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Case Report

Recurrent myoid hamartoma of the breast mimicking malignancy

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ABSTRACT

Myoid (muscular) hamartoma is a rare form of benign breast hamartoma composed of differentiated mammary glandular and stromal structures, fatty tissue and areas of smooth muscle from which its name originates. It is considered to be a variant of a mammary hamartoma. We report the clinical presentation, imaging appearances and treatment of the initial and recurrent presentation of this rare tumour in a 61-year old female, which mimicked malignancy. Although rare, myoid hamartoma's can reoccur and when they do they imaging appearances of benign and malignant tumours can overlap tend to mimic malignancy and histological diagnosis is mandatory.

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Introduction

Breast hamartoma accounts for approximately 0.7%-5% of all benign breast masses [1] Myoid hamartoma is a rare subgroup composed of differentiated mammary glandular, stromal structures and fatty tissue but with areas of smooth muscle spindle cells from which its name originates.

Most hamartomas are dismissed by radiologist when identified on mammography as they have a typical appearance of glandular and fat containing lesion, often with a well-defined thin capsule which mirrors the ultrasound appearances of normal glandular and fatty tissue. But the spindle cell muscular component of the myxoid hamartoma can cause increased density on the mammogram and ultrasound

therefore the differential becomes wider including fibroepithelial lesions and malignancy. Recurrence after successful initial surgical treatment is extremely rare with only 2 previous case reports. We describe the presentation, imaging appearances and treatment of this rare and unusual case of a recurrent myoid hamartoma mimicking breast malignancy.

Case report

A 61-year-old female initially presented with a lump in the right retro areolar area. She was normally fit and well, no prior history of breast disease or family history of breast cancer. On

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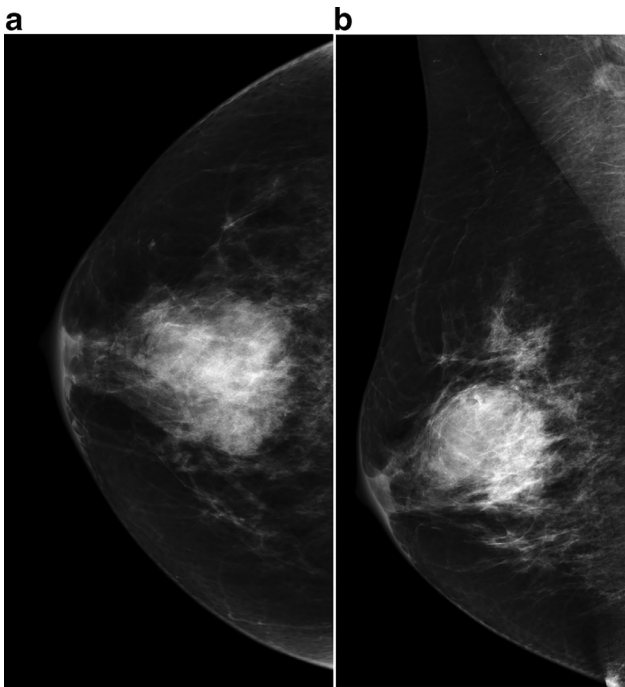


Fig. 1 – Right craniocaudal (CC) (Fig 1A) and right medial lateral-oblique mammogram (mlo) (Fig 1B) . This demonstrated the retro-areolar lobulated round mass, with indistinct margins (BI- RADS 3) that was concordant with the clinical mass at initial presentation.

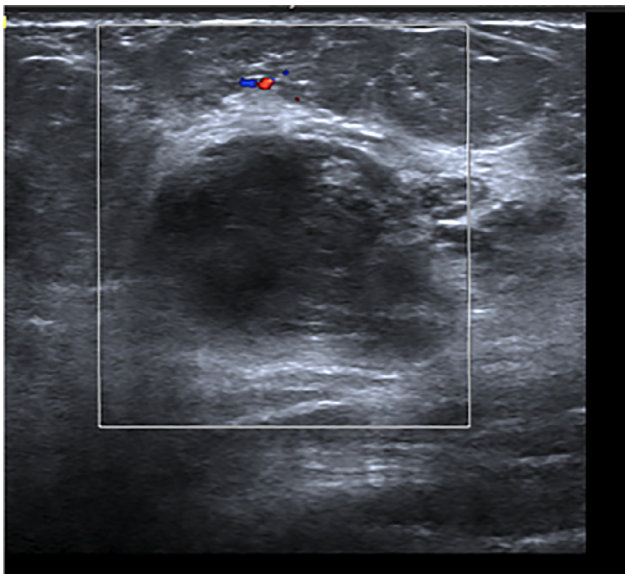


Fig. 2 – US demonstrated a demonstrated a 30 mm oval hypoechoic partially circumscribed partially microlobulated mass with no posterior features and no significant flow on colour Doppler.

