Case Report

A rare case report of malignant fibrous histiocytoma of male breast

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ABSTRACT

Malignant fibrous histiocytoma (MFH) is an extremely rare tumor of the breast that arises from the connective tissue of glands. Its complex and various histological patterns, adds difficulty to the diagnosis. We report a case of MFH of left breast in a 70-year-old male who presented with gradually increasing painless mass over a period of 1–2 years. Fine-needle aspiration cytology revealed malignant spindle cell lesion and wide local excision was performed without axillary lymph node dissection. Histopathological examination along with immunohistochemistry confirmed the diagnosis of MFH of left breast. To the best of our knowledge, this is the 6th case of MFH of left breast in an elderly male reported in the literature.

Key words: Histiocytoma, male breast, malignant

INTRODUCTION

Malignant fibrous histiocytoma (MFH) is the most common soft tissue sarcoma in adults. It is typically large deep-seated tumor which shows progressive, often rapid enlargement. The most common sites are the extremities (especially the lower limb) and less often the trunk.[1] Primary sarcoma of the breast is a rare tumor and accounts for <1% of all breast malignancies.[2] Breast sarcomas include angiosarcoma, osteosarcoma, liposarcoma, leiomyosarcoma, fibrosarcoma, rhabdomyosarcoma, and MFH. Among these sarcomas, MFH is very rare.[1] MFH is rare high-grade sarcoma and constitutes <5% of all sarcomas in adults and has been rarely seen in the breast.[3]

MFH of the breast can arise "*de novo*" from the connective tissue of the gland or develop in a breast after irradiation for carcinoma.[4] Immunohistochemistry (IHC) plays an important role in the diagnosis of MFH because it is useful to distinguish primary breast sarcoma from nonmesenchymal malignant tumors and to delineate the level of differentiation of primary breast sarcoma.

MFH of the breast is an aggressive tumor with an overall 5-year survival probability of only 50–60%.[1]

In the literature, only five cases of MFH of the breast have been reported[2,5-8] [Table 1]. Herein, we report an extremely rare case of MFH in an elderly male breast.

CASE REPORT

A 70-year-old man presented with gradually increasing painless small mass in his left breast since 1–2 years. The patient complained of enlargement of mass since 6 months. Local examination revealed a mass of 13 cm × 8 cm × 6 cm in his left breast. The mass was not fixed to the chest wall and without axillary lymphadenopathy. The right breast was unremarkable. He had no family history of any malignancy and no previous history of radiotherapy.

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Fine-needle aspiration cytology (FNAC) of the mass revealed malignant spindle cell lesion. Routine hematologic investigations were all within the normal limits. Liver function tests and hormonal assays related to the development of gynecomastia including estrogen, testosterone, prolactin, and gonadotrophic hormones were all within normal limits. With a cytological diagnosis of malignant spindle cell lesion of the breast, the patient underwent wide local excision of left breast mass without axillary lymph node dissection.

On gross examination, the specimen measured 10 cm × 8 cm × 5 cm with overlying ulcerated skin and nipple. Externally the tumor was irregular with nodular surface [Figure 1a]. The cut surface showed tumor mass was lobulated, yellowish white in color with necrotic and cystic areas [Figure 1b].

Histologically, multiple sections from breast mass showed a highly pleomorphic tumor cells arranged in storiform and interlacing fascicles. The tumor cells were spindle-shaped, ovoid and histiocyte-like with eosinophilic cytoplasm. Tumor cells showed marked hyperchromatic, pleomorphic nuclei with prominent nucleoli. Multinucleated tumor giant cells and atypical mitotic figures also shown in Figure 2. Mitotic count was 8–10/hpf with atypical forms. Surgical margins were free of tumor. Histomorphological diagnosis of pleomorphic sarcoma was made.

IHC revealed positive staining for vimentin, CD68 and negative staining for cytokeratin, muscle-specific actin, desmin, S-100 protein, MDM2, CDK4, myogenin [Figures 3 and 4]. The histological and immunohistochemical findings confirmed the diagnosis of MFH of the left breast. A computed tomography scan of the chest, abdomen, and pelvis did not reveal any distant metastasis. Postoperative period was uneventful and patient is free from local recurrence or metastasis in 6-month follow-up after surgery.

