Primary osteosarcoma of the breast with abundant chondroid matrix and fibroblasts has a good prognosis: A case report and review of the literature

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Abstract. The present study describes the case of a 77-year-old female with a recently self-detected, painless, 7-cm lump in the left breast, without evidence of metastasis clinically, who underwent mastectomy with dissection of the axillary lymph nodes. The tumor did not invade the chest wall and skin. The tumor was comprised of abundant chondroid matrix and fibrous tissue, with focal osteoid matrix, and was classified as a chondroblastic/fibroblastic variant. The tumor had a reverse zonal pattern. The tumor cells in the central portion were mainly spindle-like and sparse with minimal cytological atypia, while the remaining tumor cells in the periphery were mainly epithelioid, atypical and dense. Neoplastic osteoid woven bone or trabeculae were observed in the central portion of the tumor. No metastasis was identified in the axillary lymph nodes. The patient was alive without evidence of local recurrence or hematogenous spread at the 60-month follow-up.

Introduction

Primary osteosarcoma of the breast is a rare tumor, which is indistinguishable from conventional osteosarcoma of the bone and other extraskeletal sites using histological examination (1-6). In comparison, bone-producing spindle cell neoplasms with an epithelial origin, known as metaplastic (sarcomatoid) carcinomas, and malignant phyllodes tumors are more common (7). Primary breast osteosarcomas are considered to be highly aggressive tumors that are associated with early recurrence and a tendency for hematogenous, instead of lymphatic, spread, most commonly to the lungs (2,3,6,8-11). The tumor subtype, size and mitotic figures may be predictors of prognosis. The present study describes a case of a chondroblastic/fibroblastic variant of this rare tumor that had a good prognosis.

Case report

A 77-year-old Chinese woman presented to the Chengde Central Hospital Outpatient Department with complaints of a lump in the left breast that was self-detected a month prior to presentation. There was no history of nipple discharge, fever and pain. There was no history of breast trauma, prior local irradiation and surgery, nor any other tumor history. The patient denied using any hormonal therapy or a family history of breast disease. A breast examination showed a 7x7x6-cm irregular, firm mass in the lower inner quadrant of the left breast. The mass was poorly mobile and adherent to the skin and chest wall. No axillary lymphadenopathy was detected upon physical examination. Mammography showed that the mass was relatively well demarcated and partially calcified. The tumor did not invade the overlying skin and underlying chest wall. Breast carcinoma was thus indicated. The patient refused a needle biopsy and underwent a mastectomy.

The mastectomy specimen contained a 7-cm, relatively well-circumscribed mass, which had a gray-white cartilaginous appearance in the cross-section. Microscopically, the tumor was slightly lobular and relatively well demarcated, however, the adjacent fat tissue had been invaded and the surrounding non-neoplastic breast parenchyma revealed compressed lobular units (Fig. 1A and B). The tumor was mainly composed of cartilaginous components. The abundant cartilaginous proliferation varied from mature lacunar cartilage to poorly-differentiated areas displaying myxoid changes with no lacunar arrangement. In certain areas there was a transition from cartilaginous proliferation to fibrous cells. More than half of the tumor cells (~60%) were spindle-like and sparse with minimal cytological atypia, which were mainly observed in the central portion, while the other cells (40%), which were mainly in the periphery, were epithelioid, atypical and dense. High-power magnification revealed low mitotic activity even in the dense area (1 mitoses/10 high power microscopic fields; Fig. 1C and D). Neoplastic osteoid woven bone or trabeculae were observed in the central portion.

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Hemorrhage and necrosis foci were also observed in the central portion. No lymph-vascular invasion or neural invasion was observed. There was no histological evidence of an epithelial or carcinomatous component, despite extensive sampling of the tumor. No evidence of a preexisting malignant phyllodes tumor was present. The tumor was negative for cytokeratin, as well as for the estrogen and progesterone receptors and HER2. The tumor was classified as a chondroblastic/fibroblastic variant of osteosarcoma. The 15 axillary lymph nodes studied showed no metastasis. The patient underwent a simple mastectomy without post-operative adjuvant chemotherapy or radiation therapy. At 60 months post-mastectomy, the patient was alive and well without clinical evidence of local recurrence or distant metastasis. Written informed consent was obtained from the patient for publication of this case report and all accompanying images.

Discussion

Carcinoma is the most common malignancy of the breast and sarcomas form a minority of breast neoplasms. Primary osteosarcoma of the breast is rare and represents <1% of all primary breast malignancies. However, the actual incidence of primary osteosarcoma is difficult to determine, as a number of the ~100 previously-reported cases are likely to have included metaplastic carcinomas, as well as osteogenic sarcomas arising in association with a biphasic tumor, such as a phyllodes tumor or carcinosarcoma (7). Almost every previous reference to primary osteosarcoma of the breast in the literature is in the form of single case reports. Silver and Tavassoli reported a clinicopathological analysis of 50 cases observed over a 38-year period, the largest collection of primary breast osteogenic sarcomas to date (6). In almost all cases, the patients were diagnosed clinically as having breast carcinoma and the final diagnosis was established by histology. The histogenesis of primary osteosarcoma of the breast remains unclear, but an origin from totipotent mesenchymal cells of the breast stroma or a transformation from a preexisting fibroadenoma or phyllodes tumor has been suggested (1).

The presentation of breast osteosarcoma usually occurs at an advanced age, in contrast with skeletal osteosarcomas where the patients are younger. There has been a report of a breast osteosarcoma diagnosis at the age of 96 years, however, the usual mean age at presentation has been reported to be ~64 years, and cases are more frequently postmenopausal (1,6). Risk factors for extraskeletal osteosarcomas have not been identified to date, although certain cases have been attributed to local irradiation, trauma or the presence of a foreign body (6). The majority of patients present with a mobile, often large, irregular lump, without axillary metastases. Primary
Primary breast osteosarcomas are considered to be highly aggressive tumors associated with early recurrence and a tendency for hematogenous spread. However, the presence of a chondroblastic/fibroblastic variant, minimal cytological atypia, a low mitotic rate and good local control due to adequate resection may result in a good prognosis.

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