CASE REPORT

Primary Osteosarcoma of the Breast presenting as a Large Breast Abscess

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ABSTRACT

Introduction: Primary extraosseous osteogenic sarcoma is one of the rarest forms of malignant tumor of the breast. It can arise as a result of osseous metaplasia of a preexisting neoplasm or from a none-phylloides sarcoma of a previously normal breast. Due to its rarity, the natural history and optimal treatment methods remain unclear.

Case presentation: A 60 years old patient presented to the surgical casualty with a large breast abscess. Abscess wall histology revealed an osteosarcoma of the breast. Left total mastectomy with axillary clearance was performed. Histology and subsequent immunohistochemical studies confirmed the diagnosis of osteogenic sarcoma without lymph nodal metastasis. Patient was referred to the oncologist for further management.

Conclusion: Rare types of breast tumors can be presented as breast abscess. Incision and drainage together with wall biopsy aid to exclude associated sinister pathologies. Diagnosis of primary osteosarcoma of the breast was made using histological and immunohistochemical findings once the possible primary from the bones of sternum and ribs was excluded. Treatment is as for sarcomas affecting other locations and should comprise a multidisciplinary approach.

Keywords: Breast, Breast abscess, Primary osteosarcoma.


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INTRODUCTION

Breast cancer is the commonest cancer in females. Primary osteosarcoma of the breast is a very rare but aggressive malignancy.  
1,2 Histological differentiation, from conventional osteosarcoma of bone or other extraskeletal forms of osteosarcoma, is difficult.  
1,2 Extraskeletal forms of osteosarcoma have been reported in other organs like thyroid, kidney, bladder, colon, heart, testes, penis, gall bladder, and the cerebellum.  
2 Although primary osteosarcoma of bone is common in the young, primary osteosarcoma of the breast is seen in an older age group with a mean age of 65 years.  
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CASE REPORT

A 60-year-old unmarried lady presented to the surgical casualty ward, with a large abscess in left breast. She had noticed a lump on her left breast about 3 months ago, which rapidly increased in size over the last month showing signs of inflammation. On clinical examination, breast was grossly distended with features of inflammation. The consistency of the lump was irregular with many hard areas with cystic areas interspersed, suggestive of pus collection. Some enlarged lymph nodes were noted in the axilla. Imaging revealed a breast abscess with variable echogenic areas. Incision and drainage of the abscess done and pus sent for culture and antibiotic sensitivity test (ABST). Histological examination of abscess wall and breast tissue revealed a primary osteogenic sarcoma of the breast. Total mastectomy and level 2 axillary lymph node clearance was performed.

Macroscopic examination of the mastectomy specimen shown 100 × 80 × 90 mm size growth spreading across both upper quadrants as well as the lower lateral quadrant. Microscopy revealed a tumor composed of markedly pleomorphic spindle cells admixed with multinucleate osteoclast like giant cells with areas showing tumor osteoid formation. Scattered, round neoplastic cells were also observed. A high mitotic activity was noted [6/10 high power field (HPF)] with numerous cells with apoptotic nuclei. There was minimal tumor necrosis. The overall appearances revealed osteosarcoma of the breast (Figs 1A to C). As the tumor showed prominent osteoblastic differentiation with malignant osteoid formation, it was classified as osteoblastic osteosarcoma. Immunohistochemical assessment confirmed the diagnosis as high grade osteosarcoma of the breast. All level I and II lymph nodes revealed reactive changes only. Multiple sections of the breast lump failed to reveal any other epithelial or sarcomatous component, suggestive of carcinosarcoma. No primary tumor elsewhere was present.
Mammography of the non-affected breast was normal. Other investigations, such as ultrasound scan of the abdomen, staging contrast-enhanced computed tomography (CECT) abdomen and, chest and skeletal scintigraphy showed no evidence of metastatic disease. Following surgery patient was referred to oncologist for adjuvant chemotherapy (Fig. 2).

**DISCUSSION**

Primary osteosarcoma of the breast is a rare malignant tumor and accounts for less than 1% of all primary breast malignancies.\(^1,3,4\) It commonly affects elderly women, but has also been reported in men.\(^5\) It is a highly aggressive tumor, accounting for about 12.5% of mammary sarcoma.\(^1\)

Histogenesis of this rare tumor is not clear. Origin from totipotent mesenchymal cells of the breast stroma or as a transformation from preexisting fibroadenoma or phyllodes tumor has been suggested.\(^3,4,8\) Absence of metaplastic transformation of a preexisting fibroadenoma or phyllodes tumor suggests that this tumor arose from previously normal breast tissue.

Immunohistochemistry plays an important role to differentiate mesenchymal neoplasia from undifferentiated carcinoma. After the exclusion of epithelial neoplasia, it is necessary to define the histogenesis of the lesion.\(^9\) Tumor cells were strongly positive for vimentin, a mesenchymal marker. Pan cytokeratin (epithelial cell marker), CD34 and bcl-2 were negative in tumor cells (Fig. 1C).

