ORIGINAL RESEARCH PAPER
Volume - 12 | Issue - 04 | April - 2023 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

A CASE OF PRIMARY EXTRASKELETAL OSTEOSARCOMA OF THE BREAST: A RARE CASE REPORT AND LITERATURE REVIEW.

Oncology

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ABSTRACT

Primary osteosarcoma of the breast is a very uncommon, heterogeneous and locally aggressive neoplasm. Due to rarity of this tumor, optimal consensus about the management strategy of this entity is yet to be defined in the guidelines. Exact process of tumorogenesis is uncertain in the literature. Here we report a rare case of 41 year old female presented with a palpable lump for a duration of around 12 months. On histopathology report of surgical sample, post mastectomy revealed osteoid producing tumor with surrounding fibroadenoma. This case study traces that osteosarcoma of the breast may occur from pre-existing breast lesion like fibroadenoma.

KEYWORDS

Breast, Osteosarcoma, Fibroadenoma

INTRODUCTION

Osteosarcoma is the commonest primary bone malignancy, often seen in young adolescents and pediatric age group (1). However osteosarcoma can rarely arise from extraskeletal tissues. Very few case reports of extraskeletal osteosarcoma have been reported in the literature. Similarly, osteosarcoma from primary breast is an extremely uncommon, highly aggressive and heterogeneous neoplasm. (2,3)

Breast sarcomas form less than 1% of all breast cancers and 5% of all sarcomas. Exact tumorogenesis for Primary breast osteosarcoma (POB) is uncertain but research articles mentions that the origin is probably from totipotent mesenchymal remnants or it may occur secondary to neoplastic transformation in a pre-existing breast lesions like phyllodes or fibroadenoma or intraductal papilloma.(4,5)

Due to rarity of this tumor, definite consensus about management is yet to be defined. In this article we report a rare presentation of the primary osteosarcoma of the breast in 41 year old female.

History

41-year-old woman with no significant medical history of comorbidities presented to our OPD with a palpable breast lump of about 3cm in size in the upper outer quadrant of the left breast and it had been there for the duration of about 12 months.

Physical examination showed an enlarged mass localized in the superior outer quadrant of left breast with no axillary lymph nodes palpable. Lab parameters were normal with normal alkaline phosphatase level.

Diagnostic mammography (fig 1) described 3×3cm mass in upper outer quadrant of left breast with internal specks of dense micro calcification and adjacent parenchymal distortion. USG left breast described hyperechoic lesion with irregular margins in UOQ of left breast with perilesional and minimal internal vascularity. Staging CT and Bone scan was done to rule out primary bone disease, which was suggestive of no evidence of disease elsewhere. CT scan thorax (fig 2) revealed heterogenous soft tissue lesion in outer central quadrant region of the left breast. Lesion shows internal necrosis with calcification.

Cytological examination on biopsy was suggestive of negative for epithelial neoplastic cells and only osteosarcoma component was evident. On immunohistochemistry AE1/AE3, CD34, CK7,ER, PR, HER2 markers were negative and SATB2 was evident.

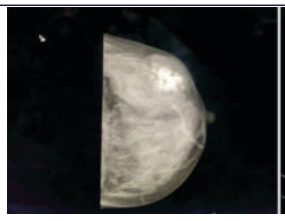

Fig 1: Mammography

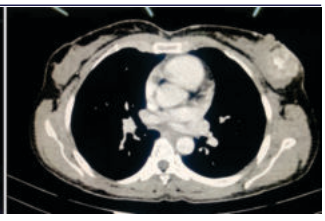

Fig 2: CT scan thorax

Patient underwent left Mastectomy and SLNB. Macroscopic gross examination of a surgical specimen revealed well circumscribed (3.8x 3.5cm) hard to firm solid tumor with infiltrating margins. Microscopic examination revealed tumor size of 3.8 x 3.5cm with multiple section showing tumor comprised of epithelioid to spindle cells showing moderate nuclear atypia. Tumor cells are seen producing lace like malignant osteoid, focal areas show cartilaginous differentiation and periphery of the tumor showed a compressed fibroadenoma (fig 3). No malignant epithelial component was identified. DCIS absent, LVI and PNI was not evident and margins were free from tumor. Axillary lymph nodes were negative (0/4). These features favored primary osteosarcoma of breast. Patient was planned for adjuvant chemotherapy Cisplatin plus Adriamycin regimen. She has received 3 cycles of chemotherapy till date.

