



The unusual suspects: A review of unusual benign and malignant male breast imaging cases



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ABSTRACT

Male breast disease is uncommon. Men presenting with breast symptoms may represent unique diagnostic challenges for the radiologist, particularly if imaging findings are not classic for gynecomastia or carcinoma. In this paper we review 10 unusual male breast cases, 5 benign and 5 malignant, including the radiologic findings, differential diagnosis, pathology and management.

1. Introduction

Male breast cancer accounts for < 1% of breast cancers [1]. As a result, men are not routinely screened with mammograms or ultrasound (US), and radiologists are less familiar with the varying imaging appearance of male breast pathology. Male breast cancer patients have a worse overall prognosis compared to women, since they are usually diagnosed at a later stage with up to 50% of men having axillary metastases at the time of presentation [2–4]. When men are referred for breast imaging, it is most often to evaluate a palpable lump, breast enlargement, or tenderness. Although male breast cancer and gynecomastia can present similarly, male breast cancer typically manifests as a unilateral, painless, eccentrically located subareolar mass while gynecomastia tends to be painful, tender, centrally located, and bilateral [5]. Secondary signs of carcinoma such as nipple retraction, skin ulceration or thickening, and axillary adenopathy may assist in differentiating carcinoma from gynecomastia [4]. Secondary signs of malignancy may occur earlier in male patients because of smaller breast size.

Understanding normal male breast anatomy is essential for understanding male breast pathology. The normal adult male breast is characterized primarily by subcutaneous fat and a remnant of subareolar ductal tissue [6]. Lobular development, which requires both estrogen and progesterone, is usually not observed in men. Therefore, lobular pathology commonly seen in women, including cysts, fibroadenomas, and invasive lobular carcinoma, is rare [6]. Male breast

disease can affect any component of the breast, including the skin, subcutaneous tissue, stroma, glands, neurovascular structures, and lymphatic tissues. Knowledge of this anatomy can help generate an informed differential diagnosis when faced with a challenging case (Table 1).

Typically, radiologic evaluation of a symptomatic male patient begins with bilateral diagnostic mammography followed by sonography if the mammogram is inconclusive or shows suspicious findings. If gynecomastia explains the patient's symptoms and can be diagnosed with mammography, ultrasound is usually not necessary. Familiarity with the presentation and imaging findings of a spectrum of male breast pathology will facilitate accurate image interpretation. This review will focus on unusual male breast cases in order to familiarize radiologists with their imaging appearance and management.

2. Malignant neoplasms

2.1. Case 1: invasive lobular carcinoma

Invasive lobular carcinoma (ILC) accounts for < 4% of all male breast cancer cases due to the absence of terminal lobules in men [7]. Estrogen exposure and Klinefelter syndrome are associated with an elevated risk. Typically the clinical presentation is a painless mass in an older male patient (Fig. 1) [7]. Mammographic findings include a spiculated mass or an area of architectural distortion, with a minority of

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Table 1
Male breast pathology by anatomic region.

	Benign	Malignant (primary)*
Skin	Epidermal inclusion cyst Sebaceous cyst Seborrheic keratosis	Paget's Disease Dermatofibrosarcoma Basal cell carcinoma of the nipple Melanoma
Subcutaneous tissues	Hematoma Fat necrosis Abscess	
Fat	Lipoma Angiolipoma Pseudogynecomastia	Liposarcoma
Stroma	Pseudoangiomatous stromal hyperplasia (PASH) Fibromatosis Myofibroblastoma Diabetic mastopathy Benign spindle cell tumor Inflammatory pseudotumor	Spindle cell sarcoma
Ducts	Gynecomastia (glands & stroma) Intraductal Papilloma	Ductal Carcinoma in Situ Invasive ductal carcinoma Papillary carcinoma Adenoid cystic carcinoma Invasive lobular carcinoma
Lobules (rare)	Fibroadenoma Cyst	
Nervous system	Granular cell tumor Schwannoma	Malignant peripheral nerve sheath tumor
Vessels	Hemangioma Venous or lymphatic malformation Glomus tumor	Angiosarcoma
Lymph node	Reactive lymph node	Lymphoma (primary or secondary)

* Secondary malignancies (metastases) may also involve the male breast.

tumors presenting as an asymmetry. In rare cases, mammography may be normal. US typically shows an irregular mass with hypoechoic or heterogeneous echotexture; however, a tumor may appear as a circumscribed mass, as isolated shadowing without a mass, or may be sonographically occult, making diagnosis difficult [8].

