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Inflammatory Myofibroblastic Tumor of the Breast Mimicking Malignancy in an Elderly Male

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Background: Inflammatory myofibroblastic tumor (IMT) is a rare, distinctive lesion composed of a proliferation of myofibroblastic spindle cells accompanied by an inflammatory infiltrate. It was first described in the lung, but its occurrence at various extrapulmonary sites has also been reported. The literature mentions only a handful of cases of IMT in the breast and only 1 case in the male breast. We report the second case of IMT in the male breast.

Case Report: A 60-year-old male presented with a large, lobulated lump in the left breast that had progressively increased in size during the past year. The lump measured 15 × 10 cm. Ultrasonography revealed a solid mass lesion with regular borders in the subcutaneous plane of the left anterior chest wall. Fine-needle aspiration cytology showed a cellular mesenchymal tumor. Macroscopically, the nodule was firm, circumscribed, and yellow. On microscopic examination, the tumor was composed of bland spindle cells arranged in sheets and short fascicles along with a rich inflammatory infiltrate comprising predominantly plasma cells, admixed with lymphocytes, neutrophils, and eosinophils. On immunohistochemistry, the tumor cells were positive for vimentin, focally positive for smooth muscle antigen, and negative for anaplastic lymphoma kinase, CD34, S100, β-catenin, and cytokeratin. Thus, a final diagnosis of IMT was rendered.

Conclusion: IMT is a rare entity with intermediate clinical behavior. Knowledge of this entity and its recurrence and metastatic potential is of paramount significance to guide appropriate treatment and follow-up.

Keywords: *Breast neoplasms–male, myofibroblasts*

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INTRODUCTION

Various benign and malignant conditions are known to occur in the male breast.[1] Inflammatory myofibroblastic tumor (IMT) is a rare, distinctive lesion composed of a proliferation of myofibroblastic spindle cells accompanied by an inflammatory infiltrate.[2] Previously known by various names such as inflammatory pseudotumor, plasma cell granuloma, histiocytoma, and fibroxanthoma, IMT was first described in the lung in 1939, but its occurrence at various extrapulmonary sites has also been reported.[2,3] The development of IMT in the breast has rarely been described in the literature. The single case documented in the male breast occurred after recent mechanical trauma.[4] However, in our case, the presentation of the tumor was spontaneous.

CASE REPORT

A 60-year-old male presented with a large, lobulated lump in the left breast that had progressively increased in size during the past 1 year. The lump measured 15 × 10 cm. Ultrasonography revealed a solid mass lesion with regular borders in the subcutaneous plane of the left anterior chest wall abutting the pectoralis muscle. Fine-needle aspiration cytology showed a cellular mesenchymal tumor. Mastectomy was performed, and the sample was sent for histopathologic assessment. Gross examination showed a tumor in the upper outer quadrant measuring 10 × 7.5 × 6 cm. The cut section of the tumor was homogeneous, grey-white, and fleshy (Figure 1). On histopathologic examination, a variably cellular tumor with well-defined margins was identified. The tumor was composed of oval to spindle-shaped cells arranged in sheets and short fascicles. The cells had vesicular nuclei with minimal atypia and a moderate amount of eosinophilic cytoplasm. Stroma showed hyalinization and a rich inflammatory infiltrate comprising predominantly plasma cells, admixed with lymphocytes, neutrophils, and eosinophils (Figure 2A). Increased vascularity, consisting of congested and dilated blood vessels, was observed. No epithelial or ductal elements were found on extensive grossing. Deep resection margins and other resection margins were free of tumor. On immunohistochemistry, the tumor cells were positive for vimentin; focally positive for smooth muscle antigen (SMA) (Figure 2B); and negative for anaplastic lymphoma kinase (ALK), CD34, β-catenin, and cytokeratin (CK). Thus, a final diagnosis of IMT was rendered. The patient is being followed and is doing well 6 months after the surgery.

