Volume 22, No 4 | International Journal of Radiation Research, October 2024

Giant pseudoangiomatous interstitial hyperplasia in adolescent breast: A case report based on imaging findings

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► *Case report*

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Received: September 2023
Final revised: February 2024
Accepted: March 2024

Int. J. Radiat. Res., October 2024; 22(4): 1099-1102

DOI: 10.61186/ijrr.22.4.1099

Keywords: *Comprehensive nursing, lung cancer, radiotherapy, respiratory function test, quality of life, self-care ability.*

ABSTRACT

Background*:** This Pseudoangiomatous stromal hyperplasia (PASH), a rare benign breast tumor, manifests as a fissured pseudovascular cavity formed by interstitial hyperplasia. Imaging often presents it as a nodular or non-mass-type lesion associated with other benign lesions. Clinical detection of PASH generally reveals a rare mass type. ***Materials and Methods: We report a case of a 13-year-old girl with a rapidly growing circumscribed mass in her unilateral breast. Mammography of the breast revealed a high-density mass without calcification; color Doppler ultrasound showed a low echo with arterial blood flow; magnetic resonance imaging (MRI) suggested that the mass was heterogeneous; diffusion-weighted imaging (DWI) results indicated a slightly high signal; T2-weighted imaging (T2WI) high-signal components showed a fissure without enhancement; and the time-signal intensity curve of solid components displayed a type I curve. Patient underwent surgical treatment, whereupon pathological results indicated PASH. ***Conclusion:*** PASH presents as a rapidly growing borderline mass without calcification in a unilateral breast. Comparative analyses of the imaging characteristics of multiple MR sequences show significant enhancement of the low-signal area in T2WI results.

INTRODUCTION

Pseudoangiomatous stromal hyperplasia (PASH), which is a rare type of breast stromal proliferative tumor, is frequently accompanied by other breast diseases but occasionally manifests alone. Previous studies have shown that PASH commonly affects premenopausal women or patients who undergo hormone replacement therapy, as a result of which most scholars believe that PASH onset is related to hormones. According to existing reports, the tumor is often limited to nodular or non-mass-like lesions, and this makes it difficult to distinguish PASH from fibroadenoma and adenopathy. Furthermore, the mass type is rare.

In this study, we report a case of PASH that presented as a rapidly growing breast tumor in an adolescent female. To this end, we summarized a complete variety of imaging data and explored the imaging features of different examination methods to help diagnostic imaging specialists gain a comprehensive understanding of the disease.

CASE DESCRIPTION

The patient was a 13-year-old female with a right breast mass that had been gradually increasing for 6 months. Physical examination revealed that the patient's right breast had a 15 cm × 20 cm tumor. The tumor had soft texture and unclear boundaries. It was moderately mobile, with no adhesion to surrounding glandular tissue. Furthermore, the tumor exhibited no redness, swelling, or ulceration, nor were there any orange peel symptom, symmetrical double nipples, depression, and discharge.

Mammography using a Selenia Panoramic Digital Mammography System (Hologic Inc., United States) revealed a huge mass in the right breast, with smooth edges and a uniform density. No malignant calcification was found, nor was thickened skin. Unfortunately, it was not possible for us to accurately measure the size of the tumor because mammography cannot fully display the tumor (figure 1). Ultrasonography (Siemens HELX OXANA Ultrasonic Diagnostic Apparatus) showed a 15.01 cm × 8.94 cm × 7.38 cm hypoechoic mass in the right breast. The imaging result was characterized by an uneven echo while displaying a clear boundary around the mass. Color Doppler flow imaging (CDFI) indicated arterial blood flow signals (figure 2). Chest computed tomography (CT) (PHILIPS 256-layer CT, the Netherlands) showed a large mass in the right breast with an uneven internal density. The maximum, minimum, and average CT values were 57 Hu, −12 Hu, and 27 Hu, respectively (figure 3). MRI (GE 1.5T Twin speed with Exite II superconducting magnetic resonance imaging system), using the contrast agent Gd-DTPA, revealed an oval-shaped mass in the right breast, 10.8 cm × 14.9 cm × 10.9 cm in size, with smooth edges and uneven signals. The mass displayed equal signals on T_1WI, low signals on

