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Case Report

Pilomatrixoma, a Rare Mimicker of Male Breast Cancer

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ABSTRACT

Pilomatrixoma or calcifying epithelioma of Malherbe is a benign skin tumor arising from the hair follicle; breast occurrence is considered a rarity. Clinically presenting as a palpable abnormality and with both benign and malignant mammographic and sonographic features, it can be easily misdiagnosed as a breast neoplasm. We report a very rare case of pilomatrixoma of the male breast in a 36-year-old male presenting with a firm, superficial nodule in the upper outer quadrant. Though the sonographic trifecta of imaging features (shape-margins-orientation/oval, circumscribed mass, parallel to the skin) is consistent with a benign lesion, a histologic diagnosis was warranted based on its most suspicious feature of internal pleomorphic calcifications. Pathologic diagnosis revealed the uncommon benign entity of pilomatrixoma in the male breast. Our patient was recommended for surgical excision based on current literature recommendations for management in most reports of pilomatrixoma. One alternative recommendation presented in a single report of pilomatrixoma in the breast supported follow-up imaging based on benign imaging characteristics.

Keywords: Pilomatrixoma, Male breast, Cancer, Benign, Surgical excision

INTRODUCTION

The workup for a palpable lump in the adult male breast usually begins with bilateral diagnostic mammograms and more frequently than not, results in a confident diagnosis of gynecomastia. At times, an ultrasound, in addition to mammography is utilized in the initial evaluation for a palpable lump in a male patient. Ultrasound can confirm the appearance of gynecomastia, or evaluate palpable lumps not confidently characterized as gynecomastia or other benign entities mammographically. Examples of benign masses, more commonly seen in the male breast, include gynecomastia, fat necrosis, oil cyst, or lipoma. Occasionally, we are surprised by a rare diagnosis in the male breast. Faced for the first time with the pathologic diagnosis of "pilomatrixoma", we conducted a literature search on PubMed applying the terms "pilomatrixoma" and "male breast" with filters set to include only full articles published in the English language. Our search yielded 9 cases of such an entity found in the male breast between 1987 and 2018, on average about 3 cases reported per decade in the English literature. Thereby, we contribute our case of pilomatrixoma in the male breast to highlight diagnostic imaging clues based on mammographic and sonographic imaging to expand the current literature. This case as described emphasizes the imaging variability in the sonographic appearance of pilomatrixoma and the pathologic appearance of keratinization within these lesions as relates to imaging. Simultaneously, we review the literature to enlist the necessary histological clues that help differentiate this benign tumor, not routinely seen in the mammographic practice, from the malignant mimicker of carcinoma and raise awareness that such diagnosis, though rare, should be considered in the differential of

a palpable superficial mass in the male breast when imaging raises such suspicion.

CASE REPORT

A 36-year-old HIV positive male presented with a painless hard palpable "pea-size" lump, in the left breast, which the patient reported had been present for many years though appeared increased in size over the most recent 6 months. No visible skin abnormalities were observed on clinical examination. The patient did not report any family history of breast cancer. Diagnostic mammography demonstrates an oval-shaped, circumscribed mass in the upper outer left breast, with smooth borders, and multiple pleomorphic calcifications [Figure 1]. Gray-scale ultrasound examination revealed a 1.0 cm × 0.8 cm × 0.6 cm superficial isoechoic circumscribed oval mass, parallel to the skin and chest wall, with internal vascularity, and multiple hyperechoic foci corresponding to the calcifications seen on mammogram [Figure 2]. Though the mass has benign-appearing features including oval shape, circumscribed margins and parallel orientation to the skin and chest wall on ultrasound examination, the presence of pleomorphic calcifications and the patient's concern of recent increase in size, resulted in a low to moderate suspicion for malignancy Breast Imaging Reporting and Data System (BIRADS) 4B.[1]

An ultrasound-guided core biopsy was subsequently performed with a 14 gauge spring-loaded core needle biopsy device with 3 tissue passes. Pathologic diagnosis revealed a pilomatrixoma [Figure 3]. Imaging characteristics are concordant with pathologic diagnosis, thereby clarifying that the calcifications within the mass correspond to the whorls of keratin present. The patient was recommended for surgical excision; however, given his limited finances, he did not undergo surgery.

