Idiopathic Granulomatous Mastitis, A Rare Cause of Male Breast Mass

Erkeklerde Memede Kitlenin Nadir Bir Nedeni, Idiopatik Granülomatöz Mastit

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ÖZET

Granülomatöz mastit erkeklerde oldukça nadir görülen, memenin kronik benign inflamatuvar bir hastalığıdır. Literatürde bildirilmiş sınırlı sayıdaki vakalar nedeniyle prognozu tam olarak bilinmemektedir.

76 yaşında erkek hasta yaklaşık bir aydır olan sağ memede ele gelen ağrısız, meme başında çekintiye sebep olmuş kitle yakınması ile başvurdu. Yapılan meme ultrasonografisinde 19x11 mm boyutunda düzensiz sınırlı, solid hipoekoik alanlar içeren kitle lezyonu saptandı.

Öncelikle yapılan görüntülemelerinde meme kanseri görüntüsüne sahip olması, memenin yarısından fazlasını kaplayan kitle nedeniyle hastaya meme karsinomu ön tanısı ile basit mastektomi + sentinel lenf nodu biyopsisi uygulandı. Postoperatif spesimen sonucu granülomatöz mastit ile uyumlu olarak patoloji tarafından raporlandı.

Literatürde bildirilen erkek granülomatöz mastit olgularında da bizim sunduğumuz olgudaki gibi belirsiz bir patogenez vardır. Risk faktörlerini tanımlamak ve tedaviyi optimize etmek için erkek granülomatöz mastit ile ilgili daha fazla vaka raporu bildirilmelidir.

Anahtar Kelimeler: granülomatöz mastit, erkek meme kanseri, erkek granülomatöz mastit

ABSTRACT

Granulomatous mastitis is a chronic benign inflammatory disorder of the breast which is extremely rare in male patients, and the prognosis is not well known mainly due to the limited number of cases reported in the literature.

A 76- year -old male patient applied with nipple discharge and a palpable painless lump on his right breast for one month. Breast ultrasonography showed a 19x11 mm sized mass with solid hypoechoic components, and irregular shape. A presumptive diagnosis of breast carcinoma was made due to the results of imaging methods and the core biopsy, which resulted in unclearly. Simple mastectomy and sentinel lymph node biopsy was performed. The patient was discharged uneventfully on the second postoperative day. The histopathological findings supported the diagnosis of idiopathic granulomatosis mastitis.

Male granulomatous mastitis, which previously reported in the literature, has unclear pathogenesis. It will be necessary to accumulate more case reports about GM in order to define the risk factors and optimizing the treatment.

Keywords: granulomatous mastitis, male breast cancer, male granulomatous mastitis

INTRODUCTION

Granulomatous mastitis (GM) is a chronic benign inflammatory disorder of the breast. Clinically, it presents as a palpable lump that has inflammatory features. Nipple discharge, nipple retraction, a tender and painful lump, skin thickening, erythema, sinus formation, and axillary adenopathy can be seen with GM (1).

Idiopathic Granulomatous Mastitis (IGM) is most commonly seen in female patients (1). To our knowledge, in the literature, there are limited case reports about IGM in male patients (2,3,4). Due to the rarity of this disease, the current management is still unclear.

Here, we aimed to present an elderly male patient diagnosed with idiopathic granulomatous mastitis that mimicked breast carcinoma both clinically and radiological.

CASE PRESENTATION

A 76- year -old male patient applied to our clinic with a palpable painless lump and nipple discharge in his right breast nearly for one month. He had no history of chronic illness, trauma, surgery, and smoking. During the physical examination of the right breast, nipple discharge, skin thickening, and erythema were seen; however, axillary lymphadenopathy was absent. Contralateral axillary lymph

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nodes and breast were normal. Laboratory tests were unremarkable. Breast ultrasound revealed a retro areolar hypoechoic lesion with a size of 19x10 mm, on the right breast. A core biopsy was performed. The core biopsy was reported as 'suspicious for malignancy'.

