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Primary Osteosarcoma of the Breast: A Rare Case Report and Review of Literature

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Abstract

Unlike skeletal osteosarcoma, primary osteosarcoma of the breast is extremely rare tumor. We report very rare case of primary osteosarcoma of the breast in a 50-year old female. Core needle biopsy of the breast lump was suggestive of metaplastic carcinoma with chondroblastic osteosarcoma component. Further immunohistochemistry confirms Chondroblastic Osteosarcoma-Extraskeletal. Treatment strategy should be considered carefully according to patient status. More data and further research are needed to develop an optimum treatment plan for this rare entity.

Key words: - Carcinoma Breast, Osteosarcoma, Extraosseous Osteosarcoma, Primary osteosarcoma of breast.

Introduction

Primary mammary osteosarcoma is an extremely rare entity and account for 12% of all mammary sarcomas [1]. Most published data consist of single cases report, with one major series of 50 cases [2]. Here, we present a case of primary osteosarcoma of breast who presented to us as right breast lump for last 1 year and treated by simple mastectomy.

Case report

50-year old postmenopausal, normotensive, non-diabetic lady presented to Mahavir Cancer Sansthan, Patna (India) on 2nd February 2018 with lump in right breast for last 1 year. Size of the lump was gradually increasing. No history of any addiction. No relevant past history. No history of any previous radiation exposure. No history of familial breast cancer. On examination, approximately 12 x 10 cm firm to hard lump was present in the right breast with small approximately 5 x 4.5 cm ulceration in upper outer quadrant. Nipple areolar complex was normal. Ultrasound of breast showed complex cystic mass seen at 10 O'clock position in right breast.

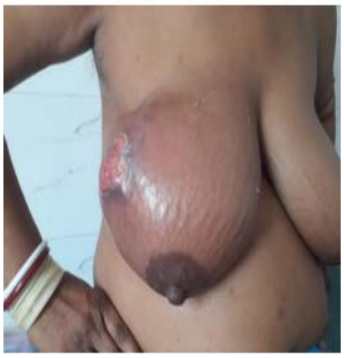

Fig 1 & 2: - Clinical picture of right breast lump

Core needle biopsy from right breast lump section showed cartilage & osteoid formation along with malignant cells having spindle nuclei with multinucleation in few of them suggestive of metaplastic carcinoma with chondroblastic osteosarcoma component.

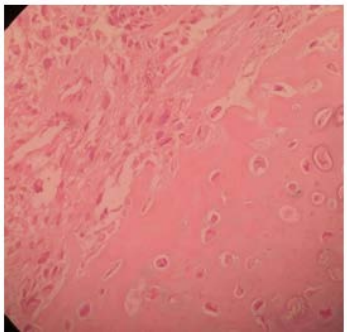
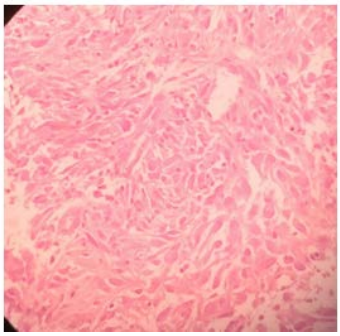

Fig 3 & 4: - Section shows cartilage & osteoid formation along with malignant cells having spindle nuclei with multinucleation in few of them. Suggestive of metaplastic carcinoma with chondroblastic osteosarcoma component.

On further immunohistochemistry CK, CK-7 and S-100 were negative and Vimentin was positive in tumor cells suggestive of Chondroblastic Osteosarcoma-Extraskeletal. F18-NaF positron emission tomography and attenuation corrected CT scan report showed large soft tissue lesion in upper outer quadrant of right breast and no any scan evidence of skeletal metastasis noted.

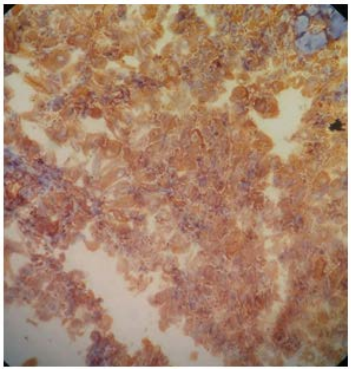
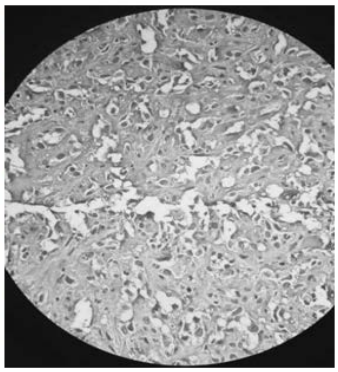

Fig 5 & 6: - Immunohistochemistry showing Vimentin positive and Ck negative respectively.

