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CASE REPORT

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Extraosseous osteoblastoma: A rare cause of breast mass in a prepubertal girl

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Abstract

Extraosseous osteoblastoma of the breast, a rare disease, was diagnosed in a prepubertal girl. After tumor excision, the patient recovered well and an optimal cosmetic result was achieved. Interdisciplinary discussions about the case are essential.

KEYWORDS

general surgery, obstetrics and gynecology, pediatrics and adolescent medicine

1 | INTRODUCTION

Extraosseous osteoblastomas are very uncommon. We present a case of osteoblastoma of the breast in a 9.8-year-old girl, its clinical and radiologic findings, treatment, and follow-up.

Osteoblastoma is a rare, benign bone tumor with a predilection for the spine, sacrum, and long tubular bones. It occurs mostly within the first four decades of life, with a greater probability of occurring in the second and third decades. Patients with osteoblastoma usually have a good prognosis and a local recurrence rate of around 15%-20%.[1,2]

We present a case of extraosseous osteoblastoma, an extremely rare disease,[2,3] in the breast in a prepubertal girl.

2 | CASE PRESENTATION

A 9.8-year-old obese (body mass index [BMI] = 24.7 kg/m^2; above the 97th percentile) female, Tanner stage 3, with normal child development, presented to the breast clinic at our institution. She reported having painless swelling of the left breast for 1 month. Clinical examination revealed a mass in the upper outer quadrant of the left breast. Magnetic resonance imaging (MRI), which had been performed a few days before presentation, detected a 27 × 18-mm lump located laterocranially in the left breast. It was well defined, with pronounced calcifications and limited diffusivity, and assessed as BI-RADS 4 (Figure 1). BI-RADS (Breast Imaging Reporting and Data System), developed by the American

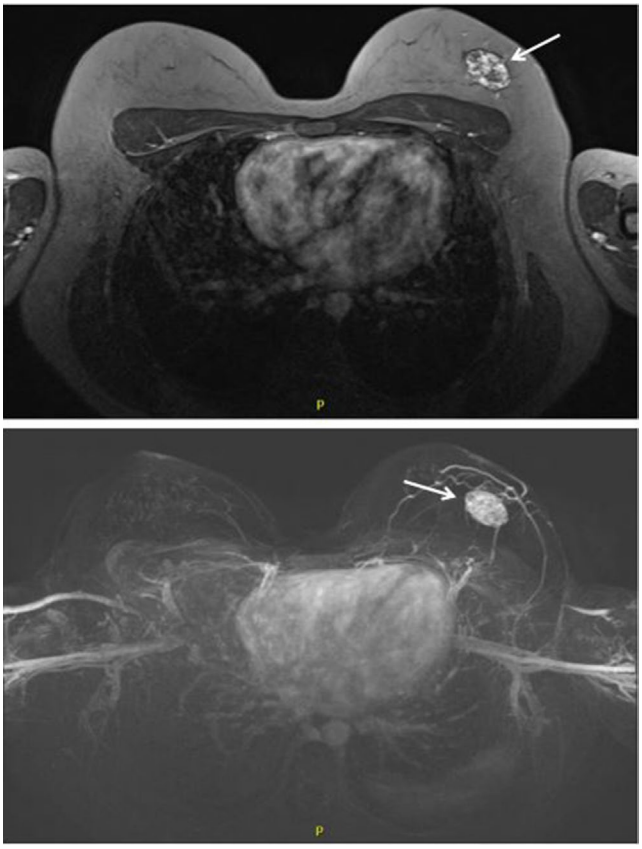

FIGURE 1 Magnetic resonance imaging (MRI) revealed a mass (arrow) consisting of calcifications located in the upper outer quadrant of the left breast, BI-RADS (Breast Imaging Reporting and Data System) 4

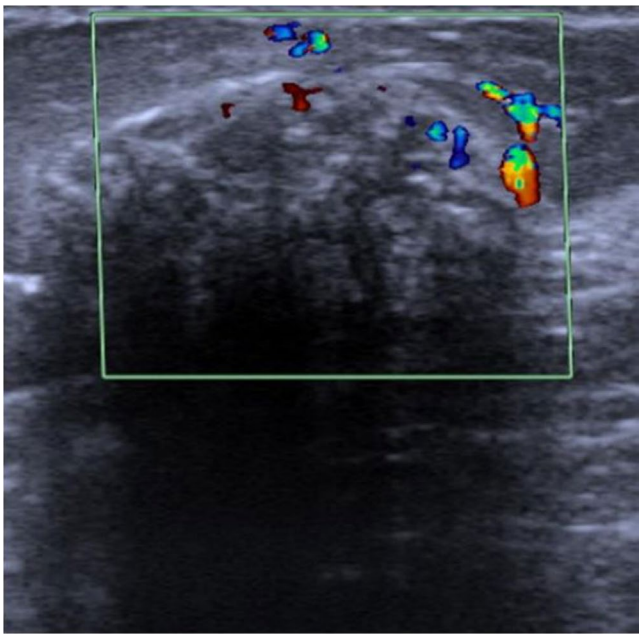

FIGURE 2 Mammary sonography showing a 28-mm lesion with multiple calcifications and remarkable hyperemia in the left breast

