CASE REPORT PEER REVIEWED | OPEN ACCESS

A large hemangioma of the breast influencing the management of breast cancer

Madeleine Louise Kelly, Marli Williams, Clement Wong, Grace T Kwok

ABSTRACT

Introduction: Hemangiomas are a rare well-demarcated proliferation of mature blood vessels lined by a single layer of endothelial cells occurring in up to 1.3% of mastectomy specimens. Accurate diagnosis often requires the incorporation of clinical, radiological, and histological features.

Case Report: A 61-year-old female presented with a self-detected right breast lesion. Initial work-up revealed a right breast lesion at 12 o'clock with core biopsy confirming intermediate grade ductal carcinoma in situ. The management was complicated by the presence of a large right chest wall congenital cavernous hemangioma. Wide local excision of the breast lesion was performed with no sentinel lymph node biopsy done due to extensive vascular malformation. Further excision of margins was required to achieve surgical clearance.

Conclusion: Despite its rarity, clinicians should be aware of the potential occurrence of a hemangioma on the breast and potential implications for the surgical management of breast cancer.

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INTRODUCTION

Vascular lesions are not commonly encountered on the breast and represent as a significant diagnostic dilemma due to the overlap in morphological features between benign vascular lesions and the malignant counterparts. Breast hemangiomas account for approximately 0.4% of all breast tumors [1]. Breast hemangiomas can be further classified into cavernous and non-cavernous subtypes with cavernous type being most common [1, 2]. Breast hemangiomas usually appear as superficial lobular masses with circumscribed margins both visually and on radiological modalities. Typically managed either non-operatively or with surgical excision, hemangiomas carry an excellent prognosis with a low recurrence rate [1]. On the histological specimen there will typically be endothelial-lined dilated vascular channels often with absence of cellular atypia [1–3]. Due to its rarity, hemangiomas on the breast can present a diagnostic challenge making it essential to recognize its pathology, thereby distinguishing it from other forms of neoplasms.

CASE REPORT

A 61-year-old female presented with a self-detected right breast lump associated with nipple puckering. Initial

work-up investigations of ultrasound and mammogram revealed a 4 cm right breast lesion at 12 o'clock, 1 cm from the nipple (Figure 1) BIRADS 6 and a left breast lesion at 10 o'clock BIRADS 4. Core biopsies of bilateral lesions showed benign pathology of the left breast lesion and a 40 × 14 × 24 mm lesion of intermediate grade ductal carcinoma in situ in the right breast lesion with no evidence of invasive carcinoma. Her comorbidities of significance include a congenital cavernous hemangioma over right chest wall, past history of radiation exposure at age 3 due to childhood treatment of leukemia, hypertension, and benign liver cysts. Her risk factor profile included a family history of breast cancer (mother and maternal aunt), no breastfeeding history and a hysterectomy and oophorectomy after diagnosis of cervical cancer.

Given the history of a large cavernous hemangioma over the right chest wall and the patient's examination revealing an ashen gray appearance and extensive venous malformation along the right posterolateral chest wall extending from the angle of scapula to sternum (Figure 2) the decision was made for further imaging and referral to the vascular team before further management. Computed tomography with intravenous (IV) contrast in the arterial phase revealed a large malformation involving the subcutaneous tissue and skin extending between the anterior 6th to 9th ribs to the lower mediastinum and various hypoattenuating lesions consistent with liver cysts (Figure 3).

After vascular consultation and magnetic resonance imaging of the chest was performed, the decision was made to proceed with operative management. A ROLLIS guided wide local excision of the right breast lesion was performed. The procedure was performed under general anesthesia. The ROLLIS seed in the specimen was identified and confirmed radiologically and the vascular team was on standby. No sentinel lymph node biopsy was performed despite the mass-forming nature of the right breast lesion due to the extensive vascular malformation in the right axilla. Radiological imaging had suggested no lymphatic involvement and with the absence of biopsy proven invasive disease this decision seemed sound. A facial skin biopsy was also performed for the patient's grayed skin appearance. The patient recovered well post-operatively and was discharged home the following day.

