

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: <http://Elsevier.com/locate/radcr>

Breast Imaging

Primary breast osteosarcoma mimicking calcified fibroadenoma on screening digital breast tomosynthesis mammogram

Debbie Lee Bennett MD^{a,*}, Gloria Merenda MD^b, Stephanie Schnepf MD^c,
Mary Catherine Lowdermilk MD^a

^a Department of Radiology, St. Louis University Hospital, 3655 Vista Ave., St. Louis, MO 63110, USA

^b Department of Pathology, SSM St. Mary's Hospital, 6420 Clayton Rd., St. Louis, MO 63117, USA

^c Department of Surgery, SSM St. Mary's Hospital, 6420 Clayton Rd., St. Louis, MO 63117, USA

ARTICLE INFO

Article history:

Received 20 March 2017

Received in revised form 9 June 2017

Accepted 18 June 2017

Available online

Keywords:

Breast cancer

Primary osteosarcoma

Fibroadenoma

Mammogram

Tomosynthesis

ABSTRACT

Primary breast osteosarcoma is a rare malignancy, with mostly case reports in the literature. The appearance of breast osteosarcoma on digital breast tomosynthesis imaging has not yet been described. A 69-year-old woman presents for routine screening mammography and is found to have a calcified mass in her right breast. Pattern of calcification appeared “sunburst” on digital breast tomosynthesis images. This mass was larger than on the previous year’s mammogram, at which time it had been interpreted as a benign calcified fibroadenoma. The subsequent workup demonstrated the mass to reflect primary breast osteosarcoma. The patient’s workup and treatment are detailed in this case. Primary breast osteosarcoma, although rare, should be included as a diagnostic consideration for breast masses with a sunburst pattern of calcifications, particularly when the mammographic appearance has changed.

© 2017 the Authors. Published by Elsevier Inc. under copyright license from the University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Introduction

Primary breast osteosarcoma is an extremely rare malignancy, accounting for less than 0.1% of all primary breast malignancies [1]. The first English-language case was reported in 1982 [2], with approximately 100 cases described since

that time. Most of the reported cases have included images from film-screen mammography showing densely calcified masses. To date, the appearance of primary breast osteosarcoma has not been demonstrated with digital breast tomosynthesis (DBT) imaging.

We present a case of a screen-detected primary breast osteosarcoma, which was initially thought to reflect a calcified

Competing Interests: The authors declare that we have no significant competing financial, professional or personal interests that might have influenced the performance or presentation of the work described in this manuscript.

* Corresponding author.

E-mail address: bennettdl@slu.edu (D.L. Bennett).

<https://doi.org/10.1016/j.radcr.2017.06.008>

1930-0433/© 2017 the Authors. Published by Elsevier Inc. under copyright license from the University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

