# Simultaneous Bilateral Male Breast Cancer: a Case Report and Review of the Literature

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## Abstract

The Male Breast Cancer (MBC) is a very rare neoplasm which accounts for 0.5-1% of the total cases of breast cancer. Bilateral involvement is reported to occur in fewer than 2% of all the diagnosed cases of MBC, and synchronous tumors are very rare. Because of its rarity, little is known about MBC etiology, its clinical findings and treatment. Most patients present with a painless breast mass. The prognosis does not seem to be poor compared to that of the females, and it probably has a similar stage by stage prognosis. Surgical treatment is the gold standard for MBC.

The authors report a case of simultaneous bilateral male breast cancer. The aim of this case report was to contribute to the available literature by this unusual presentation of the disease.

**Key words:** Breast cancer; Male; Bilateral; Chemotherapy; Radiotherapy

**Please cite this article as:** Farrokh D, Ansaripour E, Fallah Rastegar Y. Simultaneous Bilateral Male Breast Cancer: a Case Report and Review of the Literature. Iran J Cancer Prev. 2010; Vol3, No4, p.199-203.

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Received: 5 Sep. 2010
Accepted: 2 Oct. 2010
**Iran J Cancer Prev 2010; 4:199-203**

## Introduction

The first references to MBC are found in the Edwin Smith surgical papyrus from ancient Egypt dated between 3000 and 2500 BC, with the first modern description of the clinical entity being attributed to John of Arden in the 14th century [1].

Male breast cancer (MBC) is a comparatively rare neoplasm which accounts for 1.2-2% of all cancers in men, and 1% of the total cases of breast cancer [2, 3]. Bilateral breast cancers account for only 0.5-1% of MBC, and simultaneous cancers are extremely rare [4, 5].

Most of our knowledge about the biology, natural history and treatment of MBC is extracted from female breast cancer [6].

In most reports, the age detection of MBC in males is about 60–years old which is ten years older than females. MBC usually presents as a firm, painless breast mass, with or without nipple discharge and with no skin changes. The typical clinical finding in 75% - 95% of males is a breast lump [4].

Various risk factors have been proposed for MBC, including genetic factors such as a positive familial history of breast cancer, BRCA2 abnormalities, hormonal abnormalities due to obesity or testicular disease, klienfelter's and many other risk factors [7]. Most tumors are ductal carcinoma invasive, with or without ductal carcinoma in situ [3].

Presentation is usually late, with more than 46% of patients having stage III or IV of the disease at the time of the diagnosis. Males with breast cancer exhibiting the same TNM stage receive the same treatment as women. Surgical treatment is the gold standard, and adjuvant therapy has been advocated in men based on its beneficial results [4, 5].

Here a case of simultaneous bilateral breast cancer diagnosed in a 56 year old man is reported. This case is discussed in relation to the published literature.

## Case Report

A 56-year-old male presented with an ulcerative lesion in the right breast, the patient reported that the lesion had been existed for 8 months. He had positive familial history for breast cancer. His sister had a history of right sided breast cancer and radical mastectomy 5-years earlier. His past medical history was unremarkable.

Physical examination of the right breast, revealed skin lesion with ulceration, edema and a poorly defined non tender hard mass, fixed to the skin, measuring about 2*3cm in diameter adjacent to the

**Figure1.** Right breast craniocaudal view. Mammography of the right breast shows a central mass with no micro calcifications.

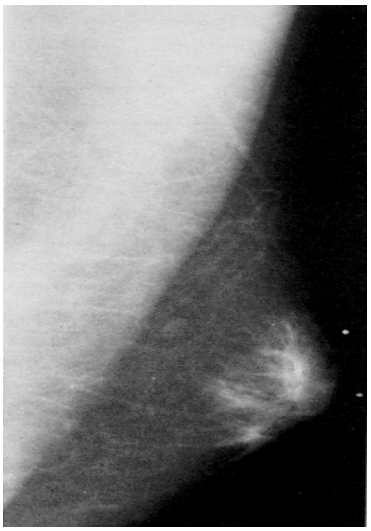

**Figure2.** Left breast lateral – oblique view. Mammography of the left breast revealed an ill – defined density with no specific mammographic appearance in the subareolar region.

nipple. Examination of the left breast showed a bloody discharge from a single duct. Nipple areola complex was normal in the left breast and there were no skin changes or nipple retraction. The axillary lymph nodes were not palpable.

Mammography of the right breast was consistent with a 2*2cm in diameter central mass with irregular margins with no micro calcifications (Fig1). Mammography of the left breast revealed an ill – defined density with no specific mammographic appearance in the subareolar region. There were no evidences of pathologic calcifications or other abnormal findings (Fig2).

Breast ultrasonography showed a hypoechoic lesion of 2*2cm in diameter with non homogeneous echogenecity in the central region, medial to the nipple in the right breast (Fig3). Ultrasonography of the left breast did not show any abnormalities. Our first diagnosis was right sided breast cancer.

