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CASE REPORT PEER REVIEWED | OPEN ACCESS

Primary osteosarcoma of the breast: A case report

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ABSTRACT

Introduction: Primary osteosarcoma of the breast is an extremely rare malignant tumor, and only few cases have been reported in the literature. It is a rare histological type of all breast tumors. Primary osteosarcoma of the breast is considered to be an aggressive tumor with a poor prognosis.

Case Report: We report a case of primary osteosarcoma of the breast in a 71-year-old woman. The patient presented a lump in the left breast, discovered on self-examination. Ultrasound and mammographic data revealed a large solid and cystic mass in the left breast, graded as highly suggestive of malignancy (Breast Imaging and Reporting Data System [BI-RADS] category 5). After a tru-cut biopsy showing a malignant mesenchymal tumor proliferation, the patient has undergone a mastectomy. The histological examination of the mastectomy specimen revealed the presence of sarcomatous proliferation with abundant osteoid substance and chondroid area but no epithelial component. The diagnosis of primary osteosarcoma of the breast was determined after ruling out the primary differential diagnosis, metaplastic carcinoma, by an immunohistochemical study. Immunohistochemically, tumor cells were non-reactive with anti-cytokeratin antibodies.

Conclusion: It is essential to recognize this rare and aggressive pathologic tumor variant in order to manage patients suffering from this pathology appropriately.

Keywords: Breast cancer, Histology, Mastectomy, Primary osteosarcoma

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INTRODUCTION

Primary osteosarcomas of the breast are very rare, aggressive, and heterogeneous neoplasms derived from non-epithelial elements of the gland and represent less than 1% of breast cancers and less than 5% of all sarcomas [1].

Its unusual presentation in the breast can put the pathologist in the dilemma of diagnostic. We report the case of a 71-year-old woman who was diagnosed with a primary osteosarcoma in her left breast. Through this

case and review of the literature, we highlight several details of this rare histological type of breast cancer.

CASE REPORT

A 71-year-old woman, with no specific history, who presented with a lump in the left breast, discovered on self-examination, progressing gradually for one year. She was in good general condition with no other associated symptoms. The clinical examination showed a 7 cm mobile rounded mass in the outer quadrant of the left breast, without skin retraction and no palpable axillary lymph nodes (Figure 1).

Mammography revealed a voluminous dense opacity, focally with indistinct margin, containing microcalcifications in the outer quadrants of the left breast (Figure 2).

Ultrasonography (US) showed a large solid and cystic mass with indistinct margin containing microcalcifications, measuring 77 mm × 62 mm. Doppler evaluation demonstrated surrounding vascularity (Figure 3). The lesion was graded as highly suggestive of malignancy (Breast Imaging and Reporting Data System [BI-RADS] category 5).

A tru-cut biopsy was taken, which showed a malignant mesenchymal tumor proliferation, suggesting a fibro-chondro-osteosarcoma. The patient underwent a mastectomy.

The gross examination of the specimen from the left breast revealed a white beige neoplasm, well-circumscribed, firm, measuring 14×9×5.5 cm, focally hemorrhagic and necrotic (Figure 4). The histological examination showed a malignant spindle cell tumor with abundant osteoid substance and chondroid area. There were scattered multinucleated osteoclastic tumor giant cells. The carcinomatous component was not found (Figure 5). Multiple sections were taken to detect an epithelial component, thus ruling out the possibility of a metaplastic carcinoma. The immunostaining for cytokeratine AE1–AE3 showed no epithelial component (Figure 6).

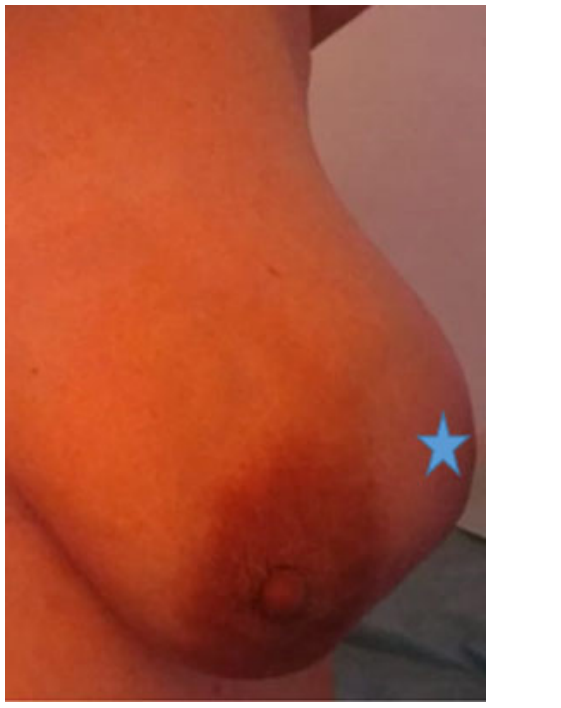

Figure 1: Clinical examination: Mobile rounded mass in the outer quadrant of the left breast (blue asterisk).