[DOI: 10.61186/ijrr.22.4.1099]

T_2WI, slightly low signals on T_1WI, high signals on T_2WI, slightly high signals on DWI, uneven signals on apparent diffusion coefficient (ADC) imaging, equal signals on T_1WI, low signals on T_2WI and a clear capsule, with uneven enhancement within a dynamic enhancement scan. The time-signal intensity curve was asymptotic, while adjacent glands were found to be compressed and displaced (figure 4). Diagnosis: Based on the imaging data, the diagnosis was a giant mass in the right breast, a type of fibroepithelial tumor, BI-RADS: 4.

Intraoperative findings: The tumor capsule, which was approximately 10 cm × 15 cm × 20 cm, was intact and soft in texture, with dilated surface blood vessels.

Pathology: Gross appearance: The tumor, 16 cm × 13 cm × 5.5 cm in size, was slightly lobulated and had a capsule on its surface. It had a gray-white section, and the texture was solid and tender. Histopathology: The glands were sparse, with spindle-shaped cells arranged like a slit in the stroma, and there were numerous visible blood vessels. Immunohistochemistry: As displayed in figure 5, the immunohistochemistry results were as follows: CD31 (partial+), CD34 (+) calretin/CR (focal+), D2-40 (–), Ki-67 (1%+), SMA (smooth muscle+), CK (AE1/AE3) (epithelial+), ER (epithelial 90%+) and PR (epithelial 90%+). Pathological diagnosis: The patient was diagnosed with pseudoangiomatous stromal hyperplasia of the right breast.

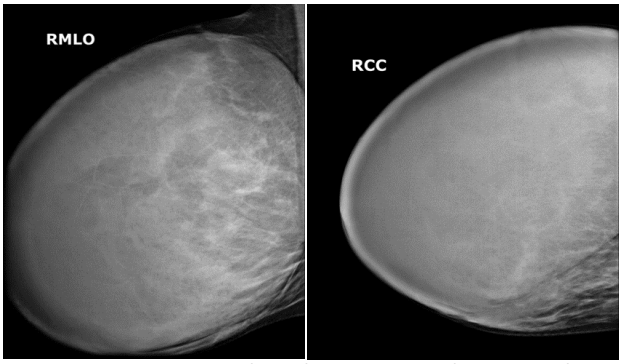

Figure 1. Mammography: A huge mass can be seen in the breast, with equal density, clear boundaries and no lobulation or malignant calcification.

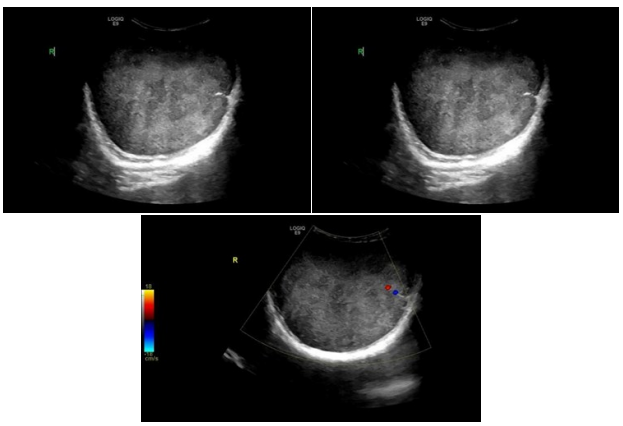

Figure 2. Two-dimensional ultrasonic imaging: **a**, the mass presents with moderate to low echogenicity and clear boundaries. **b**, CDFI shows arterial blood flow.

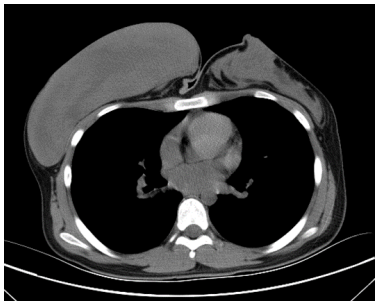

Figure 3. CT: Right breast soft tissue mass with clear boundary with surrounding tissue and uneven internal density.