DISCUSSION

Etiology and demographics

Pilomatrixoma is a rare benign slow-growing skin tumor that develops from the cellular matrix of the hair follicle.[2] Though the etiology is not determined, repeated skin trauma and inflammation are thought to be the trigger for the hair follicle matrix stimulation. Breast pilomatrixoma is very rare (1:100000 people), with only a few cases reported so far;[3] and it is exceedingly rare in the male breast with less than a handful of cases reported per decade, as per our PubMed search. It was first described by Malherbe and Chenantais in 1880, as a calcifying epithelioma of sebaceous gland. These tumors often occur in the head, neck and upper extremities with a slight predominance in female patients in the first and second decades.[2]

Clinical and imaging findings

Clinically, these tumors are firm to hard subcutaneous or intradermal solitary lesions varying from 1 to 3 cm in diameter covered by intact skin. There may be no associated skin findings, or findings ranging from purple overlying skin to the spectrum of skin inflammatory changes including signs of

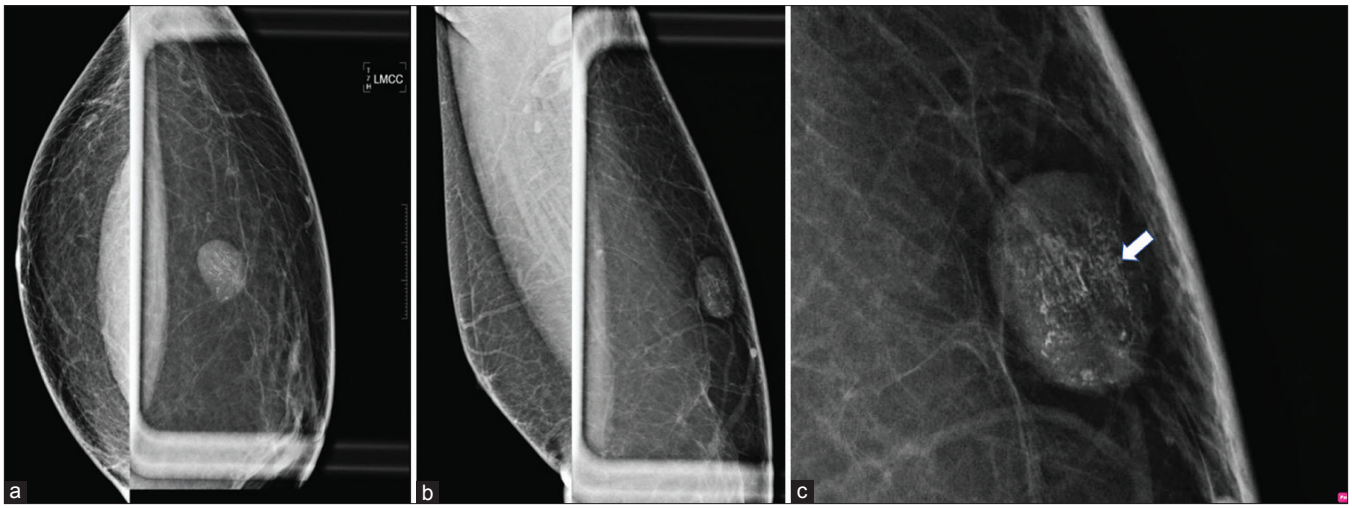

Figure 1: A 36-year-old HIV positive male presented with a painless hard palpable "pea-size" lump in the left breast, upper outer quadrant, present for many years, increased in size over the most recent 6 months. (a) Spot compression-magnification craniocaudal view of the left breast demonstrates a small circumscribed oval mass in the upper outer quadrant of the left breast. The mass contains multiple small l dystrophic calcifications. (b) Spot compression-magnification mediolateral oblique view of the left breast demonstrates a small circumscribed oval mass in the upper outer quadrant of the left breast. The mass contains multiple small dystrophic calcifications. (c) Digitally magnified mediolateral oblique view of the left breast focusing on the lesion, better demonstrating the oval shape, circumscribed margins and multiple small dystrophic calcifications (white arrow).

