

Pilomatrixoma of the Male Breast: A Rare Case

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Received: 15 November 2022

Accepted: 4 July 2022

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DOI 10.5001/omj.2023.30

Abstract

Pilomatrixoma is a rare benign skin adnexal tumor arising from the hair follicles. Its occurrence in the breast is extremely uncommon with very few cases reported internationally. It may be masqueraded as a malignancy due to foci of calcification noted in this lesion. We present a case of a 51-year-old man who had a six-month history of a firm to hard right breast mass. The mammographic and sonographic features were suggestive of skin related lesion. The lesion was excised, and histopathology confirmed the diagnosis of pilomatrixoma.

Keywords: Pilomatrixoma; Rare Case.

Introduction

After an extensive literature search, to the best of our knowledge, this is the first reported case of pilomatrixoma of the breast in Oman. We share this rare case with a brief review of the currently available literature.

Case report

A 51 years old gentleman with no obvious comorbidities presented with a right breast swelling for 6 months. Though there was no increase in size, it was associated with pricking pain. No nipple discharge was noted. There was no history of trauma. Family history revealed breast cancer in his mother. On examination, both breasts looked symmetrical with normal-looking nipple and areolar complex. A small nontender, hard 2.0 x 1.0 cm lump was noted at 2 o'clock in the right breast attached to unremarkable skin. Mammography showed a well-defined dense lesion on the medial aspect of the peri-areolar region measuring 1.6 x 1.0 cm with peripheral microcalcifications. Ultrasound of the right breast also confirmed the presence of well-defined subcutaneous, homogenous, hypoechoic oval lesion of similar dimensions located 0.5 cm from the nipple. Excision of the lesion was carried out under local anesthesia in the breast clinic and subjected to histopathological examination. Grossly, the resected specimen showed a well-circumscribed, firm, oval nodule measuring 2.0 x 1.7 x 1.7 cm with an overlying ellipse of skin measuring 1.8 x 0.6 cm. On slicing, the nodule shows a solid, yellow and gritty cut surface due to areas of calcification. Microscopic examination revealed a neoplasm in the dermis composed of solid nests of basaloid cells undergoing abrupt trichilemmal-type keratinization resulting in eosinophilic 'shadow' or 'ghost cells'. Scattered foci of calcification with foreign body reaction were also noted with infiltration by chronic inflammatory cells in the stroma. These findings

were consistent with pilomatrixoma. The patient was followed in the clinic, 10 days after the excision. His wound healed well. The patient was reassured of the benevolent nature of the lesion and its complete excision.



Figure 1: Mammography of the right breast showing well defined isodense opacities with fat content medially in the peri-areolar region and dense calcifications.

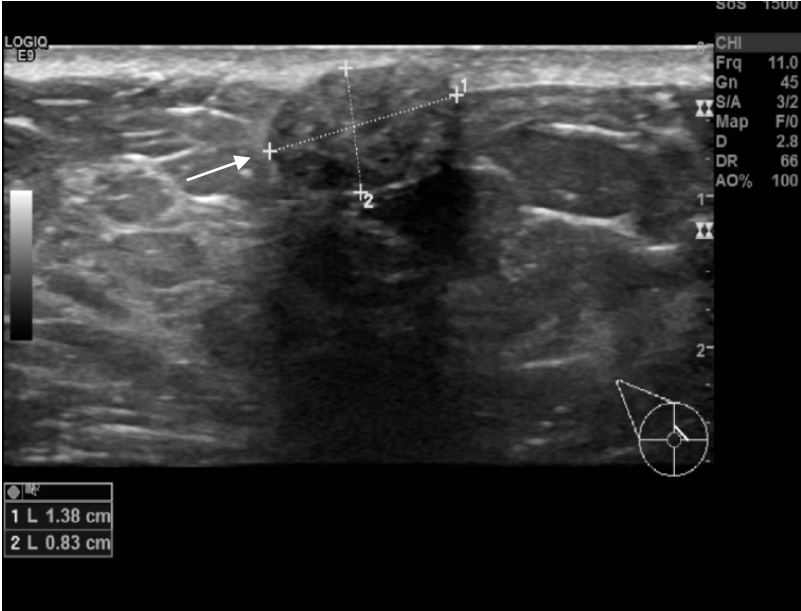


Figure 2: Ultrasound of the right breast demonstrating well defined subcutaneous homogenous hypoechoic oval lesion measuring 1.4 x 0.8 cm at 3 o'clock with a 0.5 cm distance from the nipple.

