Fibrous histiocytoma of the breast

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ABSTRACT

Two Saudi females aged 26 and 14 years presented with giant fibroadenomas. The histology after surgical excision revealed benign fibrous histiocytomas. Patient one remained well with no apparent recurrence after 5 years of the initial excision. However, in patient 2 the benign fibrous histiocytoma recurred with low-grade malignancy after a year of the initial excision. A high-grade recurrence developed after 3 months of the second excision. This was treated by radical wide excision. It was felt later that mastectomy was more appropriate and therefore was performed. The second patient highlights the fact that malignant histiocytoma can occur in the pediatric age group and that the initial surgical treatment of benign fibrous histiocytoma should be aggressive to avoid recurrence and development of malignant changes. We believe that our second patient is the youngest age ever reported in the literature with malignant fibrous histiocytoma.

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Fibrous tumor of the breast is a rare benign stromal proliferation with atrophy of the epithelial component. Fibrous histiocytoma is the most common soft tissue sarcoma of the breast, accounting for 10-60% of all breast sarcomas.[1,2] Benign fibrous histiocytoma may become malignant with a prognosis that depends entirely on the grade and the size of the tumor.[3] High-grade tumors and tumors larger than 5 cm in diameter are often associated with very poor prognosis.[2,3] It is believed that breast histiocytomas occur at an older age than fibroadenoma,[1-4] and that almost all patients who develop fibrous tumors are premenopausal.[1] We report 2 patients of breast fibrous histiocytoma occurring in 2 young Saudi females; one of them in the pediatric age group. The available treatment options are also discussed.

Case Report. ***Patient One.*** A 26-year-old Saudi nurse who presented in 1997 with a large right breast lump that was first noticed a year earlier. The lump was located in the upper outer quadrant and has suddenly increased in size soon after delivery of her baby. She was lactating for 4 months at presentation and denied any past history of benign breast disease and her family history was negative for cancer. On examination, there was a 5 x 6 x 7 cm non-tender, smooth mass occupying the entire upper outer quadrant of the right breast. She refused fine needle aspiration cytology (FNAC) and therefore surgical excision of a fleshy and vascular tumor was performed. Histology of the excised specimen revealed benign fibrous histiocytoma (**Figure 1**). She was later lost to follow-up, but reappeared 18 months later, complaining of a nodule in the hypertrophic scar of the previous excision. Fine needle aspiration cytology showed no malignancy and excision biopsy revealed cellular fibroma but no evidence of malignancy. She remained well 5 years after the initial diagnosis.

Patient 2. A 14-year-old Saudi schoolgirl presented with a left breast lump that was noticed 4 months prior to presentation. The lump was initially small (an olive size) but later increased in size especially within 2 weeks after FNAC. There was no pain, skin color changes or nipple discharge.

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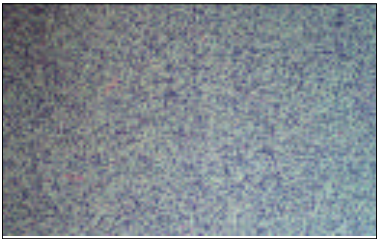

Figure 1

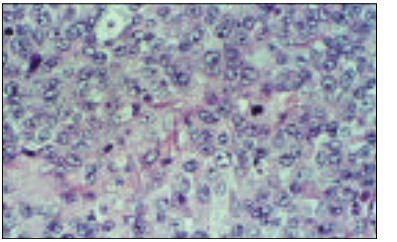

Figure 3

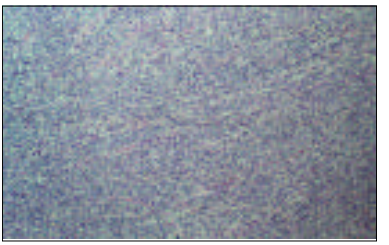

Figure 2

Figure 1 - Benign fibrous histiocytoma of the breast in patient one (hematoxylin & eosin stain x 100).

Figure 2 - High grade poorly differentiated sarcoma of the breast in patient 2 (hematoxylin & eosin stain x 100).

Figure 3 - Magnified view showing nuclear pleomorphism with mitotic figures (hematoxylin & eosin stain x 400).

