






CASUISTIC PAPER

Elżbieta Ostańska ^{1(ABCGF)}, David Aebisher  ^{2(DFG)}, Ewa Kaznowska  ^{1,3(DFG)},
Dorota Bartusik-Aebisher  ^{4(DFG)}

Primary breast angiosarcoma – a case report

¹ Clinical Department of Pathomorphology, Clinical Hospital No. 1 in Rzeszow, Rzeszow, Poland

² Department of Biochemistry and General Chemistry, Institute of Medical Sciences,
Medical College of Rzeszow University, Rzeszow, Poland

³ Department of Pathomorphology, Institute of Medical Sciences, Medical College of Rzeszow University,
Rzeszow, Poland

⁴ Department of Photomedicine and Physical Chemistry, Institute of Medical Sciences,
Medical College of Rzeszow University, Rzeszow, Poland

ABSTRACT

Introduction. Angiosarcoma is a rare breast cancer that can be primary or secondary after surgery or after breast cancer radiotherapy. It is important that breast angiosarcoma belongs to tumors with a non-specific clinical and radiological picture.

Aim. The study of the biopsies contained aggressive vasomotor hyperplasia.

Description of the case. The presented case concerns the primary angiosarcoma of the right breast in a 56-year-old woman who had never had a surgical procedure before, nor radiotherapy in the area of the breast.

Conclusion. Histopathological examination supported by immunohistochemistry is a reliable and indispensable diagnostic element in the diagnosis of vascular sarcoma.

Keywords. breast biopsy, histopathological examination, immunohistochemistry

Introduction

Breast angiosarcoma occurs in two variants as secondary cancer, associated with lymphatic edema after breast cancer surgery (Stewart-Treves syndrome) or after radiotherapy for breast cancer, usually about 3-12 years after irradiation. The dose of radiation does not correlate with the development of cancer. This type of tumor usually occurs with inflammatory changes in the skin of the breast. The second type, primary, occurs without previous diseases of the breast gland as a painless fast-grow-

ing tumor, without accompanying changes on the breast skin.¹⁻³

Aim

Therapy of a 56-year-old woman with the presence of an irregular 50 mm size change in the right breast.

Description of the case

In a 56-year-old woman who had no breast surgery or radiation therapy in the past, during the periodic mam-

Corresponding author: Elżbieta Ostańska, e-mail: eostanska@wp.pl

Participation of co-authors: A – Author of the concept and objectives of paper; B – collection of data; C – implementation of research; D – elaborate, analysis and interpretation of data; E – statistical analysis; F – preparation of a manuscript; G – working out the literature; H – obtaining funds

Received: 7.03.2020 | Accepted: 13.05.2020

Publication date: December 2020

mographic examination, asymmetric compaction in the right breast was revealed (BIRADS 4). The patient was referred for further diagnosis to the oncological clinic in the future. The completed ultrasound examination confirmed the presence of an irregular 50 mm size change. A thick-foot biocidal biosynthesis was performed to collect tissue from the change to histopathological examination. The obtained material was fixed in buffered 10% formalin, and tissue and H + E staining was performed. At the same time, in order to accurately assess the extent and size of the pathological change, a breast magnetic resonance (MRI) was performed, confirming the presence of a tumor size 55x50x46cm (BIRADS 6).

Results

In the microscopic examination, the biopsies contained aggressive vasomotor hyperplasia (Figure 1), which was confirmed by immunohistochemistry (positive reactions of endothelial markers CD31 and CD34) (Figure 2). The whole picture was complemented by a high proliferative marker Ki67 (Figure 3). Vascular sarcoma of intermediate grade (G2) was diagnosed. The patient was transferred for further treatment to the reference center.