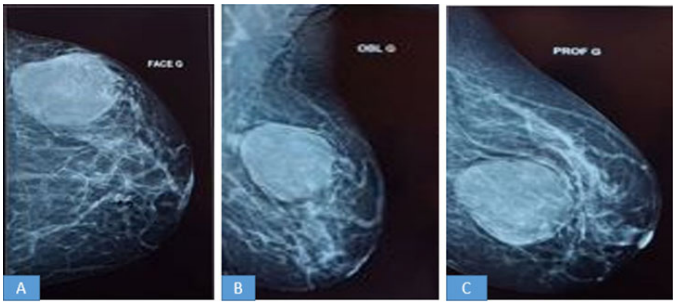

Figure 2: Left breast mammography: Face view (A), external oblique view (B), and internal profile view (C): a voluminous dense opacity, with microcalcifications in the outer quadrants.

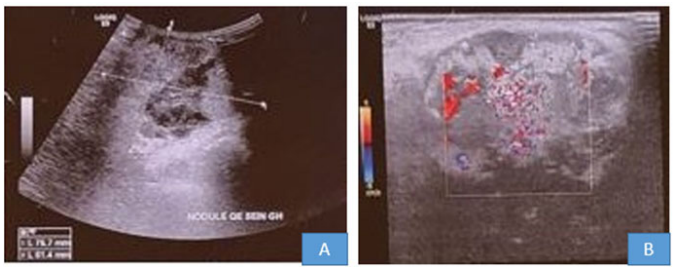

Figure 3: Left breast ultrasonography with convex probe (A) and linear probe (B): a large solid and cystic mass with microcalcifications, measuring 77 mm × 62 mm in the outer quadrants. Doppler evaluation showing a surrounding vascularity.

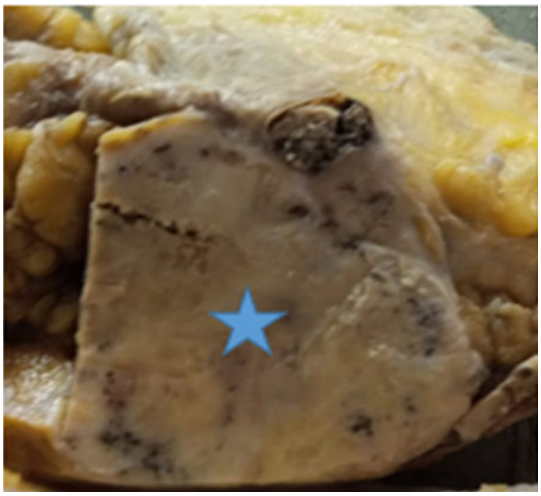

Figure 4: Gross examination: A white beige neoplasm, well-circumscribed, firm, measuring 14×9×5.5 cm, focally hemorrhagic and necrotic (blue asterisk).

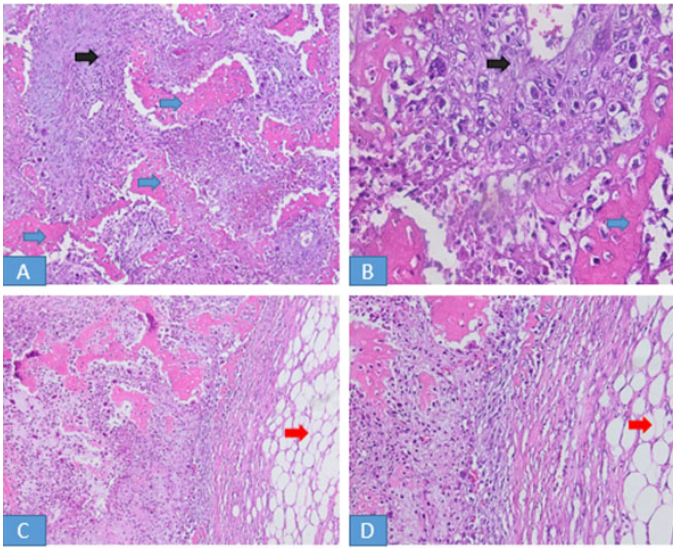

Figure 5: Histological examination, Hematoxylin and eosin stain: (A, 20×) and (B, 40×) showing a malignant spindle cell tumor (black arrow) with abundant osteoid substance (blue arrow). (C, 20×) and (D, 40×) showing a malignant spindle cell tumor with abundant osteoid substance and fat tissue (red arrow).

