

**Case report**

# Metastatic Primary Breast Leiomyosarcoma in Men: A Case Report

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## Abstract

**Introduction:** Breast malignancy is the second most common cancer world widely. Many subtypes exist including carcinomas (most common) and sarcomas (least common). In this case report we discuss a rare case of primary breast leiomyosarcoma in a male patient.

**Case report:** A 94 years old male patient presented for a rapidly evolving right breast mass. Imaging showed a hypoechoic breast mass. Thoracoabdominal CT scan revealed lung and liver lesions. Biopsies confirmed the presence of a primary breast leiomyosarcoma with metastasis to the liver and the lungs.

**Discussion:** Breast leiomyosarcoma is considered to be one of the rarest breast cancers with very few cases reported. The non-specific imaging pattern of this pathology makes histopathologic and histochemical analysis the basis of definitive diagnosis (mitotic features, necrosis, differentiation, S100, cytokeratin, desmin, SMA). Surgery with negative margin is the standard treatment for non-metastatic cases whereas palliative chemotherapy is for metastatic ones. No data on an improved prognosis or survival rate with post-operative chemo/radiotherapies.

**Conclusion:** Further detailed studies on this particular rare type of breast sarcoma are needed to establish gold standard treatment guidelines. Every contribution to the literature can help sharpen our knowledge and understanding of this entity.

## Introduction

Breast malignancy, being the second most common type of cancer, represents an imminent burden on the healthcare system and accounts for more than 10% of all cancers across both genders [1-3]. Although most commonly breast malignancies are primary tumors (more than 95% of cases), cases of metastasis from different systems (genitourinary, gastrointestinal, ...) have also been reported and account for a small percentage (less than 5%) [2,4]. In both primary and metastatic breast cancers, carcinoma is the most common type and sarcoma is the least (less than 1%), with leiomyosarcoma being the rarest, very few case reports have been documented among which less than a dozen was in men [1,5].

This is a case report of a primary breast leiomyosarcoma in a male patient presented as a rapidly evolving painless breast mass. Our patient was diagnosed with FNCLCC grade 2 primary breast leiomyosarcoma and showed hepatic and pulmonary involvement, received palliative care and deceased two weeks following admission.

In view of the rarity of this specific subtype, contributing to the literature is important to generate better knowledge of this disease progression and prognosis as well as better insight into the most appropriate medical and surgical management.

## Case report

A 94-year-old man with history of severe heart failure



presents with a painless right breast mass evolving over few months. Physical examination revealed a 7 cm non-adhering firm right breast mass, with no signs of apparent lymphadenopathy. The mammary ultrasound uncovered a well circumscribed, heterogeneous hypoechogenic formation with a probable dysmorphic nature (Figure 1). The mammography classified the mass as ACR BI-RADS 4 (Figure 2). A thoraco-abdominopelvic scan revealed several lesions consistent with pulmonary and liver metastases (Figures 3,4).

The histopathological examination done on an incisional biopsy of the right breast mass under local anesthesia displayed a tumor proliferation consisting of fusiform cells arranged in dense bundles with multiple atypia; the nuclei were hypertrophied with a mitotic activity of maximum 15 mitoses per 10 consecutive fields without necrosis. The diagnosis of a grade 2 leiomyosarcoma (Fédération Nationale des Centres de Lutte Contre le Cancer grading system, FNCLCC) was based on the immunohistochemistry test which showed a high expression of vimentin, desmin, and H-caldesmone. A scan guided biopsy of the hepatic lesions confirmed the metastatic aspect of this leiomyosarcoma. The patient deceased after two weeks in the palliative care unit.



Figure 1: Ultrasonography of the right breast mass.



Figure 2: Mammography of the right breast mass.

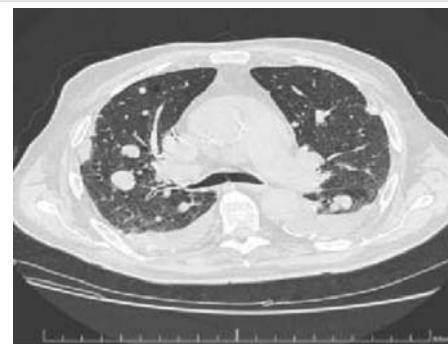


Figure 3: Thoracic CT scan showing lung metastasis.

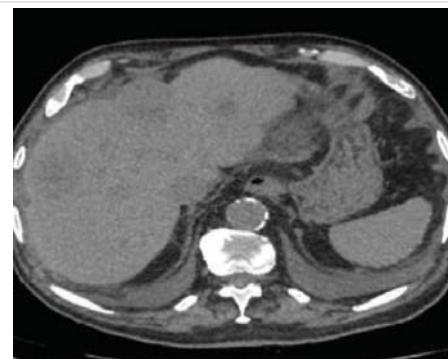


Figure 4: Abdominal CT scan showing liver metastasis

## Discussion

Among nonepithelial breast malignancies, sarcomas are the least common (less than 1%) [6-9]. Among the several histological subtypes, leiomyosarcomas are the least frequent (5 to 10% of all sarcomas) [9,10]. Due to the singularity of this subtype, only few case reports exist in the literature, even less for male patients.