Primary osteosarcoma of the breast should be differentiated from two other similar entities, namely, malignant phyllodes tumor and metaplastic carcinoma. This can be done by specific morphological features with the presence of carcinomatous component and its cytokeratin immunopositivity respectively.\(^6\)

Malignant phyllodes tumor consists of a predominant mesenchymal component and a benign epithelial component.\(^9\) Macroscopically, malignant lesions show necrotic and hemorrhagic areas with infiltrative growth pattern. Stromal features simulate a sarcoma. Although markedly pleomorphic spindle cells admixed with multinucleated giant cells are seen in this patient, the lack of epithelial component in multiple random sections examined ruled out the diagnosis of malignant phyllodes tumor.
Metaplastic carcinoma is a term used to define breast tumor that shows features different to that of epithelial or typical ductal carcinoma. This rare form of heterogenous neoplasm is characterized by mixture of adenocarcinoma with areas of spindle cells, squamous cells and other cells of mesenchymal differentiation. But none of these features were present in this specimen. This form of tumor shows more aggressive behavior than typical ductal carcinoma.

Differential diagnoses include secondary lesion from a primary osteosarcoma of bone or direct extension of an osteogenic sarcoma arising from nearby ribs or sternum. This patient revealed no radiological evidence of primary osteosarcoma of bone. No underlying bone pathology was noted during surgery of the region. The tumor was not infiltrating through the chest wall musculature and there was no evidence of microscopic infiltration of chest wall musculature by the tumor histologically.

It is very difficult to diagnose osteosarcoma of the breast from the clinical features, mammogram and ultrasound scan alone. For example, this patient presented with large breast abscess. Mammographically osteosarcoma of the breast appears as, well circumscribed dense lesions which have regular or irregular borders, with focal or extensive coarse calcification in one-third of cases. Its appearance may imitate a benign fibroadenoma.

The basic requirement for the diagnoses of a primary osteosarcoma of the breast, according to Allan and Soule include:

- The presence of neoplastic osteoid or bone
- The exclusion of origin in the bone and
- The absence of epithelial component.

In this patient, negative results of an isotope skeletal bone scan aided in excluding the possibility of a primary tumor while histological and immunohistochemical analysis showed no evidence of epithelial differentiation.

Osteosarcoma is classified into many subtypes; the commonest are fibroblastic, osteoblastic, and osteoclastic osteogenic sarcomas. Fibroblastic osteogenic osteosarcoma being associated with better survival outcome makes histological differentiation important.

Early recurrence and propensity for hematogenous rather than lymphatic spread has been noted in this highly aggressive rare tumor. Common sites for hematogenous metastasis are the lung (80%), bone (20%), and liver (7%).

As the optimal management of localized disease includes total excision of the neoplasm with adequate resection margins, mastectomy was performed for this patient in order to control local recurrence. Although axillary lymph nodes clearance is not indicated for negative nodes, as we did not have facilities to assess fresh frozen section, level 2 axillary clearance was performed. In the presence of a partial epithelial differentiation, the basic requirement for the diagnosis of a primary sarcoma of the breast is the exclusion of epithelial origin, which requires axillary lymph node dissection. In that purpose, axillary clearance was done, although biopsy report revealed it as an osteosarcoma of the breast.

In the presence of lymph node metastasis diagnosis of metaplastic carcinoma should be considered. In this patient, all lymph nodes were free of metastasis. However, axillary dissection has been generally considered unnecessary for diagnosed osteosarcoma of the breast since these tumors rarely spread through the lymphatic system.

Limited data on osteosarcoma indicate an aggressive clinical course and high incidence of recurrence and metastasis. Its management is significantly different from management of breast adenocarcinoma while it was the most important predictor of long-term survival in patients with different histological types of breast sarcoma.

Surgical management is the mainstay of treatment for breast osteosarcoma.

As with all rare tumors osteosarcoma should be managed in reference centers to determine whether treatment of choice is surgical excision or total mastectomy and whether some form of adjuvant therapy can have beneficial effects. Indications for adjuvant therapy should follow those for other soft tissue sarcoma.

The role of adjuvant chemotherapy and/or radiotherapy also has been unclear although this patient is receiving chemotherapy at the moment.

Following curative resection of the primary tumor the role of postoperative radiotherapy and chemotherapy is still controversial. Although several studies report adjuvant chemotherapy may be of value in patient management, use of adjuvant radiotherapy remains unclear. But chemotherapy is the main treatment modality for metastatic disease.
Prognostic factors for osteosarcoma include: Tumor size, number of mitosis, presence of stromal atypia, histological subtype, and resection margin involvement. The limited number of cases reported so far in the literature leaves a lot of controversy regarding the long-term prognosis of this disease.

REFERENCES