College of Radiology, is a structured system for assessing, reporting, and classifying images of the breast. Each category indicates the likelihood of malignancy and recommends further management. BI-RADS 4 indicates suspected malignancy and recommends a needle biopsy.[4,5] An ultrasound scan of our patient confirmed the presence of a 28-mm lesion. Multiple calcifications and hyperemia were identified, but no associated lymphadenopathy (Figure 2). Material from a subsequent ultrasound-guided biopsy consisted of fibrovascular stroma and bone trabeculae composed of osteoid and osteoblasts. Finally, the histopathological features were suggestive of an osteoblastoma, which was confirmed upon imaging. Abdominal sonography and a full-body MRI showed normal images.

The case was presented and discussed in an interdisciplinary meeting, and the recommended treatment was surgical excision of the tumor. After explaining the surgical procedure and the follow-up in detail and obtaining the consent of the patient and her parents, the lump was excised. The patient recovered well, and an optimal cosmetic result was achieved. A histopathological examination of the resected tumor (4 × 2.5 × 2.5 cm) confirmed the diagnosis of osteoblastoma with no evidence of malignancy (Figure 3). The resected tissue had negative margins. Adjuvant therapy was not indicated, and the first follow-up visit was scheduled for 6 months after surgery.

One year after tumor excision, the patient presented with excellent wound healing and optimal cosmetic results, and palpation of the breast did not reveal any abnormalities. Breast development was normal, and the breasts were the same size. We did not find any changes in the skin or the nipple attributable to the resected tumor. In addition, breast ultrasound (BI-RADS 2) showed normal findings. Follow-up examinations and ultrasound scans are to be performed every 6 months for another year, and then annually for 3 years.

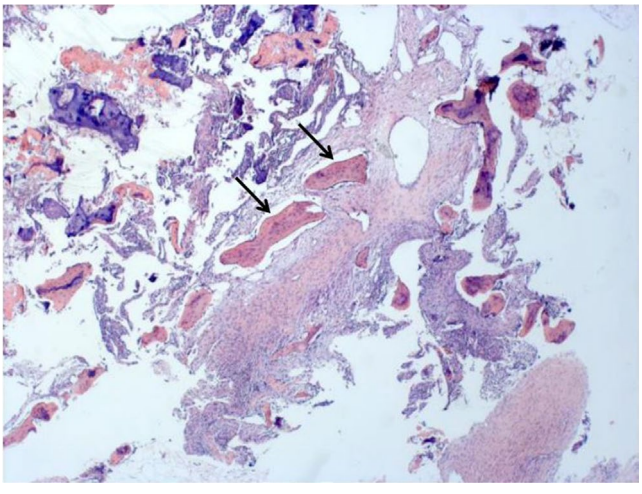

FIGURE 3 Histopathological examination of the breast tumor: The histological pattern of the bony tissue (arrows) was typical of an osteoblastoma, embedded in glandular adipose tissue. Hematoxylin-eosin stain

3 | DISCUSSION

The 8 types of benign bone tumors are osteochondroma, osteoma, osteoid osteoma, osteoblastoma, giant cell tumor, aneurysmal bone cyst, fibrous dysplasia, and enchondroma. Osteoblastoma is a rare, benign bone tumor that accounts for 1% of primary bone tumors. It arises predominantly in the axial skeleton, with spinal lesions one-third of the reported cases. It usually occurs within the first four decades of life, with a greater probability of occurring in the second and third decades.[1,6] The 73 patients with osteoblastoma included in the retrospective study of Yalcinkaya et al[7] had a mean age of 19.6 ± 9.9 years.

