# A large benign vascular neoplasm of the male breast. A case report and review of the literature

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#### **Summary**

Breast hemangiomas are extremely rare neoplasms in the male population. We report a case of a 77-year old man with a breast hemangioma which was detected in physical examination as a small nodule ten years after a chest injury. The final histological diagnosis was hemangioma of the breast, 6 cm in the largest diameter. To our knowledge, this is the largest benign vascular breast neoplasm in a male patient reported up to date. The mammographic and pathologic findings in this case are presented. The rarity of the lesion and its differential diagnosis from angiosarcoma are discussed while the problems encountered in the correct diagnosis and classification of this tumor are also presented. The need for extreme caution in the interpretation of the histological characteristics of all palpable vascular tumors of the breast is emphasized.

Key words: Vascular tumors; Breast; Angiosarcoma; Hemangioma; Mammography.

#### Introduction

Vascular neoplasms of the breast are rare lesions, comprising 0.5% of all mammary gland neoplasms [1]. The majority, especially when they are palpable tumors, are angiosarcomas [2]. Benign vascular neoplasms rarely occur in the female and only exceptionally in the male breast [3]. Their histopathologic classification has only recently been established by Rosen who was also the first to report that symptom-producing benign vascular tumors of the breast do exist [4].

Ten cases of vascular tumors of the breast, among 1,250 breast biopsies and specimens, were examined in our laboratory in the last decade (1992-2001). Nine patients were female. A case of benign vascular tumor of the breast in a male patient is presented because of its rarity and large size which makes it the largest benign vascular breast neoplasm in a male patient reported up to date. The problems encountered in the correct diagnosis and classification of this tumor are presented.

#### Case report

A 77-year-old man was examined in our Hospital Breast Unit because he had observed a slowly growing mass in his left breast. The mass was first detected as a small nodule ten years before after an injury of the chest due to a car accident. The nodule enlarged slowly over the years and on physical examination showed a large painless mass, located under the areolar region. It was a well defined mass, non tender, mobile and not attached to the chest wall. The overlying skin was bluish-tinted and the lesion simulated grossly a large bruise. (Figure 1). Mammography demonstrated a well-circumscribed breast tumor of soft tissue origin but of indeterminate nature (Figure 2). Fine needle aspiration biopsy showed a field of blood with a few connective tissue cells and a few inflammatory cells and was considered not diagnostic.

Surgical resection of the tumor was performed. The specimen measured 12 x 10 x 3.5 cm and was covered by the overlying skin measuring 11 x 12 cm. Gross examination showed a welldefined, dark reddish and hemorrhagic tumor with an elastic and spongy consistency measuring 6 x 5 x 4 cm. Multiple sections of the lesion were examined after a routine preparation of formalinfixed, paraffin-embedded and hematoxylin-eosin stained tissue. Additional sections for immunohistochemistry were obtained and processed by a streptavidin-biotin method for the investigation of estrogen receptor (ER) clone 1F6, Novocastra) and progesterone (PgR) hormone receptors (clone 1D5, Novocastra).

The histological examination showed multiple vascular spaces covered by a layer of endothelial cells without any remarkable cellular or nuclear atypia. Between these vascular spaces there were fibrous walls with sparse lymphocytic infiltrations (Figure 3).

Rare foci of endothelial papillary hyperplasia were observed but without any mitotic activity or remarkable nuclear hyperchromasia. The neoplasm was circumscribed and no evidence of invasion of the surrounding breast tissue was observed. The immunohistochemical investigation of hormone receptors in the endothelial cells was negative.

The final diagnosis was hemangioma of the breast, totally excised.

