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Primary male breast leiomyosarcoma: An unusual case successfully treated by surgery and radiotherapy

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Abstract

Background: Primary sarcoma of the breast represent less than 1% of all breast malignities, and leiomyosarcoma are extremely rare. We report an unusual case of leiomyosarcoma of the breast in a Moroccan man.

Clinical case: we report a case of a 82-year-old Moroccan man, with history of larynx cancer, who present a nodule in left breast appeared six months ago. Exploration by mammography and sonography revealed a mobile and limited nodule, measuring 33x29x17mm, containing calcifications. Staging including a thoraco-abdominal CT was normal. The Patient underwent left lumpectomy without axillary dissection, histology revealed a well demarked tumor, infiltred by a proliferation with fascicular architecture, the tumor comes into contact with limits of surgical resection. Immunohistochemistry showed positivity of actin and caldesmone, and negativity for keratin, resulting in a diagnosis of leiomyosarcoma. Complementary surgery by left mastectomy followed by adjuvant radiotherapy to the chest wall was performed. 49 months after surgery, the patient is disease free.

Conclusion: Leiomyosarcoma of the breast is an extremely rare tumor that can be a diagnostic challenge to clinicians as it has no specific clinical or radiological features. It appears to have a better prognosis than other sarcomas of the breast with good outcomes after surgery.

Introduction

Sarcomas of the breast represent a rare entity with less than 1% of breast cancer [1]. Leiomyosarcoma are less common, account for under 1% of all breast sarcoma recorded in both sexes. Only 4 cases of male breast leiomyosarcomas were reported in the literature [2].

We report a case of a man followed for primary leiomyosarcoma of left breast, he was treated by mastectomy and adjuvant radiotherapy, with a good outcome after 49 months. Our case is the first case reported in Morocco.

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Case presentation

An 82-year-old Moroccan man, with history of stage II squamous cell carcinoma of the larynx, treated by surgery nine years ago, was admitted to the National Oncology Institute of Rabat, for management of left breast lump.

History of this disease dates back to seven months before diagnosis, by appearance of a nodule in left breast. This mass had enlarged rapidly in the past three months, without inflammatory signs.

Physical examination revealed a 3 cm size mass, without a palpable axillary or supraclavicular lymphadenopathy. The controlateral breast was normal. Exploration by mammography revealed a mobile and limited nodule, with regular contours, measuring 33x29x17mm, containing calcifications. Ultrasonography confirmed suspicious malignancy (Figures 1 (a) and (b)). Staging including a thoraco-abdominal CT and bone scan was normal.

The Patient underwent left lumpectomy without axillary dissection. Histo-pathological examination revealed a well demarked tumor, infiltred by a proliferation with fascicular architecture, composed of spindle-shaped cells, with eosinophil cytoplasm and irregular neucleus nucleolated sometimes monstrous. Rare atypical multinucleated cells were observed, with few mitoses and without necrosis (Figure 2). The tumor comes into contact with limits of surgical resection. Immunohistochemistry showed positivity of actin and caldesmone, and negativity for keratin and PS100. According to this pathological analysis, the tumor was diagnosed as leiomyosarcoma. A total mastectomy was performed, with no residual tumor.

Our case was discussed in the tumor board meeting, and adjuvant radiotherapy was indicated, a total of 50 Gy to the chest wall was delivered. No chemotherapy or hormone therapy was indicated. 49 months after surgery, the patient is disease free.

