

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
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Patient: Male, 84
Final Diagnosis: Breast hemangioma
Symptoms: Feeling breast mass
Medication: —
Clinical Procedure: —
Specialty: Surgery

Objective: Rare disease

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

Case Report: 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

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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 short-term 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 well-circumscribed 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 "non-cavernous" [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

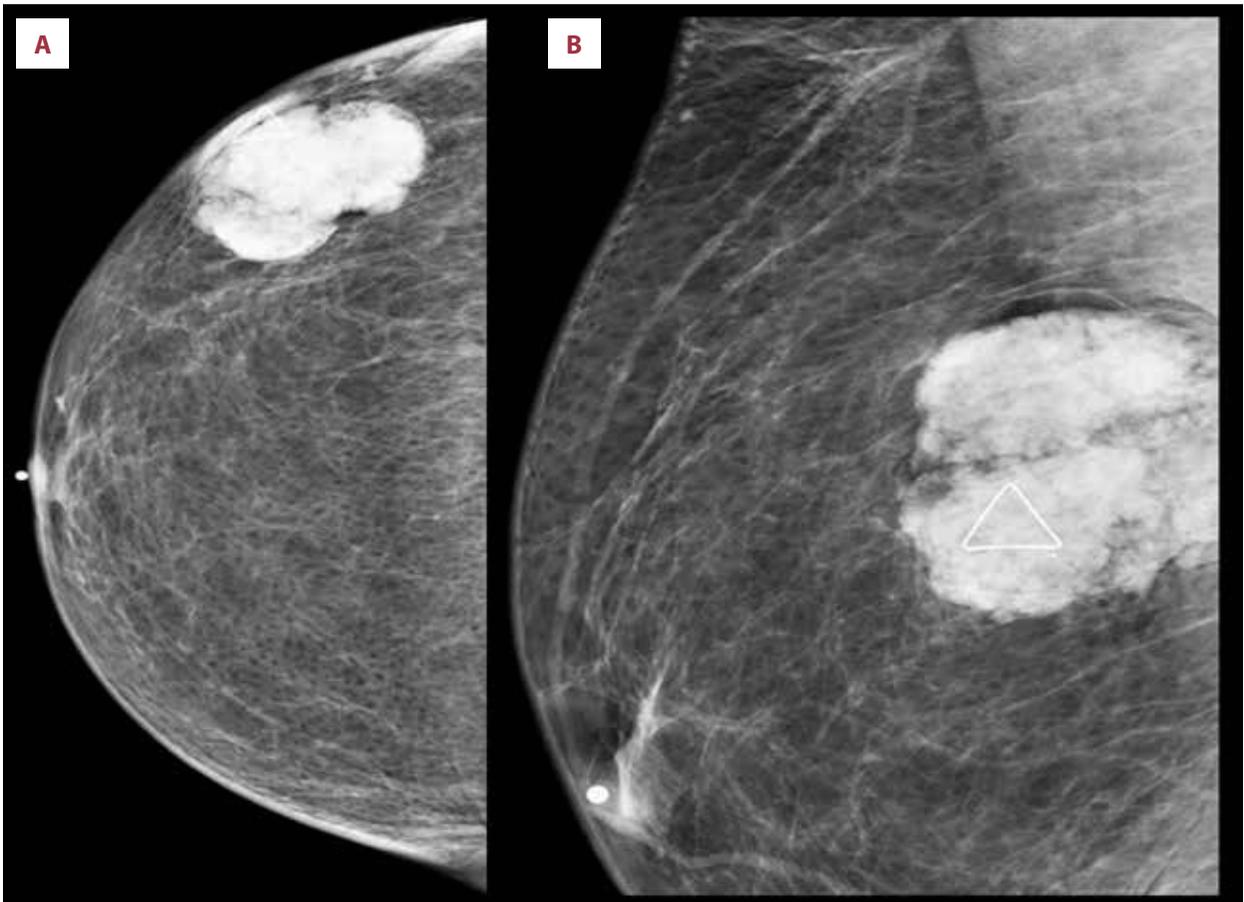


Figure 1. Right breast mammogram. Craniocaudal view (A) and mediolateral view (B) with a palpable triangular marker. A hyperdense mass with small nodules and peripheral fat was identified. Differential diagnoses included hematoma versus hamartoma.

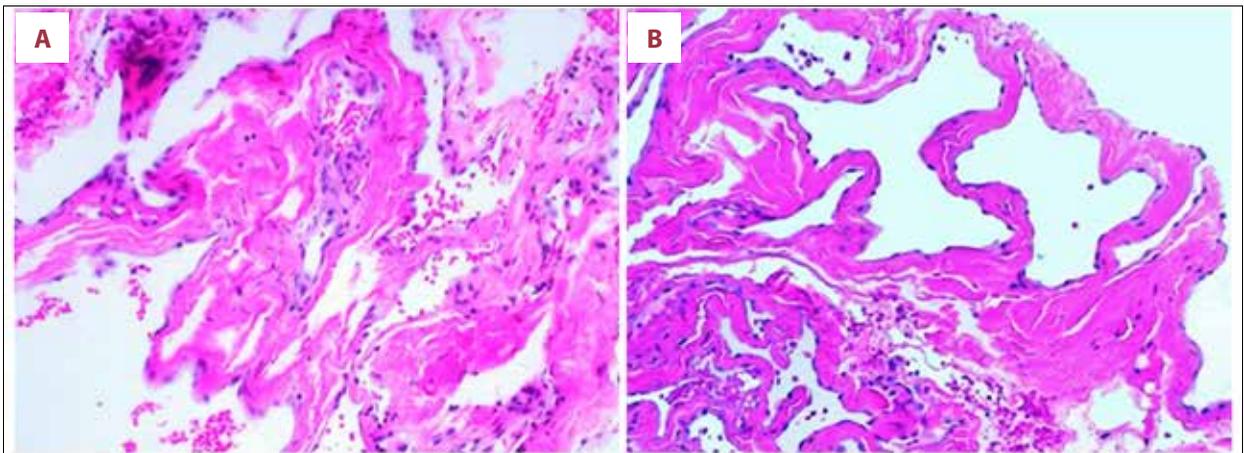


Figure 2. (A, B) Histopathologic features. Well-circumscribed proliferation of widely dilated, thin-walled blood vessels (hematoxylin and eosin; 100 \times).

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]

* 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 not 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.

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