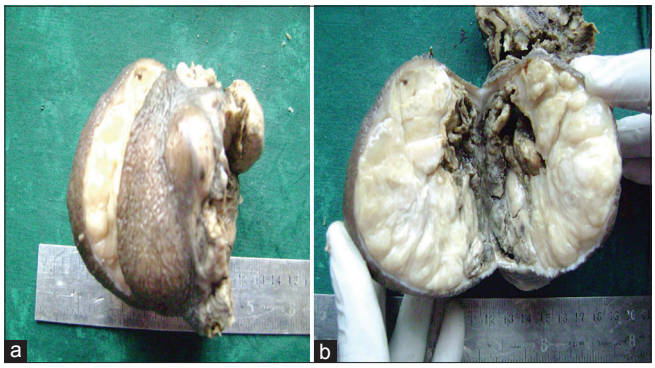

Figure 1: (a) Gross examination - tumor showing irregular with nodular surface (b) Cut section showing lobulated, solid, greyish-white tumor mass with few necrotic and cystic areas

DISCUSSION

Male breast cancer is extremely rare representing <1% of all breast cancers. The most common type of male breast cancer is invasive duct carcinoma (93.7%), followed by papillary (2.6%), mucinous (1.8%), lobular (1.5%), medullary (0.5%), and breast sarcoma (<1%).[6,8] Primary sarcoma of the breast is rare and arises from the mesenchymal tissue of the breast. According to the WHO classification, MFH is a morphological pattern rather than a distinct clinicopathological entity. Nowadays, MFH is synonymous with undifferentiated pleomorphic sarcoma which has become a diagnosis of exclusion and accounting for <5% of adult sarcomas. Primary undifferentiated pleomorphic sarcoma is defined as a group of pleomorphic, high-grade sarcomas showing no line of differentiation.[1]

Undifferentiated pleomorphic sarcoma was first introduced in 1964 by O'Brien and Stout and is the most common soft tissue tumor and mainly involves the lower extremity (49%),

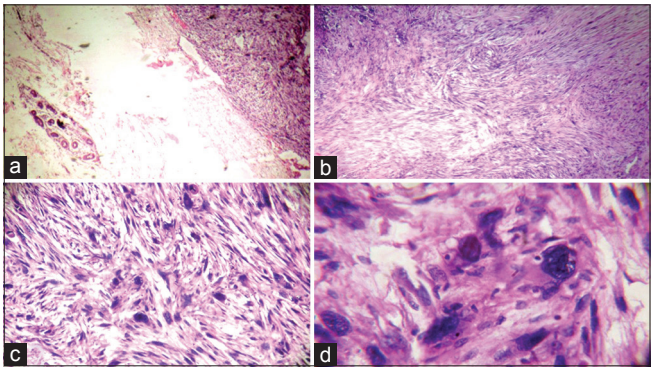

Figure 2: (a-d) Microscopy showed highly pleomorphic tumor cells arranged in storiform and interlacing fascicles with adjacent breast tissue. (a) The tumor cells were spindle shaped, ovoid and histiocyte like with eosinophilic cytoplasm. Tumor cells showed marked hyperchromatic, pleomorphic nuclei with prominent nucleoli. (b and c) Multinucleated tumor giant cells and atypical mitotic figures also seen. (d) H and E, ×400

Table 1: Comparison of case reports in the literature

Case reports	Age (years)	Tumor size	Treatment	Follow-up (months)
Lunde *et al.* 1986[5]	66	NA	Surgery + axillary lymphadenectomy + radiotherapy	18
Mahalingam *et al.* 2011[6]	72	3.1 cm × 2.3 cm	Surgery + radiotherapy	36
Hartel *et al.* 2011[7]	67	NA	NA	NA
Jeong *et al.* 2011[2]	76	1.5 cm × 1 cm	Surgery	NA
Kocak Uzel *et al.* 2013[8]	37	12 cm × 9 cm × 8.5 cm	Surgery + radiotherapy	42
Present case, 2015	70	10 cm × 8 cm × 5 cm	Surgery	6

