

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Pseudoangiomatous stromal hyperplasia in a healthy young adult male[☆]

Shahin C. Owji, BS, Neel Shroff, BS, Angelica S. Robinson, MD, Flavia E. Posleman Monetto, MD*

Department of Radiology, University of Texas Medical Branch, 301 University Boulevard, Galveston, TX 77555, USA

ARTICLE INFO

Article history:

Received 3 March 2022

Revised 9 May 2022

Accepted 12 May 2022

Keywords:

Pseudoangiomatous stromal hyperplasia

Breast

Male

Benign

Tomosynthesis

Ultrasound

ABSTRACT

This case report describes the occurrence of a rapidly enlarging pseudoangiomatous stromal hyperplasia (PASH) tumor in a 20-year-old male patient. The diagnosis was made via tomosynthesis and ultrasound-guided biopsy with pathological correlation consistent with PASH. The patient's case was discussed, and he was recommended to undergo surgical resection of the mass to alleviate symptoms due to its large size. Surgical pathology confirmed the original diagnosis and the patient had an uncomplicated postoperative course. Here, we exhibit our imaging findings; review classic presentations of PASH on mammography, ultrasound, and MRI; and discuss histological characteristics of this benign entity.

© 2022 The Authors. Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license

(<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Introduction

First described in 1986, pseudoangiomatous stromal hyperplasia (PASH) is a benign disease process of the breast that is typically observed in pre- and perimenopausal women [1]. Nontumor forming PASH has been established as an incidental microscopic finding in breast tissue, found in up to 23% routine breast biopsy specimens [2]. However, the development of a tumorous form is much more rare [3]. The development of this tumor type in men is even more rare, with

only 7 cases previously reported in the English literature [4–10]. Herein, we describe the rare presentation of a young adult male patient with a large, rapidly growing unilateral PASH tumor.

Case report

The patient was a healthy 20-year-old male with no significant past medical history who presented to the clinic with

[☆] Competing Interests: None.

* Corresponding author.

E-mail address: feposlem@utmb.edu (F.E. Posleman Monetto).

<https://doi.org/10.1016/j.radcr.2022.05.033>

1930-0433/© 2022 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)



Fig. 1 – Tomosynthesis: bilateral mediolateral oblique projections with a right breast global asymmetry encompassing the majority of the breast tissue and measuring approximately 65 mm in dimension in a 20-year-old male. The triangle corresponded to a palpable abnormality.

complaints of an enlarging right breast mass over the past 2 months. The mass had been increasing in size, but did not contribute to any other physical symptoms such as pain, erythema, discharge, or decreased range of motion. On initial physical exam, the mass measured approximately 2 × 2 cm and was nontender, nonfluctuant, and immobile. The patient was recommended to undergo bilateral mammography and sonography in order to better characterize the mass. Two months after initial evaluation, the patient underwent bilateral tomosynthesis and breast ultrasound. Tomosynthesis demonstrated global asymmetry in the right breast, which encompassed the majority of the breast tissue approximately 65 mm in greatest dimension. Findings of tomosynthesis are shown in Fig. 1. The right breast ultrasound revealed a mass in the 9:00-12:00 subareolar area of the right breast. The visualized axillary lymph nodes were unremarkable. The findings were given a category of BI-RADS 4A and biopsy of the mass was recommended. The sonographic findings are shown in Fig. 2. The patient underwent ultrasound-guided biopsy with histopathology indicating benign breast tissue with PASH. Ultrasound-guided biopsy and postbiopsy mammogram are shown in Fig. 3. Surgery was consulted and the patient's case

was discussed at interdisciplinary tumor board. The patient's mass at this point had grown into a 9 × 9 cm mass taking up most of the central breast. Partial mastectomy was suggested due to the patient's young age and the large tumor size. Surgical removal of the mass and the postoperative course were uncomplicated. Pathology of the surgical specimen confirmed the diagnosis of PASH.

