

# Pilomatrixoma of the Male Breast: Case Report and Literature Review

Maryam Al-Hssai<sup>1\*</sup>, Salim Al-Rahbi<sup>2</sup>, Anfal Al-Futaisi<sup>1</sup>, Uday Arun Gokhale<sup>3</sup> and Samya Al-Salhi<sup>4</sup>

<sup>1</sup>College of Medicine and Health Sciences, Sultan Qaboos University, Muscat, Oman

<sup>2</sup>Department of General Surgery, Royal Hospital, Muscat, Oman

<sup>3</sup>Department of Histopathology, Royal Hospital, Muscat, Oman

<sup>4</sup>Department of Radiology, Royal Hospital, Muscat, Oman

## ARTICLE INFO

### Article history:

Received: 15 November 2021

Accepted: 4 July 2022

### Online:

DOI 10.5001/omj.2023.30

### Keywords:

Pilomatrixoma; Breast Tumors; Oman.

## ABSTRACT

Pilomatrixoma is a rare benign skin adnexal tumor arising from the hair follicles. Its occurrence in the male breast is extremely uncommon with very few cases reported worldwide. Pilomatrixoma may masquerade as a malignant tumor due to the presence of foci of calcification. We present a case of a 51-year-old man with a six-month history of a firm-to-hard mass in his right breast. The mammographic and sonographic features were suggestive of skin-related lesions. The lesion was excised. Histopathology confirmed the diagnosis of pilomatrixoma.

After an extensive literature search, 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

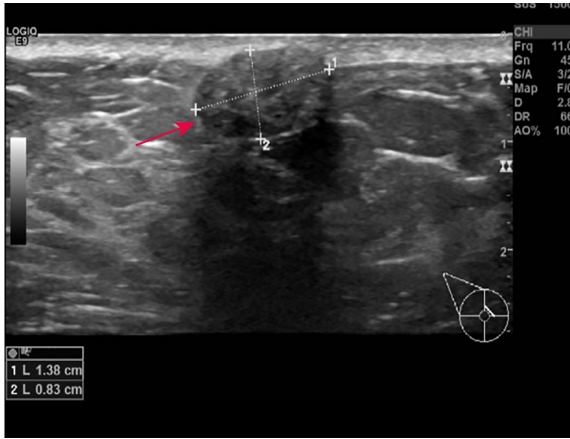
A 51-year-old man with no comorbidities presented with six months of swelling on his right breast. Though the swelling did not increase in size over time, it was associated with pricking pain. No nipple discharge was noted. There was no history of trauma. Family history revealed that his mother had breast cancer.

On examination, both breasts looked symmetrical with normal nipple-areolar complex. A small nontender, hard  $2.0 \times 1.0$  cm lump was noted at two o'clock in the right breast attached to unremarkable skin. Mammography showed a well-defined dense lesion on the medial aspect of the periareolar region measuring  $1.6 \times 1.0$  cm with peripheral microcalcifications [Figure 1]. Ultrasound of the right breast also confirmed the presence of a well-defined homogeneous, hypoechoic, subcutaneous oval lesion of similar dimensions located 0.5 cm from the nipple [Figure 2]. The lesion was excised under local anesthesia in the breast clinic.

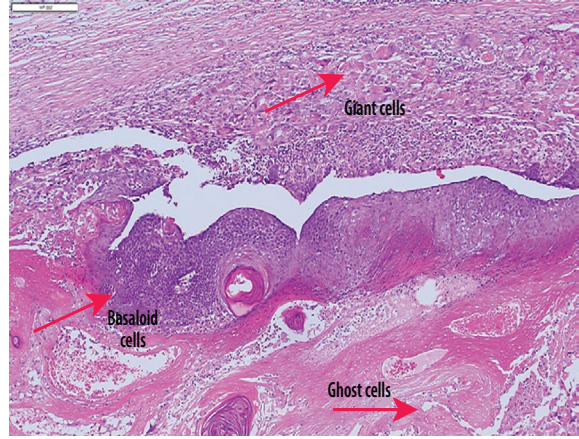


**Figure 1:** Mammography of the right breast showing well-defined isodense opacities with fat content and dense calcifications medially in the periareolar region.

