

Case Report

Myoid hamartoma of breast: a rare case report with review of literature

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ABSTRACT

Myoid hamartomas of breast are extremely rare lesions with poorly defined pathogenesis. They are composed of differentiated mammary glandular and stromal structures with areas of smooth muscle differentiation. They are postulated to arise from walls of the blood vessels and/or muscularis mamillae of the areolae. They usually present as a well demarcated lump and it is difficult to diagnose them on clinical and radiological basis. Diagnosis requires demonstration of smooth muscle phenotype using immunohistochemistry by smooth muscle actin and desmin. Surgical resection is the curative treatment. We reported a case of myoid hamartoma in a 50-year-old postmenopausal lady with its clinical, radiological and histopathological discussion. The lesion presented as a painless lump and was initially reported as BIRADS 4a on mammography. Apart from the stroma showing spindle cell component with smooth muscle differentiation, the histopathology also showed epithelial changes including epithelial hyperplasia and columnar cell change.

Keywords: Mammary hamatoma, Myoid hamartoma, Breast lump, BIRADS

INTRODUCTION

Hamartomas of the breast are benign lesions first described in 1971 by Arrigoni et al. They are also known as fibroadenolipomas, lipofibroadenomas or adenolipomas and account for 0.9-5% of benign breast masses.¹ They are characterized by a well-circumscribed lesion with a mixture of differentiated mammary tissue composed of glandular and stromal elements along with adipose tissue. Myoid hamartoma is considered to be a rare variant of mammary hamartomas and usually have a spindle cell component which shows smooth muscle differentiation. They present as painless lumps and are difficult to diagnose clinically or radiologically. There are rare case reports of ductal carcinoma arising in a previously diagnosed case of mammary hamartoma thus surgical resection is considered to be the curative treatment.^{2,3}

We reported a case of myoid hamartoma in a postmenopausal female that presented as painless lump with BIRADS 4a scoring on mammography.

CASE REPORT

A 50-year-old postmenopausal lady presented with a painless, gradually progressive lump in the upper outer quadrant of right breast for 6 months. The lump was firm and mobile with no overlying skin changes and examination of the left breast and bilateral axilla was unremarkable. Mammography revealed an irregular hypoechoic lesion with partly circumscribed margins and no focus of calcification. It was reported as BIRADS 4A (low suspicion of malignancy) (Figure 1). The patient underwent lumpectomy.

Macroscopically, the tumor was globular, measuring 3×3×2 cm and the cut section revealed a grey-white soft to firm lesion. No areas of necrosis or haemorrhage were

noted. Microscopically, hematoxylin and eosin staining showed a lesion comprised of haphazardly arranged breast ductules and lobules with focal mild to moderate epithelial hyperplasia (Figure 2). Foci of columnar cell change and columnar cell hyperplasia were also seen. The stroma was composed of spindled out cells arranged in fascicles and whorls with moderate amount of eosinophilic cytoplasm and evenly dispersed chromatin; morphologically resembling smooth muscle cells. There was no atypia or mitosis in the stromal component. These spindle cells showed strong and diffuse expression for smooth muscle actin on immunohistochemistry (Figure 3). A diagnosis of myoid hamartoma with features of epithelial hyperplasia, adenosis, and columnar cell change was rendered.

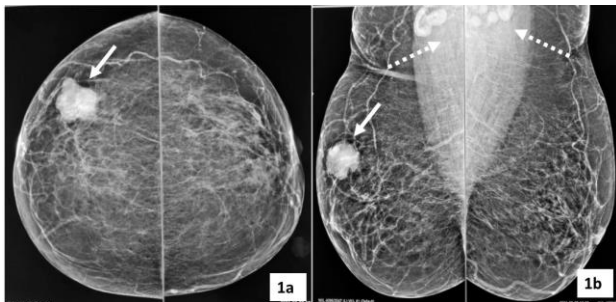


Figure 1: Craniocaudal (a) and mediolateral (b) mammographic views depict an irregular high density mass (arrows) in the upper outer quadrant of right breast with partially circumscribed margins; no focus of calcification is seen; note made of bilateral axillary lymph nodes with preserved fatty hilum (dashed blue arrows).

