhering to the lung. This method is unique to our reconstruction and has not been adopted widely [2].

Bone cement (Polymethylmethacrylate) was fixed over the mesh to provide bony support and a Vypro mesh of size 15 × 15cm was fixed over the bone cement to prevent adhesion of the flap (Fig. 2). A pedicled right latissimus dorsi muscle flap (Fig. 3) was mobilized to provide muscle cover and overlaid by split skin graft harvested from right thigh.

1.6. Follow-up and outcomes

She made an uneventful recovery. Intercostal drainage was removed on the third postoperative day and suction drain on sixth day. Histopathology was consistent with malignant phyllodes tumor. Microscopic sections demonstrated large, simple ducts surrounded by a uniform bland stroma and the stromal cells were spindle-shaped featuring elongated nuclei showing high mitotic activity. The margin of resection was negative with a tumor-free zone of 1.8cm. She is under periodical follow up and there have been no signs of recurrence or distant metastasis for the past 1 year.

2. Discussion

This case highlights the radical approach aimed at curing the radical chest wall infiltration and reconstruction. This patient was diagnosed accurately by a core biopsy initially and had a wide local excision with breast conservation. Immediate recurrence necessitated a mastectomy with subsequent radiotherapy.

Phyllodes tumor represents less than 1% of all female breast tumors. With an incidence of 2.1/million, it occurs in women aged 45–49 years [3,4]. The tumor may be benign, intermediate or malignant. Low-grade phyllodes lesions resemble fibroadenomas but are more cellular and contain mitotic figures. The frequency of chromosomal changes increases with grade and EGFR amplification is present in high grade tumors [5].

Patients typically present with a mobile, macrolobulated, and painless mass. There are no pathognomonic mammographic or ultrasound features that differentiate it from a fibroadenoma [6]. Hence resection may be suboptimal. Ultrasound findings of heterogeneous internal echo patterns, lack of microcalcifications and smooth margined fluid-filled clefts in a predominantly solid mass may suggest phyllodes tumor [7]. These tumors are usually larger and grow more rapidly than fibroadenomas [8]. A leafy internal morphology on subtraction MRI due to the enhancing cotyledonous solid portions within irregular blood-filled cystic spaces may be pathognomonic of a phyllodes tumor [9].

Cytological similarity makes distinction between phyllodes and fibroadenoma difficult and core tissue biopsy is preferred for pre-operative diagnosis [10]. A core needle biopsy may be false negative rate in 39% of patients [11]. Hence even if a biopsy is benign or indeterminate, rapid growth warrants excision. Histologic appearance may not correlate with clinical behavior as both malignant and



Fig. 1. Patient presentation.

borderline tumors can metastasize [12].

There is a minimal role for hormonal therapy as the estrogen receptors (ER) are of 'beta' subtype whereas SERMs act upon the 'alpha' subtype. Receptors are absent in the malignant stromal component of the tumor.

Phyllodes tumors uncommonly metastasize via the lymphatics (5%). The lungs are the most common metastatic site (hematogenous), followed by the skeleton, heart, and liver [13]. Lymphadenopathy is usually due to the patient's immune response to tumor necrosis but sentinel node biopsy is not accurate. Patients with clinically palpable axillary nodes need axillary dissection. Sentinel node biopsy may be done if nodes are not palpable [14].

Complete margin free excision of at least 1 cm is considered appropriate primary excision [15]. Mastectomy may be required for larger tumors between 5 and 10 cm size and tumor location [16].

Chemotherapy and radiation therapy are controversial.

Chest wall invasion is uncommon with phyllodes tumors. Reinfuss et al. reported that 2.4% of phyllodes tumors in their series clinically infiltrated the pectoralis major muscle [16]. Moore and Kinne recommend extended excision of involved pectoralis muscle, followed by reconstruction of the chest wall with Marlex mesh and methylmethacrylate [17]. Post-operative radiation for cases of chest wall infiltration has also been advocated [14].

Our patient presented with extensive chest wall infiltration involving ribs 3 and 4. Hence an appropriate resection was essential as the patient has already had a prior radiotherapy with 50Gy. However, there was a large residual defect which needed extensive reconstruction. A composite mesh (inner layer -polytetrafluoroethylene and outer layer of polypropylene) of size 15.9×21 cm was fixed all around using 1 prolene. The PTFE layer prevents the mesh



Fig. 2. Coverage of defect with composite mesh, Bone cement and Vypro mesh.



Fig. 3. Pedicled LD flap cover.

adhering to the lung. Bone cement (Polymethylmethacrylate) was prepared and fixed over the mesh to provide bony support as a replacement to the resected ribs. This is used in thoracic wall reconstruction but is not common after mastectomy. The extensive resection of a third of the rib cage required this unique alternative with interdisciplinary skill transfer. LeRoux and Shama [18] have set forth the ideal characteristics of a prosthetic material—rigidity to abolish paradoxical chest motion, inertness to allow ingrowth of fibrous tissue and decrease infection, malleability for appropriate fashioning and radiolucency to allow radiographic follow-up. Bone cement is almost ideal and has been shown to be appropriate for large chest wall defects.

A Vypro mesh of size 15 × 15 cms was fixed over the bone cement to prevent adhesion of the flap. A pedicled right latissimus dorsi muscle flap was mobilized to provide muscle cover and this was overlaid by a split skin graft harvested from right thigh.

Recurrent phyllodes tumor, post-mastectomy can be treated with aggressive resection and immediate reconstruction to ensure optimal outcome. Long term outcomes of this procedure are awaited. These patients need careful follow up to monitor for recurrence.

3. Patient perspective

This extensive resection with immediate reconstruction offered substantial emotional respite to this young patient and aimed to be curative. Mandel et al. reported a patient in whom subcutaneous mastectomy was followed by immediate prosthesis implantation. This did not prevent the detection of recurrence and benefitted the patient emotionally [19]. Though we did not perform a prosthetic implant, immediate reconstruction is aesthetically more appealing following extensive surgery. Currently this patient is under our follow up for the last 1 year and there is no detectable recurrence.

4. Informed consent

This publication is subsequent to a documented consent from the patient after adequate explanation and assurance of anonymity.

Ethical approval

Not applicable.

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Author contribution

Mohammed Farooq—Acquisition of data, performance of surgery and viewing the final draft.

Aashish Rajesh—Data analysis and writing the paper.

Both authors consent to share the post of first authors as equal contributions were made.

Conflict of interest

All authors have no conflict of interests to reveal.

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This case report has been reported in line with the SCARE criteria [20].

Appendix A. Supplementary data

Supplementary data related to this article can be found at <http://dx.doi.org/10.1016/j.amsu.2017.02.007>

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