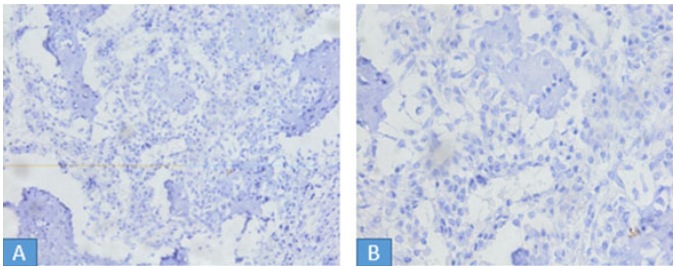

Figure 6: Immunohistochemistry: (A, 20×) and (B, 40×). The tumor cells were negative for AE1/AE3.

DISCUSSION

Carcinoma is the most common malignancies of the breast and sarcomas form a minority of breast neoplasms. Primary sarcomas of the breast account for <1% of all primary breast malignancies [2]. Among those, osteosarcoma is extremely rare.

Approximately, 150 cases of this pathology have been reported in the literature since 1957 [1]. The histogenesis of primary breast osteosarcoma remains unclear. Still, an evolution from totipotent mesenchymal cells of the breast stroma or a transformation from a preexisting fibroadenoma or phyllodes tumors have been suggested [1, 3].

The presentation of breast osteosarcoma usually occurs at an advanced age, in contrast with skeletal osteosarcomas where the patients are younger. The majority of the cases are in patients that are over 60 years old with a reported median age of 64.5 years [2]. Risk factors for extraskeletal osteosarcomas have not been identified to date, although some instances have been attributed to local irradiation, trauma, or the presence of a foreign body [2].

The present patient was 71 years old with no history of trauma or irradiation, no tumor in other sites. Clinically, breast osteosarcomas often present as rapidly enlarging masses. Rarely, patients report associated bloody nipple discharge or nipple retraction [4].

Mamographically, these tumors often present as well circumscribed, dense lesions within the breast parenchyma, with focal or extensive coarse calcifications [3, 5].

The border may be regular or irregular. The mammographic appearances may be deceptively benign and may imitate a benign fibroadenoma in a third of cases [2]. The most common macroscopic finding is the neoplasm well circumscribed, which joins the appearance found in the present case.

In almost every case in the literature, the diagnosis of primary osteocarcoma was established histologically [6]. Histologically, this tumor is indistinguishable from the conventional osteosarcomas of the bone or other extraskeletal ones [3]. The diagnosis criteria of primary breast osteosarcoma include the absence of a tumor bone, the presence of the osteoid substance or bony streaks and the lack of an associated epithelial tumor component [2]. The absence of the epithelial component must be confirmed by immunohistochemical study or by ultrastructural study [7]. In our patient, the diagnosis of primary osteosarcomas of the breast was established on the morphology by the absence of an epithelial component and confirmed by negative staining with cytokeratin antibody (AE1/AE3). The principal histological differential diagnosis in our case was metaplastic carcinoma [1]. However, this was excluded because of negative epithelial markers on immunohistochemical staining.

Malignant phyllodes with osteosarcomatous differentiation can be also raised as the differential diagnosis. The treatment is surgical by a complete removal of the tumor with negative margins or a mastectomy. An axillary lymph node dissection is essentially not indicated because an axillary node involvement is exceptional [8].

In the present case, axillary lymph node dissection was not performed.

The role of adjuvant therapy is unclear because of limited data and his efficacy has not been well established [9].

Primary osteosarcomas of the breast have a poor prognosis with a high possibility of recurrence and metastasis, especially by hematogenous route instead of lymphatic spread. The most common distant site for metastasis are the lungs [2]. Silver et al. in their study of 50 patients with primary breast osteosarcoma reported a 5-year survival of 38%, with 28% of patients developing local recurrence and 41% with distant metastases [2].

CONCLUSION

It is essential to be aware of this unusual pathologic tumor variant and to differentiate it from metaplastic carcinoma. An early diagnosis, confirmed by an immunohistochemical study and a rapid treatment mainly surgical, is necessary to improve the prognosis, which remains very poor despite everything.

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Author Contributions

Evrard Niyonkuru – Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Meriem Regragui – Conception of the work, Design of the work, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Guarantor of Submission

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.