

CASE REPORT

Leiomyosarcoma of the Female Breast

Eszter SZÉKELY,¹ L MADARAS,¹ Janina KULKA,¹ Balázs JÁRAY,¹ Lajos NAGY,²

¹2nd Department of Pathology, ³rd Department of Surgery, Semmelweis University, Budapest, Hungary

Leiomyosarcomas of the breast are rare tumors. Less than 15 such cases have been reported in the literature so far. In this paper authors describe a case of leiomyosarcoma of a female breast presenting as a firm lobulated mass, mimicking a phylloid tumor radiographically. By fine needle aspiration biopsy, on the smears discohesive malignant looking cells were conclusive to a poorly differentiated invasive ductal carcinoma of the breast. The mas-

Keywords: breast tumors, leiomyosarcoma

tectomy specimen contained a lobulated mass, microscopically showing a partly epithelioid spindle cell tumor, immunoreactive for vimentin, desmin, smooth muscle actin antibodies, and negative for epithelial markers, hormone and growth factor receptors. Axillary lymph nodes were free of tumor. A primary leiomyosarcoma of the breast was diagnosed. (Pathology Oncology Research Vol 7, No 2, 151–153, 2001)

Introduction

Leiomyosarcoma (LMSC) of the breast is rare. In the English language literature about 15 such cases have been described so far. We present a case of a 73 year old female patient, whose breast tumor had been present for several month prior to her admission. The mass appeared lobulated on ultrasonography (US). Preoperative fine needle aspiration biopsy (FNAB) was performed. Based on the cytological features, a poorly differentiated ductal carcinoma was diagnosed. The mastectomy specimen contained a firm lobulated tumor, and microscopic examination revealed a mesenchymal neoplasm, microscopically and immunohistochemically consistent with a LMSC, without the evidence of any true epithelial origin.

Clinical history

A 73 year old female patient was admitted to the surgical department, for the presence of a firm lump in her right breast, having been present for three months. X ray mammography was performed, which found a lobulated tumor

4.5 cm in diameter in the lower inner quadrant of her right breast. The lump was easily detectable by ultrasound mammography as well. An US guided FNAB was performed. According to the radiological picture, as well as the age of the patient, a phylloid tumor was suggested by the radiologist.

Materials and Methods

FNAB smears were stained with hematoxylin and eosin. Tissue blocks cut from the surgical specimen were embedded in paraffin and stained with hematoxylin and eosin. Immunohistochemical reactions were performed after microwave antigen retrieval, using monoclonal antibodies of Novocastra products.

Pathological findings

The smears obtained were rather hypocellular, and apart from necrotic debris, contained mainly discohesive epithelioid like cells, with atypical nuclei, large eosinophilic partly elongated cytoplasm, and numerous mitotic figures (*Figures 1,2*). A poorly differentiated invasive ductal carcinoma was diagnosed.

Based on the cytological diagnosis a mastectomy was performed, with supplementary axillary resection. The mastectomy specimen contained a tan-white, well circumscribed lobulated mass 4.8 cm in diameter, which was very firm. Macroscopically no area of necrosis was

Received: April 27, 2001; *accepted:* May 28, 2001

Correspondence: Dr. Eszter SZÉKELY, ²nd Department of Pathology, Semmelweis University Budapest H-1091, Üllői út 93, Budapest, Hungary; Tel: 36-1-218-28-80, Fax:36-1- 215-69-21
E-mail: szesz@korb2.sote.hu

appreciable, however, the mass contained small cysts and haemorrhage. The microscopic picture showed a spindle cell tumor composed of eosinophilic mesenchymal cells forming fascicles intersecting at right angles, with pushing margins. Tumor cells possessed hyperchromatic cigar shaped nuclei, prominent nucleoli, and numerous typical and atypical mitotic figures (20–22 mitosis/ten high power fields), (*Figures 3,4*). No epithelioid features were detectable from the first microscopic slides. Based on the clinical suspicion and the preoperative cytological diagnosis, the mastectomy specimen was reviewed, and additional blocks were cut out for further microscopic examination. One of the new slides showed a small area where discohesive epithelioid cells with round eosinophilic cytoplasm were also seen. Meticulous search, however, for any true epithelial component of the tumor by serial sections accompanied by immunohistochemical examinations -cytokeratin, (CK), (CKAE-1/AE3), epithelial membrane antigen (EMA)-, ended with negative results. Additional immunohistochemical reactions routinely performed in breast carcinomas