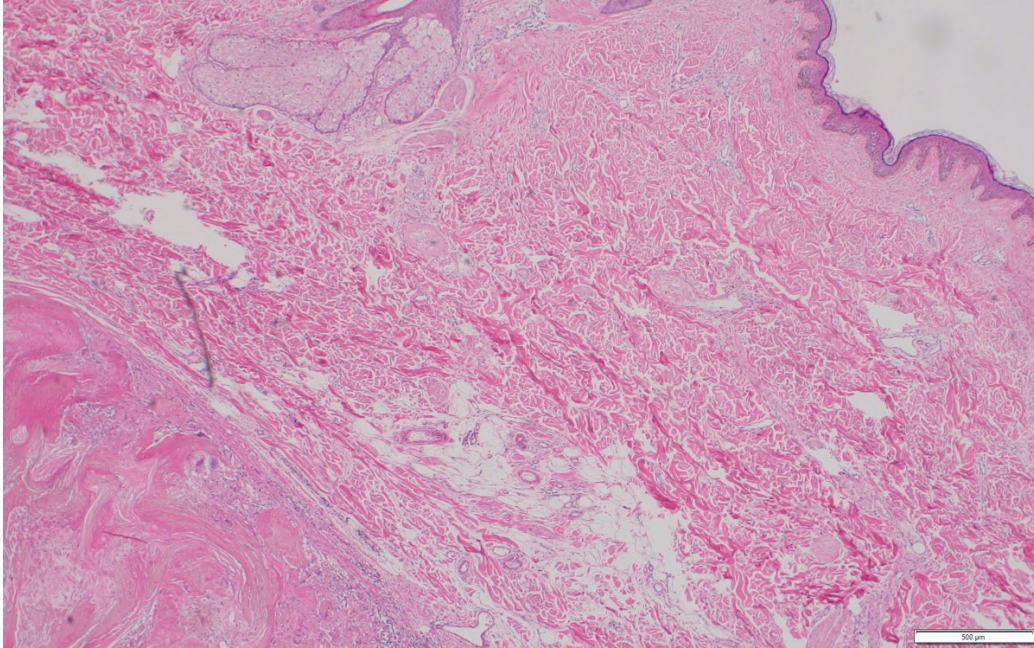


Figure 3: Overlying normal epidermis with well circumscribed dermal lesion on extreme left. (H&E x 50x).

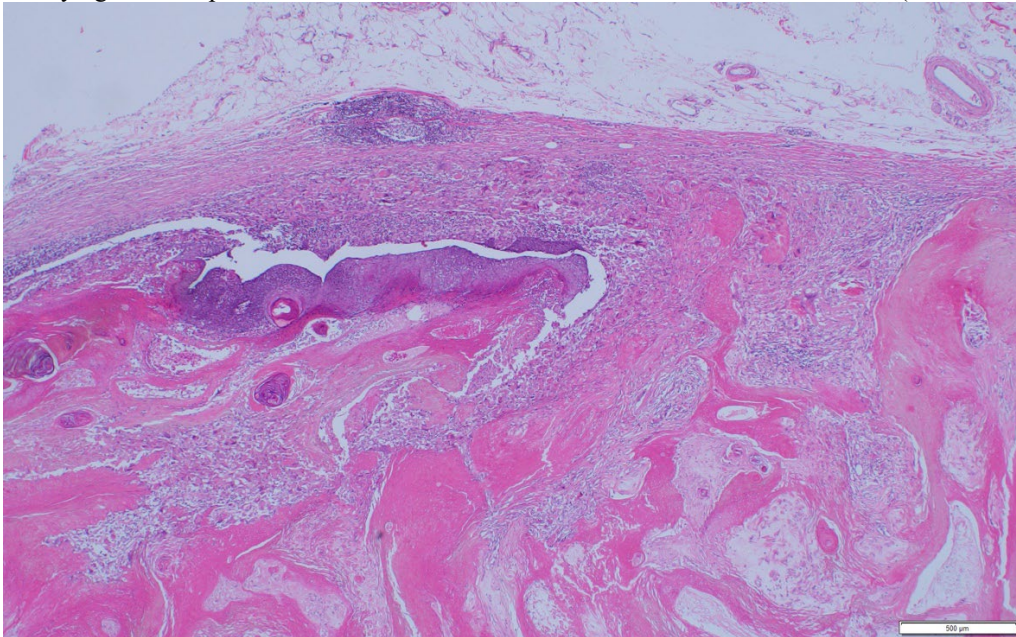


Figure 4: Scanner view of pilomatrixoma comprising all the components viz. basaloid cells, ghost cells and giant cells. (H&E 50x).