#### Discussion

Rosen has classified benign vascular neoplasms of the breast into 1) parenchymal hemangiomas and 2) subcutaneous, non parenchymal or soft tissue hemangiomas [4]. Parenchymal hemangiomas are subclassified into perilobular, capillary-cavernous, or mixed-type hemangiomas, and as tumors of venous type. The subcutaneous

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Fig. 1

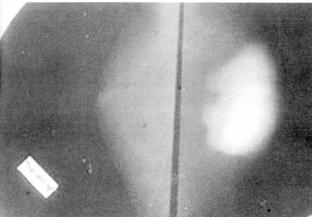


Fig. 2

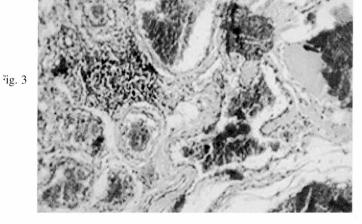


Figure 1. — Macroscopic view of breast with hemangioma.

Figure 2. — Mammography shows a large lesion, probably a soft tissue mass of indeterminate nature.

Figure 3. — Histological section of the male breast hemangioma, showing large cystic spaces.

or soft tissue hemangiomas are subdivided into hemangiomas, angiolipomas and aneurysmatic lesions [4].

According to recent studies, breast hemangiomas are usually detected by mammography or present as incidental microscopic findings in breast tissue specimens of adult women operated on because of benign as well as malignant epithelial breast diseases [5]. They measure between 0.2 and 2.5 cm, only rarely exceeding 2.0 cm in diameter [4].

In male patients, vascular tumors of the breast are extremely rare tumors. In the literature there are only five cases of hemangiomas reported up to date [6]. We believe that this low incidence could be partly attributed to low detection rates. Men are not routinely subjected to mammography, and, according to Rosen, a substantial number of hemangiomas do not produce symptoms and are of small size [4]. Therefore it is extremely possible that many small, benign and symptom-free tumors may not be ever diagnosed in the lifetime of an unknown percentage of the male population. This view is supported by the significantly higher frequency of perilobular hemangiomas found in autopsy material (11%) in comparison with the relative percentages of the same tumors detected in mastectomies for carcinoma (1.3%) or biopsies for benign breast lesions (4.5%) [1, 7].

Because of the occurrence of a subset of breast vascular tumors in young women, where 6-12% of the angiosarcomas reported occurred during pregnancy [8, 9], a hor-

monal factor may be implicated in the histogenesis of these tumors, but in our study the immunohistochemical investigation of hormone receptors was negative.

In our case, the patient reported breast injury with concommitant development of a small tumor which gradually increased in size. The role of chest injury as a precursor of a vascular lesion must be further examined. The lesion was diagnosed as hemangioma because of the absence of malignant histologic characteristics, such as atypical endothelial cells with hyperchromatic nuclei, solid tumor areas, necrosis and mitotic activity. The tumor was well circumscribed and no evidence of invasion of the surrounding breast tissue was observed. Due to its large size, it occupied the whole breast parenchyma and subcutaneous tissue and its exact classification as a soft tissue or as a breast tissue tumor was not possible.

According to most authors, any clinically palpable vascular tumor of the breast should be considered as malignant, unless proved otherwise [10]. However, it must be noted that hemangiomas may occasionally, as in our case, not only be clinically palpable, but also reach a size even greater than the mean size of angiosarcomas (5 cm). Therefore, extreme caution in the microscopic examination of all clinically palpable tumors is always needed. The pathologist should keep in mind that only the histology of the tumor, and not its size, can safely distinguish angiosarcomas from hemangiomas.

The therapy of choice for benign breast tumors is com-

plete excision of the tumor. Our patient remains well five years after surgery without any sign of recurrence of the tumor. The complete removal of the tumor is necessary, not only because it is possible that hemangiomas are precursors to angiosarcomas, but also because a thorough microscopic study of the entire lesion is required for the establishment of the diagnosis [1].

#### Conclusion

Hemangiomas of the male breast are extremely rare neoplasms, usually discovered in breast specimens as incidental microscopic findings. However, occasionally they may be clinically palpable or even reach a size greater than the mean size of angiosarcomas. Lack of malignant histologic characteristics, such as nuclear atypia, necrosis and mitotic activity is essential in order to differentiate hemangiomas from angiosarcomas, while the presence of endothelial papillary hyperplasia may pose diagnostic difficulties. However, in all cases of benign vascular neoplasms of the breast complete excision of the lesion is recommended.

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