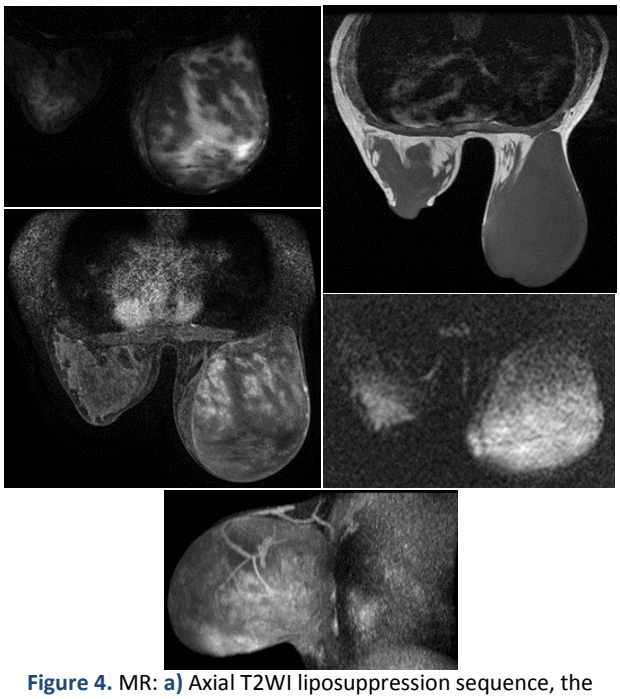

Figure 4. MR: **a)** Axial T2WI liposuppression sequence, the right breast is significantly enlarged, with an elliptical mass visible inside, clearly defined, and presenting as low signal with patchy high signal. The low signal capsule is clearly visible. **b)** Axial T1WI, the mass showed equal or slightly lower signal intensity compared to normal breast glands. **c)** Axial T1WI enhanced scan, compared with axial T2WI, the low signal components on T2WI showed significant enhancement, while the high signal components on T2WI showed no significant enhancement, the capsule showed linear and significant enhancement. **d)** DWI, the mass showed slightly high signal intensity. 4e the sagittal enhanced vascular diagram, bundant blood vessels can be seen inside the tumor.

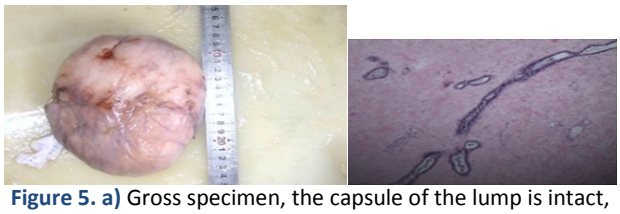

Figure 5. a) Gross specimen, the capsule of the lump is intact, with small lobules visible, solid, tender in texture and a grayish white section. **b)** HE (×100), multiple blood vessels can be seen in the mass and spindle shaped cells can be seen in the stroma arranged in a slit like manner.

DISCUSSION

First reported and named by Vuitch in 1986 [1], PASH was classified as a benign mesenchymal tumor by the World Health Organization in 2012 [2]. As of

[DOI: 10.61186/ijrr.22.4.1099]

now, the etiology and pathogenesis of PASH remain unclear. In clinical practice, PASH often presents as a single painless mass. According to Yigit B *et al.* [3], PASH is particularly common in premenopausal women and postmenopausal women undergoing hormone replacement therapy. Moreover, it can be seen in males undergoing breast development therapy [4-5]. PASH is a rare benign stromal proliferative lesion of the breast. Formed by the proliferation of myofibroblasts, it is histologically characterized by the formation of fissured pseudovascular cavities [6].