Discussion

Angiosarcoma - a malignant tumor of vascular origin 1. Angiosarcoma of the breast is a rare malignant tumor and constitutes about 0.04% of the primary malignant tumors of this organ.¹⁻⁵

Angiosarcoma in the breast may be secondary to surgery or radiotherapy usually after 3-12 years 1. This type occurs in older women (average age 67). The primary angiosarcoma occurs in younger women with an average age of around 40 years. A major role in the development of primary angiosarcoma is attributed to exposure to vinyl chloride, arsenic, thorium oxide, local injuries and inflammatory changes induced by foreign bodies.²⁻⁷ Cancer is clinically present as a non-painful tumor. Sometimes with symptoms of tissue pulsation and redness of the skin. It usually does not cause the nipple to contract. Very rarely gives metastases to the lymph nodes. Usually metastasis is present in the lungs, second breast, or in the bones.¹⁻⁸ The tumor may be visualized in mammography as a homogeneous mass. In an ultrasound scan, the picture may be heterogeneous: hypoechoic or hyperechoic. The Doppler ultrasound examination reveals a rich tumor vascularization. In the MRI examination, the extent of the tumor can be clearly visualized, especially in the secondary type after radiotherapy.¹⁻⁴ The tumor usually located deeply in the breast parenchyma. The size of the tumor usually exceeds 2 cm.¹⁻⁷ In the macroscopic image, the tumor has the appearance of a spongy congested structure or solid fibrous mass in the case of a low-differential tumor.⁴ A certain diagnosis of angiosarcoma can be determined

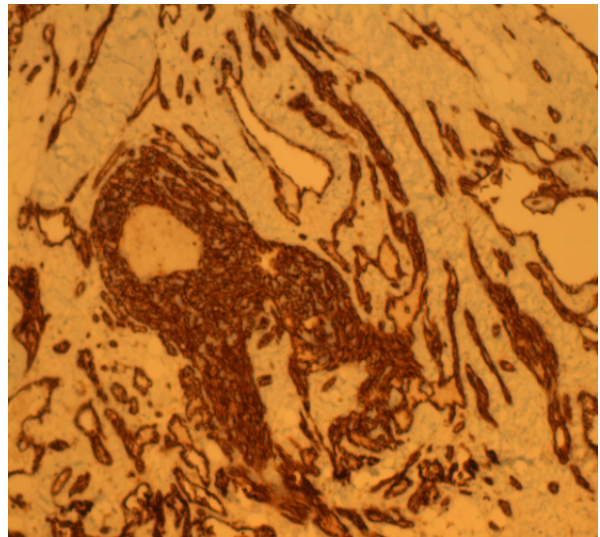


Fig. 1. Breast Angiosarcoma (H&E, 10x)

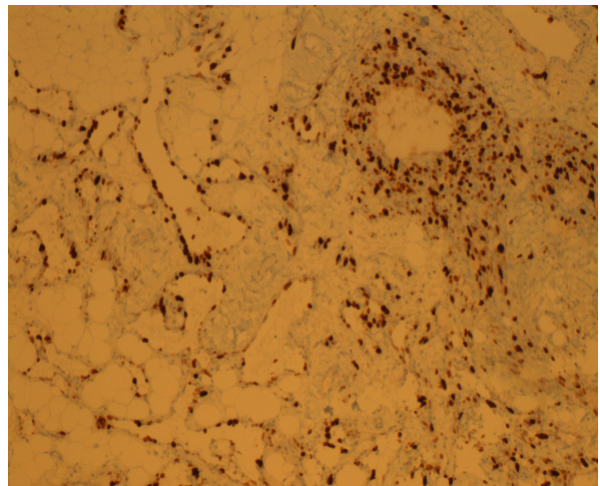


Fig. 2. Breast Angiosarcoma (CD31, 10x)

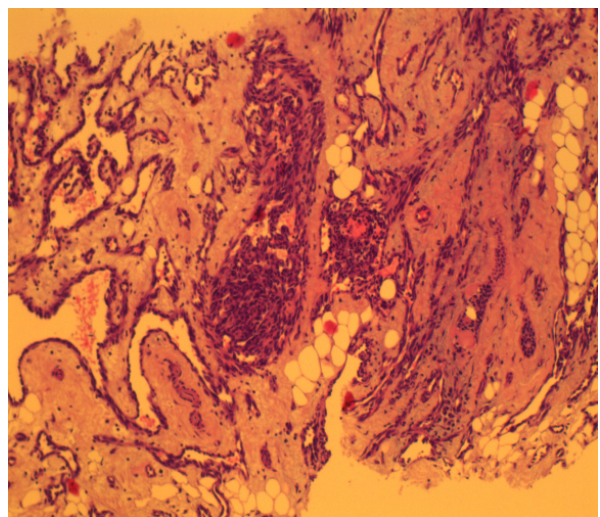


Fig. 3. Breast Angiosarcoma (Ki67, 10x)

based on histopathological and immunohistochemical examination, which exposes the vascular character of proliferation using markers to endothelial cells.

