RECURRENT PHYLLODES TUMOR OF THE BREAST WITH MALIGNANT TRANSFORMATION DURING PREGNANCY

Ahmet Bal¹, Bülent Güngör², Ayfer Kamali Polat², Tekin Şimşek³
¹Bozok University, Faculty of Medicine, Department of Surgery, Yoğurt
²Ondokuz Mayıs University, Faculty of Medicine, Department of Surgery, Samsun
³Ondokuz Mayıs University, Faculty of Medicine, Department of Plastic and Reconstructive Surgery, Samsun

Abstract
The cystosarcoma phyllodes is a rare and infrequently recurring tumor of the breast. Here we present a recurrent cystosarcoma phyllodes tumor during pregnancy involving the whole breast tissue and treated by mastectomy and silicon implantation. The fine needle aspiration cytology revealed a benign cytology-fibroadenoma and the 5.5 cm tumor was locally excised 3 years ago. The pathologic diagnosis was benign phyllodes tumor. During pregnancy, the tumor recurred and involved the whole breast. Mastectomy and reconstruction with silicon implant was the treatment choice at this time. The pathology of the mastectomy specimen was malignant phyllodes tumor. The phyllodes tumors should be locally excised with a wide margin although they are benign in nature. Mastectomy and reconstruction may be recommended as a safe surgical treatment modality in giant phyllodes tumors of the breast.

Key words: cystosarcoma phyllodes, mastectomy, silicon implantation

Introduction
The cysto-sarcoma phyllodes (CP) tumors of the breast are fibro-epithelial tumors which are rarely seen and have potential recurrence (1-4). CP tumors about which a great deal of studies have been done, was clinically identified first by Müller in 1838. Less than 1 % of all the breast tumors consist of CP tumors (4). Phyllodes tumors can appear in any age group of women, although it is seldom seen in girls (5,6). CP tumors are the ones which are not considered initially in clinical diagnosis, show slow or rapid growth pattern, and are diagnosed after biopsy. These rarely encountered tumors are typically seen as mobile masses in the diameter of 5 centimeters or more. Nevertheless, CPs with diameters of 40 centimeters are reported in the literature (7). CP tumors which are clinically similar to fibro-adenomas (FA) are distinguished from FA histopathologically by their cellular pattern, having increased cellular atypical changes and excessive stromal growth. CP tumors typically have more frequent local recurrence and higher malignity incidence in comparison with FA. Local recurrence up to 50 % after surgery has been reported in CP tumors (8). Here, we intend to share this rarely seen malignant CP case relapsing during pregnancy and having a high recurrence potential in the light of our experience with a single case and bibliographic information.

Case
A 32 years old female patient admitted to our clinic with the symptoms of hard, tender mass covering approximately the whole left breast (Figure1). In her physical examination; a hard, tender and indurated mass filling almost her entire left breast, and lymph nodes in left axilla were discovered. After reviewing the patient’s history and her previous files, it was determined that an excision of a 5.5 centimeter mass with a 1 centimeter surgical margin in the left upper quadrant of the left breast was performed 3 years ago. The result of a preoperative fine needle aspiration biopsy in an outer center was “Benign cytology-Fibroadenoma”, and the post operative pathology report was benign CP tumor. We learned that the patient noticed the mass in her left breast 18 months after the operation and soon after she became pregnant. The patient reported the mass had grown rapidly during pregnancy. During the lactation period a warm, indurated tenderness developed on her left breast. A mass filling the whole breast tissue and having solid and cystic components, in the size of 13x10 centimeters, causing dilatation in milk canals was...
A mass concordant with solid fibroadenoma with 15 millimeters diameter, located in the right inferior quadrant of the right breast was detected. Multiple lymph nodes, 2 centimeters diameter, appearing reactive in the left axilla were observed. In addition, a 13x9 cm mass (CP tumor recurrence?) which filled almost the entire left breast, including necrotic cavities, indicating hypervascular type 1 contrast dynamic in the serial examination with contrast, was determined by a magnetic resonance imaging of the breast (Figure 2). Additionally, another 15 mm mass showing similar contrast with the mass in the left breast, located in the right lower quadrant of the right breast, was determined. Recurrent CP and lactation mastitis was diagnosed in the patient. For treatment of mastitis, an appropriate antibiotic therapy was given to the patient and the progress of mastitis regressed. There were no pathological findings in the patient’s serum biochemical examinations, except an increased white blood cell count on admission, but decreased later. Because of the fact that the patient wanted her breast to be conserved, subcutaneous mastectomy and axillary lymph node sampling were done. Frozen section lymph node examination was performed during the operation in order to search for an accompanying infiltrative ductal or lobular carcinoma, despite the difficult diagnosis of a sarcomatous metastasis. The operation was completed by replacing silicone implant under the pectoralis major muscle for reconstruction. The patient had no complications in the postoperative period and was discharged with on the 5th postoperative day. As a result of the pathology of mastectomy material, it was pointed out that the tumor involving the whole breast was malignant CP, and there were some reactive changes in the axillary lymph nodes. There was no evidence of metastasis as determined by the scanning of systemic metastasis performed during the postoperative period. Neither metastasis nor recurrence was seen in the 6th month after operation. The patient is followed closely by having a control examination every three months.