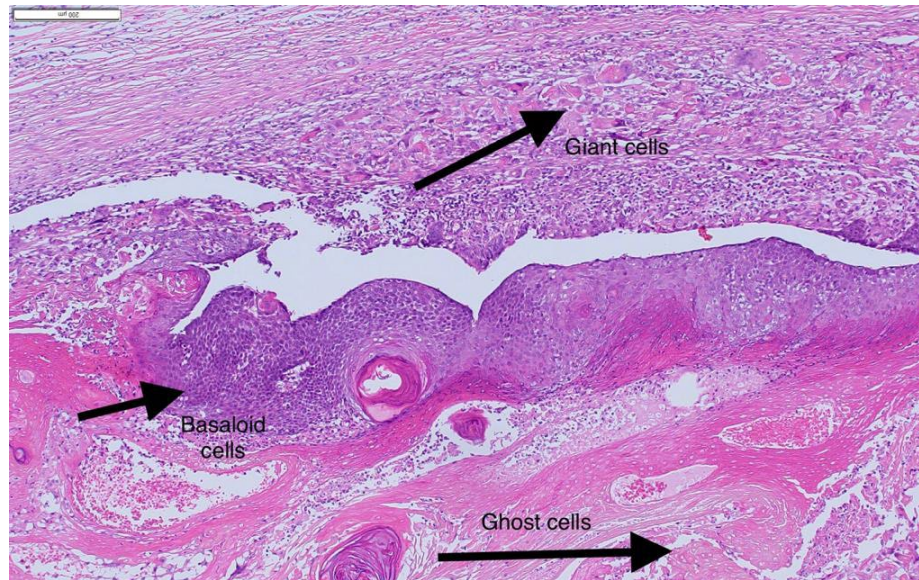


Figure 5: Microphotograph of Pilomatrixoma comprising nests of basaloid cells, ghost cells and multinucleated giant cells. (H&Ex100).

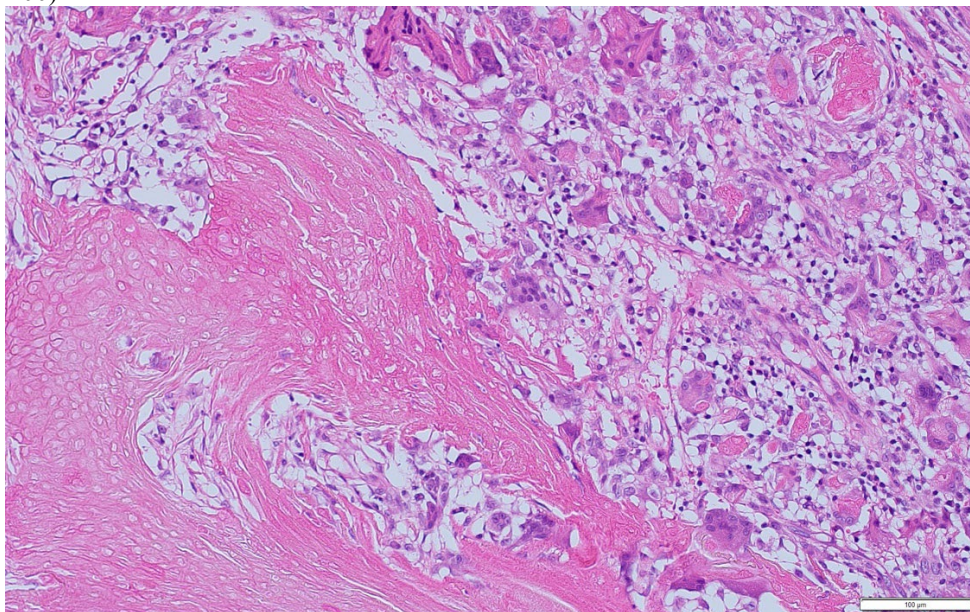


Figure 6: Ghost cells and multinucleated giant cells. (H&E x 200).

Discussion

Pilomatrixoma, also known as Pilomatrixoma or “Calcifying Epithelioma of Malherbe” was first recognized in 1880 by Malherbe and Chenanatais.¹ It is a rare, slow growing benign neoplasm originating from hair matrix that is more common in children and young adults with a slight female preponderance.² in the Pilomatrixoma can be seen in any hair bearing skin, but most commonly in the head and neck region. Less commonly they are noted in the upper extremities, trunk, and lower extremities.³ Pilomatrixoma’s appearance in the breast is considered to be very rare (1:100000 people). After extensive research, we found 9 cases of pilomatrixoma of the breast in adult males reported in the english literature (Table 1).