(oestrogen, progesteron receptors, p53, cErb B-2) also proved to be negative. Tumor cells showed strong positivity for vimentin, desmin and smooth muscle actin (SMA) antisera (*Figure 5*). Ki-67 proliferation marker showed positivity in 5% of the tumor cells. Axillary lymph nodes were free of metastases. Operation was not

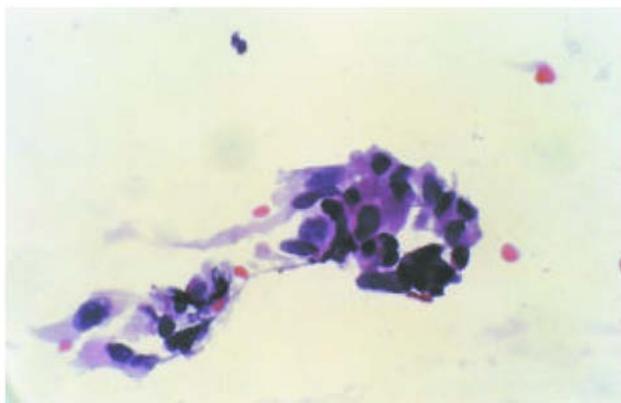


Figure 1. Hypocellular smear showing a cell cluster, composed of epithelioid cells with atypical nuclei, large eosinophilic, partly elongated cytoplasm. HE, x400

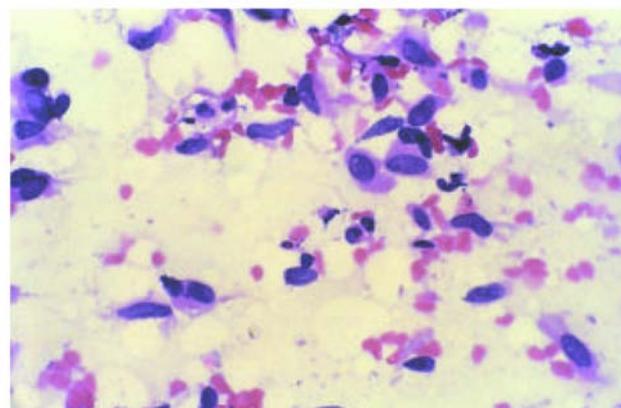


Figure 2. After smear review, discohesive cells with eosinophilic, elongated cytoplasm were clearly visible. HE, x400

