Case Report

Primary breast osteosarcoma: A diagnostic challenge

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ABSTRACT

Extraskeletal osteosarcomas account for < 1% of the soft tissue sarcomas and are known to more often localize in soft tissues of the lower extremities. Primary osteosarcomas of the breast are extremely rare. A majority of the reported cases were in fact initially erroneously diagnosed and treated as primary breast carcinomas. We recently got to treat an interesting case of a primary breast osteosarcoma and discuss its evaluation and management with added emphasis on the incremental role of technetium 99-methylene diphosphonate (Tc-99m) bone scintigraphy in its clinical diagnosis. Tc-99m uptake can occasionally be seen in the delayed bone scintigraphy images of extra skeletal malignancies, but the uptakes are considered to be typically less intense than the uptakes noted in primary skeletal malignancies. Extraskeletal osteosarcomas are however the exceptions to this rule, the intense uptake in bone scintigraphy further aided in an accurate preoperative diagnosis and management of our patient.

Keywords: Bone scintigraphy, extra skeletal osteosarcoma, primary breast osteosarcoma, prognosis

INTRODUCTION

Mammary sarcomas are extremely rare and makeup < 1% of all primary breast malignancies.[1,2] Primary osteosarcomas of the breast are extremely rare and represent about 12.5% of the mammary sarcomas.[1,2] The majority of the reported cases were initially erroneously diagnosed and treated as primary breast carcinomas, and the final correct diagnosis of osteosarcomas was established only after the histological examination of the mastectomy specimen. We recently got to treat extremely rare case of primary breast osteosarcoma and explore the incremental value of bone scintigraphy in aiding in its accurate preoperative clinical diagnosis.

CASE REPORT

A 73-year-old lady with no other comorbid illnesses presented to our center for further evaluation of a painless hard lump in her left breast for the past 5 months. Her past, medical and family histories were unremarkable. Clinical examination revealed a well circumscribed hard mobile lump of about 5 × 3 cm in the upper outer quadrant of her left breast. Examination of the right breast, both axilla and both supraclavicular fossa were unremarkable, and so was the examination of the other organ systems. Mammography revealed a large densely calcified lobulated mass with irregular margins in the upper outer quadrant of the left breast. In addition, a small noncalcified area was noted in the lower and the medial aspect of the mass [Figure 1a and b]. A computed tomography (CT) chest done subsequently also confirmed the presence and extent of the multilobulated densely calcified extra skeletal mass with normal lung parenchyma [Figure 2]. The mass was densely calcified and could not be penetrated by a 22 gauge or a Tru-cut biopsy needle. A Tru-cut biopsy was re-attempted from the noncalcified soft tissue component under ultrasound guidance, the histopathology along with immunohistochemistry (IHC) correlation of which (positive for vimentin and negative for keratin, estrogen receptor [ER], proliferation rate [PR] and human epidermal growth factor receptor 2 [HER2neu]) was reported as a malignant tumor with an osteoid matrix, suggestive of a high grade primary breast osteosarcoma [Figure 3a-d]. An intense localized uptake in the left breast on a technetium 99-methylene diphosphonate (Tc-99m) bone scintigraphy further corroborated the preoperative diagnosis of primary breast osteosarcoma [Figure 4]. The patient was taken up for a definitive radical surgery which in this case entailed a simple mastectomy [Figure 5a and b]. The final diagnosis confirmed the preoperative diagnosis of a nonmetastatic high-grade primary breast osteosarcoma, excised with free margins. The decision not to give any adjuvant therapy was taken considering the patient's elderly age and frail built and more so lack of robust evidence. The patient is presently disease free and is on regular follow-up for close to 4 months following surgery.

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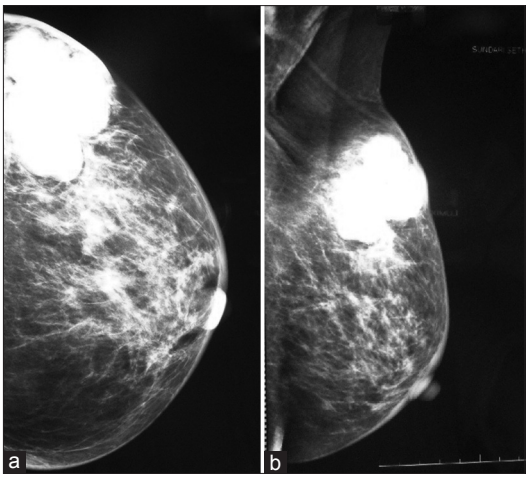