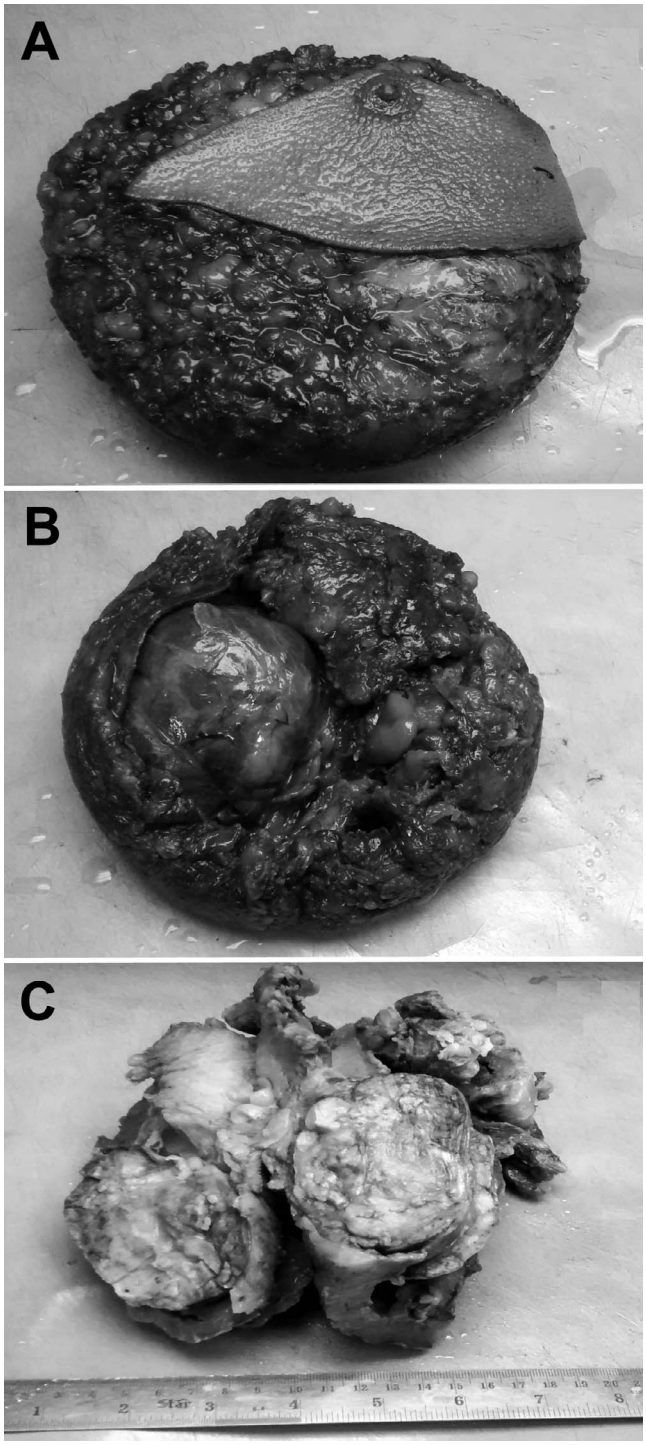

Figure 1. Mastectomy specimen (A) shows a well-defined tumor (B) that appears homogeneous, grey-white, and fleshy on cut section (C).

DISCUSSION

Diseases of the male breast encompass a myriad of benign and malignant conditions, similar to those occurring in the female breast. Most lesions are benign, gynecomastia being the most common. Benign mesenchymal tumors of the male breast include lipoma, pseudoangiomatous stromal hyperplasia, granular cell tumor, fibromatosis, myofibroblastoma, schwannoma, IMT, and hemangioma.[1,2]