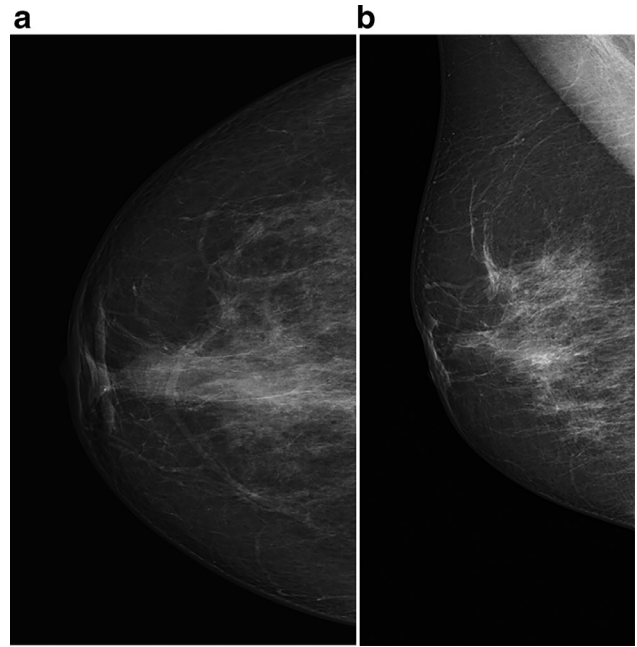


Fig. 3 – . Normal right CC (Fig 3A) and right MLO mammogram (Fig 3B) taken at screening 2 years later .

clinical examination a well-defined, round mass was palpated and clinically was indeterminate, probably benign.

Digital mammography identified a mass in the right retro areolar area measuring 37mm which was round with indistinct margins which was not seen 1 year earlier on her screening mammograms (Figs. 1A and B). This was classified as indeterminate BI-RADS 3 [2].

Ultrasound identified a 30mm oval hypoechoic partially circumscribed, partially micro lobulated mass with no posterior acoustic features (Fig. 2). Ultrasound appearances were indeterminate. The axillary US were normal. US guided core biopsy was performed using a 14 G needle. Three passes were made which identified a cellular fibro epithelial lesion, possible benign phyllodes. As per multidisciplinary team (MDT) review, a surgical excision was performed which identified a lesion with cellular stroma, prominent smooth muscle differentiation, disorganized glandular elements admixed with adipose tissue in keeping with benign myoid hamartoma with no atypia. All surgical margins were clear. The MDT decision was to discharge the patient.

A screening digital mammogram 2 years later was normal with no evidence of a mass (Figs. 3A and B).

Five years from the initial diagnosis and 3 years from the normal screening mammogram, she presented with a palpable irregular mass in the same region, which was clinically suspicious for malignancy. Digital mammography identified a 40 mm spiculated mass in the right retroareolar region suspicious for malignancy, BI-RADS 4 (Figs. 4A and B). On US there was an irregular, spiculated hypoechoic mass measuring 34 mm with no posterior acoustic features (Fig. 5). The appearances were suspicious for malignancy. The axilla was normal on US. Core biopsy using 14 G needle identified

