Case Report

Giant Myofibroblastoma of the Male Breast: A Case Report and Literature Review

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Abstract

Myofibroblastomas are soft-tissue neoplasms that are thought to arise from myofibroblasts. They are mostly observed in males 41–85 years of age; however, this lesion also occurs in women. The usual clinical presentation is a unilateral painless lump that is not adherent to overlying or underlying structures. Microscopically, myofibroblastomas can be divided into 5 subtypes: classical, epithelioid, collagenised, cellular, and infiltrative. Mammary ducts and lobules are absent in the typical histological subtypes and the adjacent breast parenchyma may form a pseudocapsule. The majority of myofibroblastomas are immunoreactive for CD34, desmin, smooth muscle actin, and vimentin and are negative for cytokeratin and S-100 protein. We present a case of a giant myofibroblastoma arising in the background of gynecomastia in an adult male.

Keywords: *breast, gynecomastia, mesenchymal, myofibroblastoma, spindle cell*

Introduction

Myofibroblastoma is a rare, benign mesenchymal tumour of the breast that is thought to arise from myofibroblasts (1). Cases in the literature are mostly reported in males 41–85 years of age; however, this lesion also occurs in women (2). In addition to the breast, myofibroblastomas have also been reported at extramammary sites such as the popliteal fossa, head, neck, vulva, buttocks, groin, and paratesticular region (3). Since its first description by Wargotz et al. (2), less than 70 cases have been reported in the literature (2). Grossly, they are usually well circumscribed and small, seldom exceeding 3 cm. Microscopically, a myofibroblastoma of the breast is a mesenchymal tumour that is well demarcated from the adjacent parenchyma, lacks epithelial breast elements, and is composed of fascicles of spindle cells separated by thick collagen bands (4). Cases of associated myofibroblastoma and gynecomastia are very rare, and only a few cases have been reported in the literature (5).

Case Report

A 62-years-old man presented with a 9-year history of a large lump in the right mammary region. He was taking oral hypoglycemics and antihypertensives. He was recently diagnosed with hypothyroidism. The patient underwent an open nephrolithotomy in 1976 for a renal calculus. In addition, he had undergone angioplasty and stenting in 1996 for coronary artery disease. There was no family history of breast cancer. The lump initially appeared as a small and asymptomatic swelling 5 years prior, and a trucut biopsy performed at a private hospital suggested gynecomastia. The patient presented to us with an extremely large lump underneath the areola measuring 18 × 14 cm in maximum diameter (Figure 1). The lump was nontender, lobulated, well defined, and freely mobile with respect to the underlying muscular plane. No regional lymph nodes were palpable. Routine laboratory investigations were within normal limits. A mammogram of the breast revealed a large dense lesion with no microcalcifications. Ultrasonography showed a well-defined heterogeneous hyperechoic mass measuring 16 × 12 cm. An ultrasound of the scrotum was suggestive of a moderately sized bilateral hydrocele. A trucut biopsy of the breast performed during the present admission showed fibroadipose tissue. Therefore, the tumour was completely excised along with the nipple and areola. The post-operative period was uneventful. A gross tissue examination revealed a 16 cm

nodular lobulated tumour with a reddish-brown to whitish-yellow cut surface. A microscopic examination indicated that the tumour cells were arranged in an ill-defined fascicular pattern and were separated by thick collagen bundles (Figure 2). The cells were mostly spindle shaped and monomorphic. Adipose tissue was observed at the periphery of the tumour. No breast epithelial elements or necrosis were identified within the tumour. The tumour cells showed scant mitoses and immunopositivity for smooth muscle actin, vimentin, Bcl-2, and CD34 (Figure 3) and were negative for cytokeratin and S-100 protein. Based on the morphology and immunohistochemical results, a diagnosis of myofibroblastoma was made.

Discussion

A myofibroblastoma is a rare, benign mesenchymal tumour of the breast that is composed of myofibroblasts (1,2). Cases in the

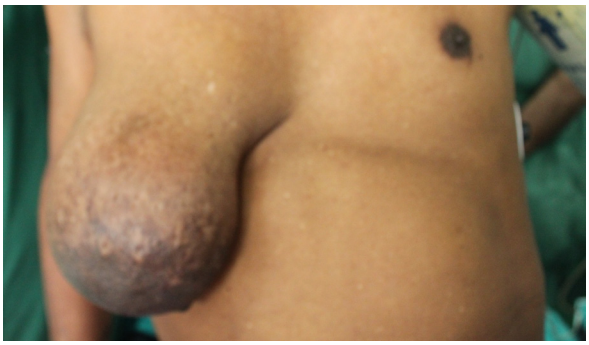

Figure 1: A large, pendulous right breast.