NA: Not applicable

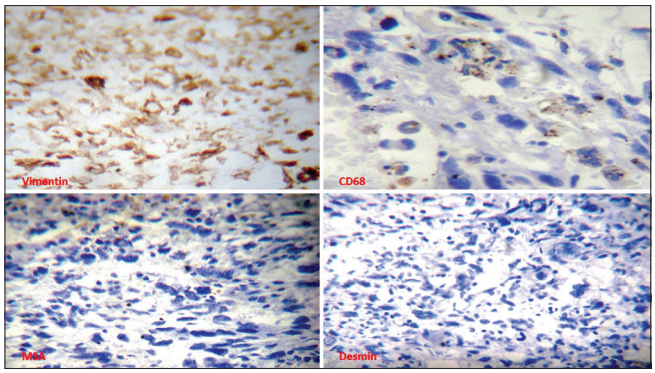

Figure 3: Immunohistochemistry revealed positive staining for vimentin, CD68 and negative staining for muscle-specific actin and desmin

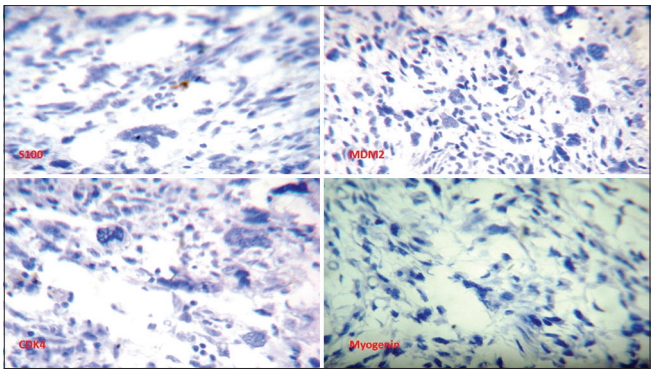

Figure 4: Immunohistochemistry revealed negative staining for S-100 protein, MDM2, CDK4, and myogenin

upper extremity (19%), retroperitoneum and abdomen (16%), whereas in the breast, it is extremely rare.[9] Histologically it is categorized into four types: Storiform-pleomorphic, myxoid, inflammatory and giant cell types.[1] Our case showed storiform-pleomorphic variant. The incidence of male breast cancer increases with age with a peak in the sixth and seventh decades. The clinically majority of patients presented with gradually or rapidly progressing breast swelling, nipple discharge, ulceration, nipple retraction and may be associated with pain. Ultrasound examination and mammography does not show specific features to obtain a differential diagnosis between various sarcomas. In our case, ultrasound examination and mammography were not done.

Our patient had a clinical presentation of progressive, painless mass in his left breast without axillary lymphadenopathy. FNAC was done and revealed as malignant spindle cell lesion. Wide local excision was carried out which histopathologically and immunohistochemically confirmed as MFH.

Microscopically MFH show marked pleomorphic spindle cells arranged in storiform pattern and admixed with bizarre giant cells, foamy cells and atypical mitotic figures. Before making the diagnosis of MFH of the breast it is necessary to rule out the epithelial differentiation based on thorough gross, microscopic and immunohistochemical examination. Differential diagnosis includes metaplastic (sarcomatoid carcinoma), malignant phyllodes tumor, inflammatory myofibroblastic tumor, and myofibrosarcoma but characteristic histological features and IHC ruled out these lesions.

Based on the review of literature treatment of choice for MFH of the breast is local surgical resection with or without axillary lymph node dissection. Axillary dissection has been considered unnecessary for MFH of the breast since these tumors rarely spread through the lymphatic system. The role of adjuvant chemotherapy and radiation also has been unclear. However, radiotherapy should be considered when surgical margins are inadequate or microscopically involved.[10] In our case, wide local excision without axillary dissection was done, and chemotherapy or radiotherapy was not given since the surgical margins were free of tumor.

MFH of the breast is aggressive, fast-growing tumor with a high rate of local recurrence (44%) and distant metastases (42%), particularly to the lungs (82%), bony skeleton, pleura, and liver; regional lymph nodes involvement ranges from 12% to 32%.[10] The adverse prognostic factors in MFH include the size of the tumor, the presence of the distant metastasis and older age.[7]

CONCLUSION

MFH of the breast is a very rare tumor, especially in males and present diagnostic difficulty. Thorough histopathological examination and appropriate IHC are mandatory for accurate diagnosis. Surgical excision of the tumor with adequate margin is the most significant prognostic factor which helps to prevent local recurrence and to improve survival rate.

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Conflicts of interest
There are no conflicts of interest.

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