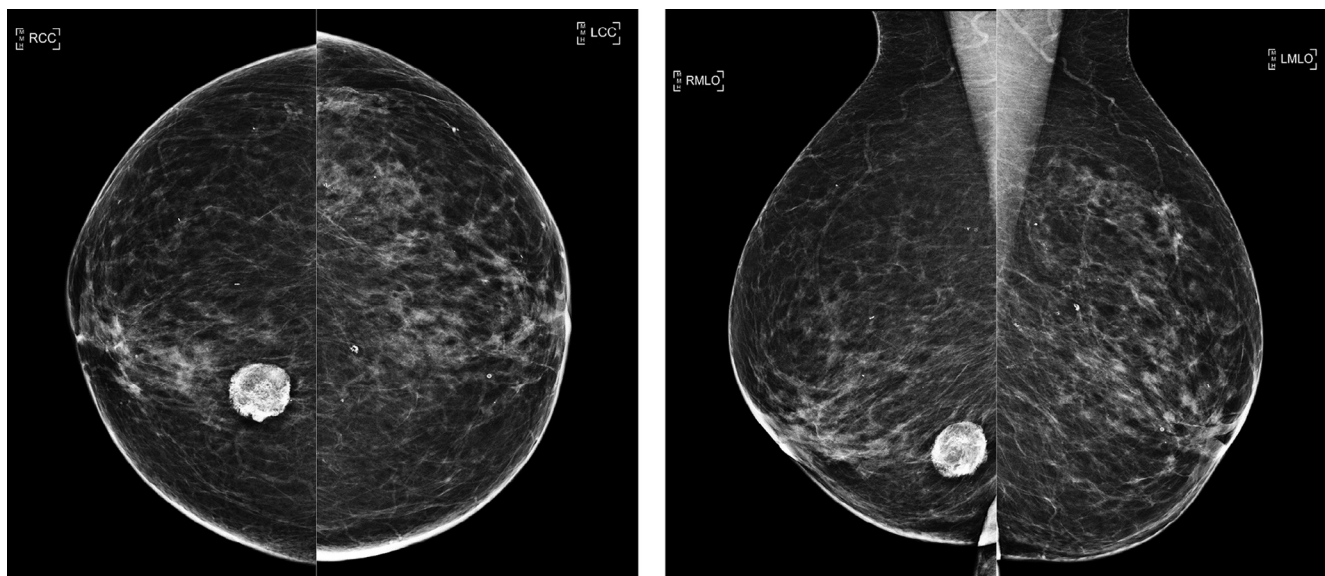


Fig. 1 – Screening mammogram. Standard craniocaudal (CC) and mediolateral oblique (MLO) views of both breasts demonstrate an oval, calcified mass in the lower inner quadrant of the right breast. The mammographic appearance of the left breast is normal. LCC, left craniocaudal; LMLO, left mediolateral oblique; RCC, right craniocaudal; RMLO, right mediolateral oblique.

fibroadenoma on previous mammograms. The patient's subsequent evaluation, treatment, and follow-up are also described.

Case report

A 69-year-old woman presented for routine screening mammography. The patient was found to have an oval, partially

calcified mass in the lower inner quadrant of her right breast (Fig. 1). Margins of the mass were circumscribed. The pattern of calcifications was “sunburst” on DBT images (Fig. 2A). When compared with the mammogram from the previous year, the mass had significantly enlarged in size (Fig. 2B and C). Calcifications were new from the mammogram performed 2 years before presentation (Fig. 2D). No suspicious mass, calcification, or distortion was seen in the contralateral left breast.

