Letter to the Editor

Imaging findings of primary breast telangiectatic osteosarcoma: a case description

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Introduction

Telangiectatic osteosarcoma (TOS) is a rare subtype of osteosarcoma which is characterized by multiple dilated aneurysmal cavities filled with blood and high-grade sarcoma cells presented at the peripheral rim and septations, and it accounts for 2–12% of all osteosarcoma cases (1,2). Most of TOS are located in the fastest growing long tubular bones and commonly affect the younger population during the first two decades of life. In contrast, the extraskeletal TOS, a rare subtype of TOS, which is defined as TOS locates within soft tissue free from any underlying bone, mainly affect adults around 50–65 years old (3,4). TOS of the mammary gland is extremely rare and to our knowledge, no reliable data of incidence is readily available. Two isolated case reports have described TOS in intimate association with recurrent phyllodes tumor or metaplastic carcinoma of the mammary gland (5,6). In this case report, we describe TOS of the breast and its remarkable findings on ultrasonography, mammography, and magnetic resonance imaging (MRI). To the best of our knowledge, this is the first case reporting the imaging features of primary TOS of the breast without preexisting lesions. The pathological characteristics have previously been described in our previous paper (7).

Case presentation

History and physical examination

A 60-year-old woman presented a painful lump in her left breast for 10 days. Physical examination found a well circumscribed, fixed mass approximately 4 cm in diameter located in her upper outer quadrant of the left breast. The patient denied any history of breast-associated disease or family history of breast cancer.

Imaging findings

Ultrasonography revealed a locally irregular solid mass with some cystic cavity, measuring 48×34 mm in her left breast (*Figure 1*). The margin was circumscribed, and the internal echo structure showed heterogeneous hypoechoic with some anechoic area. No obvious blood flow was found inside or around the mass.

Mammography showed an oval mass with even high density, measuring 50×41 mm in the upper outer quadrant of the left breast (*Figure 2*). The mass was otherwise well-circumscribed with locally spiculated margins. No calcifications were found in the mass. The patient underwent an MRI as well. A poorly defined, patchy,

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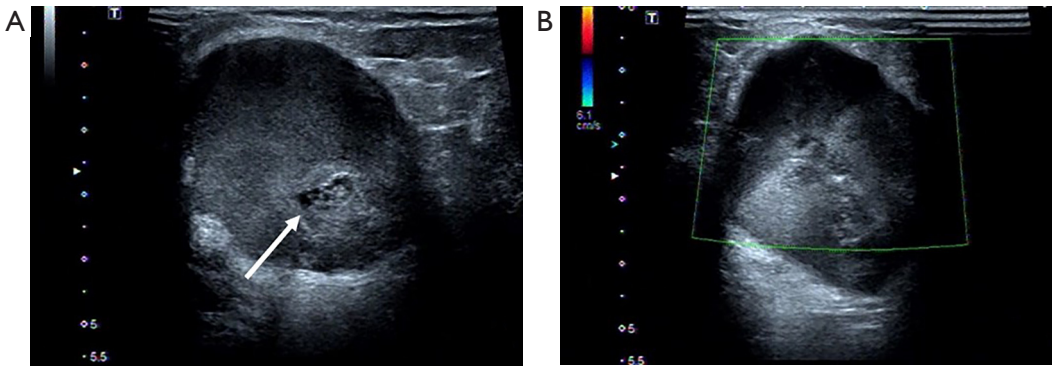

Figure 1 Ultrasonography of the mass in the left breast. (A) Grey scale ultrasonography shows a locally irregular, circumscribed, hypoechoic mass with some cystic cavity (arrow). (B) Color Doppler ultrasound shows no obvious blood flow inside or around the mass.

