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Olgu Sunumu
Case Report

Leiomyosarcoma of the breast - a case report*

Memenin leiomyosarkomu - bir olgu sunumu*

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Abstract
Primary leiomyosarcomas of the breast are extremely rare tumors. There were approximately thirty patients reported in the literature up to date. Herein, we report a 66-years old female patient who presented with an enlarging mass in her right breast which was diagnosed as primary leiomyosarcoma following previous mixed invasive ductal carcinoma and invasive lobular carcinoma in her left breast. Although primary leiomyosarcomas of the breast are very rare tumors, they should be kept in mind in the differential diagnosis of spindle cell breast tumors including metaplastic carcinoma in patients with prior breast cancer.
Keywords: Breast cancer; leiomyosarcoma

Özet
Memenin primer leiomyosarkomları oldukça nadirdir. Literatürde bugüne kadar yaklaşık otuz hasta rapor edilmiştir. Bu olgu sunumunda, daha önce sol memesinde mikst invaziv duktal karsinom ve lobüler karsinom teşhisi olan, sağ memesinde büyüme gösteren kitle ile başvuran ve primer leiomyosarkom tanısı alan 66 yaşındaki kadın hasta bildirilmektedir. Memenin primer leiomyosarkomları çok nadir tümörler olmasına rağmen, meme kanseri öyküsü olan hastalarda ortaya çıkan iğsi hücreli meme tümörlerinin metaplastik karsinomu da içeren ayırıcı tanısında akılda tutulmalıdır.
Anahtar kelimeler: Meme kanseri; leiomyosarkom

Introduction

Primary sarcomas of the breast are extremely rare, with less than 0.1% of all malignant tumors of the breast. Leiomyosarcomas belong to a less common subgroup (5% to 10%) of breast sarcomas (1). Leiomyosarcoma probably originates from blood vessels, the smooth muscle of the nipple-areolar complex or myofibroblasts (2). Herein, we report a case of primary leiomyosarcoma of the breast occuring 18-months after the diagnosis of mixed invasive ductal and lobular carcinoma of her other breast.

Case

A 66-years old female patient with a mass in her left breast was performed radical mastectomy with axillary lymph node dissection and tumor was diagnosed as mixed invasive ductal and lobular carcinoma. Investigations for tumor staging did not show any other mass at that time. Then, she had six cures chemotherapy consisting of TAC (docetaxel-adriamycin-cyclophosphamide) and she did not receive radiotherapy. During clinical follow-ups, 18-months after chemotherapy, a rapidly progressive growing mass was recognized in her right breast. Breast ultrasound revealed a 13 cm lobulated lesion in the inner half of the breast. A spindle cell tumor with cytologic atypia was seen in intraoperative examination and then right radical mastectomy with axillary lymph node dissection was performed relying on the possibility of metaplastic carcinoma. Grossly; tumor nodule was 12x10x8 cm sized, well circumscribed with a whorled and lobulated cutsurface and located in upper and lower inner quadrants. Histologically; tumor was composed of interlacing and parallel fascicles of spindle cells with pleomorphic nuclei and eosinophilic cytoplasm (Figure 1). Thirty-two mitoses per 10 high power fields were counted and focal necrosis was present in the tumor. All the surgical margins were free of the tumor. Histochemically, smooth muscle elements were recognised with Von Gieson and Masson Trichrom stains. Immunohistochemically; tumoral cells exhibited diffuse positivity with smooth muscle actin (Figure 2), and vimentin and desmin positivity was focally. The tumor was negative for pancytokeratin (Figure 3) and S-100. According to histological and immunohistochemical features, the tumor was diagnosed as leiomyosarcoma. Pursuant to grading system of French Federation of Cancer Centers Sarcoma Group (FNCLCC) (3) total score was 5 (score 3 for mitotic count, score 1 for necrosis and score 1 for tumor differentiation) and histological grade of tumor was considered as Grade II. Radiological examinations did not define any other tumor focus so that the possibility of metastatic tumor was ruled out. Tumor stage was accepted as Stage IIB based on the pathological and radiological findings. Although all the margins were free, radiotherapy and chemotherapy were recommended due to the tumor size and histological grade, but the patient did not accept to receive these therapies.

Discussion

Leiomyosarcoma is not an uncommon soft tissue tumor. It can exist particularly in uterus, retroperitoneum,

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subcutaneous tissues and gastrointestinal tract as primary sites but it can occur throughout the body.

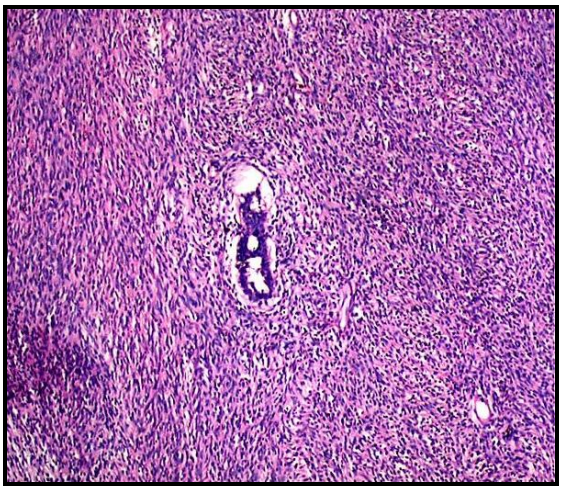

Figure 1. Tumoral spindle cells surrounding benign breast duct (H&E, ×100).