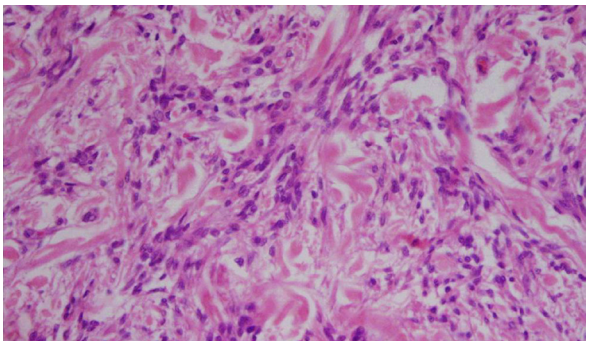

Figure 2: Haphazardly arranged short fascicles of oval-to-spindle-shaped cells separated by thick eosinophilic collagen bands (haematoxylin and eosin staining, 400 × magnification).

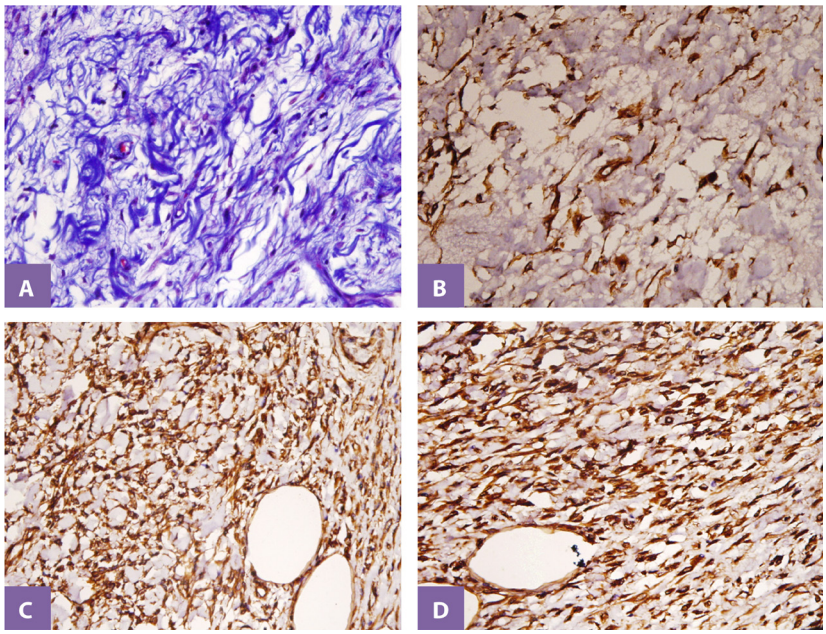

Figure 3: (A) Thick collagen bands, Masson's trichrome stain (200 × magnification). (B) Tumour cells immunopositive for vimentin (200× magnification). (C) CD34 immunopositivity in tumour cells (200 × magnifications). (D) Tumour cells immunopositive for BCL2 (200 × magnification).

literature are mostly reported in men, 41–85 years of age; however, this lesion also occurs in women (2). The usual clinical presentation is a unilateral painless lump that is not adherent to overlying or underlying structures. Bilaterality and unilateral multicentricity are rare. The association between myofibroblastoma and gynecomastia is very rare (5). Radiologically, myofibroblastomas are homogenous, lobulated, and well-circumscribed lesions, typically lacking microcalcification. Ultrasonography cannot often differentiate a myofibroblastoma from a fibroadenoma (6). Microscopically, myofibroblastomas can be divided into 5 subtypes: classical, epithelioid, collagenised, cellular, and infiltrative. Mammary ducts and lobules are absent in the typical histological subtypes and the adjacent breast parenchyma may form a pseudocapsule. Myofibroblast proliferation may also be observed in inflammatory reactions, fibromatosis, and some sarcomas (7). Microscopic analyses have also demonstrated that myofibroblasts resemble myoepithelial cells, but they can be distinguished either by immunohistochemical staining or electron microscopic characteristics (8). The majority of myofibroblastomas are immunoreactive for CD34, desmin, smooth muscle actin, and vimentin and are negative for cytokeratin and S-100 protein. The epithelioid variant may be negative or only focally positive for CD34. Rarely, a myofibroblastoma also shows nuclear positivity for the oestrogen, progesterone and androgen receptors (9). Compared to malignant spindle-cell tumours, myofibroblastomas are usually less cellular and do not show a high mitotic rate, atypical mitoses, anaplasia, or necrosis. (10). Myofibroblastomas behave in a benign fashion and an excision biopsy is usually adequate for this tumour. No recurrence or metastasis has been described in the literature.

Conclusion

A careful clinical and histopathological examination along with the use of immunohistochemical and ultrastructural techniques are necessary to correctly diagnose a unilateral, extremely large male breast lump, which may clinically simulate gynecomastia, phyllodes tumour, or carcinoma. Moreover, this diagnosis may be missed radiologically or in fine-needle aspiration cytology and biopsy specimens.

Authors' Contribution

Conception and design: KK
Analysis and interpretation of the data: VS
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Critical revision of the article for important intellectual content: AS
Provision of study materials or patients: LS

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