Discussion

The vast majority of patients who develop the tumorous form of PASH are either premenopausal or actively receiving some form of hormonal therapy (ie, hormone replacement therapy, oral contraceptives), suggesting that tumor development is regulated by sex hormones [3,11]. This is further supported by the finding of progesterone, estrogen, and androgen receptor positivity on immunohistochemical testing. Although PASH occurs predominantly in females, 7 cases have also been reported in the male population [4–10]. Generally, males who develop this condition have some extent of underlying gynecomastia, indicating that increased breast tissue and hormonal imbalance are important risk factors for disease progression [8].

The presentation of PASH tumors varies widely among patients. In a study of 57 patients with imaging evidence of PASH tumor, 44% initially presented with a palpable mass that prompted further mammographic evaluation. Conversely, in 53% of patients, the first sign of abnormality was revealed on screening mammography as a round circumscribed mass, with no preceding clinical correlation [3].

On gross examination, PASH may resemble a fibroadenoma. Similar to the clinical and imaging features of fibroadenoma, a PASH tumor is described as a single, discrete, well-demarcated mass that ranges from solid to elastic in consistency. The rate of growth and the size of PASH vary widely from one case to another, ranging from microscopic foci to lesions that measure up to 18 cm in diameter [7]. Histologically, a PASH tumor may resemble the histological presentation of a low-grade angiosarcoma or a phyllodes tumor, largely due to the slit-like spaces lined by outstretched spindle cells through the acellular mammary stroma [12,13]. PASH can be differentiated from these mimicking conditions via immunohistochemistry of the spindle cells, which are characteristically vimentin and CD34 positive and factor VIII negative [13]. Furthermore, unlike the vascular channels filled with red blood cells in angiosarcoma, the slit-like spaces in PASH do not contain erythrocytes, hence the term “pseudoangiomatous” [13].

Features of PASH may be viewed on various imaging modalities, including mammography, sonography, and magnetic resonance imaging (MRI). The most classic mammographic presentation of the tumor is a noncalcified, round, circumscribed mass. Less frequently, PASH may present as a focal asymmetry on mammography. In up to 22% of patients, PASH may present with no imaging findings despite the presence of a histopathologic diagnosis [12,14–16]. Ultrasound findings are generally nonspecific, revealing a round, circumscribed, and hypoechoic mass. Rarely, the mass may exhibit more worrisome features such as increased echogenicity, het-

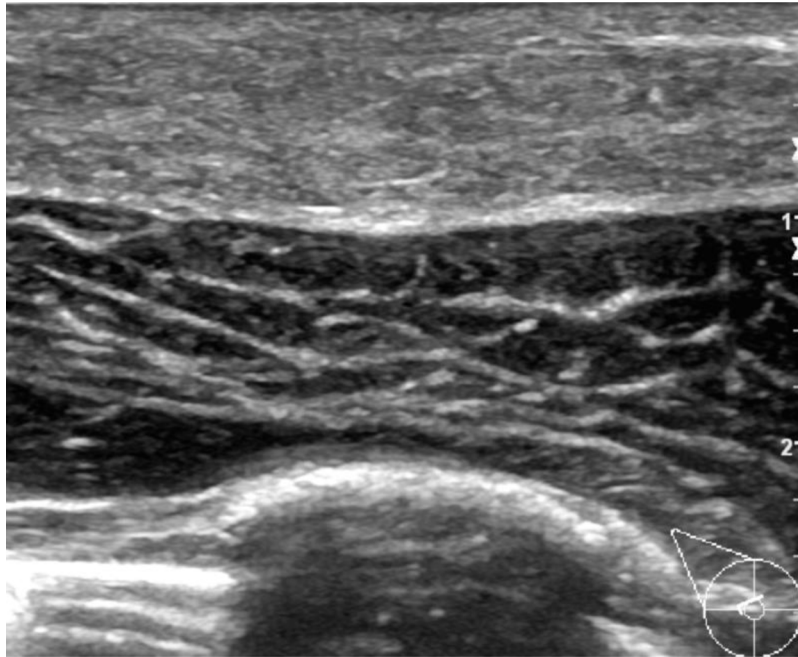


Fig. 2 – Ultrasound of the subareolar region of the right breast in the radial scanning plane, showing increased breast tissue most prominent in the 9:00-12:00 subareolar region.