Chest X-Ray and abdominal ultrasonography were normal. Blood tests were within normal limits. Hormone tests (LH, FSH, E1, E2, PRC and testosterone) were also normal. Cytopathological examination of nipple discharge in the left breast was highly suspicious for malignancy.

Excisional biopsy of both breasts was positive for breast cancer. The right sided lesion revealed a high grade invasive ductal carcinoma (Fig4), and the left sided lesion was a DCIS and low grade invasive ductal carcinoma (Fig5). Sentinel node was positive for the right breast tumor, and negative for the left breast lesion.

Immunohistochemical staining of the tumors of the both breasts showed them to be estrogen and progesterone receptor positive.

The patient underwent modified radical mastectomy on the right breast, and a simple mastectomy on the left side. He received chemotherapy, followed by endocrine therapy consisting of tamoxifen postoperatively. After 2 years of follow up our patient is still alive, without any evidence of local recurrence or distal metastasis.

## Discussion

The overall incidence of MBC is around 0.5-1% of all breast cancers. Although MBC is rare, a geographic variation in its incidence has been reported. The MBC rate across the world ranges between 0.1 and 3.4 per 100000 males [8]. In the USA and Europe the rate is at the order of 1 per 100000 males. MBC is much more common than female breast cancer in sub-Saharan Africa compared to the western countries.

It seems that the higher incidence of MBC in Africa is related to environmental and socio – economic factors rather than genetic ones [9, 10].

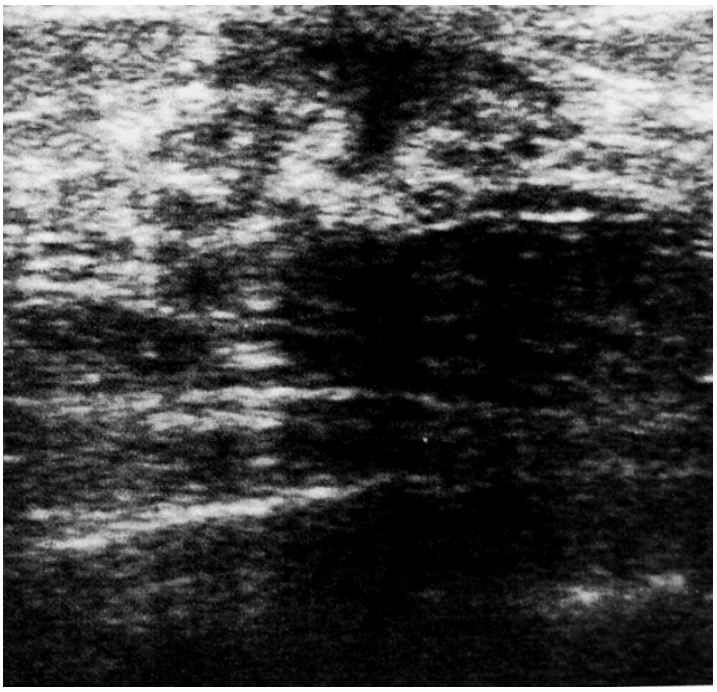

**Figure3.** Breast ultrasonography showed a hypo – echoic lesion with non homogeneous echogenecity in the central region

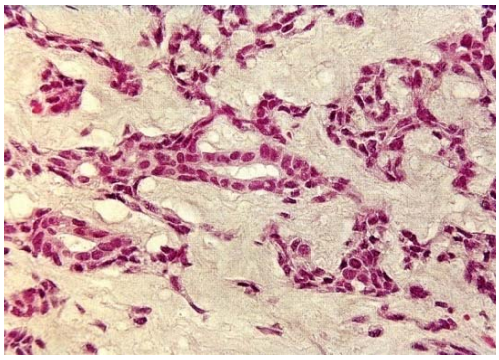

**Figure4.** Histology, Invasive ductal carcinoma

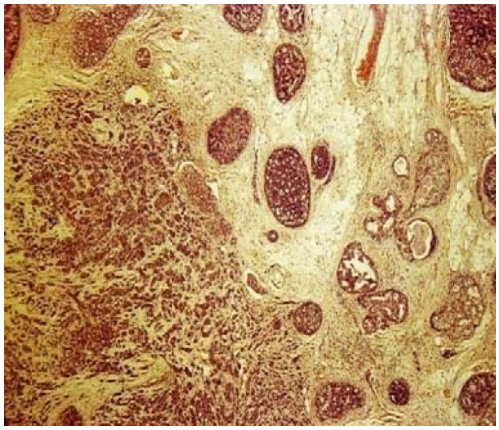

**Figure5.** Histology, DCIS and invasive ductal carcinoma

Although the etiology of MBC remains elusive, there are several predisposing factors some of which are as follows [5, 7]:

1. Positive family history and genetic factors
2. Suppuration of testicular function, as in klinfelter's syndrome, or as a sequel of orchitis, undecended testis, testicular injury, or occupational exposure to heat or toxic agents
3. Gynecomastia
4. Hyperestorogenism secondary to liver dysfunction
5. Previous radiation of breast tissue
6. Exposure to electromagnetic fields for more than 30 years
7. Epidemiological factors such as high temperatures, organic solvents, alcohol and tobacco
8. Hyperprolactinemia secondary to head trauma and ingesting prolactine elevating drugs

A family history of breast cancer is found in 5-10% of the cases. In case of having a family history of prostate cancer, the risk of developing breast cancer rises to four times compared to that of normal family histories [5].

