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Case Report

Myoid hamartoma of breast clinically masquadering as phyllodes tumor- A rare case report

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ABSTRACT

Mammary hamartoma (MH) is an infrequent form of breast tumor that accounts for approximately 4.8% of all benign breast lesions. Myoid (muscular) hamartomas are uncommon benign breast lesions that are considered to be a variant of mammary hamartomas. Since the pathogenesis of MH is poorly understood, its true incidence is under-reported by clinicians and as well as pathologists.
Thus, we report this case of MH in a middle-aged female, who presented with a large breast mass mimicking phyllodes tumor and giant fibroadenoma, however, histopathology and immunohistochemical examination aided its diagnosis as Myoid Hamartoma. We present this case to highlight this rare benign breast lesion.

1. Introduction

Mammary hamartoma (MH) are uncommon and under-recognized benign neoplasm of the breast with an incidence of 0.1–0.7%. They are focal benign malformations that are characterized by the presence of abnormal growth of mammary tissues including ducts, lobules, stroma, and adipose tissue.

Hamartomas are described as an overgrowth of mature tissues that normally occur in an area of the body with a disorderly arrangement, with often one element predominating, while on the other hand, Choristomas is a mass of tissue histologically normal for a part of the body other than the one in which it is located (heterotopic/ectopic). Both the terms (choristoma and heterotopia) are used interchangeably.[1] A rare benign breast lesion known as a myoid (muscular) hamartoma is thought to be a subtype of mammary hamartoma.[2] Clinicians and pathologists underestimate the diagnosis of hamartomas because of its unknown pathophysiology. We present a case of a rare myoid breast hamartoma. Although the precise incidence of myoid hamartoma is unknown, less than 50 cases have been documented in the literature. In order to highlight this unusual benign entity, we present a rare case of myoid hamartoma of the breast in a middle-aged female that presented as a large lump that mimicked a phyllodes tumour.

2. Case Report

A 30-year-old female presented with a 2-year history of a palpable lump in her left breast which progressively increased in size to a large mass measuring 17 x 12 x 5 cm. There was no history of breast disease in the patient's family. The leison was located in the upper inner quadrant of the left breast and was a clearly defined, hard, movable, smooth, and non-tender mass. On ultrasonography, a clearly defined solid mass with intralesional heterogeneous density was detected. No skin changes, nipple retraction or discharge was present. Axillary lymphadenopathy was

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absent. Clinical differentials considered were Phyllodes tumor and Giant fibroadenoma.

FNAC was done and the smears showed clusters of benign ductal epithelial cells. It was followed by lumpectomy. Cut surface of the lesion showed yellow-white, well-circumscribed non encapsulated mass with focal areas of myxoid changes (Figure 1). Histological examination revealed a well-encapsulated tumor mass showing glandular and stromal structures. The glands were lined by inner ductal and outer myoepithelial cells. Also seen were areas of microcystic and macrocystic alterations, apocrine metaplasia and adenosis.(Figure 2) The stroma was predominantly composed of bland spindle cells with eosinophilic cytoplasm.(Figure 3) These cells were resembling mature smooth muscle cells and were arranged in whorls and irregular interlacing bundles.(Figure 4) Immunohistochemistry was done for smooth-muscle actin (SMA) and desmin to characterize the stromal elements. It showed diffuse cytoplasmic positivity for SMA (Figure 5) and desmin (Figure 6) and was negative for cytokeratin and S-100 protein. Histology and IHC confirmed the diagnosis of Myoid Hamartoma. Post-operative complications were not observed during the Outpatient Department follow-up. In the course of 16-month follow-up period, no recurrence was noted.

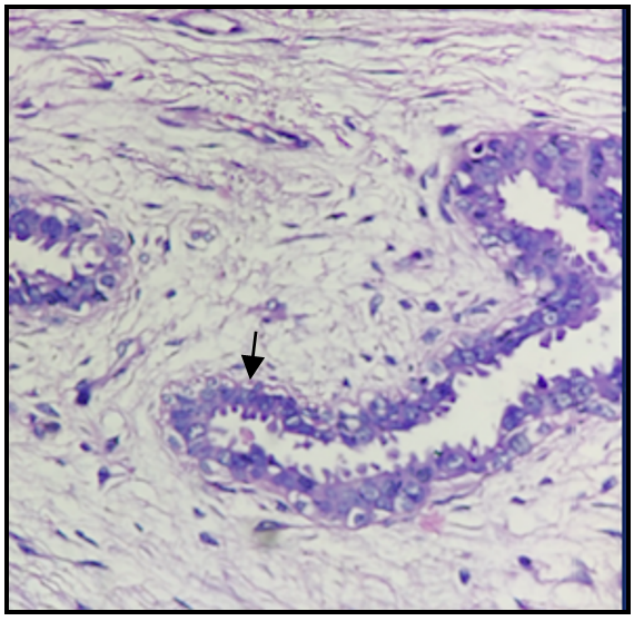

Fig. 2: Glandular elements showed apocrine metaplasia at places (arrow) [H & E 40X]