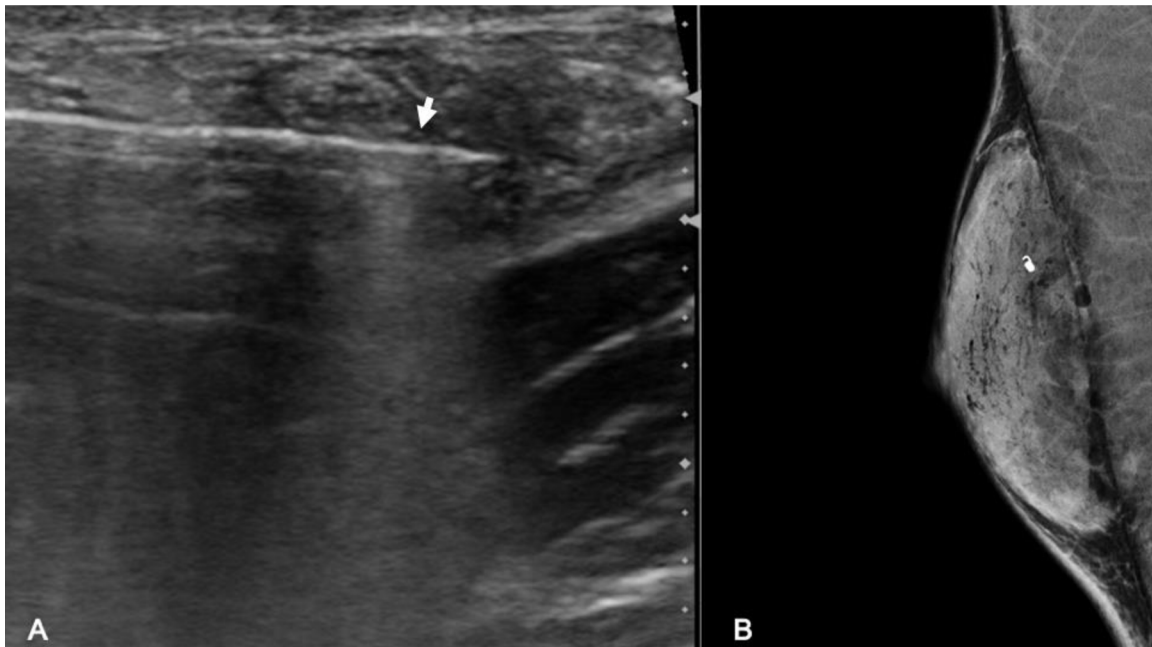


Fig. 3 – Biopsy: (A) Ultrasound-guided biopsy with a 16-gauge Bard automated core biopsy needle (white arrow); and (B) postclip mammogram in the lateromedial projection showing a postbiopsy coil clip.

erogeneous echotexture or irregularly shaped borders [12,16]. On MRI, PASH can have a wide range of presentations, with the most common imaging finding being an area of nonmass enhancement with persistent kinetics (59%). Other less common appearances on MRI include masses with persistent kinetic features or washout (28%), or foci (13%) [12,17].

Conclusion

PASH is a common benign disease process that may rarely result in the formation of a solid tumor of varying sizes. Although primarily affecting premenopausal women or those

on hormonal therapy, cases have also been reported in men with gynecomastia. Careful assessment is required in order to differentiate PASH on imaging from fibroadenoma and histologically from angiosarcoma or phyllodes tumor. Although somewhat nonspecific, the imaging findings of PASH on mammography, sonography, and MRI can serve as important clues in diagnosis.

Patient consent

Informed consent was obtained from the patient involved in this case.

