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Case report
Olgu sunumu

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Breast myofibroblastoma: Report of two cases with literature review

Meme myofibroblastom: Literatür taraması ile iki olgu sunumu

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Abstract
Myofibroblastoma (MFB) is a rare mesenchymal benign tumor that arises from the stromal structures of the breast tissue. It occurs in the elderly without sex predilection. Its clinical and radiological presentations are aspecific, thus MFB may be confounded with other malignant and benign breast lesions. However, the main histological characteristic of MFB is the presence of spindle cellsin a collagenous background. In immunohistochemistry, MFB is positive for vimentin and CD34 with a noticeable low mitotic activity. Surgical excision remains the treatment cornerstone, with excellent outcomes. We retrospectively reviewed the records of all the patients who underwent surgery of breast from 01 January 2012 to 31 December 2018. We found two cases of breast myofibroblastoma. The first was a young woman aged 17 years, and the second was a male aged 87 years. The main symptom was a palpable breast lump in both patients. The radiological work up concluded to a benign lump in the young woman, and a suspicious breast lump in the man, to whom we performed a core biopsy. Histology staining showed the features of MFB. The woman underwent a lumpectomy, and the man underwent a mastectomy. Final histological staining showed spindle cells with a collagen matrix. The cells were positive for CD34, vimentin, and actin. Those features were compatible with the diagnosis of breast MFB. The aim of this report was to describe the clinical, radiological and histological features of breast MFB.
Keywords: Myofibroblastoma, Breast, Spindle cell

Öz
Miyofibroblastom (MFB), meme dokusunun stromal yapılarından kaynaklanan nadir bir mezenkimal benign tümördür. Cinsiyet farkı olmadan yaşlılarda daha sık görülür. Klinik ve radyolojik sunumları özgündür, bu nedenle MFB diğer malign ve benign meme lezyonlarıyla karıştırılabilir. Bununla birlikte, MFB'nin ana histolojik özelliği, iş mili hücrelerinin kollajen zemininde varlığıdır. İmmünohistokimyada MFB, belirgin düşük mitotik aktiviteye sahip vimentin ve CD34 için pozitiftir. Cerrahi eksizyon, mükemmel sonuçlarla tedavi temelini korumaktadır. 01 Ocak 2012 - 31 Aralık 2018 tarihleri arasında meme ameliyatı geçiren tüm hastaların kaydını retrospektif olarak inceledik. İki meme miyofibroblastom olgusu bulduk. Birincisi 17 yaşında genç bir kadındı, ikincisi 87 yaşında erkekti. Her iki hastada da ana semptom elle tutulur bir meme yumrusuydu. Radyolojik çalışma genç kadında iyi huylu bir tümör ile sonuçlandı, ancak erkekte görüntüler şüpheli bir tümör ortaya çıkardı. Adam için çekirdek biyopsi yaptık. Histoloji boyama MFB'nin özelliklerini gösterdi. Kadına lumpektomi, erkeğe mastektomi uygulandı. Son histolojik boyama, kollajen matriksli iğ hücrelerini gösterdi. Hücreler CD34, vimentin ve aktin için pozitifti. Bu özellikler meme MFB tanısı ile uyumluydu. Bu raporun amacı, meme MFB'nin klinik, radyolojik ve histolojik özelliklerini tanımlamaktı.
Anahtar kelimeler: Myofibroblastom, Meme, İğ hücresi

Introduction

Myofibroblastoma (MFB) is a rare benign spindle cell tumor arising from mesenchymal stroma [1]. It affects older men and postmenopausal women [2]. MFB was reported in severe sites such as the head, neck, extremities, buttock, vulva, testicular region, inguinal areas, and breast [2-7]. Due to its rarity and the lack of pathognomonic clinical and radiological signs, the diagnosis may be postoperative. On the histopathological, and immunohistochemical front, MFB is characterized by the presence of spindle cells enshrined in a collagen matrix, low mitotic activity, and positivity of CD34 and vimentin [2].

We aimed to report two cases of MFB occurring in different genders with different ages and focusing on differential diagnosis.

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Case presentation

Case 1

A 17-year-old female presented with complaints of a left palpable breast mass since two years. The patient didn't have a remarkable personal or familial medical history. Physical exam revealed a well-defined, smooth and mobile mass located in the external upper quadrant of the left breast. There was no palpable lymph node in the armpit region. Ultrasonography showed a heterogeneous, hypoechoic, well-circumscribed lesion without microcalcifications (Figure 1). The mass measured 22 mm and was classified as Breast Imaging-Reporting and Data System 3 (BI-RADS). We didn't perform a core biopsy due to the probable benignity of the breast lesion. The patient underwent a lumpectomy. The histological features revealed a myofibroblastoma of the breast with free margins. The patient is currently free of disease with 8 years of regular follow-up.