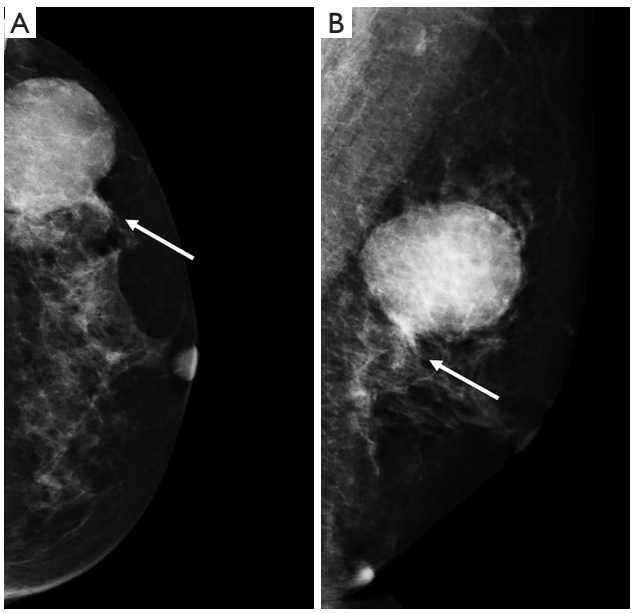

Figure 2 Mammography of the left breast showed an oval, even high-density mass. Locally spiculated margins were found in both CC view (A, arrow) and MLO view (B, arrow). CC, craniocaudal; MLO, mediolateral oblique.

abnormal signal measuring 27×25 mm was shown on MRI. The lesion had an iso-intense signal on T1-weighted sequences and iso- to hyper-intense signal on T2-weighted sequences with fat suppression, showing significantly inhomogeneous enhancement. The time-intensity curve revealed a persistent pattern. No obvious restricted diffusion was found on diffusion weighted imaging (DWI) sequences. Skin thickening and enhancement were noted adjacent to the lesion (*Figure 3*).

Operative procedure and histopathological findings

Biopsy showed a high-grade malignant tumor. Then, the patient underwent lumpectomy. The surgical specimen measured 45×40×40 mm. Under gross observation, a cystic mass containing blood clots was noticed. Microscopically, multiple dilated cavities filled with blood were observed, and the cavity wall contained highly malignant mesenchymal cells osteoid (*Figure 4*). Immunohistochemically, the tumor cells were positive for SATB2, P53, and CD99, and negative for all epithelial markers [AE1/AE3, Cam5.2, CK7, CK5/6, CK14, CK17, 34bE12, CK20, epithelial membrane antigen (EMA)]; vascular markers [CD31, CD34, v-ets erythroblastosis virus E26 oncogene homolog (ERG)]; neural differentiation markers (S100, SOX10); and melanoma markers (S100, Melan A). The Ki67 index was 30%. After thorough sampling of the tumor and according to immunostaining results, it was easy to rule out other malignant tumors such as breast cancer, carcinosarcoma, and malignant phyllodes tumor.

Mastectomy and sentinel node biopsy were performed subsequently to achieve a wide resection margin. To rule out primary bone osteosarcoma and distant metastasis, whole-body positron emission tomography-computed tomography (PET-CT) was performed. No lymph node or distant metastasis was noticed. There are no other suspected primary malignancies in the rest of the body. The tumor was finally diagnosed as a high-grade primary TOS of the breast. Detailed histopathological information of can be found in our previous paper (7).

Adjuvant therapy

Concerning the high risk of micrometastasis in TOS, we

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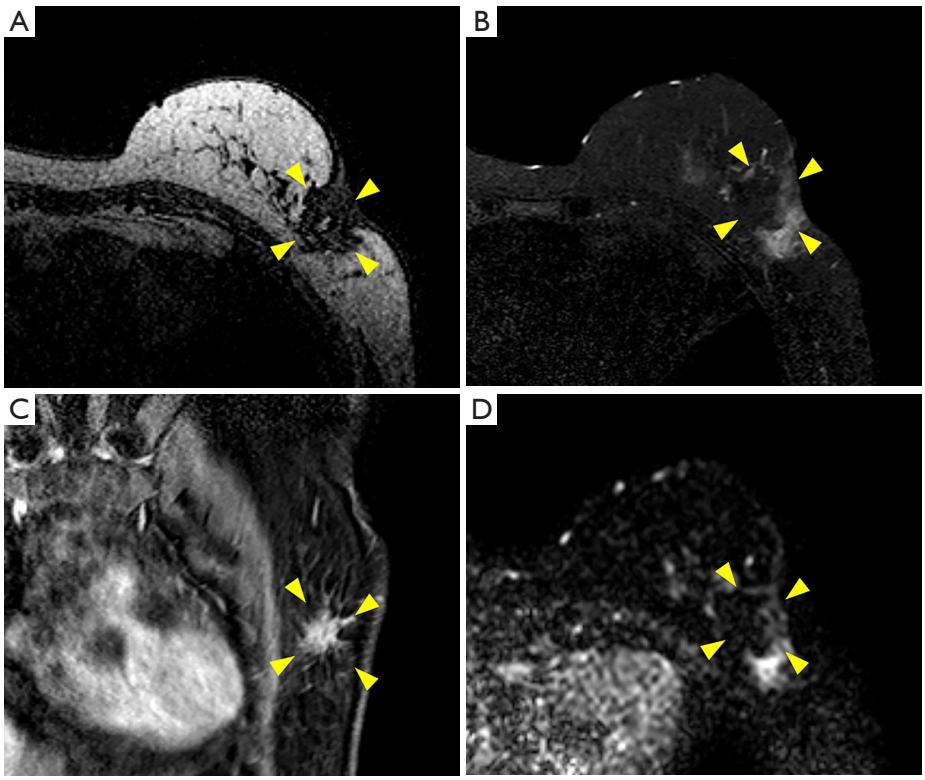