Mammography suggests that PASH mainly manifests as clearly defined noncalcified nodules [7]. Sun *et al.* [8] reported that the maximum diameter of the mass in adolescent female patients exceeds 10 cm. The 13-year-old patient in this case report exhibited a maximum mass diameter of 16 cm. The mammography result was affected by the equipment detector, thus failing to completely envelop the mass. The observed mass displayed equal density, clear boundaries and no calcification, which was consistent with previous reports [7-8]. Ultrasound examination revealed that the main manifestation of PASH was a heterogeneous hypoechoic mass or nodule, with smooth or irregular edges, without calcification, but with visible blood flow signals inside [8-10]. The ultrasound findings of this case report were consistent with those reported in the previous literature, showing a heterogeneous hypoechoic mass with clear boundaries and visible arterial blood flow signals inside. Owing to the fact that CT is not a routine examination for breast diseases, there are few reports on the CT characteristics of PASH. In the case reported in this paper, preoperative lung CT examination revealed a large and nonuniformly dense mass in the breast. The mass had soft tissue density, with a slightly lower central density. Bayramoglu Z [11] reported that PASH often presents as an uneven signal on MRI, while T_1WI mainly shows equal and low signals. In contrast, the signals are generally mixed on T_2WI. The mass displays persistent intense contrast enhancement, while dynamic enhancement curves often show inflow patterns. In the present case report, the MR plain scan showed significant enlargement of the right breast, with an oval-shaped mass visible inside. Some components showed low signals on T_1WI and T_2WI, while the enhanced scan indicated gradual enhancement. The time-signal intensity curve measured at multiple points displayed an inflow pattern. The lesion appeared to be patchy, with slightly lower signals on T_1WI and higher signals on T_2WI within the mass; however, these characteristics were not evident from contrast-enhanced scanning, which led us to consider the lesion as a vascular edema area.

In the present case report, the 13-year-old patient presented with a large tumor that was growing rapidly and had clear boundaries. Hence, it was necessary to distinguish this tumor from juvenile fibroadenoma, phyllodes tumor, medullary carcinoma, and mucinous adenocarcinoma. Juvenile fibroadenoma of the breast, which is especially common in adolescents, often manifests as a significantly enlarged breast with well-defined masses, mostly circular or oval in shape. Pathological examination shows abnormal proliferation of stroma and epithelial cells within the masses, with low T_2WI and equal T_1WI signals in the stroma, slightly higher T_2WI and equal T_1WI signals in the epithelium and weak enhancement in the stroma during enhanced scanning [12]. These characteristics distinguish juvenile fibroadenoma from PASH, which is significantly enhanced due to the presence of numerous blood vessels in the interstitial components. Phyllodes tumors of the breast often appear as rapidly growing masses, but these masses tend to bleed [13] and are distinct from PASH. Medullary carcinoma, which commonly manifests in young women, can be accompanied by lobulated, expansive growth with clear boundaries. However, DWI is limited in terms of diffusion and signal intensity and ADC imaging shows low signal intensity. The time-signal intensity curve is largely a plateau or outflow type [14], which distinguishes the curve from that in PASH. Mucinous breast carcinoma can manifest as a smooth-edged mass, with a characteristic high DWI signal and a high ADC image signal, often exceeding 2.2×10^{-3} mm^2/s. The TIC curve indicates an inflow or plateau type [15], which often leads to confusion with benign tumors. However, tumors often manifest in postmenopausal women, and due to the inconsistent tumor growth rate, these tumors often have a lobulated appearance, which can help distinguish them from PASH.

CONLUSION

Some adolescent females exhibit the clinical characteristics of PASH, which is marked by rapid tumor growth, and there is no calcification within the mass. MRI displays equal T_1WI and low T_2WI signals, indicating gradual enhancement. The low T_2WI signal area within the mass shows significant enhancement.

Ethical considerations: Ethical approval for this study was obtained from the Medical Ethics Committee of Tangshan Gongren Hospital (registration no. GRYY-LL-2018-48, registration date: November 27, 2018). The patient has signed informed consent.

Financial support: none.

Declarations of interest: none.

Contributors: L.C. collected the case and drafted the manuscript. L.S. and L.Z. performed the imaging analysis. J. W. reviewed the literature and revised the manuscript.

[DOI: 10.61186/ijrr.22.4.1099]

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[DOI: 10.61186/ijrr.22.4.1099]