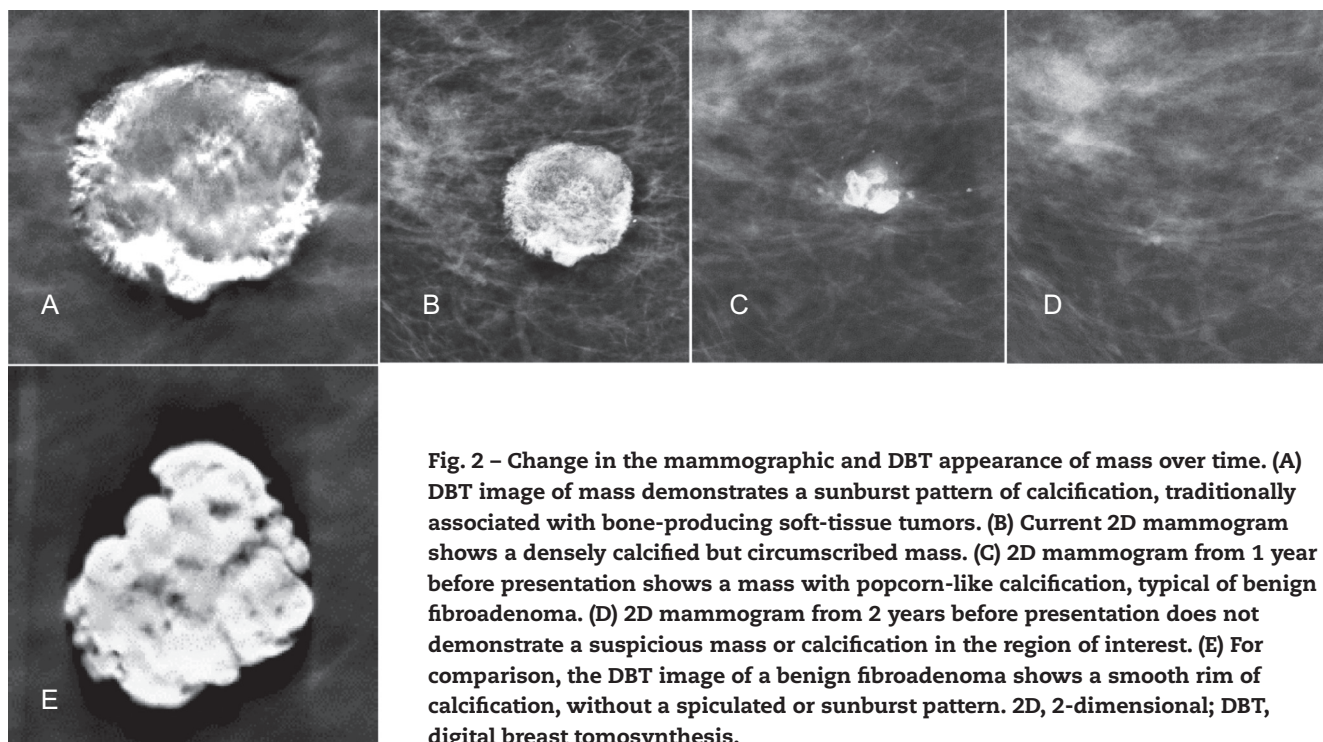


Fig. 2 – Change in the mammographic and DBT appearance of mass over time. (A) DBT image of mass demonstrates a sunburst pattern of calcification, traditionally associated with bone-producing soft-tissue tumors. (B) Current 2D mammogram shows a densely calcified but circumscribed mass. (C) 2D mammogram from 1 year before presentation shows a mass with popcorn-like calcification, typical of benign fibroadenoma. (D) 2D mammogram from 2 years before presentation does not demonstrate a suspicious mass or calcification in the region of interest. (E) For comparison, the DBT image of a benign fibroadenoma shows a smooth rim of calcification, without a spiculated or sunburst pattern. 2D, 2-dimensional; DBT, digital breast tomosynthesis.

The patient had no relevant past medical history and was generally healthy with the exception of type II diabetes, hypertension, and hypothyroidism. The patient had no previous breast biopsies or surgeries, and had never received therapeutic radiation to her chest wall.

The patient was asked to return for additional evaluation and targeted ultrasound was performed in the medial right breast, demonstrating a partially calcified mass measuring $2.9 \times 2.3 \times 2.5$ cm (Fig. 3). The mass showed indistinct margins, antiparallel orientation, and posterior acoustic shadowing. The mass was palpable at time of ultrasound, although the patient was not aware of it. Mammographic and sonographic features of the mass were deemed suspicious and biopsy was recommended.

Ultrasound-guided biopsy of the mass was performed with a Bard Max-core 14G biopsy device (Bard Biopsy Systems, Tempe, AZ). Pathology from the biopsy showed a malignant spindle cell neoplasm with an osteoid matrix formation. The differential considerations were given as primary breast osteosarcoma, matrix-producing metaplastic carcinoma, and malignant phyllodes tumor with stromal overgrowth. After discussion with the breast surgeon, a decision was made to proceed with lumpectomy and sentinel node biopsy, with the goal of achieving wide resection margins.