Case 2

A 87-year-old male consulted for a right breast mass that had been evolving for 3 years. On physical examination, there was a voluminous, firm and mobile mass in the right breast that measured 7 cm. The contralateral breast was free of abnormalities and the lymph nodes were normal.

On breast ultrasound, a well-defined, hyperechoic, heterogeneous, voluminous mass of suspicious appearance was detected in the right breast. On mammography, the mass was hyperdense and involved the whole right breast (Figure 2). The lesion was considered BI-RADS 4c.

A micro biopsy of the mass was performed, and histological findings revealed a breast myofibroblastoma. The patient underwent a mastectomy for the large tumor's size. The definitive histological assessment showed the features of MFB of the breast, with free margins. The patient didn't experience any recurrence within a follow-up of 6 months.

Histological findings

Macroscopically, the lesions were well-defined, with a grayish color. We did not observe hemorrhage in any of the sections.

Microscopically, spindle cell was the predominant cell type. In the second case, we detected mature adipocytes without any epithelial structure. The spindle-shaped cells were arranged in irregular short bundles separated by hyalinised collagen tissue. In both cases, we found rounded cells with a pseudo-epithelial appearance without clear cytonuclear atypia (Figure 3). We didn't observe mitosis or necrosis.

The immunohistochemistry was established in both cases. These fusiform cells were positive for actin, vimentin, and CD34, and negative for Cytokeratin and βCatenin (Figure 4). The proliferation index Ki 67 was estimated at 5% in both cases. These morphological and immunohistochemical findings were compatible with the diagnosis of breast myofibroblastoma.

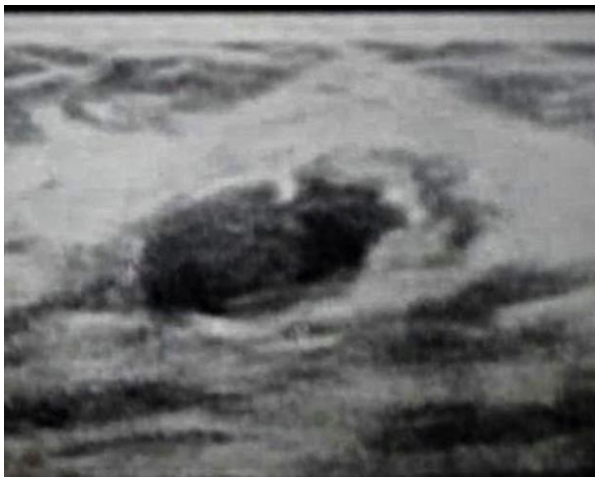

Figure 1: Ultrasonography image showing a heterogeneous, hypoechoic, well-circumscribed lesion of left breast, and measured 22mm

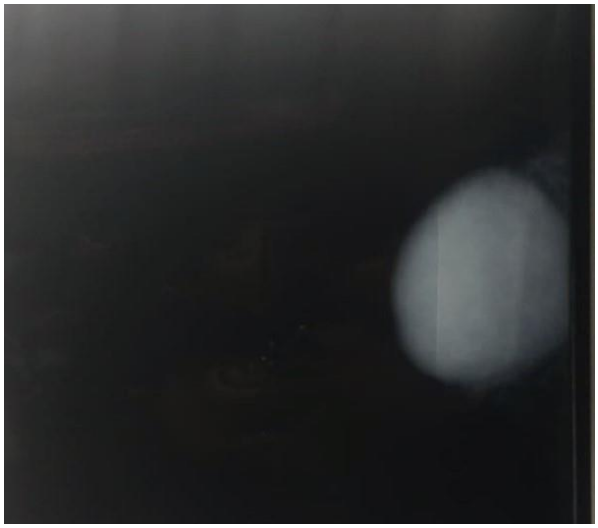

Figure 2: Mammographic image: hyperdense mass involving the whole right breast

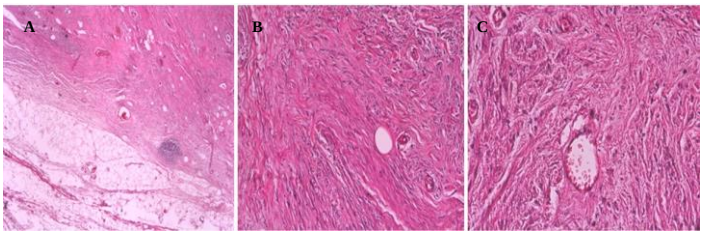

Figure 3: Microscopic image, hematoxylin-eosin staining (HES), (A) - Unencapsulated tumor with circumscribed border, (HES x 50) (B) - Fascicles of spindle cells separated by dense collagen bundles and entrapped adipocytes (HES x 100); (C) - Fibroblastic-like cells with scanty cytoplasm and elongated nuclei (HESX200)

