



Contents lists available at ScienceDirect

## International Journal of Surgery Case Reports

journal homepage: [www.casereports.com](http://www.casereports.com)

## A rare etiology of a large tumoral mass of the breast - Case report primary leiomyosarcoma and osteosarcoma of the breast

Rita Galama\*, João Matoso, Germano Capela, Carlos Bôto, Cristina Duarte, António Ribeiro Mendes

Department of Surgery, Centro Hospitalar do Médio Tejo, Tomar, Portugal

### ARTICLE INFO

#### Article history:

Received 31 October 2020

Received in revised form

14 December 2020

Accepted 15 December 2020

#### Keywords:

Breast sarcoma

Leiomyosarcoma

Osteosarcoma

Breast malignancy

Case report

### ABSTRACT

Breast sarcomas are a rare group of malignant tumors accounting for less than 1% of all malignant neoplasms of the breast and fewer than 5% of all sarcomas.

We report a case of an 87-year-old caucasian female who recurred to the emergency department with complaints of a painful mass of the left breast with purulent discharge. Observation revealed a volumous mass in the inferior quadrants of the breast, ill defined, with petrous consistency, areas of necrosis, and inflammatory signs. She was admitted to Surgery ward for further study and therapy of a probable inflammatory tumor of the breast. Magnetic resonance image was obtained, raising suspicion on papillary carcinoma and classified the breast as BIRADS5. Microbiological and cytological exams of the exudate were negative. An incisional biopsy of the tumoral mass was also obtained, and the patient discharged while waiting for surgery. Histological exam and immunohistochemical essay were compatible with leiomyosarcoma. Left mastectomy was performed and the patient was discharged with no morbidities on the 5th day after surgery. Histological exam of mastectomy piece showed a metaplastic carcinoma, with osteosarcomatous and focal leiomyosarcomatous differentiation. The lesion was classified as pT4N0M0 and subsequent radiotherapy was performed. Twenty months after surgery the patient was being followed-up on Oncology and Senology consultations and remained asymptomatic.

Consensus on ideal management of this diseases is still on debate. Some authors defend the treatment of this entity in a similar way to sarcoma of the breast. More studies are needed to better understand this entity.

© 2020 The Author(s). Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## 1. Introduction

Breast sarcomas are a rare group of malignant tumors originating in mesenchymal breast tissue, accounting for less than 1% of all malignant neoplasms of the breast and fewer than 5% of all sarcomas. Leiomyosarcoma and osteosarcoma are exceedingly rare subtypes of breast sarcoma, the risk factors are not clarified and no specific treatment has been established yet. Macroscopically, leiomyosarcoma is a necrotic and hemorrhagic mass. We report a case of an 87-year-old female with giant leiomyosarcoma and osteosarcoma of the breast. Regarding the rarity of this entity, it is of paramount importance to identify and report these cases in order to establish the ideal therapeutic interventions, either surgical resection or radiotherapy.

## 2. Case report

An 87-year-old caucasian female, followed in Senology consultation in a community hospital, with a mass of the breast, recurred to the emergency department of the same hospital, with complaints of pain and purulent discharge of the left breast. She had no relevant medical history nor did take chronic medication.

Observation at admission revealed a volumous mass in the inferior quadrants of the left breast, ill defined, petrous consistency, with areas of necrosis and inflammatory signs. The patient had an ultrasound made 4 months earlier, where it was noted a volumous mass of polycyclic contours and surrounding oedema, with cutaneous thickening, consubstantiating a mixed pattern mass, with its multiseptated cystic component (interpreted as cystadenoma/cystadenocarcinoma) measuring 80 × 44 mm – RB = BiRADS2, LB = BiRADS4. Biopsy of the mass was taken and the result was inconclusive for malignant disease. She was then admitted to surgery ward for further study and therapy of a probable inflammatory tumor of the breast (Fig. 1). A swab sample was obtained and empiric antibiotic therapy with amoxicillin plus clavulanic acid (875 mg/125 mg 8/8 h per os) was initiated.

\* Corresponding author at: Department of Surgery, Centro Hospitalar do Médio Tejo, E.P.E., Av. Maria de Lourdes de Mello Castro – Ap. 118, 2304-909, Tomar, Portugal.

E-mail address: [rita.branquinho@chmt.min-saude.pt](mailto:rita.branquinho@chmt.min-saude.pt) (R. Galama).



**Fig. 1.** Tumoral growth of left breast, with central ulcerative necrosis.



**Fig. 2.** Day 2 after left radical mastectomy.

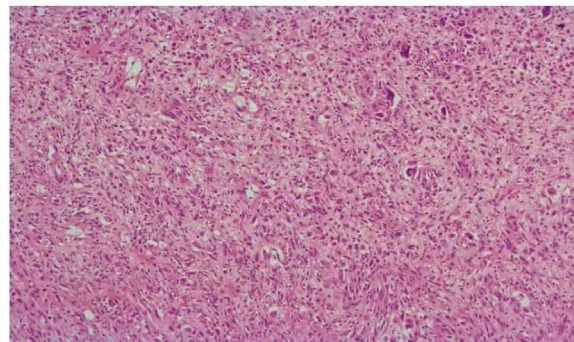
Breast magnetic resonance image (MRI) was obtained on the 5th day after admission, raising suspicion on possible papillary carcinoma and further raised classification to BIRADS5. Microbiological and cytological exams of the exudate were negative, and so empiric antibiotic treatment was suspended. By the 8th day of admission, an incisional biopsy of the tumoral mass was obtained, and the patient was discharged to follow-up consultation with no sign of inflammation, awaiting date of surgery, two weeks after discharge.

