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Myofibroblastoma of Male Breast: A Rare Case Report

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Abstract: ***Myofibroblastoma (MFB) is an extremely rare benign tumour that can arise in various organs, but commonly in the breast. Myofibroblastoma of the breast is a rare benign stromal tumour seen predominantly in men in older age. The gross appearance is that of a well-circumscribed nodule, characteristically small, seldom exceeding 3 cm. Furthermore, myofibroblastoma can arise in extra mammary sites, along the milk-line. . Radiological imaging is nonspecific in MFB, and pathological examination of needle biopsy or surgically resected specimen is necessary for the diagnosis. Surgery is recommended and considered curative without additional treatment; however, patients should be followed-up. One case of MFB of the breast is described here since it is a very rare case and pre operative diagnosis is challenging.***

Keywords: Male breast tumour, Rare tumour of male breast, Benign tumor of male breast

1. Introduction

Myofibroblastoma (MFB) is an extremely rare benign tumour that can arise in various organs and tissues, but most commonly in the breast. It accounts for less than 1% of breast tumours [1]. It was first described by Wargotz et al in 1987, who reported 16 cases of this disease predominantly in men [2]. MFB is usually identified as an asymptomatic, slow-growing mass, well-defined, nontender and mobile on clinical examination [3–5]. MFB can arise in extra-mammary sites, along the milk-line and have been reported in the literature different sites including axilla, tonsil, lung, rectum, meninges, prostate, parotid gland and tongue [3–5]. Although it is rare, it can add real challenges and difficulties to the preoperative planning and diagnosis due to its similarities to other benign and malignant breast lesions [6].

To our knowledge only 70 cases were described in the literature (7). We herein report the clinical, radiological and pathological findings observed in one MFB case treated in the Department of General Surgery.

2. Case Presentation

In 2021, a 75-year-old man presented at the Department of General Surgery of KPC Medical College (Kolkata, India) with a slowly enlarging painless palpable mass in his Right breast of 2 years and pain for 6 months. There was no history of breast trauma, associated breast tenderness or nipple discharge. The patient had no previous history of previous breast pathology, nor any family history of breast diseases. He is a known case of systemic hypertension.

Clinical examination revealed no skin changes or nipple retraction, and there were not lymphadenopathies at the axillary sites. The lump was smooth, non tender, round and well-defined, adherent to the overlying skin, freely mobile, located in the upper retroareolar site of his right breast. Axillary lymph nodes were not palpable and abdominal and testicular examinations were both unremarkable.

Ultrasonography examination revealed a well circumscribed heterogenous SOL (61x49x23mm) noted in the outer quadrant of Right breast without obvious internal vascularity (Fig.1). A subsequent fine needle aspiration cytology (FNAC) study of the lesion has shown this to be gynecomastia.

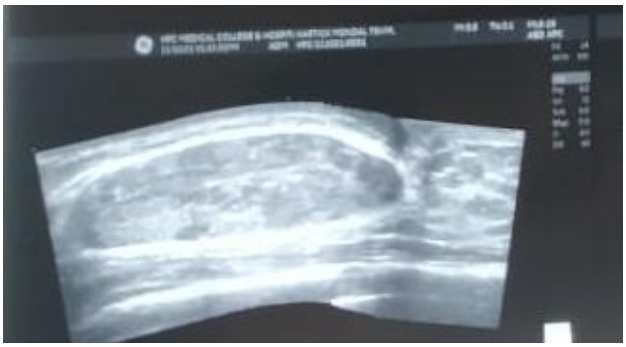

Figure 1: Ultrasonography showing a well circumscribed heterogenous SOL (61X49X23mm)

Following multidisciplinary team review, a decision was made to perform a wide local excision of the tumour. Subsequently, macroscopic histopathology revealed a well circumscribed soft specimen measuring 6X6X2cm surrounded by adipose tissue (Fig.2 and 3) and cut section showed whitish nodular myxoid appearance.