Figure 3 MRI of the left breast: a poorly defined, patchy, abnormal signal was shown on MRI. Arrowheads denote the location of the lesion. (A) The tumor had an intermediate signal intensity on T1-weighted sequences (axial). (B) The tumor had an iso- to hyper-intense signal on T2-weighted sequences with fat suppression skin thickening and enhancement were noted adjacent to the lesion (axial). (C) The tumor showed significantly inhomogeneous enhancement on contrast-enhanced T1-weighted sequences (coronal). (D) The tumor had a hypo-intense signal on DWI sequences (axial). MRI, magnetic resonance imaging; DWI, diffusion weighted imaging.

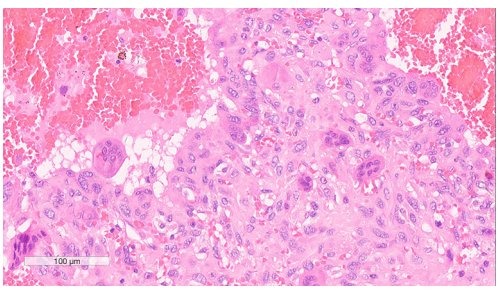

Figure 4 Histopathological examination of the tumor. Multiple dilated cavities filled with blood were noticed. Highly malignant mesenchymal cells with osteoid were located in the wall of the cystic cavity or grown in sheets (hematoxylin and eosin staining).

advised neoadjuvant chemotherapy, but the patient refused.

Follow-up

The patient has been regularly followed-up for 5 years after surgery and thus far, no recurrence or metastasis has been found.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

Primary sarcomas of the breast account for less than 1%

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Table 1 Summary of case reports of TOS in the breast

Author	Age of the patient	Medical history	Imaging findings	Treatment	Histological findings
Graadt van Roggen *et al.* (5)	50 years	Two-time local excision of phyllodes tumor	Ultrasound: a predominantly cystic mass with an irregular luminal outline and numerous fluid levels Mammography: an oval, well-circumscribed mass without calcification	Complete excision and chemotherapy	Focal remnants of the previously excised phyllodes tumor in continuity with areas of widespread differentiation towards TOS
Matsumoto *et al.* (6)	73 years	Gastric cancer	–	Mastectomy, sentinel lymph node dissection and chemotherapy	Metaplastic carcinoma with widespread TOS differentiation

TOS, telangiectatic osteosarcoma.

of all primary breast malignancies (8). Among those, osteosarcomas occurring in the breast are extremely rare. No data is yet readily available on the incidence of mammary osteosarcoma, and a proportion of approximately 0.1% of breast malignancies was estimated (9). TOS is a special subtype of osteosarcoma, and accounts for 2–12% of all osteosarcomas (1).

Different from TOS of the long tubular bones which usually occurs in children, adolescents, and young adults, primary TOS located in the axial skeleton or soft tissues is more commonly seen in middle to late adulthood (3,4). Prior to this case, only two other cases of mammary TOS have been reported before, and neither of them were primary mammary tumor. One of them was a local recurrence of a phyllodes tumor with differentiation to TOS in the breast in a 50-year-old woman with a two-time history of phyllodes tumor excision (5). The other one was a mammary metaplastic carcinoma with telangiectatic form of osteosarcomatous differentiation in a 73-year-old woman (6). The overview of these two cases is shown in *Table 1*. Including our case, all three patients were at the susceptible age of extraskeletal TOS.