Mainly CD31 and CD34 and factor VIII. A proliferative marker Ki 67 showing a high proliferative index is also helpful. Based on the histopathological picture, three angiocarcinoma groups can be defined depending on the degree of cancer maturity. A well-differentiated cancer (G1), an intermediate-grade tumor (G2), a poorly differentiated tumor (G3). The prognosis of cancer and the form of therapy depend on the degree of differentiation.¹⁻⁸ In the treatment of cancer, the main role is played by surgical complete removal of the tumor as a result of quadrantectomy or mastectomy. Adjuvant treatment with radiotherapy and chemotherapy after surgery is usually not effective. There are, however, cases where such combined treatment gives good results and prolongs the time free from tumor recurrence.²⁻⁸ Supplementary treatment may bring benefits in the G3 and large tumors, over 5 cm. Recently, research on vascular endothelial growth factor (VEGF) and the VEGF-R1 and VEGF-C receptor, give hope for targeted anti-angiogenic treatment.^{9,10} In histopathological diagnosis it is very important to differentiate this tumor with other malignant, benign and non-neoplastic tumors. Angiosarcoma in G1 and G2 must be differentiated with angiomatosis, pseudoangiomatous stromal hyperplasia (PASH) and angiomas. This is especially a challenge when we have a thick-needle biopsy material. Helpful then is the overall picture including imaging tests (benign tumors are less than 2.0 cm), histopathological and immunohistochemical.¹¹⁻³⁰

Conclusion

In histopathological diagnosis it is very important to differentiate this tumor with other malignant, benign and non-neoplastic tumors. Angiosarcoma in G1 and G2 must be differentiated with angiomatosis, pseudoangiomatous stromal hyperplasia (PASH) and angiomas. This is especially a challenge when we have a thick-needle biopsy material. Helpful then is the overall picture including imaging tests (benign tumors are less than 2.0 cm), histopathological and immunohistochemical.

References

- O'Neill AC, D'Arcy C, McDermott E, et al. Magnetic resonance imaging appearances in primary and secondary angiosarcoma of the breast. *J Med Imaging Radiat Oncol.* 2014;58(2):208-212.
- Azizun-Nisa, Zeeshanuddin, Kayani N. Malignant vascular tumours associated with the breast: a study of 7 cases. *J Pak Med Assoc.* 2013;63(5):646-649.
- Bennani A, Chbani L, Lamchahab M, et al. Primary angiosarcoma of the breast: a case report. *Diagn Pathol.* 2013; 22;8:66.
- Savage R. The treatment of angiosarcoma of the breast. *J Surg Oncol.* 1981;18(2):129-134.
- Adem C, Reynolds C, Ingle JN, Nascimento AG. Primary breast sarcoma: clinicopathologic series from the Mayo Clinic and review of the literature. *Br J Cancer.* 2004;91:237-241.
- Donnell RM, Rosen PP, Lieberman PH, et al. Angiosarcoma and other vascular tumors of the breast. *Am J Surg Pathol.* 1981;5(7):629-642.
- Mahdi Y, Rouas L, Malihy A, Lamalmi N, Alhar Z. Diagnostic difficulties of primary angiosarcoma of the breast: a case report *J Med. Case Rep.* 2018;12:228-230.
- Bhosale SJ, Kshirsagar AY, Patil MV, et al. Primary angiosarcoma of the breast: a case report. *Int J Surg Case Rep.* 2013;4(4):362-364.
- Zelek L, Llombart-Cussac A, Terrier P, et al. Prognostic factors in primary breast sarcomas: a serie patient with long-term follow – up. *J Clin Oncol.* 2003;21(13):2583-2588.
- Yin M, Mackley HB, Drabick JJ, Harvey HA. Primary female breast sarcoma: clinicopathological features, treatment and prognosis. *Sci Rep.* 2016;6:31497.
- Cassou-Mounat T, Champion L, Bozec L, et al. Primary and Secondary Breast Angiosarcoma: FDG PET/CT Series. *Clin Nucl Med.* 2019;44(1):e33-e35.
- Tang T, Li H. Repeated resection-associated breast angiosarcoma: A case report. *Medicine (Baltimore).* 2018;97(39):e12513
- Lyou Y, Barber E, Mehta R, et al. Radiation-Associated Angiosarcoma of the Breast: A Case Report and Literature Review. *Case Rep Oncol.* 2018;11(1):216-220.
- Lokanatha D, Anand A, Lakshmaiah KC, et al. Primary breast angiosarcoma - a single institution experience from a tertiary cancer center in South India. *Breast Dis.* 2018;37(3):133-138.
- Gervais MK, Burtenshaw SM, Maxwell J, et al. Clinical outcomes in breast angiosarcoma patients: A rare tumor with unique challenges. *J Surg Oncol.* 2017;116(8):1056-1061.
- Cantile M, Di Bonito M, Cerrone M, et al. Primary breast angiosarcoma in young women from the same geographic region in a short period of time: Only a coincidence or an increased risk? *Breast J.* 2018;24(1):91-93.
- Leon-Castillo A, Chrisinger JSA, Panse G, et al. Index report of cutaneous angiosarcomas with strong positivity for tyrosinase mimicking melanoma with further evaluation of melanocytic markers in a large angiosarcoma series. *J Cutan Pathol.* 2017;44(8):692-697.
- Wang L, Lao IW, Yu L et al. Primary Breast Angiosarcoma: A Retrospective Study of 36 Cases from a Single Chinese Medical Institute with Clinicopathologic and Radiologic Correlations. *Breast J.* 2017;23(3):282-291.
- Wang L, Lao IW, Yu L, Yang W, Wang J. Breast Angiosarcoma with Exophytic Growth. Farrokh D, Modoodi E, Falah Rastegar Y. *Arch Iran Med.* 2016;19(11):812-815.