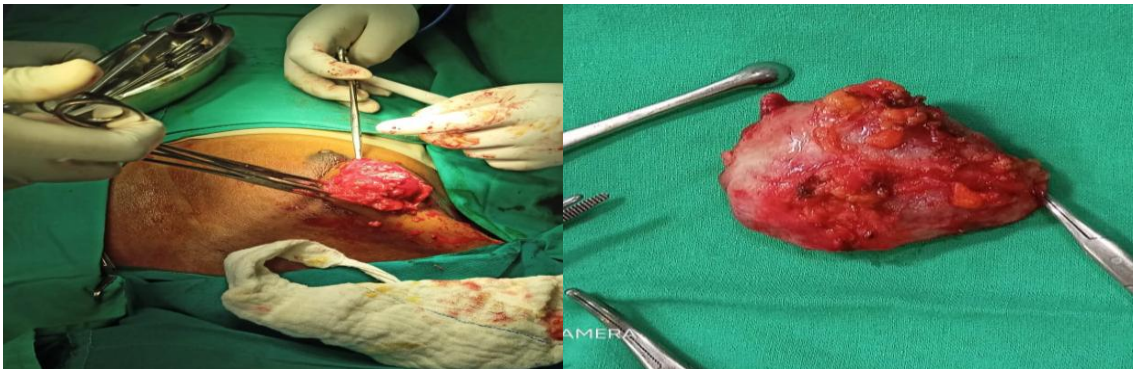

Figure 2 and Figure 3: Macroscopic appearance of the breast tumour

Microscopically, show sheets of stellate and fusiform, spindle shaped neoplastic cells in stroma having myxoid areas along with prominent arborizing plexiform vasculature at places. Stroma also shows areas of collagenisation and mixed inflammatory cell infiltration (Fig 4 and 5). The circumferential resection margins show involvement of tumour. All these features are suggestive of Low grade fibromyxoid sarcoma.

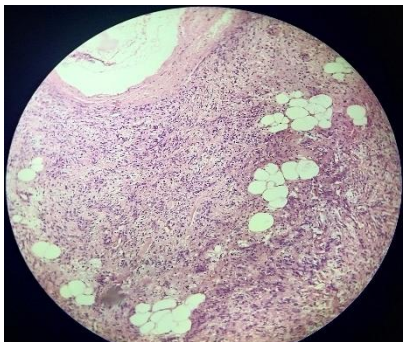

Figure 4: Uniform looking spindle shaped cells with bland nuclear features arranged in short fascicles admixed with variable amount of fat

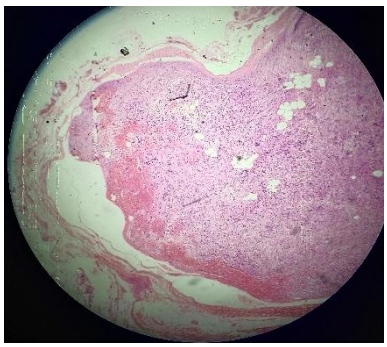

Figure 5: Well circumscribed tumour (4X magnification)

Immunohistochemistry of HP slides showed immunoreactivity for CD34. Tumour cells were negative for S100, p63, Beta catenin and cytokeratins. The Ki67 proliferative index was <1%. The overall Histomorphological and immunohistochemical profile is in favour of myofibroblastoma.

Although wide local excision was performed with a curative intent, the neoplasm abutted the circumferential margin of the resection, and therefore complete excision of the tumour could not be guaranteed. Follow-up ultrasound scans of breast post-operatively are planned to detect any evidence of recurrence.

3. Discussion

MFB was first described in 1981 and was named by Wargotz *et al*. [8] in 1987, using a series of 16 cases in which 11 patients were men and the average age was 63 years. In 2001, a case of extramammary MFB was described by McMenamin and Fletcher [4]. Subsequently, few other cases of MFB arising in older men (between 60 and 70 years old) have been described in the literature, as spindle cell lesion of the male breast associated with gynaecomastia [5]

Soft tissue neoplasms of the breast that are composed of myofibroblasts have been classified as myofibroblastomas [2]. Myofibroblasts are spindle-shaped or fusiform mesenchymal cells derived from fibroblasts and are present in small numbers in all tissues. Proliferation of myofibroblasts is seen in various conditions including inflammatory reactions, fibromatosis and some sarcomas [9]

Non-specific imaging of this type of tumour necessitates the support of histopathological analysis for correct diagnosis. In our case, patient underwent wide local EXCISION OF THE LUMP after the only execution of fine needle aspiration cytology. Myofibroblasts are distinguished from spindle myo-epithelial cells largely on the basis of their distribution, immunohistochemical staining and electron microscopic characteristics [10]. Depending on their phenotypic state, both these types of cells may be reactive with anti-actin antibodies. Myoepithelial cells are typically positive for protein S-100 and cytokeratin in their epithelial phenotype, but myofibroblasts are negative. Their most common immunoprofile is diffuse desmin and CD 34 positivity [11]. Histological features and immunohistocytochemical features in this case were those of a myofibroblastoma. Microscopically, myofibroblastomas can be divided into five sub-types; classical, epitheloid, collagenised, cellular and infiltrative. In this case, the myofibroblastoma had features of mixed cellular, infiltrative and collagenised type.

4. Conclusion

Myofibroblastoma is a rare benign tumour of the breast. The tumour described in this report is unusual owing to its presentation, with gradual enlargement but rapid development of pain mimicking a malignant tumour.

Malignant neoplasms, such as stromal sarcoma, malignant fibrous histiocytoma and spindle-cell sarcoma, or metaplastic carcinoma should not be confused with a myofibroblastoma. The clinical significance of this entity lies primarily in its recognition as a distinctive benign neoplasm.

Additionally, malignant transformation has not been reported yet. There is only one case in the literature of recurrence of MFB at the previous excisional site [5]. However, annual follow-up with ultrasonography and bilateral mammography (above 50 years old) is recommended. Further studies with larger number of patients and longer follow-up are necessary to draw more validated conclusions.

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