These tumors are usually sporadic but around 75% of the cases have mutations in the CTNNB1 gene which regulates beta-catenin thereby triggering quick, uncontrolled division of matrix cells leading to pilomatrixoma. Some cases were also found to be related to syndromes such as Gardner syndrome and myotonic dystrophy.⁴

The exact pathogenesis of Pilomatrixoma is still unclear, however, some suggest that recurrent skin trauma induces an inflammatory response leading to hair matrix overgrowth while others considered them to be hamartomas.⁵

It usually presents as an asymptomatic, single, subcutaneous, firm nodule attached to the skin with a size varying from 1-3 cm, but rarely they may attain a size more than 5 cm, often designated as giant pilomatrixoma. Some of these lesions may be associated red-blue discoloration of the skin and ulceration.⁶

Mammography commonly demonstrates a dense mass with multiple, irregular, pleomorphic microcalcifications.⁷ In addition, Sonography shows a well-circumscribed, oval mass with internal echogenic foci and a peripheral hypoechoic rim which is comparable to the US findings in our case. If these lesions get calcified, they would appear as hyperechoic mass with posterior acoustic shadowing.

Although imaging studies are useful in determining the size and location of the lesion, a core needle biopsy is the modality of choice for confirmation of diagnosis. Its histopathological features typically consist of two types of cells surrounded by foreign-body giant cells and calcifications. These are “shadow” or “ghost” cells which are located centrally and basophilic cells at the periphery.⁷

Pilomatrixomas are benign and the tendency to develop into malignancy is extremely rare with very few reported cases of pilomatrix carcinoma. The majority of the literature agrees on surgical excision as a treatment of choice.⁸ It not only serves the cosmetic purpose but also prevents the rare possibility of malignant transformation of the tumor. On the other hand, some suggest a conservative approach for patients with a small superficial breast mass showing benign imaging characteristics. These include masses of BI-RADS 3 category without pathologic diagnosis or benign biopsy results.³

Table 1: Reported cases of pilomatrixoma of the breast in adult males.

Reference	Age	Clinical features	Radiological findings	Treatment
Hubeny,et al. 2011	53	Painless, hard, right sided breast lump	Mammography: “well-circumscribed, oval-shaped mass of the medial right breast, with smooth borders, and multiple small punctuate calcifications” Ultrasound: ” small superficial isoechoic well-marginated oval mass parallel to the skin, with smooth borders, and multiple hyperechoic foci”	Surgical excision
M.M.Martins, et al.2014	47	Painful, firm, right sided breast nodule	Mammography: “nodule with indistinct limits and microcalcifications” Ultrasound: “lobulated node, isoechoic, parallel to the skin, with multiple hyperechoic foci”	Surgical excision
S.Alsharif, et al.2015	43	Painless, firm, left sided breast mass with bluish skin discoloration	Mammography: ” single, superficial, well-defined, and encapsulated 2.3-cm mass containing amorphous microcalcifications in the inferomedial aspect of the left breast” Ultrasound: “well-defined, subcutaneous isoechoic mass containing multiple bright foci”	Surgical excision with wide margins

Jea Ryoung Gil, et al.2016	69	Painless, palpable, erythematous, ulcerative left sided breast mass	Mammography: “irregular, exophytic high-density mass with dystrophic calcifications and coarse heterogeneous calcifications” Ultrasound: ” irregular, indistinct, exophytic hyperechoic mass”	Surgical excision
Kapoor, et al.2018	48	Painless, firm, left sided breast lump	Mammography: “solitary oval, well-circumscribed, high-density focal mass in the lower inner quadrant of left breast with a radiolucent halo” Ultrasound: ” well-defined, oval, hypoechoic solid mass measuring 1.0 3 0.8 cm in the parareolar region at the 6–7 o’clock position in the subcutaneous tissues”	Surgical excision
Ward, et al.2018	56	Painless, firm, left sided breast lump	Mammography: ” circumscribed mass, within superficial subcutaneous fat, abutting the skin undersurface, within the left breast at 9 o’clock, 12 cm from the nipple” Ultrasound: ” 1.8 cm oval circumscribed and mildly hypoechoic mass, inseparable from the skin undersurface”	-
Clark, et al.2019	36	Painless hard, left sided breast lump	Mammography: ” Oval-shaped, circumscribed mass, in the upper outer left breast, with smooth borders, and multiple pleomorphic calcifications “ Ultrasound: “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”	Surgical excision was recommended (not done due to limited finances)
Tokur, et al.2021	42	Painless, firm, right sided breast mass	Mammography: “well-circumscribed, approximately 3 2-cm sized, oval density with coarse and fine pleomorphic calcifications in the centrally located lesion in the right breast” Ultrasound: ” heterogeneous echo pattern with significant acoustic shadowing due to calcifications in the mass”	Surgical excision
Sood N,et al.2021	48	Painless, firm, left sided breast lump.	Mammography: “BIRADS II”	Surgical excision

Conclusion

Pilomatrixoma is a rare benign lesion seen rarely in males and when it presents in the breast it can mimic breast carcinoma because of its associated calcifications. Therefore, it is prudent to consider pilomatroxoma as a differential diagnosis in patients presenting with superficially located breast lumps.

Disclosure

No conflict of interests.

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