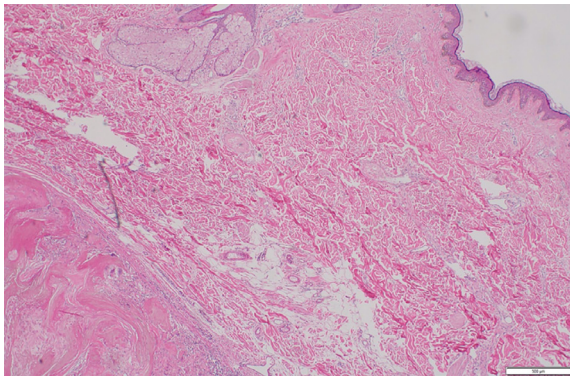
The resected specimen was subjected to histopathological examination. Grossly, it was a well-circumscribed, firm, oval nodule measuring  $2.0 \times 1.7$



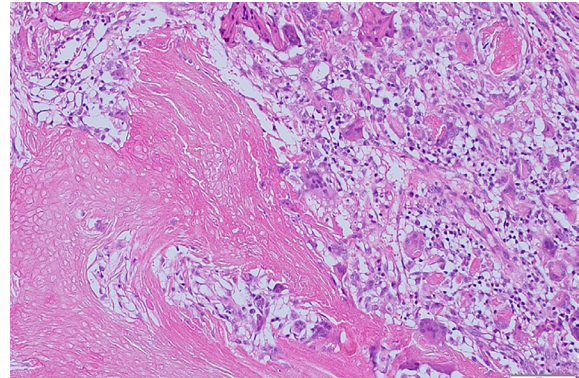
**Figure 2:** Ultrasound of the right breast showed a well-defined homogeneous, hypoechoic, subcutaneous oval lesion measuring 1.4 × 0.8 cm at three o'clock, 0.5 cm away from the nipple.



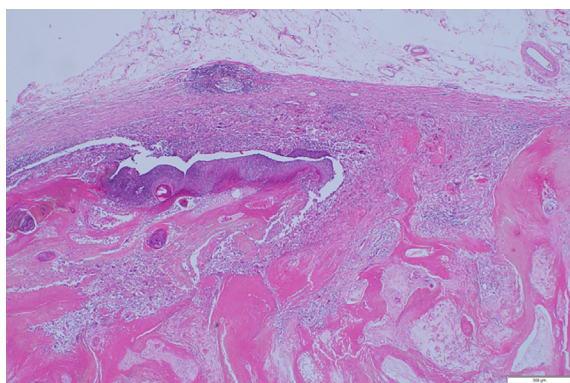
**Figure 5:** Microphotograph of pilomatrixoma comprising nests of basaloid cells, ghost cells, and multinucleated giant cells (red arrows) (hematoxylin and eosin stain, magnification = 100 ×).



**Figure 3:** Overlying normal epidermis with a well-circumscribed dermal lesion on the extreme left (hematoxylin and eosin stain, magnification = 50 ×).



**Figure 6:** Ghost cells and multinucleated giant cells (hematoxylin and eosin stain, magnification = 200 ×).



**Figure 4:** Scanner view of pilomatrixoma comprising all the components viz. basaloid cells, ghost cells, and giant cells (hematoxylin and eosin stain, magnification = 50 ×).

× 1.7 cm with an overlying ellipse of skin measuring 1.8 × 0.6 cm [Figure 3]. On slicing, the nodule showed 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' [Figure 4]. Also noted in the stroma were scattered foci of calcification with foreign body reaction and infiltration by chronic inflammatory cells [Figures 5 and 6]. These findings were consistent with pilomatrixoma. When the patient visited the clinic 10 days after the excision, his wound had healed well. He was reassured that the lesion was benign and completely excised.

## DISCUSSION

Pilomatrixoma, also known as pilomatricoma or 'calcifying epithelioma of Malherbe' was first recognized in 1880 by Malherbe and Chenanatais.<sup>1</sup>

It is a rare, slow-growing benign neoplasm originating from the hair matrix, more common in children and young adults with a slight female preponderance.<sup>2</sup> While pilomatrixoma can develop in any hair-bearing skin, it is most commonly seen in the head and neck regions. Less commonly, they are noted in the upper extremities, trunk, and lower

extremities.<sup>3</sup> Pilomatrixoma's appearance in the breast is considered rare (1:100 000 people). After an extensive literature search, we found nine cases of pilomatrixoma of the breast in adult males reported in the English language [Table 1].