Figure 1: Mammography. (a) Cranio-caudal (b) medio-lateral view: Showing a large densely calcified lobulated mass measuring 5 × 3 cm with irregular margins in the upper outer quadrant of the left breast

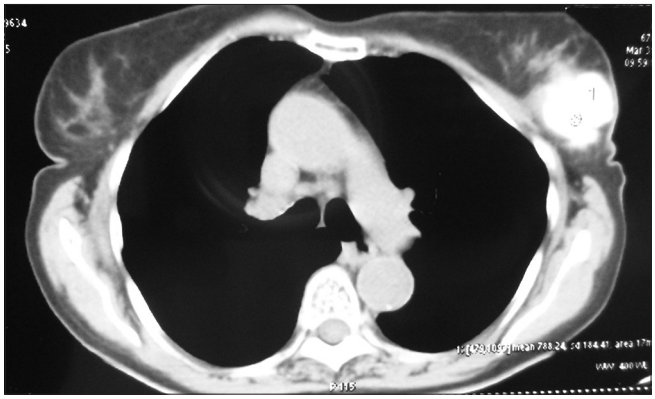

Figure 2: Computed tomography scan axial view showing the 5 × 3 cm multi lobulated densely calcified extra skeletal mass with a normal lung parenchyma

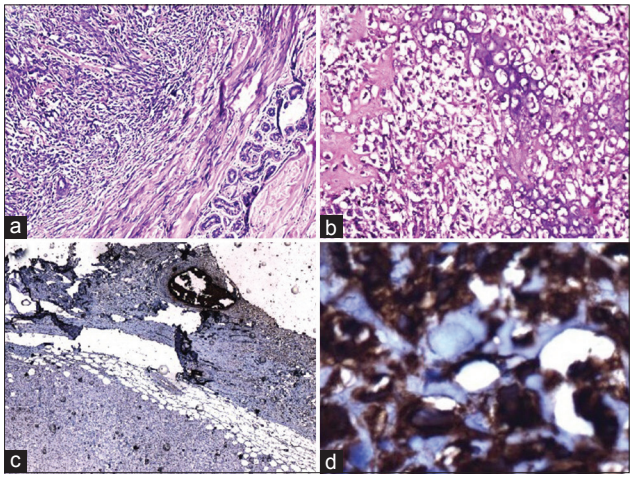

Figure 3: (a and b) Breast tissue with adjacent fragment showing malignant osteoid surrounded by ovoid to spindled plump cells with hyperchromatic nuclei and scant cytoplasm along with a few multinucleated giant cells. Mitosis was also noted (H and E, ×10). (c) Tumor cells showing immune negative for keratin (immunohistochemistry [IHC], ×40). (d) Tumor cells showing immune positive for vimentin (IHC, ×40)

DISCUSSION

Extraskeletal osteosarcomas account for < 1% of soft tissue sarcomas and are more often located in soft tissues of the lower extremities. It generally tends to affect the middle and older aged women in contrast to the younger age group of patients with skeletal osteosarcoma.[3] No specific etiological factor is reported to be associated with its occurrence.[1,3]

Skeletal osteosarcomas typically show increased uptake in Tc-99m bone scintigraphy.[4] The role of nuclear imaging in the evaluation and staging of extraskeletal osteosarcomas have been less studied. Localization of Tc-99m in extra osseous neoplasms is well-documented, but is typically considered less intense than in skeletal neoplasms, the postulated causes being tumor vascularity, local pH factors, inflammation, hormonal influences, altered calcium metabolism and cell wall damage.[4] The malignant extraskeletal new bone formation is the reason for the intense uptake in primary breast osteosarcomas. Bone scintigraphy is also useful in excluding multifocal disease, skip lesions and at times pulmonary metastases. Some authors have reported fludeoxyglucose-positron emission tomography-CT to be useful in staging and restaging of extraskeletal osteosarcomas.[5]

Histo-pathological evaluation remains fundamental in the diagnosis of primary extraskeletal osteosarcomas. The histological criteria for extraskeletal osteosarcomas include-exclusion of a bony origin, presence of malignant osteoid, absence of an epithelial component and lack of association of a benign tumor.[6] Primary osteosarcomas of the breast are extremely rare. A diagnosis of metaplastic breast carcinoma should also be excluded prior to making a diagnosis of primary breast osteosarcoma.[1,7] IHC plays an important role in differentiating primary breast osteosarcomas from sarcomatoid or metaplastic carcinomas, the latter being immuno-positive to keratin, whereas extraskeletal osteosarcomas are not.[1,3,7] The cells from a primary breast osteosarcoma are positive for vimentin and further display negativity for ER, PR and HER2neu.[1,3] A secondary lesion from a primary osteosarcoma of the bone is also an important differential as the two entities have different histological behaviors and require different treatments.[8]