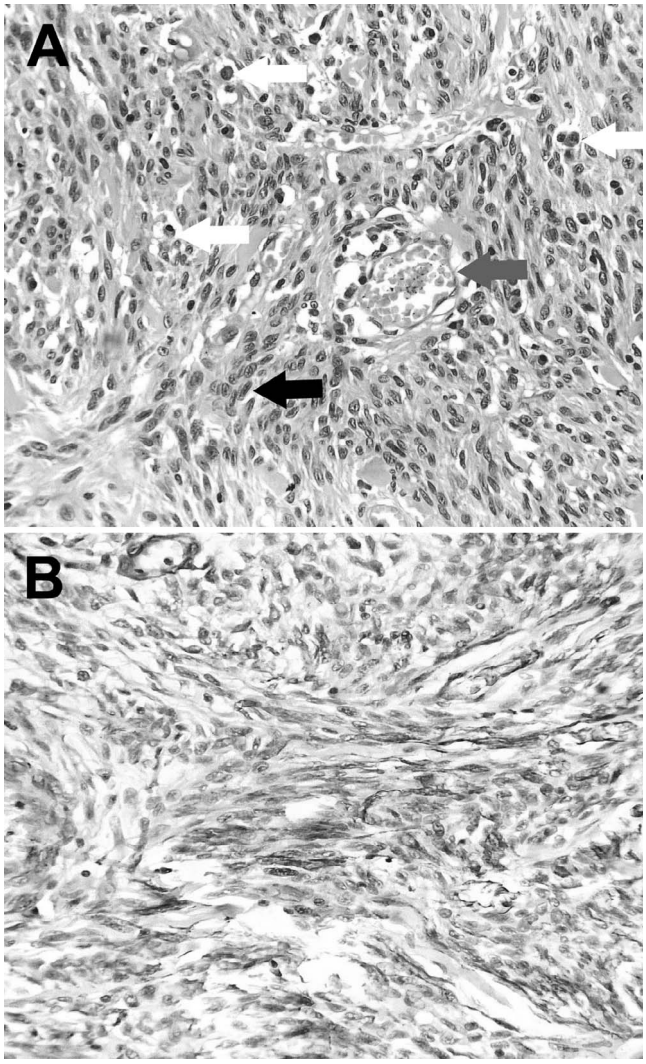

Figure 2. A: Spindle cells are arranged in sheets and short fascicles admixed with plasma cells (hematoxylin and eosin stain, ×400). The white arrows show lymphoplasmacytic infiltrate, the black arrow shows spindle tumor cells, and the grey arrow shows a congested blood vessel. B: Immunohistochemistry shows focal smooth muscle antigen positivity in tumor cells (×400).

IMTs are tumors of unknown origin that fall into the category of tumors with intermediate malignant potential according to the World Health Organization classification of soft tissue tumors.[2] The most common site of involvement is the lung. Extrapulmonary sites include the mesentery and omentum, soft tissue, pelvis, mediastinum, bone, larynx, central nervous system, and rarely the breast.[2,5,6] So far, only 1 case of IMT occurring in the male breast has been reported.[4]

These tumors usually show a predilection for children and young adults, but cases have also been reported as late as the eighth decade of life.[2] Patients generally present with nonspecific symptoms, depending on the site of involvement. Pulmonary tumors may cause cough, chest pain, or, less often, hemoptysis, while abdominal tumors are associated with abdominal pain or intestinal obstruction.[2,5] In the breast, they generally present as a palpable, slightly tender mass that is closely adherent to

skin.[7] Because this presentation overlaps with that of malignancy, which is more commonly seen in older men, it was our first differential diagnosis on clinical examination. However, the imaging finding of a solid mass with regular margins lowered the suspicion of malignancy. IMTs predominantly have well-defined borders on radiology. However, cases with ill-defined masses and with focal areas of irregularly marginated acoustic shadowing without a mass configuration on ultrasonography have been reported.[7]

On gross examination, IMTs are circumscribed tumors ranging in size from 1-20 cm. The cut surface is firm and fleshy, with areas of calcification, hemorrhage, and necrosis in a few cases.[2,5] Histopathologic examination shows myofibroblasts and inflammatory cells arranged in 1 of 3 patterns: a myxoid/vascular pattern, a compact spindle cell pattern, and a hypocellular fibrous (fibromatosis-like) pattern.[2] Our case demonstrated a compact spindle cell pattern comprising a cellular proliferation of uniform spindle cells with a fascicular or storiform architecture in a collagenous stroma. Differential diagnoses that were considered with the present histomorphology included desmoid fibromatosis, nodular fasciitis, myofibromatosis, phyllodes tumor, and metaplastic carcinoma.[2] The absence of mitoses, necrosis, and atypia ruled out malignancy. Negative expression of CD34, β-catenin, and CK ruled out phyllodes tumor, fibromatosis, and metaplastic carcinoma, respectively. Coexisting prominent inflammatory infiltrate and SMA positivity in tumor cells favored the diagnosis of IMT instead of other benign differential diagnoses. Seldom, pleomorphism, atypical mitotic figures, necrosis, and vascular invasion may be seen, and these features do not necessarily favor malignancy.[2,6]

The pathogenesis of IMT is uncertain. Initially thought to be a reparative postinflammatory condition or an autoimmune condition, IMT is now defined as a neoplastic process.[4,8] Rearrangements involving the ALK locus on chromosome band 2p23 are found in 50% of IMTs by fluorescence in situ hybridization, supporting the neoplastic nature of IMT.[8] Clonality has also been demonstrated by cytogenetic analysis, with the finding of 9p deletion in 3 cases of IMT.[9]

Treatment for most patients with circumscribed IMTs consists of wide excision with negative resection margins.[4,5,7] The recurrence rate varies by anatomic site, ranging from 2% for pulmonary tumors to 25% for extrapulmonary lesions.[5] Tumors with ill-defined morphology, large tumor size (>3 cm), and incomplete resection have been associated with higher recurrence rates.[2,4] Distant metastasis of IMT is rare, occurring in 5% of cases.[2,5,6] Thus, regular follow-up for detection of recurrence, metastasis, or malignant transformation is highly recommended.[10,11]

CONCLUSION

IMT is a rare entity composed of a proliferation of myofibroblasts and plasma cells. The literature mentions only a handful of cases of IMT in the breast and 1 case in the male breast. Knowledge of this entity is of paramount significance to avoid overdiagnosis and aggressive therapy as a malignant tumor.

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