REFERENCES

- [1] Vuitch MF, Rosen PP, Erlandson RA. Pseudoangiomatous hyperplasia of mammary stroma. *Hum Pathol* 1986;17(2):185–91. doi:10.1016/s0046-8177(86)80292-1.
- [2] Bowman E, Oprea G, Okoli J, et al. Pseudoangiomatous stromal hyperplasia (PASH) of the breast: a series of 24 patients. *Breast J* 2012;18(3):242–7. doi:10.1111/j.1524-4741.2012.01230.
- [3] Jones KN, Glazebrook KN, Reynolds C. Pseudoangiomatous stromal hyperplasia: imaging findings with pathologic and clinical correlation. *AJR Am J Roentgenol* 2010;195(4):1036–42. doi:10.2214/AJR.09.3284.
- [4] Vega RM, Pechman D, Ergonul B, Gomez C, Moller MG. Bilateral pseudoangiomatous stromal hyperplasia tumors in axillary male gynecomastia: report of a case. *Surg Today* 2015;45(1):105–9. doi:10.1007/s00595-014-0892-6.
- [5] Maciolek LM, Harmon TS, He J, Sadruddin S, Nguyen QD. Pseudoangiomatous stromal hyperplasia of the breast: a rare finding in a male patient. *Cureus* 2019;11(6):e4923 Published 2019 Jun 17. doi:10.7759/cureus.4923.
- [6] Komforti MK, Fineberg S, Koenigsberg T, Harmon BE. Unilateral pseudoangiomatous stromal hyperplasia in a transgender (male to female) woman on estrogen therapy. *Breast J* 2020;26(2):265–6. doi:10.1111/tbj.13547.
- [7] Jonckheere J, Vanhoeij M, Garkalne I, Antic M, Schiettecatte A, de Mey J. A rare cause of unilateral breast swelling in a male infant caused by fibrous hamartoma of infancy combined with pseudoangiomatous stromal hyperplasia. *Radiol Case Rep* 2019;15(3):234–6. doi:10.1016/j.radcr.2019.11.015.
- [8] Mizutou A, Nakashima K, Moriya T. Large pseudoangiomatous stromal hyperplasia complicated with gynecomastia and lobular differentiation in a male breast. *Springerplus* 2015;4:282. doi:10.1186/s40064-015-1083-7.
- [9] Val-Bernal, Celeiro-Muñoz C, Linares E, Gallardo E, García-Somacarrera E. Aberrant axillary breast tissue with pseudoangiomatous stromal hyperplasia in a man. *Cesk Patol* 2018;54(3):143–6.
- [10] Shehata BM, Fishman I, Collings MH, et al. Pseudoangiomatous stromal hyperplasia of the breast in pediatric patients: an underrecognized entity. *Pediatr Dev Pathol* 2009;12(6):450–4. doi:10.2350/08-09-0528.1.
- [11] Ferreira M, Albarracin CT, Resetkova E. Pseudoangiomatous stromal hyperplasia tumor: a clinical, radiologic and pathologic study of 26 cases. *Mod Pathol* 2008;21(2):201–7. doi:10.1038/modpathol.3801003.
- [12] Drinka EK, Bargaje A, Erşahin ÇH, et al. Pseudoangiomatous stromal hyperplasia (PASH) of the breast: a clinicopathological study of 79 cases. *Int J Surg Pathol* 2012;20(1):54–8. doi:10.1177/1066896911418643.
- [13] Raj SD, Sahani VG, Adrada BE, et al. Pseudoangiomatous stromal hyperplasia of the breast: multimodality review with pathologic correlation. *Curr Probl Diagn Radiol* 2017;46(2):130–5. doi:10.1067/j.cpradiol.2016.01.005.
- [14] Holloway TL, Jatoi I. Tumorous PASH presenting as rapid unilateral breast enlargement. *Mayo Clin Proc* 2013;88(7):e75. doi:10.1016/j.mayocp.2013.02.014.
- [15] Mai C, Rombaut B, Hertveldt K, Claikens B, Van Wettere P. Diffuse pseudoangiomatous stromal hyperplasia of the breast: a case report and a review of the radiological characteristics. *JBR-BTR* 2014;97(2):81–3. doi:10.5334/jbr-btr.41.
- [16] Celliers L, Wong DD, Bourke A. Pseudoangiomatous stromal hyperplasia: a study of the mammographic and sonographic features. *Clin Radiol* 2010;65(2):145–9. doi:10.1016/j.crad.2009.10.003.
- [17] Nia ES, Adrada BE, Whitman GJ, et al. MRI features of pseudoangiomatous stromal hyperplasia with histopathological correlation. *Breast J* 2021;27(3):242–7. doi:10.1111/tbj.14154.