Breast leiomyosarcomas can arise from blood vessels smooth muscle cells, nipple-areola smooth muscle cells or mesenchymal stromal cells [4,8,11]. They usually present as a painless slow growing well circumscribed breast mass with an average size of 5 cm without skin invasion nor axillary lymphadenopathy [7,10,12,13]. Ultrasonography and mammography typically show a heterogeneous hypoechoic non calcified mass without specific signs [5-7]. Fine needle biopsy can be contributive to the diagnosis, pre-operative excisional or core biopsy can confirm diagnosis since on clinical examination and imaging studies leiomyosarcoma is not very specific and can be mistaken for other benign or malignant tumors [2,7,13]. Usually but the definitive diagnosis is often made through histopathological and histochemical analysis post-surgical resection; they are characterized by fusiform or spindles shaped cells arranged in intersecting bundles, atypical cells, and pleomorphic nuclei [2,7,13]. As for immunohistochemistry, leiomyosarcomas are usually positive for SMA (like all the presented cases and ours), desmin (like our case), vimentin (like our case) and negative for S100 and cytokeratin [2,7,13]. These cells frequently



present with a high mitotic index per field (up to 24) and rarely with necrosis [8,9,13]. The preferred grading system is done using the National Federation of Centers for the Fight Against Cancer (FNCLCC) classification system which accounts for the number of mitoses, necrosis and the level of differentiation, and this represents a major prognostic factor and helps refining treatment approaches in each individual case [5,7,12].

Arsalane, et al. reported a case of an inflammatory painless right breast mass invading adjacent skin and causing significant weight loss in a 68 years old previously healthy man [12]. Unlike our case, their patient had a 8x9 mass invading the pectoralis major muscle with axillary lymphadenopathy and no signs of lung, liver or bone metastasis [12]. Histopathology showed necrotic cells with a fusiform appearance and an atypical mitosis which were positive for Ki 67, smooth muscle actin SMA and h-caldesmone while negative for protein S100 and cytokeratin [12]. The patient was diagnosed with FNCLCC grade 2 leiomyosarcoma without metastasis and underwent radical mastectomy with pectoralis major resection and axillary lymph nodes dissection followed by adjuvant radiation therapy and had no local or distal recurrence during nine months of follow-up [12].

Osorio et al reported the first case of a 15 cm fungating right breast leiomyosarcoma in a 39 years old female patient which presented with lung, pleural, bone and skin metastasis [10]. On histochemistry cells were positive for SMA and vimentin and negative for cytokeratin and protein S100 [10]. The patient received palliative care and deceased one month post diagnosis [10].

Ely Cheikh, et al. discuss a case of bilateral breast leiomyosarcoma in a 65 years old male patient who had a previous history of an unknown breast mass resected twenty years before the reported case [9]. Post resection, immunohistochemical studies on both masses (right and left breasts) revealed positive expression of smooth muscle action (SMA) and h-caldesmone and negative expression of desmin, S100, CD34 and CD68, which confirmed the diagnosis of leiomyosarcoma [9]. Unlike our case, their patient had no metastases at the time of presentation and post-resection, and he received radiotherapy after negative margins excision for local control and they report a period of eleven months recurrence free survival [9].

Additionally, Yang, et al. report a case of an 84 years old male patient who presented for a slow growing painless breast mass which appeared ten years before presentation [4]. Pre-operative imaging was suggestive of a BI-RADS type 4a mass and tumor markers were not elevated [4]. Contrary to our patient, no distal metastases were seen on CT scan [4]. They opted for a total resection of the mass and histochemical studies confirmed the diagnosis of primary breast leiomyosarcoma; SMA, Ki 67 and desmin positive, CD34 and S100 negative [4]. Post-operative radiotherapy

was recommended but not done and a six months follow-up check showed no signs of recurrence [4]. This unusual case of leiomyosarcoma (painless slow growth, no local or distal invasion) can be related to the senescent cellular components in a seventy plus individual, knowing that leiomyosarcomas typically have an aggressive presentation, and if untreated can be rapidly fatal [4].

Thus far, surgical resection with negative margins (3cm free margins or 2cm for breast conservation) is the most adopted therapeutic intervention for non-metastatic cases and presents a better prognosis and higher survival rate despite the risk of local or metastatic recurrence [4,6,12,13].

There is no clear data on the benefit of post-surgical chemotherapy and radiotherapy in decreasing local or distal recurrence or improving survival rates but are being performed in case of metastatic pathology or failure to reach negative margins during surgery or evidence of lymph nodes involvement [3,10,12]. Axillary lymph nodes dissection is not recommended unless there is evidence of lymphadenopathy as no evidence of leiomyosarcoma lymphatic spread has yet been documented [3,10,12]. Although rare, it is important to distinguish primary breast lesions from metastatic ones; even clinical findings might guide this differentiation (absence of inflammatory signs in metastatic diseases i.e. orange peel sign, nipple retraction) [2]. Palliative chemotherapy is usually adopted for metastatic non-surgical cases [2,13]. However, due to the scarce available case reports and the lack of definitive guidelines, the approach to treatment is still relatively subjective to each case hence the need for further investigations and studies to assess and determine a clear course of action in treating this rare pathology.

## Conclusion

Among breast cancers, primary leiomyosarcomas are a rare entity and even rarest in men. The lack of sufficient evidence and reported cases limits our understanding of this cancer's progression and adequate treatment approaches. Both localized and metastatic cases have been documented. World widely, wide excision with healthy margins is the standard treatment for non-metastatic cases. Exclusive management of this exceptional disease by specialized centers will help develop well-defined recommendations and improve diagnosis methods as well as prognosis and survival rates.

**Informed consent:** Signed written informed consent was obtained.

## Authors' contributions

MILAN Joelle, JRADI Ahmad, AZAR Anthony: Methodology, data collection and writing.

MATTA Clemence, ELIAS Bachir: Critical revision and supervision.



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