

Accepted: 2018.08.13 Published: 2018.11.30

e-ISSN 1941-5923 © Am J Case Rep. 2018: 19: 1425-1429 DOI: 10.12659/AJCR.911842

# Hemangioma of a Male Breast: Case Report and **Review of the Literature**

Authors' Contribution-Study Design A Data Collection B Statistical Analysis C Data Interpretation D Manuscript Preparation E Literature Search F Funds Collection G

ABCDEF 1,2 Arthy Yoga

ABCDEF 3,4 Kirill A. Lyapichev BCDEF 3,5 Donghwa Baek

ABCDEF 3,6 Carmen R. Gomez-Fernandez

- 1 Department of Breast Surgery, Texas Oncology, Houston, TX, U.S.A.
- 2 Division of Breast Surgery, Department of Surgery, Bethesda Health System, Boynton Beach FL USA
- 3 Department of Pathology and Laboratory Medicine, University of Miami, Miami, FL. U.S.A.
- 4 Department of Hematopathology, The University of Texas M.D. Anderson Cancer Center, Houston, TX, U.S.A.
- 5 Department of Pathology and Genomic Medicine, Methodist Hospital, Houston,
- 6 Sylvester Comprehensive Cancer Center, Miami, FL, U.S.A.

Corresponding Author: Conflict of interest: Arthy Yoga, e-mail: arthyyoga@gmail.com

None declared

**Patient:** Male, 84

**Final Diagnosis: Breast hemangioma Symptoms:** Feeling breast mass

**Medication: Clinical Procedure:** 

**Case Report:** 

Specialty: Surgery

**Objective:** Rare disease

**Background:** Male breast hemangioma is a rare benign neoplasm that is usually excised for definitive diagnosis.

> In our case report, we present a male octogenarian with multiple comorbidities who presented with a large palpable mass in his right breast. The diagnostic imaging studies were suggestive of a benign tumor, with a BI-RADS (Breast Imaging Reporting and Data System) score of 3. Subsequent core needle biopsies were diagnostic of benign hemangioma. The patient was managed with observation due to his comorbidities. Benign vascular tumors in the male breast are exceptionally rare, and in our review of the literature we found only 14 previously published cases. Historically, fine needle aspiration has been found to be unreliable in making a

definitive diagnosis and surgical excision has been the standard treatment.

Conclusions: Recent studies and our case report indicate that core needle biopsy may be a reliable diagnostic tool and

observation is a possible option for hemangiomas in male patients who cannot undergo surgery.

MeSH Keywords: Breast Neoplasms, Male • Hemangioma • Patient Care Management

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/911842

1523

2 1

1 2 2

39



# **Background**

Hemangioma is a benign tumor consisting of a mass of blood vessels [1]. It is a common neoplasm that can be found in different parts of the body. There are multiple types of hemangiomas identified based on different histological appearance, age, presentation, and location: capillary (juvenile and lobular), cavernous, intramuscular, spindle cell, epithelioid, and cardiac hemangioma which is often associated with Kasabach-Merritt syndrome.

Breast hemangiomas are benign vascular lesions that occur in breast parenchyma [2]. Although breast hemangiomas are well described in female patients [3], male breast hemangiomas are exceptionally rare, and only 14 cases have been previously reported [4-17]. In this case report, we describe an unusual breast hemangioma in an octogenarian male patient.

### **Case Report**

An 84-year-old male with an extensive medical history presented to our institution due to a recently identified lump of the right breast. He had no previous history of breast-related problems, radiation treatment, or family history of breast or ovarian cancer. The patient was wheelchair bound and was prone to easy bruising. He was on aspirin for cardiovascular disease. The patient believed the breast lump might be a hematoma caused by trauma.

On clinical examination, he had thin pale skin with a bluish discoloration associated with an underlying mass in the upper outer quadrant of the right breast. The mass was soft, mobile, and not attached to the skin or chest wall. The mammogram study showed a 4.22 cm hyperdense mass with small nodules and peripheral fat (Figure 1). These findings were thought to be related to a breast hamartoma or hematoma. The ultrasound revealed a 3.2×3×1.9 cm oval isoechoic lesion with heterogeneous appearance, smooth margins, and no posterior shadowing. BI-RADS (Breast Imaging Reporting and Data System) score 3 was given. Due to the benign appearance on imaging studies and the multiple comorbidities of the patient, a shortterm follow-up in 3 months was recommended.