Pathologic features include an irregular or ill-defined tumor consisting of non-cohesive small cells throughout fibrous connective tissue or single-file cells arranged in linear cords which infiltrate the stroma [7]. This single-file arrangement contributes to decreased conspicuity on imaging. Cells tend to be E-cadherin negative or reduced [7].

ILC is usually treated with mastectomy and axillary dissection, with adjuvant chemotherapy and tamoxifen utilized in some cases [7].

2.2. Case 2: intraductal papillary carcinoma presenting as mastitis

Malignant papillary lesions of the breast include micropapillary DCIS, noninvasive papillary carcinoma and invasive papillary carcinoma [9]. Invasive papillary carcinoma is the second most common subtype of invasive cancer in men, after invasive ductal carcinoma not otherwise specified, and is more common than benign papillomas in males [10]. Invasive components, if present, are usually at the lesion periphery which may result in biopsy sampling error which usually targets the center of the lesion [9]. Noninvasive papillary carcinoma may be intraductal or intracystic [9]. Tumors are typically subareolar in location and can have a circumscribed or spiculated margin on mammography with poorly defined indistinct margins suggestive of invasive disease. Calcifications are uncommon and are often seen only microscopically [11]. Occasionally patients may initially present with

symptoms of mastitis including breast erythema. In the case presented here (Fig. 2), the patient had symptoms of mastitis but short interval follow up revealed an underlying mass. Biopsy revealed an intraductal papillary carcinoma. This highlights the importance of close imaging follow up of apparent infection to resolution even if clinical symptoms improve with antibiotic treatment [12].

On ultrasound noninvasive papillary lesions are often associated with a cyst or dilated duct. Solid masses or papillary projections arising along a cyst wall are characteristic of papillary carcinoma [11]. Simple cysts rarely occur in males, therefore a cyst or dilated duct must be viewed with caution with real time scanning recommended to evaluate for a subtle mural nodule [13]. Surgical excision is required for full evaluation of papillary lesions in men, as aspiration or biopsy may be prone to sampling error and histologic underestimation of disease [13–14].

2.3. Case 3: metaplastic breast carcinoma (MPBC)

Metaplastic breast carcinoma (MPBC) accounts for < 1% of breast cancers and is aggressive with larger tumor size at presentation and higher grade than invasive ductal carcinomas no special type [15]. MPBCs are a histologically diverse group of tumors characterized by two or more cell types. Ductal carcinomatous epithelium transforms to squamous or spindle cell components or mesenchymal elements [16]. The most common variant of MPBC is squamous cell carcinoma, of which basaloid squamous carcinoma is a variant (Fig. 3). Typically, tumors are estrogen and progesterone-receptor negative. The metaplastic basaloid triple-negative breast cancer is one of the most aggressive, therapy-resistant, and metastasizing tumors [17]. This is a particularly lethal subtype with a 5-year survival rate as low as 40% [18].

MPBC typically appears on mammography as a mass with circumscribed or microlobulated margins, usually without spiculations and rarely with associated microcalcifications or architectural distortion [19]. MPBCs with circumscribed margins are more likely to be composed solely of spindle cells [19]. US of metaplastic breast cancer often demonstrates microlobulated or circumscribed oval masses. Some MPBC show heterogeneous echogenicity, which might indicate the presence of both cystic and solid components. At other times the echogenicity might be homogeneous, suggesting more of a solid mass [19].

In most cases, MPBC is treated with mastectomy, with or without axillary node dissection [15]. A vast majority of cases are treated with adjuvant chemotherapy. Radiation therapy is used less commonly, in 50%–60% of cases [20].

2.4. Case 4: chronic lymphocytic leukemia/small lymphocytic lymphoma

Breast lymphoma may be primary or, more commonly, secondary, with multiple subtypes [12,21]. On mammography breast lymphoma usually appears as circumscribed or indistinctly marginated intramammary masses without calcification, although solitary masses (Fig. 4), architectural distortion, and normal mammographic findings have also been reported [13]. US findings often include a hypoechoic or heterogeneous mass with posterior acoustic enhancement, a hyper-echoic margin, and hyper-vascularity on Doppler images [12]. Magnetic resonance imaging (MRI) shows variable morphology and enhancement, with the majority of cases demonstrating type 2 kinetics [22].

Lymphadenopathy on mammography is classically associated with lymphoma. The lymphadenopathy may be unilateral or bilateral, characterized by multiple enlarged, dense, axillary lymph nodes. US

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