Figure 1a. Epitheloid histiocytes, hematoxylin and eosin staining, magnification=40x

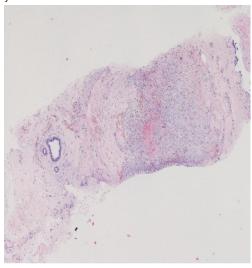
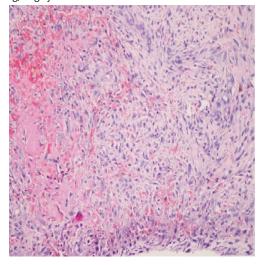


Figure 1b. Epitheloid histiocytes, hematoxylin and eosin staining,magnification= 200x

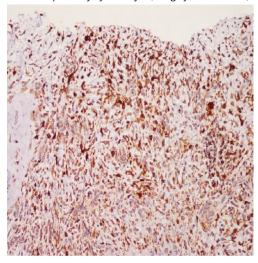


The presumptive diagnosis of breast carcinoma due to the results of the imaging methods and the core biopsy, simple mastectomy, and sentinel lymph node biopsy was performed. Perioperative frozen section examination was reported reactive for sentinel lymph node biopsy. The patient was discharged uneventfully on the second postoperative day.

Postoperative histopathology examination reported ductal epithelial hyperplasia accompanied by a ductal reaction

consistent with granulomatous mastitis in multiple serial cuts containing CD68 positive histiocytes (Figure 1a, Figure 1b, Figure 2).

Figure 2. CD68 positivity of histiocytes, magnification= 200x,



After the diagnose of GM, tuberculosis skin test with PPD and Mycobacteria tuberculosis PCR (Tuberculosis PCR) were performed, and results were negative for both. The tissue was cultured, and no microorganisms were detected. Also, there was no sign of malignancy.

Written informed consent was obtained from the patient for treatment, surgery, and publication..

DISCUSSION

Granulomatous mastitis (GM) is a chronic benign inflammatory disorder of the breast, which is mostly seen in women during the reproductive age period (4). Kesler and Wolloch described IGM in 1972, among female patients (5).

There are many accused etiological factors presented in IGM, like infections, ethnicity, autoimmune response, hormonal factors such as high levels of estrogen, progesterone, pregnancy, breastfeeding, diabetes mellitus type 2, obesity, hyperprolactinemia, exogenous hormone replacement treatment, smoking, etc. (3,4).

Male IGM is an infrequent clinical entity that was first described in 2005 by Reddy et al. (2). To our knowledge, only five cases were reported between the ages of 26-46 in the literature (2,3,4). Reddy et al. reported a 46-year old male patient who had a subareolar mass on his right breast, which was treated with local excision. Al Manastra

reported a 29-year-old male patient with retro-areolar mass treated with an excisional biopsy that no recurrence was observed(3). Barreto reported one of the most extensive male IGM series in the literature with three male patients (4).In his study, one of the patients had cryptorchidism surgery. Also, one of them was a male-to-female transgender patient who was on anti-androgen therapies and estrogen replacement therapy for eight years (4). The third male patient with IGM was HIV positive, and Mycobacterium tuberculosis complex DNA was present in the breast biopsy material. However, our presented patient had no risk factors such as a history of chronic illness, obesity, surgery, trauma, smoking, and medical or hormonal replacement therapy and also negative for HIV and tuberculosis.

GM and breast cancer have similar appearance with the imaging methods; imaging studies cannot accomplish a definite differentiation. For diagnosis of GM breast ultrasonography, mammography, and breast MRI can be used. A study shows that in GM patients most seen pathological finding with breast USG was a hypoechoic mass which was irregularly shaped (4). Moreover, increased vascularity, skin thickening, ipsilateral axillary lymphadenopathy can be seen, and even also, there can be no masses (4). Both breast carcinoma and GM have similar appearances in clinical and radiological studies. Our case had features like breast carcinoma with the performed breast USG; therefore, surgical excision was performed.

Many studies show that IGM has comprehensive different treatment approaches including oral antibiotics, oral corticosteroids, limited or wide surgical excisions or mastectomies, and observation. In female patients with GM, generally, the first preferred treatment is medical treatment without surgery (4). However, for male IGM, in literature, the treatment options in male patients were limited as mass excision and as in our case, simple mastectomy. On the other hand, the effects of oral corticosteroids and antibiotics in the treatment of male granulomatous mastitis are not yet clearly known.

In conclusion, male granulomatous mastitis is a sporadic inflammatory disease with unclear pathogenesis and limited cases. We should not forget that it is challenging to differentiate breast carcinoma from granulomatous mastitis especially in elderly male patients. A better

understanding of the pathogenesis and treatment of GM, with more extensive case series, may lead to a better-addressed management.

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