Familial risk is reported in a few clusters of MBC with the recent availability of BRCA1, and BRCA2 genetic testi ng. Familial cases usually have BRCA2 rather than BRCA1 mutation, and it has been found that 10% of male with a mutation in the BRCA2 gene develop breast cancer. It is well known that P53 and C – erbB – are expressed in MBC [7, 11].

With regard to hormonal abnormalities, it has been reported that MBC is caused by imbalance between estrogen and testosterone. Estrogen is thought to play an important role in the development of MBC since the plasma concentration of this hormone is higher in MBC patients than in normal males.

With regards to testosterone, castration or prostate gland therapy is said to increase the risk of MBC while testicular dysfunction due to mumps or cryptorchidism alters the level of estrogens, androgens and gonadothropins and increases the risk of MBC [3, 7].

The common clinical presentation in MBC is a painless mass, usually located beneath or adjacent to the nipple areola complex, or in the upper outer quadrant of the breast. The tumor is usually poorly defined, non tender on palpation and should be differentiated from gynecomastia. The masses in gynecomastia are usually bilateral, well defined, tender and just beneath the subareolar region [2, 4]. Other frequent signs are nipple retraction and nipple discharge. Bloody discharge, is reported in 75% of the MBC cases. Other symptoms may include skin lesions such as retraction, edema, erythema and ulceration. It may present with just an axillary mass or with a remote metastasis [4, 12].

The prevalence of MBC increases with age with the peak incidence in the late sixth and early seventh decades of life. The incidence of MBC rises logarithmically with age, in contrast to the slight decrease at the time of menopause and subsequent increase in women. Frequent age distribution of breast cancer for females is bimodal with early onset at the age of 50 and late onset at the age of 70 [2, 4].

As in females, most breast cancers found in males are infiltrating ductal carcinomas, with or without ductal carcinoma in situ (DCIS) that accounts for 7-10% of the cases [3, 13]. Invasive lobular carcinoma and other histological types occur rarely. The rate of occurrence of a second breast primary is similar to that observed in female breast carcinoma. Diagnosis is based on clinical findings, mammography and ultrasonography.

The most common finding in mammography is the presence of a well – defined mass usually lobulated, speculated or an ill-defined lesion characteristically eccentric to the nipple or beneath the nipple areola complex [12]. Calcification is a less common feature in males than in females and is often larger, rounder, fewer and more scattered in males [14, 15]. Ultrasonic features are similar to those observed in females, typically a hypo echoic poorly defined mass with or without acoustic shadowing [12, 14]. Occasionally the tumor presents as swelling or slight distortion of subcutaneous fat.

Excisional biopsy with an immediate intraoperative pathology examination confirms the malignancy and makes wider excision possible during the same procedure.

The prognosis of MBC remains uncertain because of the late diagnosis, unpredictable course and high potential for metastases. The prognosis does not seem to be poor compared to females when age and stage are matched [6].

Due to the absence of screening protocols in men and the limited amount of mammary tissue, allowing rapid local invasion, a late diagnosis with a poor survival rate is often made [4, 5, 16].

Surgical treatment remains the gold standard in MBC. Surgery is usually mastectomy with axillary clearance or sentinel node biopsy. Modified radical mastectomy is a principal surgical treatment. In cases with pectorals muscle involvement, part or all of the muscle should be resected to obtain local regional control. Lumpectomy with postoperative radiotherapy is not usually used for MBC, as the tumor is usually adherent to pectorals fascia and as the size of the male breast is small [2, 4].

Hormonal manipulation and chemotherapy constitute an essential part of the adjuvant therapy. Some patients could benefit from systemic chemotherapy and the most common regimens are CMF (Cyclophosphamide, Metothreaxate, 5-fluorouracil), or other anthracyclin – based regimens [17].

Indications for radiotherapy, by stage are similar to female breast cancer. Post mastectomy radiotherapy should be proposed in case of lymph node positivity, cancer infiltrating the pectorals muscle, and advanced T stage [2, 4, 6]. In cases of diffuse recurrence, hormonal therapy, chemotherapy or radiotherapy can be administered for palliative purpose [16, 17].

There is a belief that male breast cancer has a poorer prognosis based on its tendency to be diagnosed at a later and more advanced stage in most cases. However, breast cancer does occur in males, and patients must become aware of its existence and the importance of surgical evaluation for any lump in their breasts.

## Acknowledgment

None

## Conflict of Interest

The authors declare that they have no conflict of interests in this case report.

## Authors' Contribution

All of the authors contributed in all parts of the article.

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