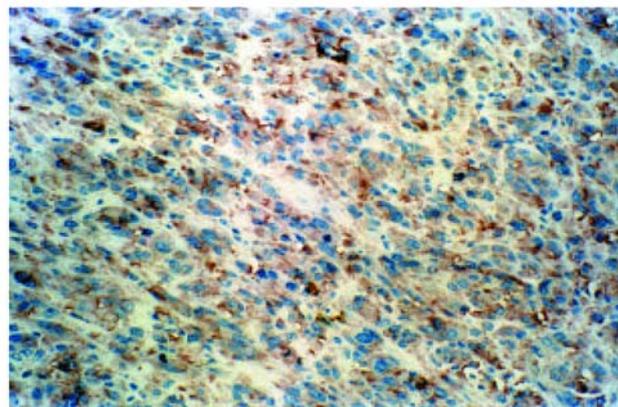


Figure 3. The tumor had pushing margins. HE, x200

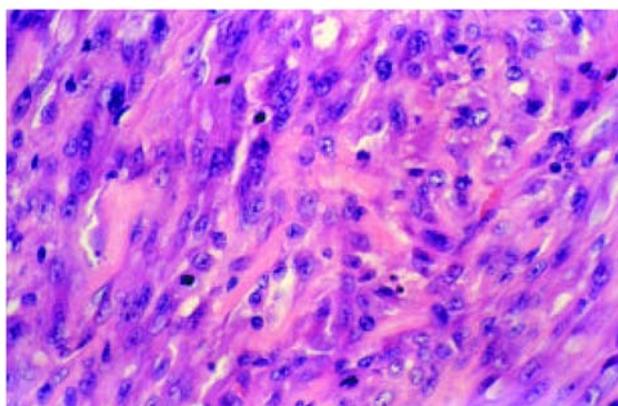


Figure 4. Tumor cells possessed hyperchromatic cigar shaped nuclei, prominent nucleoli, and numerous typical and atypical mitotic figures. HE, x400

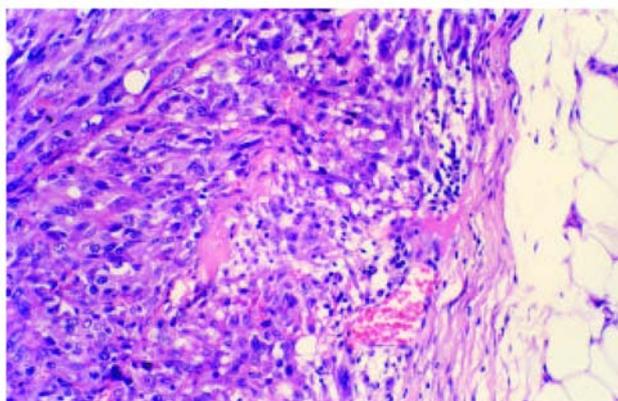


Figure 5. Tumor cells showed strong positivity for smooth muscle actin (SMA) antiserum. x200

followed by chemotherapy or irradiation. One year after the operation the patient is still well.

Following the histological examination of the mastectomy specimen, a review of the smears found no equivocal signs of a pure, mesenchymal neoplasm, however, discohesive cells with eosinophilic, elongated cytoplasm were clearly visible among the small epithelioid cell clusters.

Discussion

Leiomyosarcoma is not uncommon among soft tissue tumors. The most common primary site is the uterus, the retroperitoneum, subcutaneous tissues, and the GI tract, but any part of the body may harbour this histological type of soft tissue tumor.^{8,13} Even the great blood vessel might be the primary site.^{2,3,5} A general agreement on treatment is still lacking, since there are tumors which respond well to chemotherapy, while others show no chemo- or radiosensitivity.^{1,7,9,10,12,14,15} Leiomyosarcomas of the breast are particularly rare. They should be differentiated from leiomyomas and other spindle cell tumors of the breast. Metaplastic carcinomas may be large, well circumscribed tumors, which show CK positivity. It is noteworthy though that there have been reports on focal CK and/or EMA positivity reported in leiomyosarcomas as well. Myoepitheliomas express SMA and CK14, a distinctive feature not found in other spindle cell tumors. Myofibroblastomas of the breast are benign stroma rich tumors with characteristic morphology and desmin positivity. The origin of a true LMSC in the breast is thought to be the pluripotent mesenchymal cell of the mammary stroma, rather than the smooth muscle cells of the vascular wall. These tumors arise mainly in the nipple region. There is even more uncertainty about the treatment of breast LMSC-s, since there are only a few number of cases having been described, and no general conclusions have been drawn so far.^{4,6,16} The mitotic rate and clinical stage are the only reliable prognostic factors that have influence on survival and relapse.¹¹ However, according to some authors, even the reliability of the mitotic count is debatable, since there are reported cases with low mitotic count and early recurrence, while there are others with high mitotic rate and no metastasis.¹⁶ The few cases with long term follow up show that albeit these tumors initially have a favourable prognosis, later on metastases occur and may cause the death of the patients.^{4,16} It is questionable, however, whether post-operative treatment (radiation, chemotherapy) would be of any benefit since metastasis might appear decades later without any treatment and so a conservative attitude could save the patient from the disagreeable and even harmful consequences of these ancillary treatment methods.¹⁶ Axillary block dissection is not recommended, since none of the reported cases showed lymph node metastases. However, it is very important to differentiate these tumors from

