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Case Report

Osteoblastoma of the breast: An uncommon and easily overlooked location ☆☆☆

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ABSTRACT

Osteoblastoma is a rare benign osseous neoplasm that accounts for 1%–3% of all primary bone tumors. Osteoblastomas can involve any part of the skeleton, but mainly occurs in the spine and other long bones, rarely in extra-skeletal areas. Extra-skeletal osteoblastomas arise from tissues outside of the bone, and only a few cases have been reported previously. To our knowledge, only one case of osteoblastoma in the breast has been described in the English literature. Here, we report another case of a breast osteoblastoma in a middle-aged woman, which was initially detected by ultrasound examination and digital mammography, and then was confirmed by histopathology. In this report, the imaging features and differential diagnosis of breast osteoblastoma are discussed.

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Introduction

Osteoblastoma was first described as an osteogenic fibroma of the bone by Lichtenstein and Jaffe in 1956 [1]. This disease occurs preferentially in males than in females at a ratio of 2.5:1. The age of onset of patients ranges from 3 to 78 years old, with a peak incidence between 20 and 30 years [2]. The most commonly involved bone is the vertebrae, followed by the long tubular bones of limbs [3], and rarely, it has been reported in the temporal [4], parietal [5] or occipital bones [6]. The manifestations of skeletal osteoblastoma in ultrasound and radiological examinations vary greatly. The typical image presents

as a “lytic” zone, which is surrounded by bony condensation and minimal osteosclerotic reaction peripherally.

Extra-skeletal osteoblastoma originates from extraosseous tissues. It is rather rare and is more difficult to diagnose due to the non-bone location. García Callejo and Ledebor reported an osteoblastoma of the thyroid and cricoid cartilage [7–8]. Another report claimed that an intraarticular osteoblastoma of the hip joint was similar to osteoblastoma according to its pathological features [9]. In 2017, Li et al. reported one case of a breast osteoblastoma and its transformation to an aggressive osteoblastoma in situ as a recurrence after local excision. Here, we describe another breast osteoblastoma to provide more information about extra-skeletal osteoblastomas to radiologists and clinicians.

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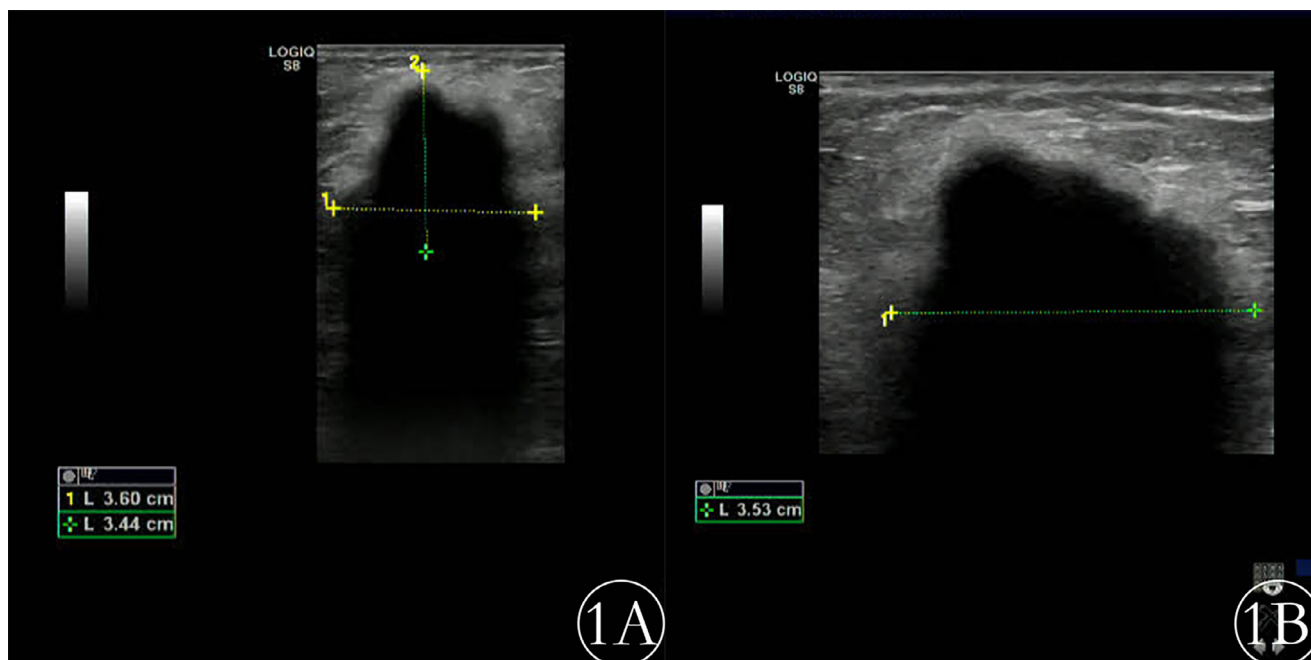