Surgery in the form of wide excision aimed at achieving negative margins remains the cornerstone of management of primary breast osteosarcomas.[1,3,7-11] Lymph node dissection is not indicated, except when clinically involved. The effectiveness of adjuvant radiotherapy and chemotherapy however remains unclear. It has been used either singly or in combination by various authors in an attempt to reduce the local and systemic recurrences.[8,9] The prognosis of breast osteosarcomas unlike breast sarcomas and breast carcinomas is poor.[9] Primary breast osteosarcomas are considered to be highly aggressive neoplasms with early recurrences and propensity for hematogenous spread, the 5-year survival rate has been reported at a dismal 38%.[3]

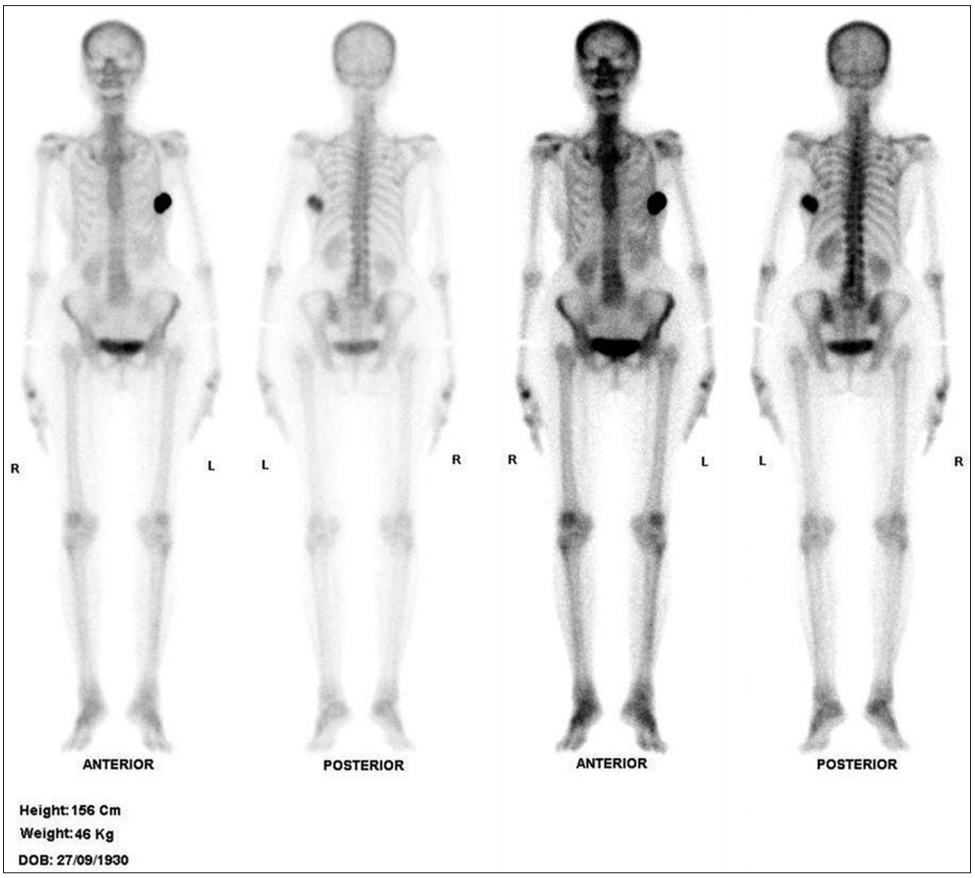

Figure 4: Technetium 99-methylene diphosphonate bone scintigraphy showing an intense uptake in the left breast corresponding to the primary tumor

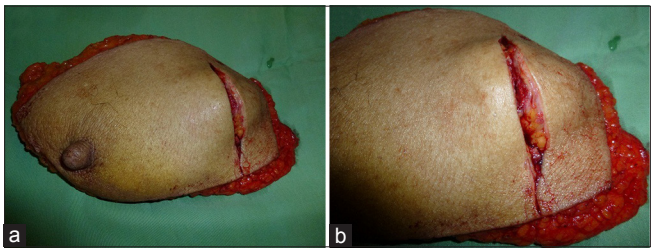

Figure 5: (a and b) Mastectomy specimen showing the tumor excised with wide margins

CONCLUSION

An accurate clinicopathological correlation is vital in the management of primary breast osteosarcomas, an intense uptake in a Tc-99m bone scintigraphy further aids in an accurate preoperative diagnosis of this often erroneously diagnosed condition.

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