Pathological analysis of the specimen showed 45 mm of intermediate grade ductal carcinoma in situ (DCIS), with no invasive carcinoma. Receptor status was ER+, PR+, HER2–. The posterior and medial margins were involved in DCIS. The patient was taken to theater to re-excise the cavity to achieve surgical clearance. After re-excision, the posterior margin was still involved but no further resection was deemed possible due to the extensiveness of the hemangioma. As such the decision was made to proceed with adjuvant radiation therapy and hormonal therapy. Interestingly, the facial skin biopsy showed features consistent with argyria, with no features of vascular malformation.

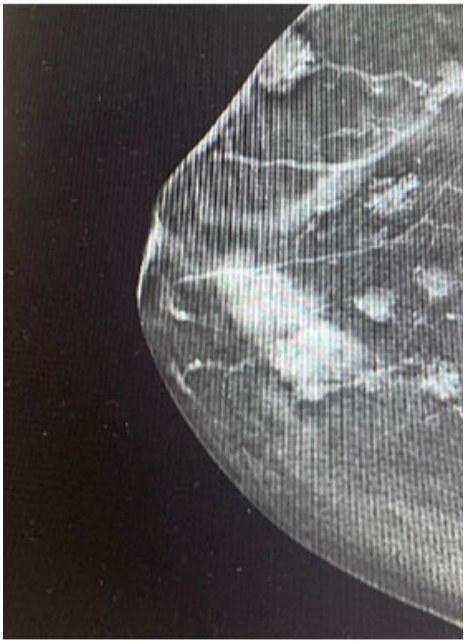

Figure 1: Mammogram revealing a 40 mm. Right breast lesion with extensive cavernous hemangioma.

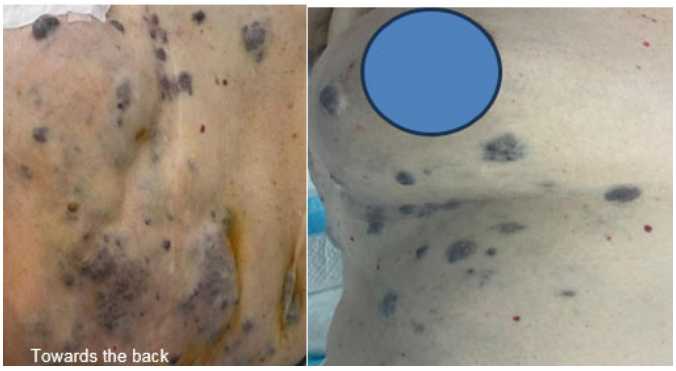

Figure 2: Extensive congenital venous malformation along posterolateral chest wall extending from angle of scapula to sternum.

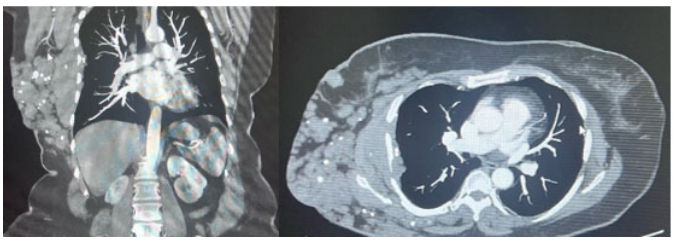

Figure 3: Large malformation involving the subcutaneous tissue and skin extending between the anterior 6th to 9th ribs to lower mediastinum and various hypoattenuating lesions consistent with liver cysts.