Fig. 1 – A and B: Ultrasound examination showed a hypoechoic mass with "eggshell-like" calcifications boundary.

Case report

A 56-year-old woman came to our hospital with a slightly painful non-inflammatory mass in her right breast. She had touched the mass accidentally one month before. Since then, without any treatment, the mass had not increased significantly. Physical examination found a hard, mobile and tender mass with a clear boundary. Ultrasound showed a hypoechoic mass approximately 35 mm × 36 mm × 34 mm, with an irregular shape and poorly defined borders. There was a hyperechoic halo at the edge of the right breast at 8–10 points 40 mm to the nipple (Fig. 1A, B). Color Doppler ultrasound displayed blood flow signals in the periphery. Full-field digital mammography in cranial-caudal and mediolateral oblique views revealed a dense mass (close to calcified density) measuring 38 mm × 42 mm in the upper right external breast. Mammographic appearance of spiculated breast lesion with lobular margins, without obvious calcification within it. The blood vessels around the mass were compressed. A few enlarged lymph nodes were detected in the bilateral axillae (Fig. 2A, B). A digital breast tomosynthesis double projection showed a sclerotic border around the mass (Fig. 2C, D).

The mass was excised after the patient signed an informed consent. The mass was located in the glandular layer and was poorly demarcated from the surrounding area. Finally, the mass, approximately 5 cm × 6 cm × 5 cm (Fig. 3A) was completely removed and photographed using a molybdenum target (Fig. 3B). A cross-section showed a grayish-white tough area surrounded by osteoblastic tissue (Fig. 3C). The specimen was decalcified and observed with a microscope. There was significant hyperplasia of woven bone trabeculae surrounded by overlying osteoblasts and interspersed with a fibrovascular

interstitium (Fig. 4A, B). The patient was pathologically diagnosed with breast osteoblastoma.

Discussion

Extra-skeletal osteoblastomas, namely, osteoblastoma arising from tissues outside of the bone, are very rare. Their etiology is unknown and is presumed to be partly due to trauma [10], which results in the differentiation of embryonic mesenchymal cells into bone tissue or the ossification of fibroblasts in the mammary gland mesenchyme in response to external or internal stimuli. In our case, the patient denied any breast-related trauma. The presentation of osteoblastoma imaging depends on the degree of ossification. Ultrasound can be valuable in the diagnosis of breast disease, but is less sensitive in the diagnosis of bone tumors. In our case, ultrasound showed a hyperechoic halo around the tumor. That result was similar to a previous report, which showed an eggshell-like calcification [11]. In that case report, the author also observed blood flow around the tumor that recurred 7 months after surgical resection and transformed into a progressive osteoblastoma. Whether the appearance of blood flow signals is related to the benign or malignant nature of the tumor and/or to future tumor recurrence is not clear. In our study, ultrasound also suggested the presence of blood flow around the tumor. Therefore, although the histopathological results showed a benign tissue, a close follow-up is necessary.

Digital mammography is the most commonly used tool for the diagnosis of breast disease and is very sensitive for diseases containing calcifications. In this case, the high-density image suggested the presence of ossification or calcification.