Pathologic analysis of the resection specimen confirmed the diagnosis of breast osteosarcoma. Examination of the gross surgical specimen showed a circumscribed mass, with the microscopic examination showing malignant osteoid (Fig. 4). Immunohistochemical staining was performed for epithelial markers and was negative, confirming that this was not a metaplastic breast carcinoma. Surgical margins



Fig. 3 – RT breast ultrasound. Transverse (TRV) ultrasound image of the medial RT breast at 3:00, 4 cm from the nipple (CM FN), shows a corresponding partially calcified mass with indistinct margins, antiparallel orientation, and posterior acoustic shadowing. Mass was biopsied percutaneously with ultrasound guidance. RT, right.

were widely negative (closest margin was 0.7 cm). Sentinel node biopsy was negative for nodal metastasis.

Bone scan was then performed, which showed postsurgical changes in the right breast and degenerative changes, but

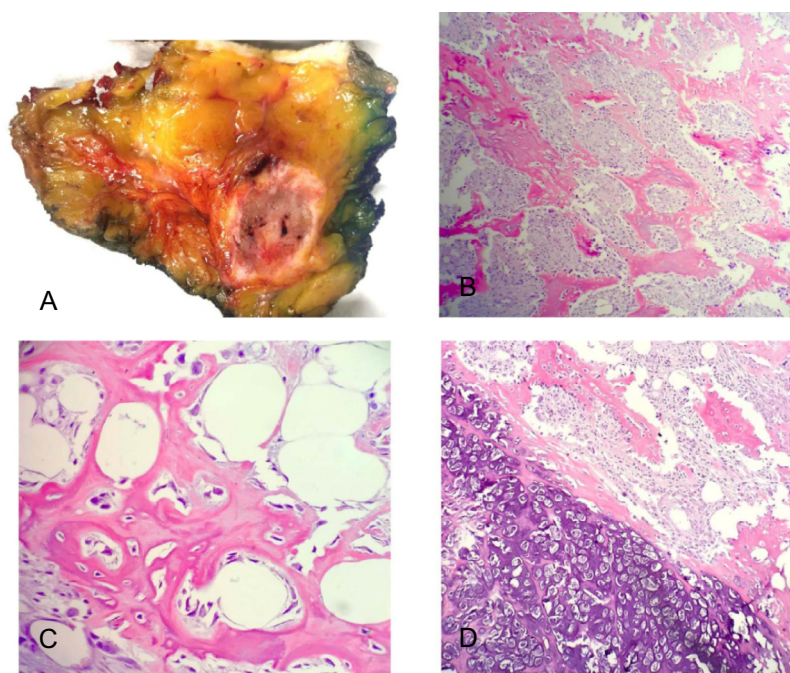


Fig. 4 – Gross and microscopic pathology of lumpectomy specimen. (A) Gross specimen shows a well-circumscribed, firm nodule measuring $2.5 \times 2.0 \times 1.5$ cm. (B) Low-power view of the specimen shows sarcomatous stroma and malignant osteoid formation. (C) High-power view of specimen shows tumor osteoid production by anaplastic cells. (D) Low-power view of the specimen shows both malignant chondroblastic (lower left) and osteoblastic (upper right) matrix production.