metaplastic breast carcinomas or malignant phylloid tumors, since carcinomas argue for prompt aggressive post-operative treatment to give any chance for the patient to escape early death from metastases after surgery.⁴ Our case also highlights the responsibility of preoperative FNAB. Though being rare, in cases of hypocellular smears obtained from breast lesions radiographically suggestive of a phylloid tumor, the possibility of a tumor of mesenchymal origin in the lesion should be raised. If there is any doubt about the nature of the lesion, intraoperative frozen examination can save the patient from unnecessary axillary lymph node dissection. Although preoperative FNAB led to a misdiagnosis in our case, we think it merits presentation in order to extend the short list of cases with a further example.

References

- ¹Blanchard DK, Budde JM, Hatch GF: 3rd Tumors of the small intestine. *World J Surg* 24:421-429, 2000.
- ²Burke AP, Virmani R: Sarcomas of the Great Vessels. A Clinicopathologic study. *Cancer*, 71:1761-1773, 1993.
- ³Burke AP, Virmani R: Tumors of the heart and great vessels, AFIP, 1996.
- ⁴Falconieri G, Della Libera D, Zanconati F, Bittesini L: Leiomyosarcoma of the Female Breast. *Am J Surg Pathol* 108:19-25, 1997.
- ⁵Glock Y, Laghzaoui A, Wang J: Fissured leiomyosarcoma of the descending thoracic aorta. A propos of a case and review of the literature. *Arch Mal Coeur* 90:1317-1320, 1997.
- ⁶Palacios G: Leiomyosarcoma of the Female Breast. *Am J Surg Pathol* 109:650-651, 1998.
- ⁷Majeski J, Crawford ES, Majeski EI: Primary aortic intimal sarcoma of the endothelial cell type with long term survival. *J Vasc Surg* 27:555-558, 1998.
- ⁸Mentzel T, Calonje E, Fletcher CDM: Leiomyosarcoma with prominent osteoclast-like giant cells *Am J Surg Pathol* 18:258-265, 1994.
- ⁹Miracco C, Laurini L, Santopietro R: Intimal type primary sarcoma of the aorta. Report of a case with evidence of rhabdomyosarcomatous differentiation *Virchows Arch* 435:62-66, 1999.
- ¹⁰Patel KR, Niazi TBM, Anthony P: Griffiths. Massive osteolytic bone metastases from a primary aortic sarcoma: a case report. *Hum Pathol* 28:1306-1310, 1997.
- ¹¹Pautier P, Genestie C, Rey A: Analysis of clinicopathologic prognostic factors for 157 uterine sarcomas and evaluation of a grading score validated for soft tissue sarcoma. *Cancer* 88:1425-1431, 2000.
- ¹²Rochais JP, Icard P, Coffin O: Intimal sarcoma of the thoracic aorta: a case report. *Eur J Vasc Endovasc Surg* 18:181-182, 1999.
- ¹³Roncaroli F, Eusebi V: Rhabdomyoblastic differentiation in a leiomyosarcoma of the retroperitoneum *Hum Pathol* 27:310-313, 1996.
- ¹⁴Shimoda H, Oka K, Otani S: Vascular leiomyosarcoma arising from the inferior vena cava diagnosed by intraluminal biopsy. *Virchows Arch* 433:97-100, 1998.
- ¹⁵Soh LT, Chew SH, Ang L: Uterine leiomyosarcoma-a Singapore experience. *Aust NZJ Obstet Gynecol* 39:246-248 1999.
- ¹⁶Ugras S, Dilek ON, Karaayvaz M, et al: Primary Leiomyosarcoma of the Breast. *Surg Today* 27:1082-1085, 1997.