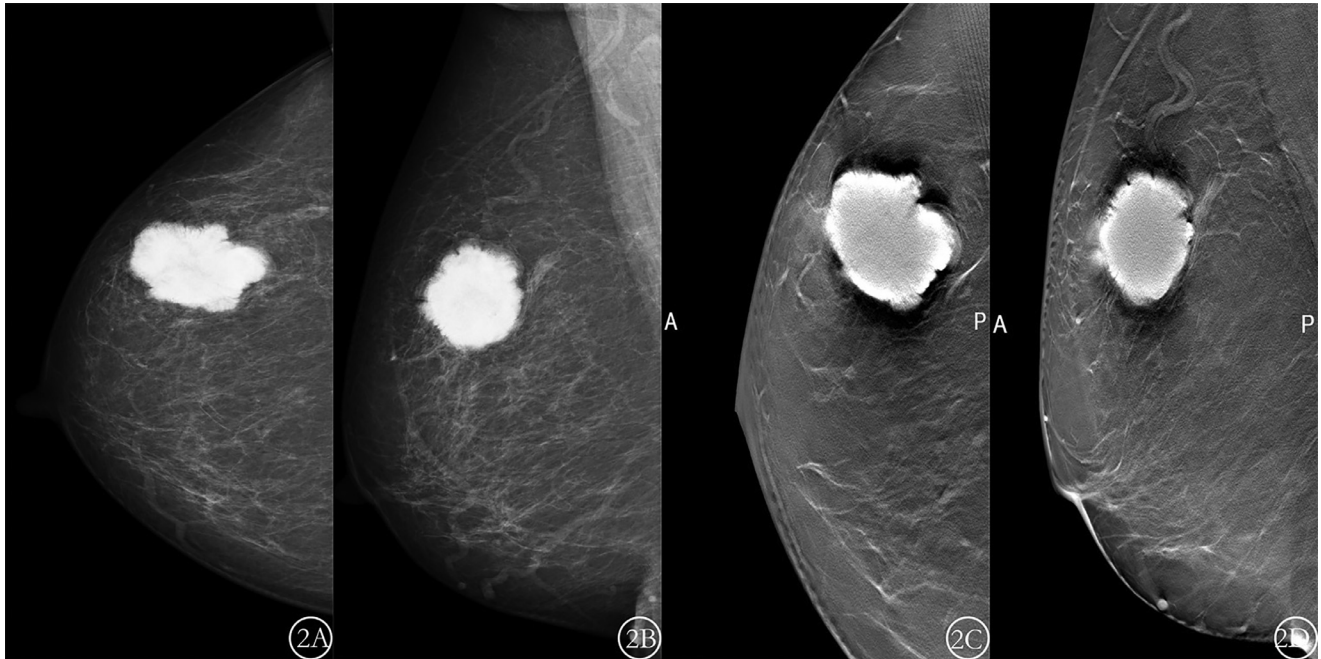


Fig. 2 – The mass was clearly dense with lobulation and burr visible (2A:Mammography CC; 2B: MLO), as well as hypodense areas around the mass (2C: Digital breast tomosynthesis with CC; 2D: MLO).