Because the lesion appeared to persist and increased in size to 4.9×3.4×1.7 cm on the follow-up ultrasound evaluation, a core needle biopsy was performed.

The gross pathology revealed multiple cores of soft, yellow and tan, fibrofatty tissue, measuring 1.7 cm in length by 0.2 cm in diameter to 0.7 cm in length by 0.2 cm in diameter. On the histological examination, the tumor was composed of a wellcircumscribed proliferation of widely dilated, thin-walled blood vessels lined by flattened endothelial cells without cytologic atypia or mitoses (Figure 2). The diagnosis of cavernous hemangioma without atypia was made.

The pathologic findings were concordant with the imaging and clinical findings. After further discussion of the radiologic and pathologic findings with the patient and his daughter, they did not wish to pursue a surgical excision due to the patient's advanced age, multiple comorbidities, and the benign nature of this lesion. Follow-up was recommended, but the patient was lost to follow-up.

# **Discussion**

Although multiple classifications of benign vascular lesions of different organ systems have been described in the literature, the classification of benign vascular tumors in the breast was first described by Rosen et al. [3,18]. Most breast hemangiomas are microscopically and incidentally found tumors, and they are designated as perilobular hemangiomas. The larger, clinically or mammographically detectable tumors are hemangiomas, which are further classified into cavernous, capillary, complex, or venous types. The noncavernous hemangioma was initially termed "atypical" hemangioma, but as this entity does not predispose to the development of angiosarcoma, this type of hemangioma is now simply designated as "noncavernous" [3,19].

Because of its rarity, the incidence of breast hemangioma was variably reported. Perilobular hemangiomas were present in 1.3% of mastectomies performed due to breast cancer and present in 11% of autopsy specimens [20,21]. One relatively recent study reported 12 hemangioma cases in 1362 surgical specimens (0.8%), only one of which occurred in a male [22]. To the best of our knowledge, the first case report of hemangioma in the male breast was described in "Cancer Research" by Dr. Johnston from University of Maryland in 1936 [15] and, only 10 male breast hemangiomas have been reported in the English literature and available on PubMed search using keywords "male," "breast," and "hemangioma" [4-13,22]. An additional 4 publications were found in Google Scholar, which presented similar cases of male breast hemangiomas [14–17]. The clinicopathologic features of these cases, in addition to our case, are summarized in Table 1.

There are several differences in clinical manifestations between male and female breast hemangiomas. Male tumors tend to be larger. All 15 male hemangiomas, including the case described here, presented with a palpable mass or nodule while only about 40% of female hemangiomas present as a palpable mass. The tumor size in men ranged from 0.7 to 14 cm with a mean diameter of 5.4 cm (versus 1.1 cm in women). Three

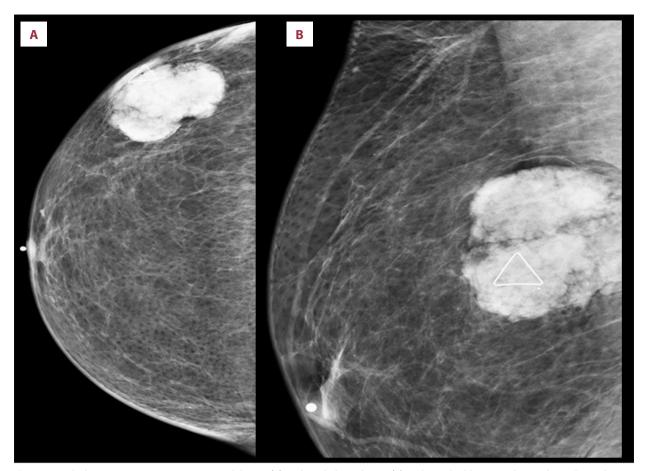


Figure 1. Right breast mammogram. Craniocaudal view (A) and mediolateral view (B) with a palpable triangular marker. A hyperdense mass with small nodules and peripheral fat was identified. Differential diagnoses included hematoma versus hamartoma.