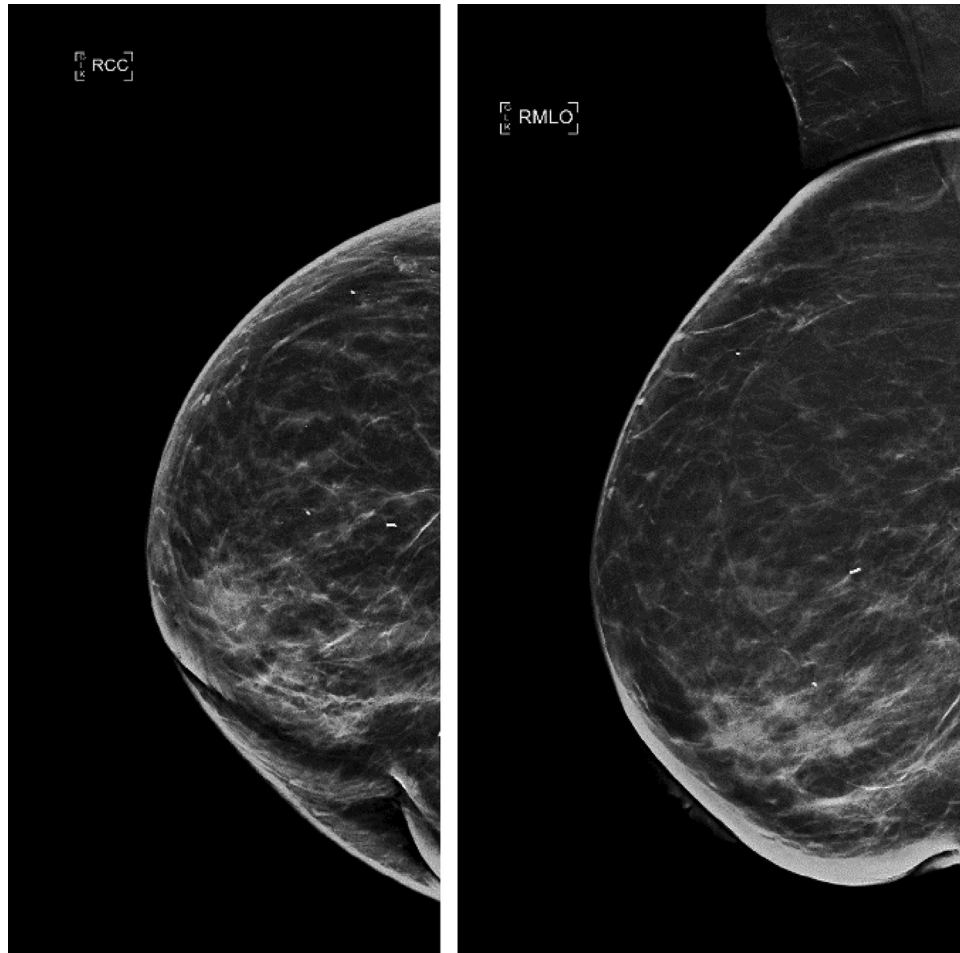


Fig. 5 – Post-treatment mammogram. Craniocaudal (CC) and mediolateral oblique (MLO) views of the right breast from a surveillance mammogram performed 1 year after diagnosis demonstrates post-treatment change from lumpectomy and radiation. There is no evidence of recurrent malignancy. RCC, right craniocaudal; RMLO, right mediolateral oblique.

no site of abnormal uptake to suggest a primary bone-forming malignancy elsewhere. This confirmed the diagnosis of primary extraskeletal breast osteosarcoma. The patient was then seen by a medical oncologist and a radiation oncologist; adjuvant radiation was recommended and was completed without complication. Baseline staging computed tomography of the chest, abdomen, and pelvis did not show any evidence of metastatic disease. Chemotherapy was not recommended.

The patient was seen for follow-up 1 year after her initial diagnosis. Post-treatment changes were evident in the patient's right breast, but there was no evidence of residual or recurrent malignancy (Fig. 5). Follow-up surveillance computed tomography was also normal.

Discussion

As mentioned in the Introduction, primary breast osteosarcoma is a rare malignancy, with approximately 100 cases described in the existing literature through isolated case reports and small case series [3-6]. Many of the published

case reports have shown large calcified masses that presented as palpable abnormalities. Our case shows the mammographic and DBT appearance of a screen-detected primary breast osteosarcoma.

Some authors have hypothesized that breast osteosarcoma may develop from malignant transformation of the stromal component of a pre-existing fibroadenoma; this hypothesis is based on the finding of fibroadenomatous tissue in the periphery of some osteosarcomas on pathologic analysis [3,7]. This case shows the mammographic progression of a pure breast osteosarcoma, without the presence of an underlying fibroadenoma, as an illustration of the imaging overlap between the 2 entities. Although other authors have mentioned this diagnostic dilemma, only 1 other report to date has illustrated this potential imaging pitfall [3].