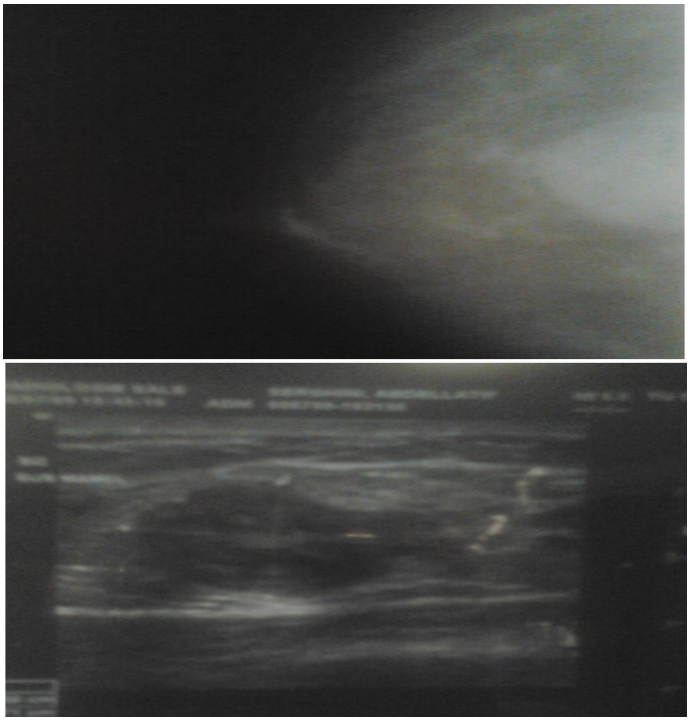

Table 1: (A&B) preoperative mammography (a) and sonography (b), showed a limited nodule, with regular contours, measuring 33x29x17mm, containing calcifications.

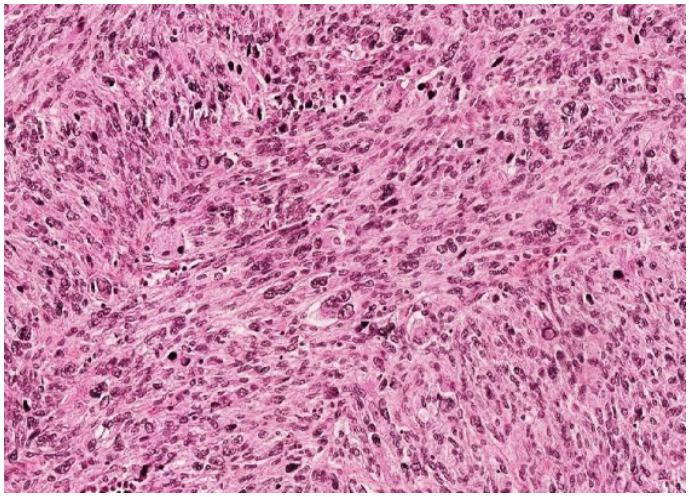

Table 2: histophatology: spindle-shaped cells, with eosinophil cytoplasm and irregular neucleus, and few mitosis (x200).

Discussion

Primary sarcomas of the breast are a rare tumors, it represent less than 1% of all breast malignancies [1]. Leiomyosarcoma are uncommon, only 30 cases was reported in the literature including 4 cases in men [2].

Origin of leiomyosarcoma of the breast is controversial. Pathologists suggest that may originate from the muscular blood vessels or from the smooth muscle of the nipple [3,4].

Diagnosis of breast primary leiomyosarcoma can be a challenge because the rarity of this entity, and of the nonspecific clinical and radiological findings. The role of the pathologist is important, and the diagnosis is mostly confirmed by the IHC [5]. Differential diagnosis includes leiomyoma, sarcomatoid carcinomas, and other soft sarcomas. Immunohistochemical staining is essential of diagnosis, leiomyosarcomas are usually positive for desmin, and actin, and are negative for S100, cytokeratins, and epithelial markers [6,7].

Optimal treatment of primary breast leiomyosarcoma is controversial due to the rarity this entity. Only a large excision with wide margins can provide good results. Axillary lymph node dissection is unnecessary because of low risk of node involvement [8]. Chemotherapy, radiotherapy and hormone therapy have not improved disease free or overall survival [1]. For our patient, surgery and adjuvant radiotherapy was sufficient to ensure control of the disease.

Leiomyosarcoma appears to have a better prognosis than other sarcomas of the breast, and the prognostic factors are not known because of the limited number of described cases. Local recurrence and lung metastasis are not rare; so the close follow-up is justified [9,10].

Conclusion

Primary male breast leiomyosarcoma is an uncommon tumor with good outcomes. The treatment is based on large excision, adjuvant radiotherapy can be used if breast conserving surgery. Prognosis seems better than other breast sarcomas; however, there is a need for further cases to determine the prognostic factors.

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