Case Report

Primary osteosarcoma of the breast presented as a large breast abscess

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ABSTRACT

Primary extra osseous 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 - phyllodes sarcoma of a previously normal breast. Due to its rarity, natural history and optimal treatment methods remain unclear. A 60-year-old patient presented to the surgical casualty with 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. The patient was referred to the oncologist for further management. Rare types of breast tumors can be presented as breast abscess. Incision and drainage together with wall biopsy help 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 sternum and ribs were excluded. Treatment is as for sarcomas affecting other locations and should comprise a multidisciplinary approach.

Key words: Breast, breast abscess, primary osteosarcoma

INTRODUCTION

Breast cancer is the most common cancer in females. Primary osteosarcomas of the breast are extremely rare and represent about 12.5% of the mammary sarcomas and <1% of all primary breast malignancies.[1] Histological differentiation, from conventional osteosarcoma of bone or other extraskeletal forms of osteosarcoma, is difficult.[1] 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 among the young, primary osteosarcoma of the breast is seen in an older age group with a mean age of 65 years.[1,3]

CASE REPORT

A 60-year-old unmarried lady presented to the surgical casualty ward, with a large abscess in the left breast. She had noticed a lump on her left breast about 3 months ago, which had 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. Histological assessment of the abscess wall and the breast tissue revealed a primary osteogenic sarcoma of the breast. Total mastectomy and level 2 axillary lymph node clearances were performed.

Macroscopic examination of the mastectomy specimen revealed a 100 mm × 80 mm × 90 mm size growth spreading across both upper quadrants and the lower lateral quadrant.

Microscopy revealed a tumor composed markedly of pleomorphic spindle cells admixed with multinucleate osteoclast-like giant cells with areas showing tumor osteoid formation. Scattered, round neoplastic cells were also observed. High mitotic activity was noted (6/10 HPF) with numerous cells with apoptotic nuclei. There was a

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minimal tumor necrosis. The overall appearances revealed osteosarcoma of the breast [Figure 1a and b].

Because 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 level 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 was present elsewhere.

Mammography of the nonaffected breast was normal. Other investigations, such as an ultrasound scan of the abdomen, staging contrast enhanced computerized tomography of abdomen and chest and skeletal scintigraphy did not show evidence of metastatic disease. Following surgery, the patient was referred to the oncologist for adjuvant chemotherapy [Figure 2].

DISCUSSION

Primary osteosarcoma of the breast is a rare malignant tumor and accounts for <1% of all primary breast malignancies.[1-3] It commonly affects elderly women but has also been reported in men.[1,3] It is a highly aggressive tumor, accounting for about 12.5% of the 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.[2,4] Absence of metaplastic transformation of a preexisting fibroadenoma or phyllodes tumor suggests that this tumor has risen from previously normal breast tissue.

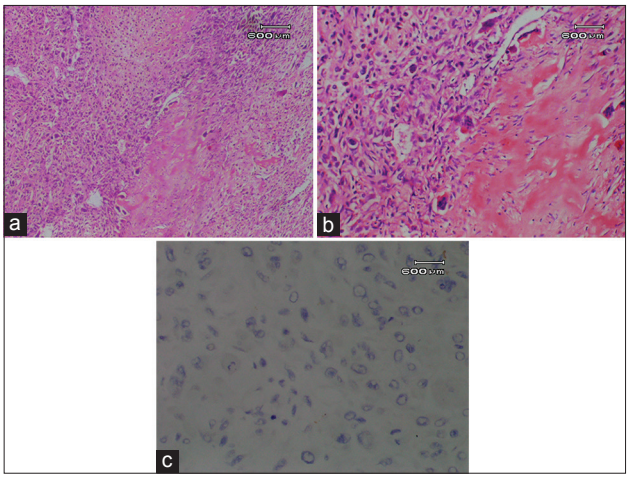

Figure 1: (a) Primary osteosarcoma of the breast showing tumor osteoid, osteoblast, and osteoclast-like giant cell proliferation (×10) (b) primary osteosarcoma of the breast-high power to show tumor osteoid formation (×40) (c) pan cytokeratin (CK) stain-tumor cells do not express epithelial marker pan CK