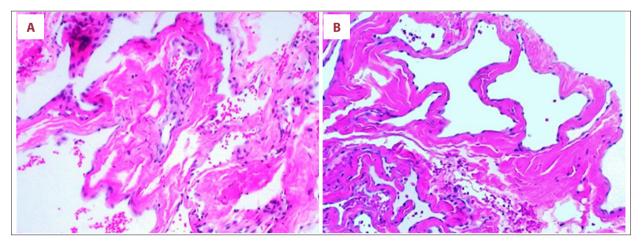


Figure 2. (A, B) Histopathologic features. Well-circumscribed proliferation of widely dilated, thin-walled blood vessels (hematoxylin and eosin; 100×).

male patients experienced breast pain, which might be related to the large tumor size. Another 4 patients had bluish discoloration of the overlying skin. Although the presence of a large vascular tumor in the breast with overlying skin discoloration might be more suggestive of angiosarcoma in female patients,

these features were not necessarily indicative of malignancy in male patients.

Except for the size, the radiologic features in male hemangiomas appeared similar to those that have been described in

**Table 1.** Clinical presentation and histological types of male breast hemangiomas.

Mean age (years)	62.2 (range: 30–84)
Clinical presentation	
Incidental	0/15 (0%)
Mammographic finding*	2/8 (9%)
Mean size (cm)	5.4 (range: 0.7–14)
Overlying skin discoloration	4/15 (27%)
Breast pain	3/15 (20%)
Histopathology	
Perilobular hemangioma	0/15 (0%)
Cavernous hemangioma	13/15 (87%)
Noncavernous hemangioma	2/15 (13%)
References	[4–17]

<sup>\*</sup> Only 8 cases had information about mammographic study.

females [23,24]. Eight case reports described mammographic findings, and most of them showed a well-circumscribed high-density mass with or without lobulation. Three cases demonstrated calcifications, which represented phlebolith formation [4,10,16]. Breast sonography revealed variable features. Some were well-circumscribed oval masses while others were described to be irregular with indistinct borders. The echogenicity of the reported masses was also variable. This heterogeneity might stem from vascular channels, phlebolith formation, and surrounding fatty tissue.

Fine needle aspiration (FNA) was performed in 6 of the male breast hemangioma cases, and biopsies typically yielded no more than a few benign mesenchymal cells with bloody background [4,6,7,10,12,14]. Surgical excision with pathological examination revealed 13 cavernous hemangiomas and 2 capillary hemangiomas. Six case reports did not provide descriptions of the specific subtypes of the hemangiomas, but based on the histological descriptions and figures presented, we categorized 5 cases as cavernous type [7,10,12,15,17], and the other case as capillary type [9]. Most of the cavernous hemangiomas were associated with a gross appearance of a brown to dark red hemorrhagic tumor with spongy consistency. Microscopically, they showed dilated vascular channels lined by flat endothelial cells. None exhibited cellular or nuclear atypia, mitoses, "blood lakes," or necrosis, confirming the benign nature of the tumors. Perilobular hemangiomas were not reported in any of the male patients (Table 1).

The 2 most important differential diagnoses of breast hemangiomas are angiosarcoma and pseudoangiomatous stromal hyperplasia (PASH). Although extremely rare, angiosarcoma has been described in the breast of male patients and should be considered as a plausible differential diagnosis [25–33]. Although large (>3 cm), palpable, intraparenchymal vascular tumors are usually indicative of angiosarcoma in the female breast [24], these features will not help differentiating angiosarcoma from hemangioma in the male breast, mandating histologic diagnosis. Imaging studies might be helpful in larger lesions where magnetic resonance features are more typical but cannot completely replace the histopathological studies due to the difficulty in differentiation from angiosarcomas [34]. Histologic features suggestive of angiosarcoma include infiltrative margins, anastomosing vascular channels, lobular invasion, endothelial tufting, papillary formations, solid and spindle cell foci, mitoses, "blood lakes," and necrosis [18,30,35].

PASH should also be considered in the differential diagnosis. It is relatively common and found in about half of male breast specimens, 24% to 98% of which are associated with gynecomastia [36,37]. Its mammographic and ultrasonographic appearance is nonspecific, and pathologic examination is essential [38]. Augmented collagenous stroma and the absence of a true endothelial lining differentiates it from true vascular channels.