Our case shows the natural history of primary breast osteosarcoma, developing first as a benign-appearing mass with popcorn-like calcifications, with a marked change in the size and pattern of calcification the following year. DBT may be helpful for distinguishing the 2 entities, as osteosarcoma shows a spiculated or a sunburst margin of calcification, rather than a smooth calcified margin as seen with fibroadenomas.

Regardless of whether a patient presents with a screen-detected or palpable abnormality, biopsy of the suspicious mass should be the first step in establishing a diagnosis. Although previous authors have relied on excisional biopsy for diagnosis, it was possible to perform a percutaneous ultrasound-guided biopsy in this case, despite the presence of dense calcification.

After a pathologic diagnosis of osteosarcoma is made, bone scan should be performed to exclude a primary bone-forming malignancy elsewhere [8–10]. Treatment should include surgical excision of the mass. One study reported that 60% of patients with primary breast osteosarcoma developed either locally recurrent or metastatic disease, with a higher local recurrence rate for patients treated with lumpectomy (67%) vs mastectomy (11%) [6]. Metastatic disease was most commonly via hematogenous spread, rather than nodal spread, and was apparent in the first year after diagnosis in almost half of the patients [6]. The overall 5-year survival was estimated at 38%, with better survival rate for patients with smaller (<4.6 cm) tumors.

Given the improved survival with detection of smaller tumors, radiologists should be aware of the imaging appearance of primary breast osteosarcoma on both screening and diagnostic mammograms. The change in mammographic appearance over time should raise suspicion for malignancy. Furthermore, the pattern of osteoid formation (in a sunburst formation) should also raise concern for malignancy, particularly with DBT imaging.

REFERENCES

- [1] Adem C, Reynolds C, Ingle JN, Nascimento AG. Primary breast sarcoma: clinicopathologic series from the Mayo Clinic and review of the literature. *Br J Cancer* 2004;91(2):237–41.
- [2] Mertens HH, Langnickel D, Staedtler F. Primary osteogenic sarcoma of the breast. *Acta Cytol* 1982;26(4):512–6.
- [3] Crevecoeur J, Jossa V, Gennigens C, Parmentier J-C, Crèvecoeur A. Primary osteosarcoma of the breast: a case report. *Clin Case Rep* 2016;4(1):62–6.
- [4] Al Samaraee A, Angamuthu N, Fasih T. Primary breast osteosarcoma: a case report and review of literature. *Scott Med J* 2014;59(4):e1–4.
- [5] Krishnamurthy A. Primary breast osteosarcoma: a diagnostic challenge. *Indian J Nucl Med* 2015;30(1):39–41.
- [6] Silver SA, Tavassoli FA. Primary osteogenic sarcoma of the breast: a clinicopathologic analysis of 50 cases. *Am J Surg Pathol* 1998;22(8):925–33.
- [7] Killick SB, McCann BG. Osteosarcoma of the breast associated with fibroadenoma. *Clin Oncol (R Coll Radiol)* 1995;7(2):132–3.
- [8] Lee JK, Sun SS. Primary osteogenic sarcoma of the breast demonstrated by Tc-99m MDP scintigraphy. *Clin Nucl Med* 1998;23(9):619.
- [9] Ellmann A, Jawa ZM, Maharaj M. Primary osteogenic sarcoma of the breast detected on skeletal scintigraphy. *Clin Nucl Med* 2006;31(8):474–5.
- [10] Coussy F, Le Scodan R, Guinebretiere J-M, Langer A, Lerebours F. Breast mass with intense 99mTc-diphosphonate uptake revealing primary breast osteosarcoma. *J Clin Oncol* 2011;29(15):e428–30.