Menarche started at 12 years and she had no past medical or surgical history. Her family history was negative for benign or malignant breast diseases. On examination, there was a 3 x 4 cm non-tender well-defined and mobile lump in the inner upper quadrant of the left breast. Fine needle aspiration cytology was acellular and revealed fatty tissues only. Tru-Cut biopsy was refused by the patient and a provisional diagnosis of fibroadenoma was made. She underwent surgical excision and the histology showed completed excision with free resection margins and was consistent with benign fibrous histiocytoma. She remained well and asymptomatic with no evidence of recurrence at 6-month follow-up. She presented 6 months later with a small lump underneath the operative scar, which was mobile, non-tender with no evidence of lymphadenopathy. Surgical excision of what looked like a scarred tissue from the previous operative site was performed. Histology showed fibrous histiocytoma of low-grade malignancy. This was confirmed by immunohistochemistry. The patient failed to appear for follow-up, but she presented 3-months later with a larger recurrence along the entire length of the scar in the upper inner quadrant of the left breast, which was mobile and seemingly not adherent to the pectoralis muscles. There was no axillary lymphadenopathy. The chest was clear and no hepatomegaly. Mammography of the right breast, ultrasonography of the liver, chest x-rays and bone scan were all negative of metastatic spread. She underwent a wider excision of this recurrence. At surgery, it was apparent that the lump was infiltrating the pectoralis muscles, which was partially excised together with the specimen. Histology revealed high-grade fibrous histiocytoma with clear resection margins (**Figures 2 & 3**). She was referred to the King Faisal Hospital and Research Center (Oncology Center), Riyadh, Kingdom of Saudi Arabia for possible postoperative radiotherapy, but it was felt that mastectomy was more appropriate. Therefore, mastectomy was performed and she remained well at 12-month follow-up.

Discussion. Soft tissue sarcomas of the breast accounts for <1% of all malignant breast tumors.[3-5] They may arise de novo or after radiotherapy.[2,4,5] Most common histological type is malignant fibrous histiocytoma of the breast, which are often classified into low and high-grade malignant tumors.[2] It is on this grading and the size of the

tumor at the initial presentation that the prognosis is determined. Therefore, high-grade malignant tumors and tumors larger than 5 cm are associated with poor prognosis and more radical and aggressive treatment option should be adopted.[2,3] Older age (>60 years) and grossly involved resection margins are also associated with poorer survival.[2] Lymphatic spread is rare but hematogenous spread is common; most common metastatic sites are lung, bone, brain and subcutaneous tissue. Such distant metastasis often proves to be fatal.[4] Extensive radiological survey of patient 2 revealed no distant metastases. Almost all patients who develop fibrous tumors of the breast are premenopausal. But, it has been reported in a postmenopausal woman who has been on intensive oestrogen therapy.[1] This report is of benign fibrous histiocytoma of the breast in 2 young Saudi females; one of them in the pediatric age group. In both patients the initial clinical presentation was indistinguishable from giant fibroadenoma. In patients 2, the tumor became of low-grade malignancy a year after the initial excision and became of high-grade malignancy within 3 months of the second excision. Such presentation resembles that of 45-year-old woman who presented with a breast lump 52 months following breast conserving surgery and radiotherapy. Surgical excision revealed nodular fasciitis. However, the tumor recurred locally 4 times before the diagnosis of malignant fibrous histiocytoma was finally reached.[6] An alarming feature is the sudden increase in size and rapid growth especially after delivery as in patient one or after FNAC as in patient 2. Such symptoms should alert the clinician to the fact that the lump is unlikely to be an ordinary fibroadenoma. It is believed that breast histiocytomas occurred at an older age than fibroadenoma; 10-20 years over the age of incidence of fibroadenomas with a peak age of sarcoma 40-50 years.[1-4] However, this has not been the case in our 2 patients especially in patient 2. Another cause for rapid tumor growth is intense hormonal therapy.[1] The role of FNAC in diagnosing fibrous histiocytoma is less clear and poses a diagnostic dilemma, but it should be the initial diagnostic procedure for investigating such lesions, as a specific diagnosis is rendered in the majority of cases.[7] The diagnosis can be considered when a cytologic examination reveals a hypercellular, spindle cell smear with osteoclast-like giant cells in the absence of ductal epithelial or myoepithelial cells. The diagnosis should be supported by immunohistochemical and electron microscopic studies.[7] It was unfortunate that FNAC was refused by the first patient and therefore, was deferred. In the second patient, FNAC was acellular; hence Tru-Cut biopsy was offered, but also refused!