Discussion

CP tumors are one of the rare diseases of women usually seen between the ages of 35-55 (9). Our patient is 32 years old, and is in this age range. In radiological studies, certain criteria in mammography, ultrasonography (USG) and magnetic resonance imaging (MRI) are not available to distinguish FA and CP tumors (9). In our patient, the lesion was identified in patient’s USG, but no information was given about the fact that the tumor was CP. According to our opinion, the probability of CP in the breast especially with 5 centimeters mass in diameter should be taken into consideration and the treatment should be planned appropriately. We think that the comment of recurrent CP tumor in our patient’s MRI report is connected to the first pathological result and clinical information given. The main treatment of CP tumors is surgical excision. The type of surgery is defined according to the expectation of the patient, size of tumor, localisation and type of tumor (benign/borderline/malignant or recurrent) and tumor/breast ratio. We preferred the treatment type as mastectomy because of the recurrent tumor; the tumor covering almost all the breast; discordance of breast/tumor size; severe mastalgia; poor cosmetic appearance; and the consensus between the patient and clinician for the mastectomy. It is known that local recurrences occur frequently in CP tumors. The rate of recurrence is reported 28-50 % in literature (8,10). The most important risk factor for recurrence is the resection done within 1-2 cm negative surgical margins (11). The age, type of surgery, increased mitotic activity and excessive stromal activity are considered among the risk factors for local recurrence by Chen et al. (12). Asoglu determined that size of tumor, excessive stromal growth and negative surgical margin smaller than 1 cm are risk factors for local recurrence (13). Moreover, the reliability and confidence of the preoperative diagnosis is very important for the success of treatment. Our case supports high recurrence rates for CP tumors. The possible risk factors for local recurrence...
in our patient are the diagnosis of FA in aspiration biopsy before the first operation and 1 cm negative surgical margin in her first operation. The adequate surgical margin couldn’t be provided. The common approach in surgical treatment of CP tumors is that there is no indication of axillary dissection (9,14). Since there are clinically palpable lymph nodes in our patient’s axilla, sampling was done; the histopathologic result was “reactive lymphadenopathy” concordant with the literature. Although it is not certain, excessive hormonal activity is also a risk factor in growth and differentiation of the tumor. As it is known, FAs are similar to CP in pathologic characteristics and tend to grow in pregnancy. The number of CP tumors reported during pregnancy is very small (15). CP tumors occurring during pregnancy tend to grow rapidly. It was reported that proliferation in lobular and alveolar tissue of the breast is related with increased progesterone, estrogen, human chorionic gonadotropin hormone, angiogenic factors and vascular endothelial growth factor during pregnancy (15,16). The hormonal sensitivity of phyllodes tumors was not exactly determined. We think that the growth and malignant differentiation of the tumor with pregnancy can be related to increased hormonal activity and especially with estrogen in our case. Although cystosarcoma phyllodes tumors are rarely seen, they have high recurrence potential if appropriate treatment is not performed. The excised material should be within the safety margins. The fact that it can grow rapidly during pregnancy should be taken into consideration because of the reproductive age group of these patients. Mastectomy and breast reconstruction should be considered as an appropriate surgical treatment in giant CP tumors.

References

Correspondence
Bülent Güngör
Tel : +90(362) 3121919
E-mail : bgungor@omu.edu.tr