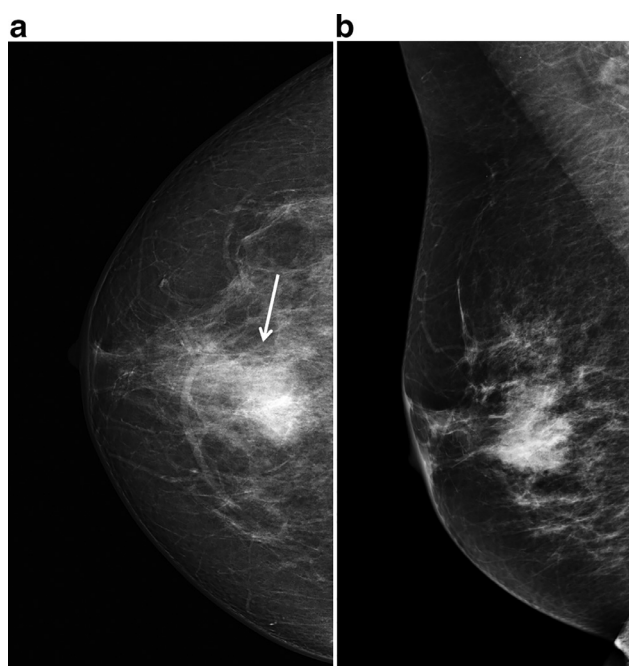


Fig. 4 – A & 4B. Right CC (Fig 4A) and right MLO mammogram (Fig 4B) demonstrated a 40mm irregular mass, partially spiculated mass in the right retroareolar region BI-RADS 4.

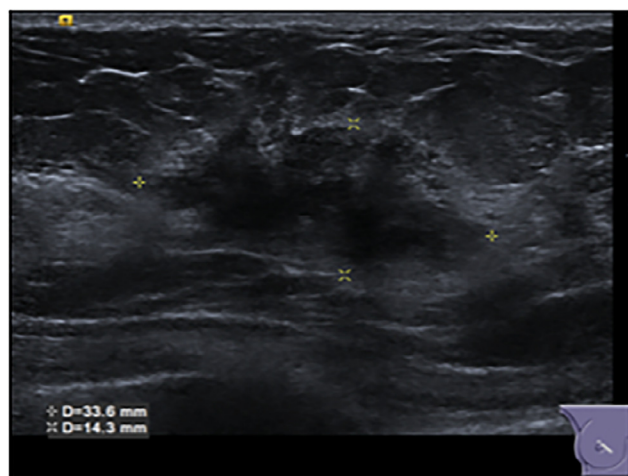


Fig. 5 – US demonstrated a demonstrated an irregular hypoechoic mass measuring 34mm with no posterior features with US appearances suspicious for malignancy.

fibroepithelial lesion consistent with the previous diagnosis of myoid hamartoma with no evidence of malignancy.

The MDT decision again was surgical excision of the lesion, which confirmed benign myoid hamartoma with no atypia and clear surgical margins (Figs. 6 A-D). Although there is no

evidence for follow up in benign tumors, due to this unusual recurrence, and quick growth of the tumour, MDT decision was to perform a mammogram in 1 year.

Discussion

Myoid hamartomas are characterized by the presence of histologically normal, but irregular and randomly distributed smooth muscle cells [3] and first described in 1973 by Davies and Riddell [4]. The mean age of diagnosis is 41 years and although usually an exclusively female tumor, Ravakhah et al [5] described a male case confirmed by excision biopsy. Myoid hamartoma have a wide range of appearances with classical benign circumscribed lesion similar to fibroadenomas to irregular lesions suspicious of malignancy.

The literature usually describes the lesions as well-circumscribed, oval or round opacity on mammography, with or without a thin capsule. Ultrasonography reveals an internally iso or hypoechoic, solid mass with regular margins often similar to fibroadenoma [6]. Few reports have described dynamic MRI findings of early and significant rise of signal intensity curves although this is not specific [7]. Both the initial presentation and recurrence in our patient were suspicious of malignancy. In this case the initial presentation had more benign features on imaging, but more malignant features on the recurrent imaging.

The histological differential diagnosis of myoid hamartoma should include various benign and malignant spindle cell tumors and tumor-like lesions. Lesions that should be considered include fibroadenoma, adenomyoepithelioma, leiomyosarcoma, and metaplastic breast carcinoma [8]. Immunohistochemical studies are helpful in making a diagnosis with several reports suggesting spindle and epithelioid tumor cells show strong positive staining for SMA, desmin and vimentin and the absence of staining for cytokeratin as well as S-100 protein [9]. Some case reports have reported ER and PR positivity in epithelial and stromal cells in most cases of breast hamartoma's suggesting impact of female sex hormones on cellular growth [10]. Good volume histological cores are needed in these cases as the presence of spindle and epithelial cells can mimic phyllodes tumours which initially occurred in this case. This is why surgical excision for these fibroepithelial lesions is recommended to reach a definitive diagnosis rather than Vacuum excision [11].

Breast hamartomas are rarely associated with malignancies, and excision is considered curative with no adjuvant therapy needed. Recurrence is seen in approximately 8% of reported breast hamartomas although specific data for recurrence of myoid hamartoma are not available [12].