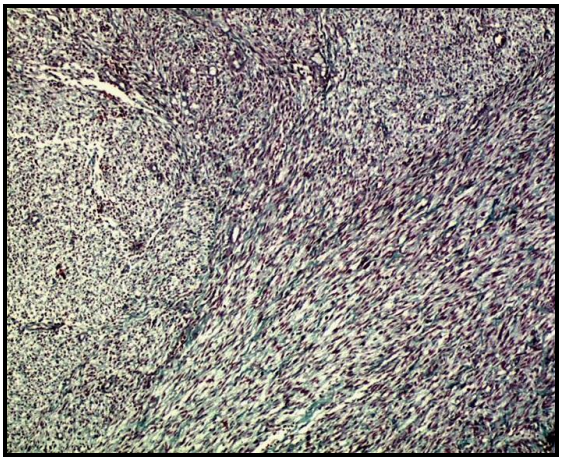

Figure 2. Smooth muscle antigen positivity in spindle cells (SMA, ×200).

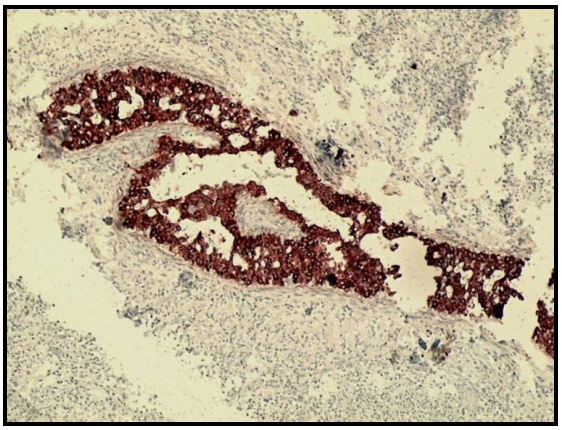

Figure 3. Pancytokeratin negativity in spindle cells (Pancytokeratin, ×200).

Although, leiomyosarcoma of the breast can be seen as a primary or secondary (metastatical or postradiation) tumor (4), primary type is extremely rare (1). The smooth muscle component of leiomyosarcoma might be originated from the smooth muscles around the nipple or blood vessels. Also, sarcomatous change from leiomyoma or other spindle cell tumors can be the source of the tumor (5). Most of the cases are female and the median age of presentation is 56 (range 24-86) (2,6). Nearly half of the tumors are in or near the nipple-areola complex, but any quadrant can be affected (2). Clinically, they present with a progressively enlarging tumor. Radiological findings are nonspecific. In fine needle aspiration cytology, these tumors can be misdiagnosed as poorly differentiated carcinoma, metaplastic carcinoma, or other sarcomas such as malignant phylloides tumor. Immunohistochemical studies are essential for the differential diagnosis of leiomyosarcomas from other spindle cell tumors, especially metaplastic carcinoma and soft tissue sarcomas. These tumors usually exhibit positivity with desmin, smooth muscle actin, muscle specific actin and they are negative for S100 (neural tumor marker), cytokeratins and epithelial markers. The principal treatment is surgical excision with free surgical margins. Procedures of axillary node dissection are not necessary. Chemotherapy and/or radiotherapy are not suggested for smaller than 5 cm grade I tumors having free margins. If the margins are involved or closer than 2 cm, histological grade II or III and tumor is >8-10 cm in size, chemotherapy and/or radiotherapy can be essential. Therefore, the tumor staging and status of surgical margins of the resection specimen defines the therapy procedures following surgery for leiomyosarcomas as in other soft tissue sarcomas (7). Long-term follow-up is important because recurrence or metastasis were reported more than 10 years after treatment (8).

Postradiation sarcomas including leiomyosarcomas of the breast following cancer therapy can occur in previously irradiated field and several years after radiotherapy. Radiation induced sarcoma diagnosis depends on the criteria established by Huvos et al (9). These tumors are associated with poor prognosis because the diagnosis is late due to the effects of prior therapy. Some authors reported that a good response to chemotherapy was observed for postradiation leiomyosarcomas and high survival rates for patients treated with chemotherapy associated surgery. So that they suggested neoadjuvant chemotherapy a potential part of treatment (10). Cyclophosphamide is an alkylating agent used commonly for the treatment of malignancies as well as for autoimmune disorders (11). Cyclophosphamide has been linked to secondary tumors such as bladder cancer, osteosarcoma, and leukemia. Therefore, it is unclear whether this type of exposure causes the development of breast leiomyosarcoma (12). De la Pena and Wapnir (13) reported a case of leiomyosarcoma of the breast with a 10-year-history of cyclophosphamide exposure. A case demonstrated two separate breast cancers and metastatic leiomyosarcoma of the thigh from retroperitoneum was reported as multiple synchronous tumors (14). The present case was

diagnosed as leiomyosarcoma after excluding the possibility of metaplastic carcinoma by demonstrating pancytokeratin negativity in several tumor sections. This tumor should be regarded as primary leiomyosarcoma of the breast, since we could not identify any other focus as a primary tumor and she had no radiotherapy. In addition, she had only received six cures of cyclophosphamide therapy for only 18 months and this time was shorter than the reported previous case in which exposure time was approximately 10 years (13). To the best of our knowledge, primary breast leiomyosarcoma with contralateral breast cancer has not been described before.

To conclude, in recent years with the increasing survival rates in breast cancer, radiation induced leiomyosarcomas seems to become more common. Postradiation leiomyosarcomas have worse prognosis and chemotherapy accompanied by surgery is suggested for these tumors rather than primary ones. Although primary leiomyosarcomas are very rare tumors, they should be kept in mind in the differential diagnosis of spindle cell breast tumors including metaplastic carcinoma in patients with prior breast cancer. Since postradiation leiomyosarcomas show worse prognosis than primary tumors, to distinguish breast leiomyosarcoma as primary or postradiation is essential.

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