20. Ito T, Tanaka K, Suzumura K, et al. Angiosarcoma arising in the non-operated, sclerosing breast after primary irradiation, surviving 6 years post-resection: A case report and review of the Japanese literature. *Int J Surg Case Rep.* 2016;24:26-31.
21. Akrami M, Mohammadipour M, Mokhtari M, Dayani M. Exsanguinating Hemorrhage during Open Biopsy in a Primary Breast Angiosarcoma: A Case Report. *Iran J Med Sci.* 2016;41(2):154-156.
22. Bordoni D, Bolletta E, Falco G, et al. Primary angiosarcoma of the breast. *Int J Surg Case Rep.* 2016;20S:12-15.
23. Tanaka Y, Uchida A, Umemoto T, et al. Spontaneous regression of breast angiosarcoma after conservative treatment with radiotherapy: a case report and review of the literature. *J Med Ultrason.* 2015;42(3):427-432.
24. Masai K, Kinoshita T, Jimbo K, Asaga S, Hojo T. Clinico-pathological features of breast angiosarcoma. *Breast Cancer.* 2016;23(5):718-723.
25. Kilic F, Kandemirli SG, Er ME, et al. Primary angiosarcoma of the breast: Diagnosis with computer-assisted MRI-guided radio-guided occult lesion localization (ROLL) technique. *Diagn Interv Imaging.* 2015;96(11):1203-1206.
26. Wang L, Huang C, Yang X, Xiao H, Zou L. Primary Angiosarcoma of the Breast with Pulmonary Metastasis. *Breast J.* 2015;21(4):435-437.
27. Taghipour Zahir S, Sefidrokh Sharahjin N, Rahmani K. Primary breast angiosarcoma: pathological and radiological diagnosis. *Malays J Med Sci.* 2014;21(5):66-70.
28. Laé M, Lebel A, Hamel-Viard F, et al. Can c-myc amplification reliably discriminate postradiation from primary angiosarcoma of the breast? *Cancer Radiother.* 2015;19(3):168-174.
29. Pandey M, Sutton GR, Giri S, Martin MG. Grade and Prognosis in Localized Primary Angiosarcoma. *Clin Breast Cancer.* 2015;15(4):266-269.
30. Zemanova M, Rauova K, Boljesikova E, et al. Analysis of radiation-induced angiosarcoma of the breast. *Bratisl Lek Listy.* 2014;115(5):307-310.