DISCUSSION

Hemangiomas are a well-demarcated proliferation of mature blood vessels lined by a single layer of endothelial cells and pericytes lacking atypia. Breast hemangiomas are a rare benign vascular lesion occurring in patients as

young as 18 months [1–3]. There are multiple types of hemangiomas identified based on different histological appearance, age, presentation, and location. Most breast hemangiomas are small, incidentally found tumors and are designated as peri-lobular hemangiomas [3–5]. These are found in up to 1.3% of mastectomy specimens and 11% of postmortem breast specimens [5]. The larger clinically or radiologically detectable tumors are hemangiomas, which are further classified into cavernous, capillary, complex, or venous types [4, 5]. Hemangiomas rarely present as a palpable breast mass. In order to get an accurate diagnosis it is important to incorporate clinical and radiological information, morphological features, and immunohistochemical results [1–5].

Gross examination of specimens of breast hemangiomas often show a circumscribed red or dark brown lesion with a spongy cut surface [1, 2]. The lesions are mostly located in the subdermal or subcutaneous tissue and are rarely intraparenchymal [2–4]. These lesions usually cause slight discoloration to the overlying skin due to large draining veins. On imaging there are no pathognomonic features, however, on mammography or ultrasound, hemangiomas are often oval or lobular solid masses with varied echotexture which may contain calcifications [5]. Hypovascularity has been reported in benign lesions, and hypervascularity in malignant lesions. On magnetic resonance imaging, breast hemangiomas usually present as an ovoid mass with circumscribed borders isointense to the surrounding fibroglandular tissue on T1-weighted images and homogenously hyperintense on T2-weighted images [5]. The lesions tend to have slow, delayed enhancement, indicating slow flowing blood. On the histological specimen there will be endothelial-lined dilated vascular channels often with absence of hemorrhage, necrosis, destruction of lobules, or cytologic atypia [1–5].

Atypical hemangiomas may have vascular proliferation with atypical features such as conspicuous vascular anastomoses, papillary endothelial hyperplasia, ill-defined or invasive borders, nuclear atypia, and the presence of rare mitotic figures [1–3]. Immunohistochemical markers to confirm the vascular nature of the lesion include ERG, CD31, CD34, FLI-1, D2-40, VEGFR, and Factor VIII. ERG is a nuclear specific stain and is one of the best available nuclear endothelial markers with a sensitivity of 98–100% and a nearly 100% specificity for identification of vascular origin [1–4].

Management varies from a non-operative/expectant approach with imaging and follow-up to excisional biopsy for definitive diagnosis or complete surgical excision with very low recurrence rate [6]. Significant overlap between benign vascular lesions and malignant lesions, often leads to excisional biopsies being performed. In the past, the diagnosis of hemangiomas on breast core biopsy resulted in surgical excision to exclude well-differentiated angiosarcoma but in an article by Sebastiano et al. it was found that in 22% of targeted and 23% of incidental lesions, no change in pathological classification was made [6]. Despite this surgical excision is still recommended if the lesion is larger than 2 cm, has atypical features on core biopsy, or has high rate of growth or development which can be concerning for a malignant vascular tumor, such as an angiosarcoma which has a very poor prognosis [6]. Other differentials between angiosarcomas include angiolipomas.

Despite its rarity, clinicians should be aware of the potential occurrence of a hemangioma on the breast and potential indications for surgical excision [1–5]. In the setting of intercurrent breast cancer management, clinicians may need to modify standard of practice treatment aspects including surgical nodal assessment and margin management with extended adjuvant treatment options including local systemic hormonal therapy. In other circumstances, the extent of the hemangiomas may limit oncoplastic options due to the subdermal and subcutaneous nature of the lesions [2, 3].

CONCLUSION

Breast hemangiomas are a rare, benign lesion that maybe discovered incidentally or on macroscopic examination of the breast. Typically managed either non-operatively or with surgical excision, hemangiomas carry an excellent prognosis. Lesions larger than 2 cm or with atypical features should be monitored as they are likely to be malignant vascular tumors such as angiosarcoma. Surgical treatment remains a viable option in suspicious lesions or if required for clear margins in malignant breast patients.

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Author Contributions

Madeleine Louise Kelly – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Marli Williams – Conception of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Grace T Kwok – Conception of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.