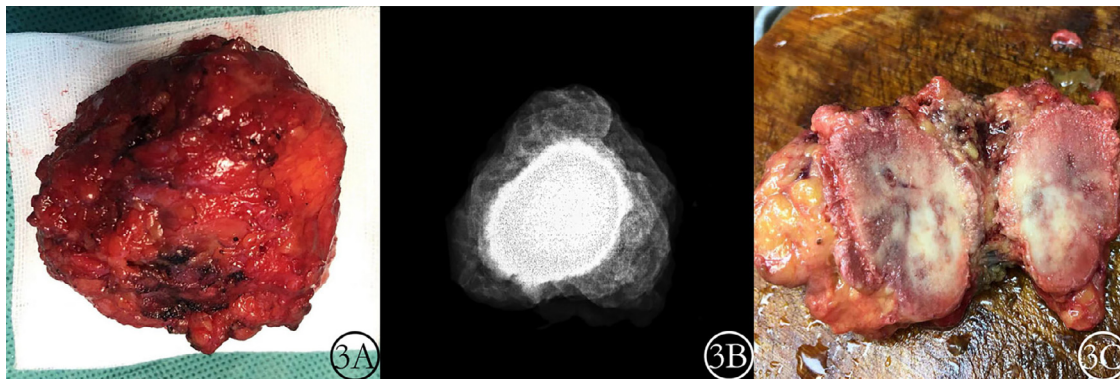


Fig. 3 – The mass with rough surface was completely removed (3A) and photographed by using a molybdenum target (3B). Its cross section showed a grayish-white tough area surrounded by osteoblastic tissue (Fig. 3C).

However, the images were overlapping in two dimensions and did not show the details of the lesion. We added a tomosynthesis examination to eliminate the interference of the overlapping images and show the boundaries of the tumor. The spiculated and sclerotic margins were displayed clearly.

Although the location and shape of osteoblastomas can be clearly shown by imagings, the final diagnosis still depends on the pathological findings due to the presence of heterogeneous images. The differential diagnosis of imaging includes: osteosarcoma, calcified fibroadenoma and breast carcinoma with calcification. Osteosarcoma arising in the breast typically appears on X-ray as a well-defined, dense mass, which may be lobulated at the margins. A highly heterogeneous density with calcification is one of the features of osteosarcoma. Our patient had several imaging features, namely,

a clear border around the mass and no soft tissue in the area, which helped rule out the diagnosis of osteosarcoma. Calcified fibroadenoma displays a lobulated or regular lesion with a different size of popcorn calcifications inside. The typical calcified breast cancer has an irregular shape with indistinct or speculated margins and pleomorphic or amorphous calcification.

A clear and correct diagnosis is particularly important for the choice of treatment options. If a malignant tumor is suspected on imaging, further pathological examination should be done to determine the nature of the lesion. Most osteoblastomas are benign, but they do have the potential to cause local destruction and degenerate to a more malignant tumor such as an osteosarcoma, as well as having a tendency to recur. Radical and complete and careful follow up are required. In

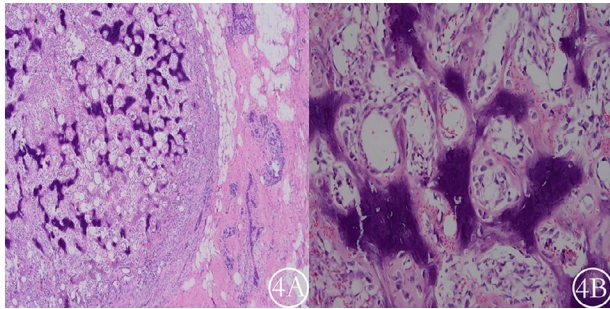


Fig. 4 – Well-defined tumor tissue in the breast lobules (4A: HE x 40). Plentiful proliferating lamellar woven bone trabeculae surrounded by mature osteoblasts, slightly hyperplastic interstitium and blood vessels were observed (4B: HE x 200)

addition, early detection and treatment generally leads to resolution of the related pain and other discomforts.

Conclusion

In general, osteoblastomas are relatively rare tumors and are difficult to diagnose when they occur in extraosseous tissues, especially in soft tissues including breast tissue. Ultrasound images and digital mammography may show the existence of suspicious tumors and, the final diagnosis should be confirmed by pathological findings. Early diagnosis, appropriate treatment, and close follow-up are very important for patients with osteoblastoma.

Author contributions

Lu Chen wrote the main part of the manuscript, Shu-hai Zhang also wrote parts of the manuscript. Xiao-min Tang took part in the inspection of Mammography. Cheng-cheng Ma and Li Yang carried out literature search and manuscript editing. Zhi-zhen Gao reviewed the manuscript. All authors read and approved the final manuscript.

Patient consent statement

Written informed consent has been obtained from the patient for the publication of this case report and any accompanying photographs.

This case report is an incidental finding in the course of clinical work and has no ethical implications.

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