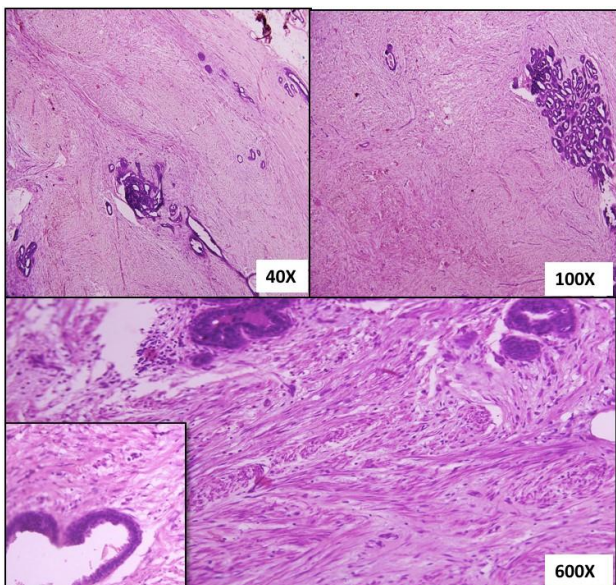


Figure 2: Closely admixed smooth muscle bundles in interlacing fascicles and bundles and breast ducts. Inset shows columnar cell change with apical cytoplasmic snouts (H&E).

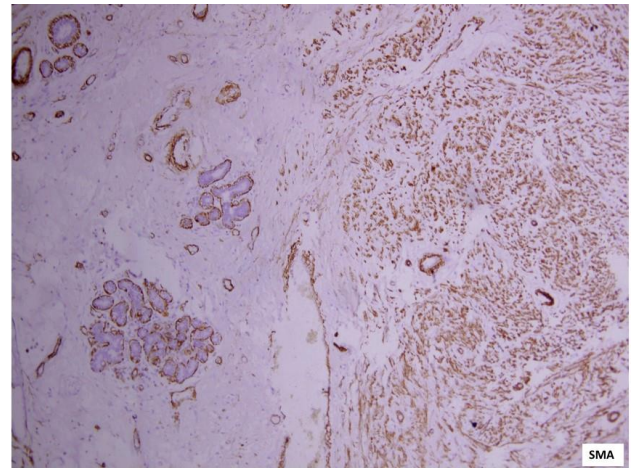


Figure 3: Strong immunoexpression of smooth muscle actin in stroma and the myoepithelial cells in the breast lobules and ducts (100×).

DISCUSSION

Mammary myoid hamartomas were first described by Davis and Riddell in 1973 via two case reports.⁴ Though the exact incidence of myoid hamartomas is unknown, they constitute an uncommon entity with unclear etiopathogenesis so far. They are more commonly seen in post-menopausal women, though a case has been reported in a 18-year-old female too.⁵ There also exists a case report of myoid hamartoma in a 35-year-old male.⁶ Clinically, they are indistinguishable from other benign breast diseases on account of their fibroelastic consistency and free mobility. Some of them might be asymptomatic and diagnosed incidentally. On sonography and mammography, they are hypo to isoechoic and may show characteristics of increased tissue density.

In the present case, additional changes were seen in the ductal component i.e., epithelial hyperplasia of mild to moderate degree, columnar cell change, and adenosis. Similar changes in epithelial and stromal components have been reported by other investigators. Su et al and Khoo et al reported foci of chondromyxoid metaplasia on histopathological evaluation.^{7,8} Also seen frequently are foci are adenosis, fibrocystic changes, epithelioid cell morphology and pseudoangiomatous stromal hyperplasia.^{7,9} The differential diagnoses on histology include adenosis, fibroadenoma and benign and malignant spindle cell lesions of breast, including leiomyoma, fibromatosis, myofibroblastoma, adenomyo-epithelioma, metaplastic breast carcinoma and benign tumours of peripheral nerve sheaths. Immunohistochemical analysis of the spindle cell component shows positive expression with smooth muscle actin, desmin and vimentin confirming the cells to have smooth muscle cell phenotype. Immunoreactivity with estrogen and progesterone receptor (ER and PR) has been reported in the stromal as well as epithelial cells suggesting the causative role of female hormones in the

progression of this entity.¹⁰ This is supported by the fact that they are almost invariably seen in females.

The etiopathogenesis of myoid hamartomas is unclear with varied theories. The myogenic phenotype in the breast hamartomas has been variably attributed to arise from smooth muscle cells of vessels, the nipple, undifferentiated breast tissue stroma, or myoepithelial cells.^{5,11} Some authors have also hypothesised metaplastic transformation of stromal cells into smooth muscle cells. This was supported with focal CD34 immunoreactivity in these cases.^{11,12} However, others studies do not concur.⁵ As for biologic behavior, recurrence is seen in 8% of mammary hamartomas and malignant transformation is an uncommon event.^{13,14} While there are rare reports of malignant transformation of myoid hamartomas, recurrence can be seen leading to strong clinical suspicion of malignancy, as reported by Prabhu et al and Ko et al.¹⁴⁻¹⁶ However, on account of rarity of mammary hamartomas, overall data on prognosis and biologic behaviour is insufficient. The patient in the present case underwent complete excision with no evidence of recurrence.

CONCLUSION

Myoid mammary hamartomas are rare benign breast lesions and are an uncommon morphologic variant of breast hamartomas. They have an unclear origin and need to be studied extensively with clinicoradiologic and pathologic correlation and patient follow up in order to ascertain their true nature.

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