The imaging signs of breast malignancies, such as ill-defined border, posterior echo attenuation, microcalcifications and rich blood flow were not found in ultrasound images of our case. The tumor showed a well demarcated, hypoechoic solid mass with no calcifications. No obvious blood flow was found inside or around the mass. According to the ultrasound features mentioned above, it may be confused with benign tumors of the breast such as fibroadenoma. The heterogeneous internal echo and cystic cavity and locally irregular shape, nevertheless, could be hints of TOS. On mammography, the mass was high-density and well-circumscribed with no calcifications, which is in accordance with the previous report (5). It seems to be difficult to identify TOS from benign occupies of breast such as fibroadenoma and breast cyst via mammography. However, locally spiculated margins in our case implied the possibility of malignancy, which can be distinguished from benign tumors of breast. The MRI results showed that our case shared some features with osteosarcomas located in soft tissues of other body parts (10,11). The tumor was ill-defined, had mixed signal intensity, and was significantly inhomogeneously enhanced. The skin thickening and enhancement adjacent to the mass implied the possibility of malignancy. The time-intensity curve revealed a persistent pattern and no obvious restricted diffusion was found on DWI sequences distinguished this case from invasive ductal carcinoma.

Other mixed solid and cystic masses in the breast such as abscess, intraductal papilloma, and other benign and malignant breast tumors with cystic degeneration should be differentiated from TOS. Symptoms, course of disease, and blood supply of the mass could be helpful to distinguish the lesions. Patients with breast abscess and intraductal papilloma may suffer from pain, swelling or nipple bloody discharge respectively, while patients of TOS have no obvious symptoms except for breast lump. Patients with benign breast tumors may have longer duration of breast lump. Malignant breast tumors usually grow faster and show rich blood flow in color doppler ultrasound. In this case, no obvious blood flow was found inside or around the tumor. However, sometime the diagnosis is still confusing. Chang *et al.* reviewed the ultrasound findings and pathological diagnosis of 175 cystic breast lesions and found that half (40 of 80 cases) of the cystic masses with a

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solid component were malignant. Hence, they suggested that breast masses of this kind should undergo biopsy with pathologic confirmation, regardless of the regular shape or well-defined margins (12).

It was once thought that the prognosis of TOS was poor. However, multiple studies have found that osseous TOS responded well to chemotherapy (13,14). An estimated 5-year event-free survival rate was 58.3%±11.9% according to Weiss *et al.* (13). For patients with metastasis, chemotherapy is a very important therapeutic option. However, patients without metastasis do not benefit substantially from chemotherapy (15). No studies about chemotherapeutic response of extraskeletal TOS, including mammary TOS, can be found in literature. We have reviewed the case reports of TOS located in soft tissue, and found most of the patients (6 in 7) received chemotherapy. The follow-up period ranges from 4 to 62 months. One patient was only treated with surgery, and the follow-up period was 10 months (11,16-18). Our patient underwent mastectomy and sentinel node biopsy, but no evidence of lymph node metastasis was found. She did not receive pre- or postoperative chemotherapy, or any other further treatment. At the time of generating this report, she was disease-free and being regularly followed-up even 5 years after surgery.

Conclusions

We presented the imaging findings of a rare case, which is the first primary TOS of the breast reported in literature. It is difficult to identify mammary TOS based only on imaging features which overlap with both benign and malignant breast tumors. Irregular shape, existence of cystic component, spiculated margins and changes of tissues adjacent to the tumor could be helpful in differentiation between this case and benign breast tumors. On the other hand, poor blood flow signal, a persistent pattern time-intensity curve, hypo-intense signal on DWI sequences, and no calcifications distinguish this case from other malignant breast tumors. In conclusion, the imaging morphology of the case was not specific, pathological biopsy is necessary for conclusive diagnosis.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://qims.amegroups.com/article/view/10.21037/qims-22-1372/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are 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. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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