Treatment is surgical aiming mainly at complete radical resection of the malignant tumor with clear resection margins.[2] Local recurrence after radical surgery approaches 8%.[8] Axillary clearance is offered in presence of clinically or radiologically detected lymphadenopathy.[8] In patient 2, as the tumor was located in the upper medial quadrant of the breast and in absence of detectable lymphadenopathy, no axillary dissection was performed and a wide local excision was apparently adequate. Later, a more radical surgical treatment was felt more appropriate and therefore mastectomy was performed. The role of postoperative adjuvant therapy remained uncertain. However, radiotherapy seems to reduce the incidence of local recurrence and improves local control[9] and for this reason our second patient was referred for radiotherapy in a tertiary oncology center. Preoperative downstaging may be possible with a neoadjuvant chemoradiation.[9] Chemotherapy is used for widespread sarcomas or for high-grade malignancy, but it does not influence survival rate when administered at an early phase.[9]

Due to the biological and clinical behavior of malignant fibrous histiocytoma, some authors advise a wide excision or even a radical or modified radical mastectomy with axillary clearance.[2,4,5] Others believe that such tumors may arise primarily from the chest wall.[10] This belief is supported by the operative finding of the total presence of the tumor within the pectoralis muscles and the complete absence of mammary glandular tissue in the excised specimen.[10] However, this was not noticed in our 2 patients.

Histologically, the tumor is composed of spindle-shaped tumor cells having polygonal nuclei and plump eosinophilic cytoplasm. Some scattered lymphocytes among the tumor cells and many bizarre multinucleated giant cells may be seen. The nuclei show prominent nucleoli with nuclear pleomorphism and abnormal mitoses (**Figure 3**). On immunohistochemical staining, the cytoplasm of tumor cells are diffusely positive for vimentin and lysozyme, and negative for cytokeratin, epithelial membrane antigen, S-100 protein, and smooth muscle actin.[4]

Our second patient highlights the dilemma facing the treating breast surgeon when encountered with such rare tumors especially in young girls. It is advisable to have a high index of suspicion when faced with what clinically look-like giant fibroadenoma or phyllodes cystosarcoma even in the pediatric age group. Rapid growth of a breast lump after FNAC, hormonal manipulation or changes as that occurring during pregnancy and lactation should raise suspicions that the lump could be a fibrous histiocytoma. Full base-line radiological survey is advised once the diagnosis is made. Initial wide excision and close long-term

follow-up are recommended even in cases of histologically proven benign fibrous histiocytoma, as those may become malignant at any time after the initial surgical excision.

References

1. Miyagawa A, Yuba Y, Haga H, Nishimura S, Kobashi Y, Miyagawa A. Fibrous tumor of the breast in a postmenopausal woman receiving estrogen. ***Pathol Int*** 2001; 51: 123-126.
2. Cha C, Antonescu CR, Quan ML, Maru S, Brennan MF. Long-term results with resection of radiation-induced soft tissue sarcomas. ***Ann Surg*** 2004; 239: 903-909.
3. Jones MW, Norris HJ, Wargotz ES, Weiss SW. Fibrosarcoma- malignant fibrous histiocytoma of the breast. A clinicopathological study of 32 cases. ***Am J Surg Pathol*** 1992; 16: 672-674.
4. Oh SJ, Kim KM, Hong TH, Park WC, Kim JS, Jung SS. Giant cell malignant fibrous histiocytoma of the breast: a case report. ***J Korean Med Sci*** 2004; 19: 477-480.
5. Pollard, SG, Marks PV, Temple LN, Thompson HH. Breast Sarcoma. A clinicopathologic review of 25 cases. ***Cancer*** 1990; 66: 941-944.
6. Pandey M, Mathew A, Abraham EK, Rajan B. Primary sarcoma of the breast. ***J Surg Oncol*** 2004; 87: 121-125.
7. Chhieng DC, Cangiarella JF, Waisman J, Fernandez G, Cohen JM. Fine-needle aspiration cytology of spindle cell lesions of the breast. ***Cancer*** 1999; 87: 359-371.
8. Wiriosuparto S, Krassilnik N, Gologan A, Cohen JM, Wenig B. Malignant fibrous histiocytoma, giant cell type, of the breast mimicking metaplastic carcinoma. A case report. ***Acta Cytol*** 2003; 47: 673-678.
9. Mazanet R, Antman K. Adjuvant therapy of sarcomas. ***Semin Oncol*** 1991; 18: 603-612.
10. Tamir G, Nobel M, Hauben DJ, Sandbank J. Malignant fibrous histiocytoma of the breast. ***Eur J Surg Oncol*** 1995; 21: 210-211.