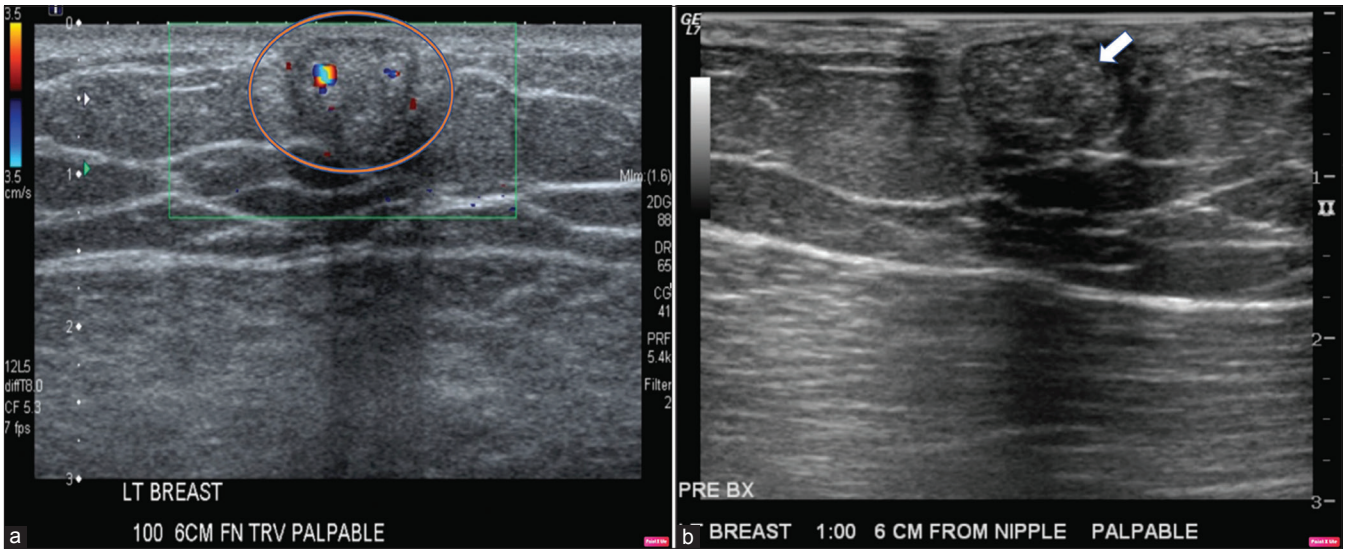

Figure 2: A 36-year-old HIV positive male presented with a painless hard palpable "pea-size" lump in the left breast, upper outer quadrant, present for many years, increased in size over the most recent 6 months. (a) A color doppler ultrasound image of the lesion shows internal vascularity. (b) A grayscale ultrasound image pre-biopsy better demonstrating the small hyperechoic foci of calcifications (white arrow) observed on the mammogram.