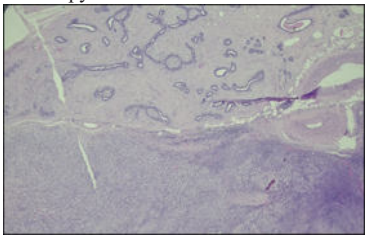

Fig 3: Cellular tumor with a compressed fibroadenoma at the periphery

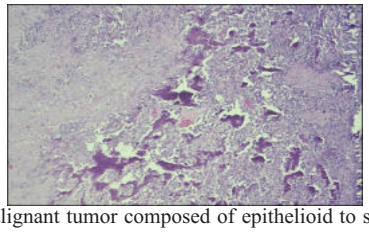

Fig 4: A malignant tumor composed of epithelioid to spindled cells

showing moderate nuclear atypia. Tumor cells are seen producing lace like malignant osteoid.

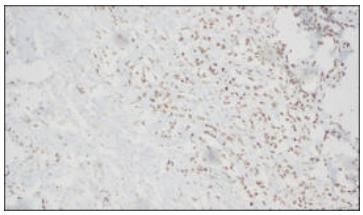

Fig 5: Tumor cells showing immunopositivity for SATB2

DISCUSSION

Primary breast sarcomas form less than 1% of all breast malignancies and POB forms about 4-12.5% of all primary breast sarcomas. (3,6)

POB usually affects women with advanced age in contrast to skeletal osteosarcoma which usually affects young population however POB has been seen in wide range from young adults to elderly females of age more than 80 years (6,7)

Literature has reported trauma, artificial implants and professional exposure to vinyl chloride as potential predisposing factors (8). Some case reports have shown irradiation for breast carcinoma as a triggering factor (9,10). Etiology is still not very clear.

POB usually presents as slow growing painless lump but some case reports have mentioned it as a rapidly growing tumor (11). In our case patient presented with painless lump for duration of about 12 months.

Unlike skeletal osteosarcoma POB rarely has high alkaline phosphatase. In POB, It is usually normal or slightly high, probably due to some amount of pathological neoplastic bone formation. In our case serum alkaline phosphatase level was normal.

In POB diagnostic mammography often shows variable size mass with well defined margins and lobulated borders with coarse or fine calcification (12). Imaging features of US and CT scan cannot clearly differentiate between benign and malignant calcified breast lesion, so use of above modalities in diagnosis is limited.(13) Tc99m MDP specificity for osteoid neoplasm is high ,so should be used to rule out primary skeletal osteosarcoma. Whole body Tc 99m scan with high uptake in breast mass outside bone is highly suggestive of extraskeletal osteosarcoma over breast carcinoma. (14)

Pre-op diagnosis is usually difficult and mostly accurate diagnosis is made on histological examination of complete surgical specimen. Tumors arising from neighboring structures such as underlying rib, sternum and pectoralis major should be ruled out before making diagnosis of primary breast osteosarcoma (15). POB is a close differential diagnosis of two similar entities, metaplastic carcinoma and Cystosarcoma phyllodes. Cystosarcoma phyllodes has specific morphological features (biphasic pattern with leaf like architecture and epithelial component. Metaplastic carcinoma can be excluded based on presence or absence of carcinomatous component (cytokeratin positivity on immunohistochemistry) (6).In our case, tumor lacks epithelial differentiation and was SATB2 positive on immunohistochemistry favoring diagnosis of POB.SATB2 is a highly sensitive and specific marker for osteoid differentiation. Some authors have emphasized the type of preexisting lesions like fibroadenoma, phyllodes. (16) In this case on microscopic examination, periphery of tumor revealed fibroadenoma component. So we hypothesize that there could be a malignant neoplastic transformation of fibroadenoma to POB.

Optimal consensus about treatment strategy is not yet clearly defined due to rarity of this entity. Surgery is still considered as the most important part of treatment. Axillary lymph node dissection is usually not advised as reports have shown no axillary lymph node metastasis on axillary lymph node dissection. POB is a locally aggressive neoplasm and usually metastasize through hematogenous spread (2). Role of chemotherapy is not well established .Benefit of perioperative chemotherapy in terms of overall survival is not determined in studies.(17).Some studies have shown improved survival rates with adjuvant chemotherapy in high grade ,size more than 5cm tumors.(7,18).Role of radiotherapy is controversial as osteosarcoma is a radio resistant tumor.

5 and 10 year estimated survival is 38% and 32% with 28% developing local recurrence ,41% developing distant metastasis. Hematogenous spread to lungs 80%, bone20%,liver17% (1,3). Prognostic factors include tumor size, number of mitoses, presence of stromal atypia.

CONCLUSION

POB is a rare, heterogeneous and locally aggressive neoplasm with exact tumorogenesis mechanism is still not certain. Before labeling POB, tumors arising from neighboring structures should be excluded. Close differential diagnosis of POB are metaplastic carcinoma and Cystosarcoma phyllodes. With keen pathological examination and immunohistochemistry diagnosis of POB can be made. Till now surgery is the main stem treatment and role of chemo radiotherapy is still unclear. Further research about treatment and mechanism of tumor development is much needed in view of improving outcomes of this rare and aggressive entity.

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