Immunohistochemistry plays an important role in differentiating the mesenchymal neoplasia from undifferentiated carcinoma. After exclusion of epithelial neoplasia, it is necessary to define the histogenesis of the lesion. Tumor cells were strongly positive for vimentin, a mesenchymal marker. Pan cytokeratin (CK) (epithelial cell marker), CD34 and bcl-2 were negative in tumor cells [Figure 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 CK immunopositivity respectively.[5]

Malignant phyllodes tumor consists of a predominant mesenchymal component and a benign epithelial component.[6] 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 the 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 a mixture of adenocarcinoma with areas of spindle cells, squamous cells, and other cells of mesenchymal differentiation.[7] But none of these features was present in this specimen. This form of tumor

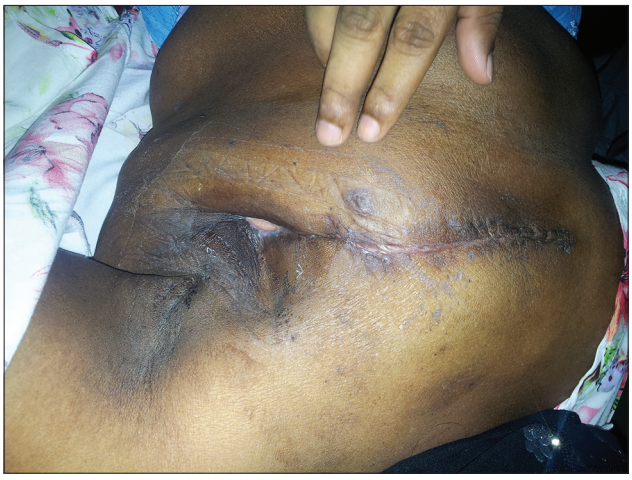

Figure 2: Macroscopic appearance following surgery: During recovery

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 the chest wall musculature histologically.

It is very difficult to diagnose osteosarcoma of the breast by clinical features, mammogram, and ultrasound scan alone; the present 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.[8]

The basic requirement for the diagnosis of a primary osteosarcoma of the breast, according to Allan and Soule include: (a) The presence of neoplastic osteoid or bone (b) the exclusion of origin in the bone and (c) the absence of epithelial component.[9]

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

Osteosarcoma is classified into many subtypes; the most common are fibroblastic, osteoblastic, and osteoclastic osteogenic sarcomas. Fibroblastic osteogenic osteosarcoma being associated with better survival outcome makes histological differentiation important.[10,11]

Early recurrence and propensity for hematogenous rather than lymphatic spread have been noted in this highly aggressive rare tumor. Common sites for hematogenous metastasis are lungs, bone, and the liver.[12]

As the optimal management of localized disease includes total excision of the neoplasm with adequate resection margins, mastectomy was performed on this patient in order to control local recurrence. Although axillary lymph nodes clearance is not indicated for negative nodes, since we do 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.[12,13]

Limited data on osteosarcoma indicate an aggressive clinical course and a high incidence of recurrence and metastasis. The management is significantly different from the management of breast adenocarcinoma while it was the most important predictor of long-term survival in patients with different histological types of breast sarcoma.[13] 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.[14] 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.[15]

Prognostic factors for osteosarcoma include tumor size, number of mitosis, presence of the 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.

This was another unusual presentation of osteosarcoma of the breast where definitive diagnosis was made based on histology and immunohistochemistry reports after excluding an osteogenic sarcoma arising from underlying ribs and sternum. To date, adequate surgical excision is the best form of treatment that offers the best outlook for patients. However, its rarity, should not prevent clinicians from following up on the literature of the disease to keep

abreast of new cases and possibly new developments of its management.

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