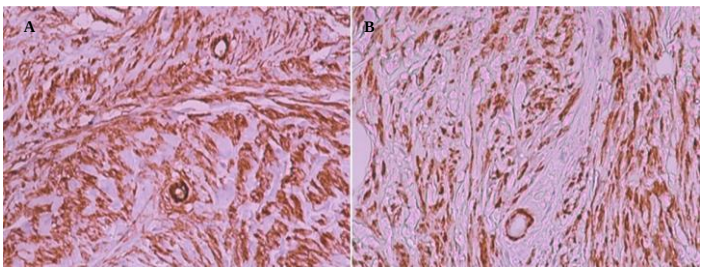

Figure 4: Immunohistochemistric image (IHC), (A) - Myofibroblast showed positivity for vimentin (IHC X200); (B) - Myofibroblast showed positivity for CD34 (IHC x 200)

Discussion

Breast myofibroblastoma is a rare, mesenchymal benign tumor firstly described by Wrgotz in 1987, who reported 16 cases [1]. Until now, only single case reports or small case series have been described in the literature.

Extra-mammary lesions are most common in older men [2]. Several extra-mammary locations have been reported, including the head, neck, extremities, buttock, vulva, and

testicular region [3-6]. The most frequent site is the inguinal region [5,7].

Magro et al. [2] studied 70 cases of breast MFB, and they witnessed that the tumor has a postmenopausal and older men's predilection. At presentation, the age ranged from 40 to 87 years. In our study, we reported a case of a 17-year-old woman, without any remarkable personal or familial history.

The risk factors for developing MFB are uncertain. However, in some reports, there was an association between renal and prostatic neoplasms with breast MFB [8]. Furthermore, some patients presented with gynecomastia, but our patient didn't have it [2,8,9]. MFB was also reported after breast cancer excision at surgical scar sites or after chest wall trauma [8,10,11].

So far, many authors suggested physiopathological explanations to understand mechanisms leading to the onset of this rare entity. Some authors suggested the role of steroid sex hormones as estrogen, progesterone and androgen receptors which were present in severe cases and associated with gynecomastia [2,9]. The second explanation was the disruptions in cytokine secretions and inflammation after trauma, leading to a transformation of tumor growth factors (TGF) in TGFβ. These findings are supported by the presence of tumor necrosis factor (TNF) and fibroblastic peptide-trophic growth factors in MFB [9]. The third explanation was the migration and transformation of fibroblasts to the surgical site after surgery [8].

Clinically, MFB may be asymptomatic and diagnosed by accident due to mammographic screening in postmenopausal women or in the context of gynecomastia [9]. However, it is usually described as a solitary, firm, slowly growing, and well-defined lump [2]. There is no documentation of axillary lymph nodes or changes in the skin and nipple.

Radiologically, there is no pathognomonic sign for the diagnosis of MFB. Ultrasonography is the first-line examination, and shows a well-defined mass, with mixed echoic patterns [10]. Mammography currently shows a heterogeneous well-defined tumor without microcalcifications. Magnetic resonance imaging (MRI) is not common to diagnose MFB, only a few cases were studied, and there were no specific signs [12].

In that regard, many differential diagnoses may arise including benign lesion as fibroadenoma, neurofibroma, lymphangioma, angiolipomas, hematoma, abscess, and malignant lesion as phyllodes tumor, carcinoma and sarcoma [10].

In our second case, we suspected a malignant lesion due to the hyperdense mass involving the entire breast. For the purpose of defining the surgical procedure, we performed a core biopsy in order to acquire a histological diagnosis.

Macroscopically, MFB is a fairly limited, firm, a whitish greyish tumor of variable size. Histologically, it consists of a proliferation of fusiform cells arranged in irregular short beams separated by thick ropes of hyalinised collagen, without atypia with a low mitotic index [1]. These cells correspond to myofibroblasts and intermediate cells between fibroblasts and smooth muscle cells [9]. In immunohistochemistry, these cells express CD 34, vimentin, actin, estrogen receptors and progesterone receptors [2,9]. Cytokeratins, c-kit, and S-100 proteins are always negative [9,11], even though the expression of desmin, SMA, bcl-2, and CD99 is variable [13].

The prognosis of MFB is excellent. Treatment remains as surgical excision. Malignant transformation of myofibroblastoma wasn't previously reported. That leads to reconsider surgical procedures in this benign neoplasm [10].

Conclusions

Myofibroblastoma is a benign, slowly growing tumor. Many differential diagnostic problems are raised and rectified by pathological and immunohistochemical examination. The treatment is based on surgical excision with excellent outcomes.

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