Histological exam of aforementioned biopsy revealed “morphological characteristics compatible with tumoral proliferation, spindle cells with elongated or irregular nuclei, non-prominent nucleolus and vacuolated cytoplasm with mitosis shape changes and foci of necrosis; scarce multinucleated giant cells and inflammatory cells”. Immunohistochemical essay for antiSMA, AE1/AE3, ALK, CD31, CD34 and S100 antibodies revealed marked positivity for SMA, compatible with leiomyosarcoma.

Thoracic computerized tomography scan (CT-scan) was negative for nodes or pulmonary metastasis. Therapeutic options were discussed with the patient, who agreed with mastectomy of the left breast.

Left mastectomy was performed by a surgeon dedicated to breast surgery, and the patient was discharged with no morbidities on 5th day after surgery (Fig. 2).

Histological exam of mastectomy piece showed focal positivity for p63 and cytokeratin 5/6, within metaplastic carcinoma, with osteosarcomatous and focal leiomyosarcomatous differentiation (Fig. 3), with negative margins. Lesion was classified as pT4N0M0 and subsequent radiotherapy with a total dose of 50 Gy (2Cy/cycle/day) was performed.



**Fig. 3.** 100x H&E. Set of atypical oval and spindle shaped cells amidst multinucleated giant cells.

The patient was followed-up on Oncology and Senology consultations, for 20 months after discharge. First assessment was at 1-month post-discharge and then every 3 months for the first year and every 6 months for the second year. She remained asymptomatic, with no signs of malignancy on imaging surveillance (MRI and CT-scan) or laboratory studies.

Data was gathered through *Scinico* software (in hospital and post-discharge consultation records), Operating theater and histological exam photographs. This case has been reported in line with the SCARE 2018 criteria [1].

### 3. Discussion/Conclusions

Given the exceeding rarity of breast leiomyosarcoma and osteosarcoma, there is very little literature on this subject and consensus on ideal management is still on debate. Leiomyosarcoma frequently presents as a large tumoral mass, well delimited, in postmenopausal women, 5th to 6th decade [2], although few tumors were as large as the one we described. Typically occurring in the uterus, retroperitoneum, subcutaneous tissues, and gastrointestinal tract [3], the breast is an uncommon primary localization [4]. Osteosarcoma may have a rapid growth pattern with no other clinical features distinguishable to other malignant breast neoplasms [2]. The characteristic histological appearance of leiomyosarcoma is that of spindle cells with elongated, pleomorphic nuclei, and definite diagnostic obtained through immunohistochemical exam reactive to desmin, vimentin and SMA and negative for cytokeratin, myoglobin and S100 [2]. Most authors establish complete surgical excision of tumoral mass with free margins as standard treatment for breast sarcomas. On this case, our experience is that a female patient with a voluminous leiomyosarcoma and osteosarcoma of the breast, without relevant medical history, treated in a community hospital by surgical resection with mastectomy and adjuvant treatment with radiotherapy (total dose of 50 Gy), similarly to the treatment of other sarcomas of the breast, had very good results, with no signs of malignancy or morbidity at 20 months after surgery. Despite our experience, more research is needed regarding leiomyosarcoma and osteosarcoma of the breast to better understand them, including risk factors and behavior of this malignancy, and define the best way of treatment.

### Declaration of Competing Interest

None.

### Funding

None.

### Ethical approval

The informed consent used was previously approved by the local Ethics Committee (Centro Hospitalar do Médio Tejo, E.PE – Tomar, Portugal).

The local Ethics Committee exempt case reports of an individual approval.

### Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

### Author's contribution

Rita Galama gathered the clinical data, wrote, reviewed and submitted the manuscript.

João Matoso and Carlos Bôto critically reviewed the study proposal and served as scientific advisors.

All authors approved the final version to be submitted.

### Registration of research studies

Not applicable.

### Guarantor

Rita Galama.

### Provenance and peer review

Not commissioned, externally peer-reviewed.

### Non-Author Contributions

Germano Capela and Cristina Duarte provided and cared for the patient.

António Ribeiro Mendes served as scientific adviser.

### References

- [1] R.A. Agha, M.R. Borrelli, R. Farwana, K. Koshy, A. Fowler, D.P. Orgill, For the SCARE Group, SCARE 2018 statement: updating consensus Surgical Case Report (SCARE) guidelines, *Int. J. Surg.* 60 (2018) 132–136.
- [2] S.Y. Lee, N.R. Lee, Positive response of a primary leiomyosarcoma of the breast following salvage hyperthermia and pazopanib, *Korean J. Intern. Med.* 33 (2) (2018) 442–445, <http://dx.doi.org/10.3904/kjim.2015.242>.
- [3] B.R. Kim, J.H. Lee, E. Cho, D.C. Kim, Y.M. Park, D.-H. Ha, S. Choi, Primary breast leiomyosarcoma located in the premammary zone: a case report, *J. Clin. Imaging Sci.* 39 (2015) 1105–1107, <http://dx.doi.org/10.1016/j.clinimag.2015.06.014>.
- [4] A. Bahrami, E. Resetkova, J.Y. Ro, J.D. Ibanez, A.G. Ayala, Primary osteosarcoma of the breast: report of 2 cases, *Arch. Pathol. Lab. Med.* 131 (5) (2007) 792–795, [http://dx.doi.org/10.1043/1543-2165\(2007\)131\[792:POOTBR\]2.0.CO;2](http://dx.doi.org/10.1043/1543-2165(2007)131[792:POOTBR]2.0.CO;2).

### Open Access

This article is published Open Access at [sciencedirect.com](https://www.sciencedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.