These tumors are usually sporadic but around 75% of the cases have mutations in the *CTNNA1*

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

Reference	Age	Clinical features	Radiological findings	Treatment
Hubeny et al. <sup>3</sup> 2011	53	Painless, hard, right sided breast lump	<b>Mammography:</b> well-circumscribed, oval-shaped mass of the medial right breast, with smooth borders, and multiple small punctuate calcifications. <b>Ultrasound:</b> small superficial isoechoic well-marginated oval mass parallel to the skin, with smooth borders, and multiple hyperechoic foci.	Surgical excision
Tokur et al. <sup>4</sup> 2021	42	Painless, firm, right sided breast mass	<b>Mammography:</b> well-circumscribed, approximately 3 × 2 cm, oval density with coarse and fine pleomorphic calcifications in the centrally located lesion in the right breast. <b>Ultrasound:</b> heterogeneous echo pattern with significant acoustic shadowing due to calcifications in the mass.	Surgical excision
Alsharif et al. <sup>5</sup> 2015	43	Painless, firm, left sided breast mass with bluish skin discoloration	<b>Mammography:</b> single, superficial, well-defined, and encapsulated 2.3 cm mass containing amorphous microcalcifications in the inferomedial aspect of the left breast. <b>Ultrasound:</b> well-defined subcutaneous isoechoic mass containing multiple bright foci.	Surgical excision with wide margins
Kapoor et al. <sup>6</sup> 2018	48	Painless, firm, left sided breast lump	<b>Mammography:</b> solitary oval, well-circumscribed, high-density focal mass in the lower inner quadrant of the left breast with a radiolucent halo. <b>Ultrasound:</b> well-defined oval, hypoechoic solid mass measuring 1.0 × 3 × 0.8 cm in the periareolar region at the 6–7 o'clock position in the subcutaneous tissues.	Surgical excision
Gil et al. <sup>7</sup> 2016	69	Painless, palpable, erythematous, ulcerative left sided breast mass	<b>Mammography:</b> irregular, exophytic high-density mass with dystrophic calcifications and coarse heterogeneous calcifications. <b>Ultrasound:</b> irregular, indistinct, exophytic hyperechoic mass.	Surgical excision
Martins et al. <sup>8</sup> 2014	47	Painful, firm, right sided breast nodule	<b>Mammography:</b> nodule with indistinct limits and microcalcifications. <b>Ultrasound:</b> lobulated node, isoechoic, parallel to the skin, with multiple hyperechoic foci.	Surgical excision
Ward et al. <sup>9</sup> 2019	56	Painless, firm, left sided breast lump	<b>Mammography:</b> circumscribed mass, within superficial subcutaneous fat, abutting the skin undersurface, within the left breast at 9 o'clock, 12 cm from the nipple. <b>Ultrasound:</b> 1.8 cm oval circumscribed and mildly hypoechoic mass, inseparable from the skin undersurface.	-
Clark et al. <sup>10</sup> 2019	36	Painless hard, left sided breast lump	<b>Mammography:</b> oval-shaped, circumscribed mass, in the upper outer left breast, with smooth borders, and multiple pleomorphic calcifications. <b>Ultrasound:</b> 1.0 × 0.8 × 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 financial constraints)
Sood et al. <sup>11</sup> 2021	48	Painless, firm, left sided breast lump	<b>Mammography:</b> breast imaging-reporting and data system category 2.	Surgical excision



gene (which regulates beta-catenin), thereby triggering quick, uncontrolled division of matrix cells leading to pilomatixoma. Some cases were also found to be related to Gardner syndrome and myotonic dystrophy.<sup>12</sup> The exact pathogenesis of pilomatixoma is still unclear, however, some suggest that recurrent skin trauma induces an inflammatory response leading to hair matrix overgrowth while others consider them to be hamartomas.<sup>11</sup> It usually presents as an asymptomatic, single, subcutaneous, firm nodule attached to the skin, sized 1–3 cm. Rarely, they may grow > 5 cm (giant pilomatixoma). Some of these lesions may be associated with red-blue discoloration of the skin and ulceration.<sup>13</sup>

Mammography commonly demonstrates a dense mass with multiple, irregular, pleomorphic microcalcifications.<sup>10</sup> In addition, Sonography may show a well-circumscribed, oval mass with internal echogenic foci and a peripheral hypoechoic rim, as it did in the present case. If these lesions get calcified, they would appear as hyperechoic masses with posterior acoustic shadowing.

Although imaging studies are useful in determining the size and location of the lesion, core needle biopsy is the modality of choice for confirmation of diagnosis. 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.<sup>10</sup>

Pilomatixomas are benign and the tendency to develop into malignancy is extremely rare with very few reported cases of pilomatix carcinoma. The majority of the literature agrees on surgical excision as the treatment of choice.<sup>9</sup> It serves the cosmetic purpose and 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 breast imaging-reporting and data system category 3 without pathologic diagnosis or benign biopsy results.<sup>3</sup>

## CONCLUSION

Pilomatixoma in the male breast is a rare benign lesion. It can mimic breast carcinoma because of associated calcification. Therefore, it is prudent to

consider pilomatixoma as a differential diagnosis in patients presenting with superficially located breast lumps.

## Disclosure

The authors declared no conflicts of interest. A written consent was obtained from the patient.

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