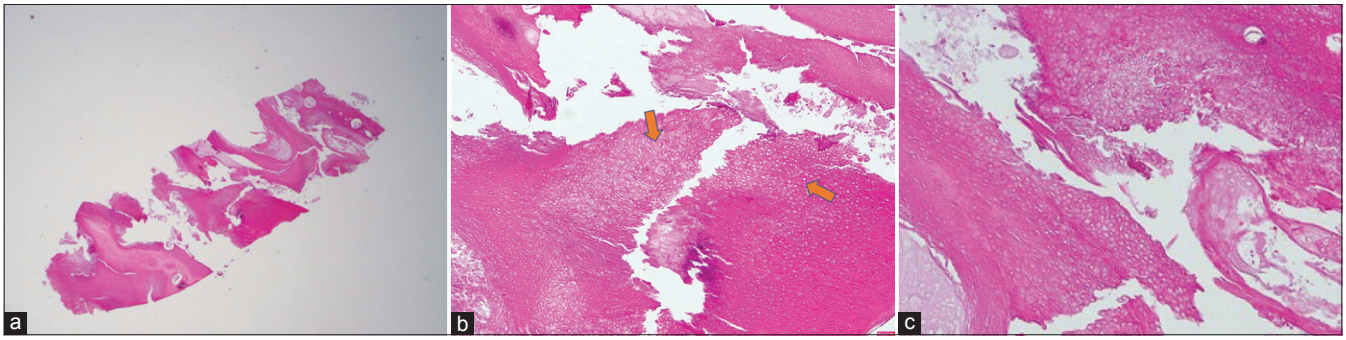

Figure 3: A 36-year-old HIV positive male presented with a painless hard palpable "pea-size" lump in the left breast, upper outer quadrant, present for many years, increased in size over the most recent 6 months. (a) Low power core biopsy specimen of a predominantly keratinizing neoplasm. (Hemotoxylin and eosin, 20´). (b) Eosinophilic keratin debris with the characteristic finding of "ghosted" nuclei (orange arrow) that is diagnostic and classically seen in pilomatrixoma. (Hematoxylin and eosin, 100´). (c) Basaloid cells with abrupt trichilemmal keratinization towards the center forming the characteristic "ghost cells." (Hematoxylin and eosin, 100´).

rubor, calor, dolor and rarely ulceration.[2,3] Mammographically, they demonstrate pleomorphic and coarse irregular calcifications (American College of Radiology BI-RADS IV–V) whose number can increase gradually, simulating the microcalcifications often associated with breast cancer.[1] Sonographically, these lesions appear as hypoechoic or isoechoic masses, with circumscribed or non-circumscribed margins, and hyperechoic internal foci representing calcifications. Posterior features may be present or absent, with the most commonly reported posterior acoustic shadowing.[2,4]

Histopathological and cytological features

These lesions can originate from the peri-areolar piliferous bulbs, and all case reports to date, report that it can mimic a breast malignancy.[2-8] Histologically pilomatrixoma is made of epithelial cells organized in nodular aggregates in a connective tissue matrix with peripheral scattered inflammatory-like elements. Each nodule is characterized by two types of epithelial cells with different organization: peripherally are densely packed viable basophilic cells producing keratin, while centrally, are the non-viable eosinophilic cells with clear spaces indicating previous nuclei, also known as "ghost" or "shadow" cells. Moreover, hair, calcifications, foci of necrosis and multinucleated giant cells can be often found.[5]

Cytological diagnosis can, however, be challenging, resulting in cases of misdiagnosis. Cytologically these tumors demonstrate a pathognomonic pattern of typical clusters of

basaloid cells surrounded by a delicate red fibrillar material, calcium deposits, and naked nuclei.[6] The first report on the cytology of pilomatrixoma was published by Woyke *et al*. in 1982. The results of this paper show that it is possible to arrive at a conclusive diagnosis of pilomatrixoma on fine needle aspiration (FNA) smears after a careful analysis of all cytological features, even in cases with an uncommon clinical presentation.[9] Literature emphasizes the need for a core needle biopsy. It is our opinion and experience that utilizing a 14 Gauge needle with a minimum of 3 passes ensures adequate sampling.

Differential diagnosis

Differential diagnosis of breast pilomatrixoma can be very challenging. The differential is based on masses with calcifications and includes skin calcifying lesions (seborrheic keratosis and inclusion cysts), fibrocystic changes (usual ductal hyperplasia, adenosis, apocrine metaplasia), lobular neoplasias, papillomas, calcified fibroadenomas, fat necrosis, and invasive ductal carcinoma.

Prognosis

Despite a very low predisposition for malignant degeneration, with only a few occasional reported cases of pilomatrixoma carcinomas of the breast, surgical excision is the treatment of choice, mainly for symptomatic and cosmetic reasons.[6] Conservative management has been reported and has been suggested in cases of benign mammographic or sonographic features, similar to our case, specifically describing a superficial circumscribed oval mass, parallel to the skin.[10] These cases can be assigned into a BI-RADS 3 category with a short term usually 6, 12 and 24 months interval to ensure stability.[11]

CONCLUSION

Pilomatrixoma of the male breast is a very rare breast lesion resembling breast carcinomas, and though rare, it should be considered in the differential of male breast mass.[9,10] This mass cannot be confidently diagnosed by mammographic and sonographic features alone and a core tissue biopsy is almost always necessary to obtain a diagnosis. Knowledge of the imaging features is helpful to establish concordance to the histological/pathological analysis and to decide whether surgical excision is necessary. Deferring biopsy or excision can be an option when benign ultrasound characteristics are present. FNA has been deemed to be diagnostic and may be considered as an option in the presence of an astute pathologist when index of suspicion is high and appropriate differential information has been relayed.

Declaration of patient consent

Patient's consent not obtained as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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