Historically, concerns over diagnostic accuracy with FNA and close morphologic overlaps between hemangioma and angiosarcoma can lead to surgical resection in male patients with vascular tumors. However, with clinical, pathological, and radiological concordances, core needle biopsy is a reliable diagnostic tool for hemangiomas. One recent study stated that breast hemangioma diagnosed by core needle biopsy does not require further excision [13], although this study contained only tumors less than 2 cm and only 1 male patient. Even though excision of a large lesion from a male breast is still supported, observation is an option in patients who cannot undergo surgery. Because our patient was not a good surgical candidate, multiple biopsy cores were taken to have a representative sample of the lesion to secure the diagnosis of hemangioma. Ki-67 immunostaining could be helpful in atypical vascular lesions [39], where Ki-67 index of more than 20% is highly suggestive of angiosarcoma with 90% sensitivity and 95% specificity. However, our case showed obvious benign histology of the hemangioma and Ki-67 staining was no obtained.

## **Conclusions**

Male breast hemangiomas are exceptionally rare. Although their radiological appearance does not differ from that in female, they tend to be larger and more likely to present with a palpable nodule. They should be differentiated from angiosarcoma and PASH. With clinical, radiologic, and pathologic concordance, core needle biopsy is a reliable diagnostic tool. In patients who cannot undergo surgery, observation is an option for this benign lesion.

#### **Conflict of interest**

None.

#### **References:**

- 1. Mosby's Dictionary of Medicine, Nursing & Health Professions Seventh edition Mosby Mosby's Dictionary of Medicine, Nursing & Health Professions Seventh edition 2272 Mosby 9780723433934 0723433933. Nurs Stand, 2006; 20(22): 36
- Hicks DG, Lester SC: Diagnostic pathology. Breast. Second edition. ed. Philadelphia, PA: Elsevier, 2016; xiii, 673, xxi
- 3. Jozefczyk MA, Rosen PP: Vascular tumors of the breast. II. Perilobular hemangiomas and hemangiomas. Am J Surg Pathol, 1985; 9(7): 491–503
- Carreira C, Romero C, Rodriguez R et al: A cavernous haemangioma of breast in male: Radiological-pathological correlation. Eur Radiol, 2001; 11(2): 292–94
- 5. Franco RL, de Moraes Schenka NG, Schenka AA, Alvarenga M: Cavernous hemangioma of the male breast. Breast J, 2005; 11(6): 511-12
- 6. Kinoshita S, Kyoda S, Tsuboi K et al: Huge cavernous hemangioma arising in a male breast. Breast Cancer, 2005; 12(3): 231–33
- 7. Kondi-Pafitis A, Kairi-Vassilatou E, Grapsa D et al: A large benign vascular neoplasm of the male breast. A case report and review of the literature. Eur J Gynaecol Oncol, 2005; 26(4): 454–56
- Leddy R, Cluver A: Mammographic and sonographic characteristics of a cavernous hemangioma in a male patient. J Ultrasound Med, 2010; 29(4): 645–59
- Schwartz IS, Marchevsky A: Hemangioma of male breast. Am J Surg Pathol, 1987; 11(9): 739
- Shi AA, Georgian-Smith D, Cornell LD et al: Radiological reasoning: Male breast mass with calcifications. Am J Roentgenol, 2005; 185(6 Suppl.): \$205-10
- Shousha S, Theodorou NA, Bull TB: Cavernous haemangioma of breast in a man with contralateral gynaecomastia and a family history of breast carcinoma. Histopathology, 1988; 13(2): 221–23
- Vourtsi A, Zervoudis S, Pafiti A, Athanasiadis S: Male breast hemangioma

   a rare entity: A case report and review of the literature. Breast J, 2006;
   12(3): 260–62
- Sebastiano C, Gennaro L, Brogi E et al: Benign vascular lesions of the breast diagnosed by core needle biopsy do not require excision. Histopathology, 2017: 71(5): 795–804
- 14. Chidambaram A, Shetty MK, Atula A: Cavernous haemangioma of male breast. Int J Res Med Sci, 2013; 1(4): 571
- 15. Johnston CC: Hemangioma of the male breast: A Case Report. Am J Cancer, 1936; 27(2): 341–43
- 16. Alikhassi A, Omranipour R, Alikhassy Z et al: Retropectoral hemangioma in a male patient: Report of a case. Arch Breast Cancer, 2015; 2(2): 69–72
- 17. Mojtahedi F, Bulut T, Oosterhof-Berktas R, Bezooijen R: Hemangioma in the male breast. J Med Cases, 2016; 7(8): 323–25
- Rosen PP, Hoda SA, Brogi E, Koerner FC: Rosen's breast pathology. Fourth edition. ed. Philadelphia: Wolters Kluwer Health/Lippincott Williams & Wilkins, 2014; 1379
- Hoda SA, Cranor ML, Rosen PP: Hemangiomas of the breast with atypical histological features. Further analysis of histological subtypes confirming their benign character. Am J Surg Pathol, 1992; 16(6): 553–60