To our knowledge, case reports of tumor recurrence are very rare with only 2 cases reported by Linell et al [13] and Ko Myung-So et al [14] in which the recurrent lesion also mimicked a malignancy as in our case.

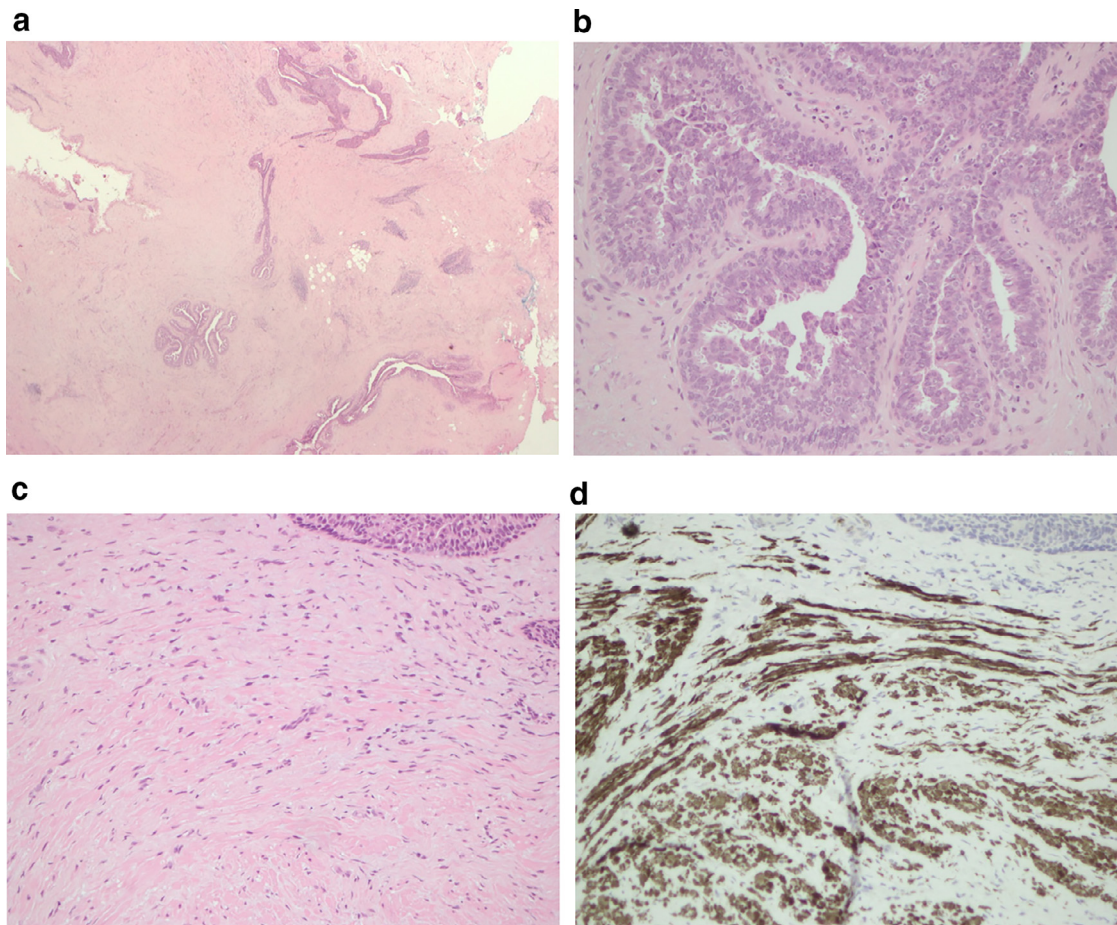


Fig. 6 – A Histology shows a fibroepithelial lesion with a proliferation of disorganised glandular elements embedded in variably cellular stroma. **B** The epithelial component shows gynaecomastoid type of usual epithelial hyperplasia. **C** and **D**. The stroma is composed of bland spindle cells (**C**) that are actin and desmin (**D**) positive in keeping with smooth muscle differentiation.

Conclusion

Myoid hamartoma although an exceedingly rare subset of benign breast hamartoma can have a wide range of imaging appearances from benign to malignant and can reoccur even with clear surgical margins. Biopsy and surgical excision are mandatory to manage the patient pathway correctly in line with national guidelines, not miss malignancy. When recurrence occurs, it appears to frequently mimic a malignancy

Patient consent

Patient consent for publication of this case was obtained.

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