Fig 7: - F18-NaF positron emission tomography and attenuation corrected CT scan – showing large soft tissue lesion in upper outer quadrant of right breast and no any scan evidence of skeletal metastasis noted.

Discussion

Primary sarcoma of the breast is a rare problem and accounts for less than 5% of all soft-tissue sarcomas and less than 1% of all breast malignancies [3]. At the Mayo Clinic, 27 881 malignant breast tumours were seen between 1940 – 1999 and 18 breast sarcomas were diagnosed accounting for 0.0006% of breast malignancies [4]. Mammary osteosarcomas account for 12% of all mammary sarcomas [1].

The mechanism of tumorigenesis is not clear, it is postulated that it is formed from totipotential cells of the stroma. It is postulated that the tumor is preceded from a fibroadenoma or a phyllodes tumor. It also has been described in the literature that radiotherapy may induce formation of breast sarcoma like osteosarcoma [5]. Our patient had none of these conditions. Primary osteosarcoma of the breast without an epithelial component present or prior irradiation is extremely rare.

Most reports in the literature are limited to presentation of single cases but Silver and Tavassoli [2] have reported a series of 50 cases of primary osteogenic sarcoma of the breast, focusing mainly on the clinicopathological findings of this rare entity. In contrast to skeletal osteosarcoma which occurs in younger patients, mammary osteosarcoma occurs in age ranged from 27 to 89 years, most commonly observed in older patients with a median age of 64.5 years [2]. Main symptoms include pain and the presence of a growing palpable breast mass that is typically movable, hard, and irregular without axillary lymphadenopathy. These are not specific findings but common for breast neoplasms. Mammographically, osteosarcomas present as a well circumscribed mass with focal to extensive coarse calcification. Because of their predominantly circumscribed nature, they may be misinterpreted as a benign lesion. An MRI showed low signal intensity of the

central mineralised matrix with rapid enhancement of the peripheral mass.

Skeletal osteosarcomas typically show increased uptake in Tc-99m bone scintigraphy. Localization of 99mTc-MDP in extra osseous neoplasm is well-documented, but is typically much less intense than skeletal neoplasm, the postulated causes being tumor vascularity, inflammation, local pH factors, altered calcium metabolism, hormonal influences, and cell wall damage [6]. In our case F18-NaF positron emission tomography and attenuation corrected CT scan report showed large soft tissue lesion in upper outer quadrant of right breast and did not detect any bony lesion elsewhere.

Histo-pathological evaluation remains fundamental in the diagnosis of primary extraskeletal osteosarcomas. Immunohistochemistry plays an important role to differentiate mesenchymal neoplasm from undifferentiated carcinoma. In our case, core needle biopsy was suggestive of metaplastic carcinoma with chondroblastic osteosarcoma component. On further immunohistochemistry CK, CK-7 and S-100 were negative and Vimentin was positive in tumor cells, suggestive of Chondroblastic Osteosarcoma-Extraskeletal.

In a study of 50 patients, a 5-year survival of 38% was observed, 28% developed local recurrence and 41% distant metastasis. In contrast to primary carcinoma of breast, where the spread is mainly through lymphatics, primary breast osteosarcoma spreads more commonly through hematogenous route. Haematogenous metastasis metastases most commonly occur to the lungs (80%), bone (20%) and liver (17%). Prognostic factors include tumor size, number of mitoses, presence of stromal atypia, histological type and resection margin involvement [2].

There are no current treatment guidelines due to limited data. In Silver and Tavassoli's analysis of 50 cases, patients were treated by excisional biopsy, tylectomy or mastectomy. 73% patients in whom local recurrence developed were treated with excisional biopsy or tylectomy. 20 cases received axillary node evaluation while the others did not. There was no positive axillary node on all axillary nodes [2]. Some author also performed modified radical mastectomy including axillary lymph node dissection [7,8]. But, most authors suggest wide local surgical excision or mastectomy depending on the size of the tumor and remaining breast tissue is the standard surgical management. It is important to achieve a complete resection with negative resection margins as margin status is a major factor deciding local disease recurrence. Axillary clearance is not necessary as these tumors generally do not spread by lymphatic route. We have treated with simple mastectomy.

The role of adjuvant radiotherapy and chemotherapy however remains unclear because of limited data and its efficacy has not been well established [7,9]. Most commonly used drugs are doxorubicin, cisplatin or ifosfamide. It has been used either singly or in combination by various authors in an attempt to reduce the local and systemic recurrences. In cases of positive margin after surgery or a huge mass that cannot be excised well, radiotherapy may be performed.

Conclusion

Primary osteosarcoma of the breast is a very rare malignancy. Signs and symptoms are nonspecific but common for breast neoplasm. Definitive diagnosis should be made by histopathology and immunohistochemistry. Mastectomy with or without axillary dissection appears to be the most adapted primary treatment for this tumor and the use of an adjuvant therapy still requires more data and further research.

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