- 20. Rosen PP, Ridolfi RL: The perilobular hemangioma. A benign microscopic vascular lesion of the breast. Am J Clin Pathol, 1977; 68(1): 21–23
- Lesueur GC, Brown RW, Bhathal PS: Incidence of perilobular hemangioma in the female breast. Arch Pathol Lab Med, 1983; 107(6): 308–10
- Kondi-Pafiti A, Dellaportas D, Myoteri D et al: Rare non-epithelial primary breast neoplasms: A ten-year experience at a Greek University Hospital. J BUON. 2013: 18(1): 70–76
- Mesurolle B, Sygal V, Lalonde L et al: Sonographic and mammographic appearances of breast hemangioma. Am J Roentgenol, 2008; 191(1): W17–22
- Glazebrook KN, Morton MJ, Reynolds C: Vascular tumors of the breast: Mammographic, sonographic, and MRI appearances. Am J Roentgenol, 2005; 184(1): 331–38
- Fraga-Guedes C, Gobbi H, Mastropasqua MG et al: Primary and secondary angiosarcomas of the breast: A single institution experience. Breast Cancer Res Treat, 2012; 132(3): 1081–88
- Granier G, Lemoine MC, Mares P et al: [Primary angiosarcoma of the male breast]. Ann Pathol, 2005; 25(3): 235–39 [in French]
- Kamat L, Rosa M, Weinfurtner R et al: Primary breast angiosarcoma in a male. Breast J, 2015; 21(5): 545–48
- Mansouri H, Jalil A, Chouhou L et al: A rare case of angiosarcoma of the breast in a man: Case report. Eur J Gynaecol Oncol, 2000; 21(6): 603–4
- Rainwater LM, Martin JK Jr., Gaffey TA, van Heerden JA: Angiosarcoma of the breast. Arch Surg, 1986; 121(6): 669–72
- Rosen PP, Kimmel M, Ernsberger D: Mammary angiosarcoma. The prognostic significance of tumor differentiation. Cancer, 1988; 62(10): 2145–51
- Wang L, Lao IW, Yu L, Wang J: Clinicopathological features and prognostic factors in angiosarcoma: A retrospective analysis of 200 patients from a single Chinese medical institute. Oncol Lett, 2017; 14(5): 5370–78
- 32. Wang ZS, Zhan N, Xiong CL, Li H: Primary epithelioid angiosarcoma of the male breast: Report of a case. Surg Today, 2007; 37(9): 782–86
- Yadav RV, Sahariah S, Mittal VK, Banerjee AK: Angiosarcoma of the male breast. Int Surg, 1976; 61(9): 463–64
- Ameen R, Mandalia U, Marr AA, McKensie P: Breast hemangioma: MR appearance with histopathological correlation. J Clin Imaging Sci, 2012; 2: 53
- Donnell RM, Rosen PP, Lieberman PH et al: Angiosarcoma and other vascular tumors of the breast. Am J Surg Pathol, 1981; 5(7): 629–42
- 36. Badve S, Sloane JP: Pseudoangiomatous hyperplasia of male breast. Histopathology, 1995; 26(5): 463–66
- Milanezi MF, Saggioro FP, Zanati SG et al: Pseudoangiomatous hyperplasia of mammary stroma associated with gynaecomastia. J Clin Pathol, 1998; 51(3): 204–6
- Celliers L, Wong DD, Bourke A: Pseudoangiomatous stromal hyperplasia: A study of the mammographic and sonographic features. Clin Radiol, 2010; 65(2): 145–49
- Shin SJ, Lesser M, Rosen PP: Hemangiomas and angiosarcomas of the breast: Diagnostic utility of cell cycle markers with emphasis on Ki-67. Arch Pathol Lab Med, 2007; 131(4): 538–44