Osteoblastomas are benign bone-forming tumors that histologically are characterized by the proliferation of osteoid and woven bone in the stroma and closely resemble osteoid osteomas. Osteoblastomas can be distinguished by their significantly larger nidus (>2 cm in diameter) compared to that of osteoid osteomas. In addition, differential diagnoses include aneurysmal bone cyst, well-differentiated osteosarcoma, giant cell tumor, ossified hematoma, and heterotopic ossification.[1,2,7] The histological characteristics of osteosarcoma can strongly resemble those of osteoblastoma, making them difficult to differentiate.[8] *FOS* and *FOSB* are members of the AP (activated protein)-1 family of transcription factors.[9] Fittall et al[10] described the recurrent rearrangement of *FOS* and its paralogue *FOSB* in osteoblastoma and osteoid osteoma. They concluded that changes in *FOS* and *FOSB* can be used as diagnostic markers for these tumors. Amary et al[9] demonstrated that most osteoid osteomas (73%) and osteoblastomas (83%) were positive for c-FOS protein expression, indicating that c-FOS immunohistochemistry is a useful diagnostic marker of these tumor types. However, immunoreactivity, usually focal or patchy, was found in 14% of patients with osteosarcoma. We did not perform immunohistochemistry for FOS rearrangement for our patient.

Progressive pain is the most common symptom of osteoblastoma[6]; however, it was not present in our patient. In a single-center study, pain was reported by 45% of patients with osteoblastoma, and the treatment of choice was conservative surgery.[7] The clinical presentation of extraosseous osteoblastoma depends on the affected area; for example, an osteoblastoma arising from the vocal fold may present as acute airway obstruction[2] and extraosseous thoracic or lumbar foraminal osteoblastoma may present as back pain[11,12] or radiculopathy.[12] In general, the prognosis for those with osteoblastomas is good.[1] However, some osteoblastomas have a more aggressive clinical course with local recurrence but no distant metastases.[2,13,14] Berry et al[15] analyzed 99 cases of osteoblastoma and found that the local recurrence rate in most patients following curettage was 24%. This relatively high recurrence rate could be minimized by surgical resection. In a retrospective review by Wu et al[16] of 13 patients with osteoblastoma in the spine, only one patient had local recurrence in the average follow-up period of 43.8 months. Follow-up examinations were performed every 3 months in the first year of follow-up, every 6 months in the second year, and then annually. The authors concluded that complete resection is essential for preventing recurrence.

The extraosseous growth of osteoblastomas is extremely rare.[2,3] Extraosseous osteoblastomas have been found in the larynx and soft tissue (axilla), and as extraosseous thoracic foraminal osteoblastoma and lumbar extraosseous intraforaminal osteoblastoma.[2,11,12,17] Li et al[18] described an osteoblastoma of the breast in a 65-year-old woman, which recurred 7 months after initial resection. In addition, cancellous osteoma in the breast and osseous metaplasia of the breast have been described in the literature.[19-21] Joshi et al[22] reported a case of an HIV-positive woman with benign osseous metaplasia of the breast that presented as a breast lump. Alyami et al[3] also reported a case of benign osseous metaplasia of the breast that presented as a breast lump, but their patient had no chronic illness.

To the best of our knowledge, this is the first report of extraosseous osteoblastoma of the breast in a prepubertal female. Following clinical and radiologic assessment, an ultrasound-guided biopsy was performed, and the histopathological examination of this specimen suggested an osteoblastoma. After surgical excision of the tumor, the patient recovered well and an optimal cosmetic result was achieved. No recurrence was observed 1 year after excision of the tumor. Follow-up visits will occur every 6 months for the first 2 years and then annually for the next 3 years. Because extraosseous osteoblastoma is a rare disease, the factors associated with local recurrence are unknown. To date, observation of a clinically asymptomatic breast osteoblastoma is not indicated. There are no data on the natural progression of this disease.

In conclusion, extraosseous osteoblastoma in the breast is an extremely rare disease, of which only two cases have been reported: a 65-year-old woman and a 9.8-year-old girl (our patient). Correct diagnosis and multidisciplinary management of this ultra-orphan disease are essential for the optimal treatment of the patient, and careful follow-up is required to ensure early recognition of recurrence.

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Published with written consent of the patient.

CONFLICT OF INTEREST

The authors declare that there are no conflicts of interest regarding the publication of this article.

AUTHOR CONTRIBUTIONS

Sabine Danzinger: collected the data, was the main contributor to the manuscript, and reviewed and approved the manuscript; Leo Kager: treated the patient, contributed to writing

the manuscript, and reviewed and approved the manuscript; Maria Bernathova and Susanna Lang: provided the findings, contributed to writing the manuscript, and reviewed and approved the manuscript; Werner Haslik and Christian F. Singer: operated on the patient and reviewed and approved the manuscript.

ETHICAL STATEMENT

All procedures performed for our study met the standards of the ethics committee of our institution and those of the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. Written informed consent for the publication of this case report and any accompanying images was obtained from the patient's mother. A copy of the written consent is available for review by the editor in chief of this journal.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analyzed during the current study.

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