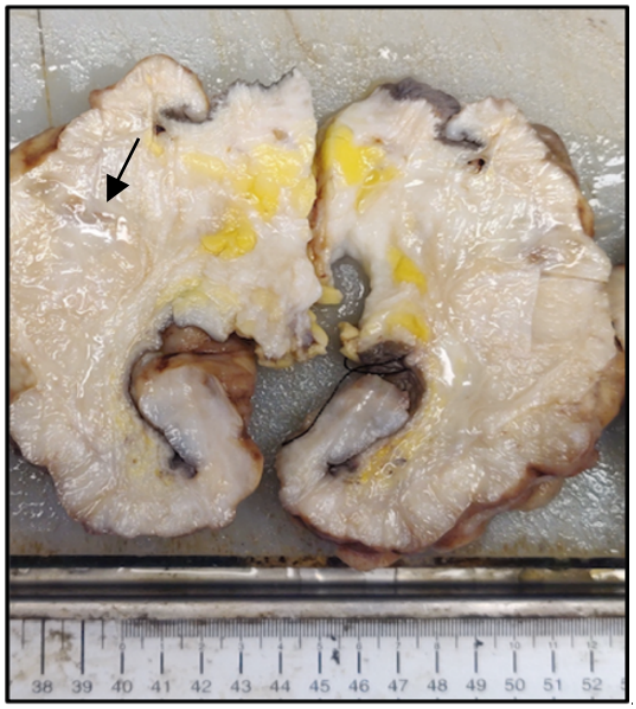

Fig. 1: Cut surface showed a yellow white non encapsulated mass with irregular margins. Focal myxoid areas also seen (arrow)

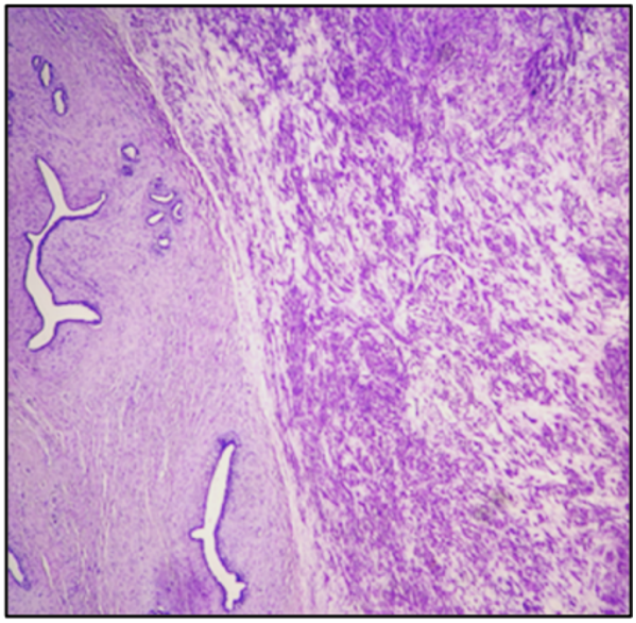

Fig. 3: Glandular and stromal elements seen (fasicular pattern) with a clear demarcation (H & E- 10X)

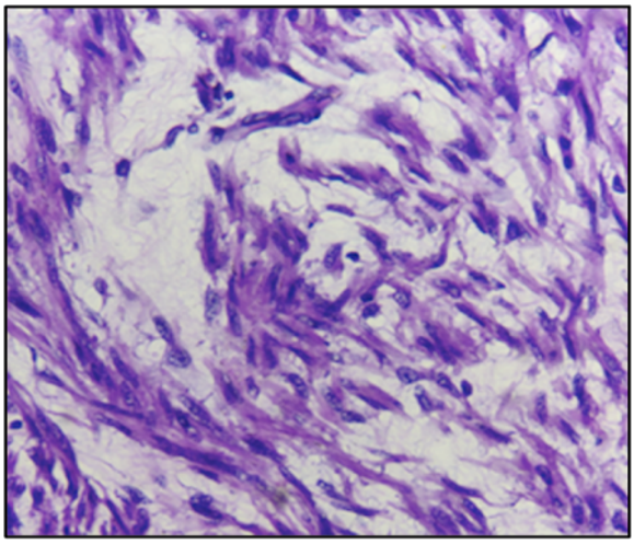

Fig. 4: Elongated spindle cells with wavy nuclei and moderate amount of eosinophillic cytoplasm (H & E-40X)

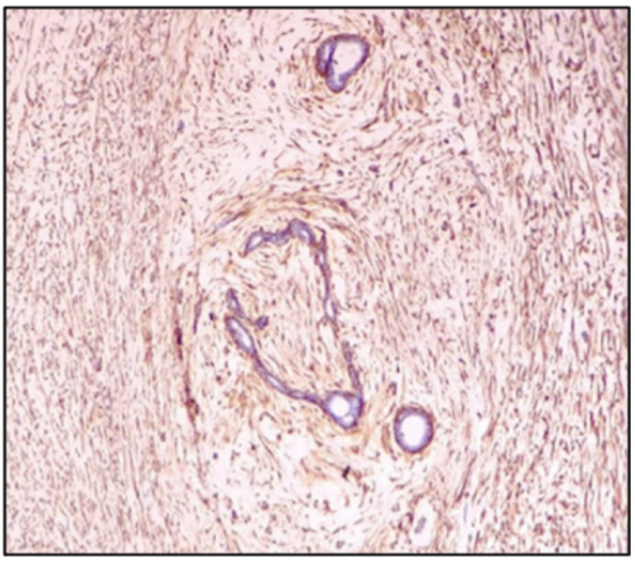

Fig. 5: IHC showing diffuse immunoreactivity for Smooth Muscle Actin (SMA) [H & E-40X]

3. Discussion

Breast lesions known as fibroadenolipoma or MH are relatively uncommon. They are benign, focal malformations that are characterized by the aberrant growth of mammary tissues, such as ducts, lobules, stroma, and adipose tissue, and they account for 0.7% to 5.0% of all benign breast neoplasms. Hamartomas' component proportions can vary, their arrangement may be haphazard, and/or their distinction may be poor. When hamartoma exhibits a significant smooth muscle component, it is known as myoid hamartoma, an uncommon type.[3] Davies and Riddell provided the initial description of it in 1973.[2]

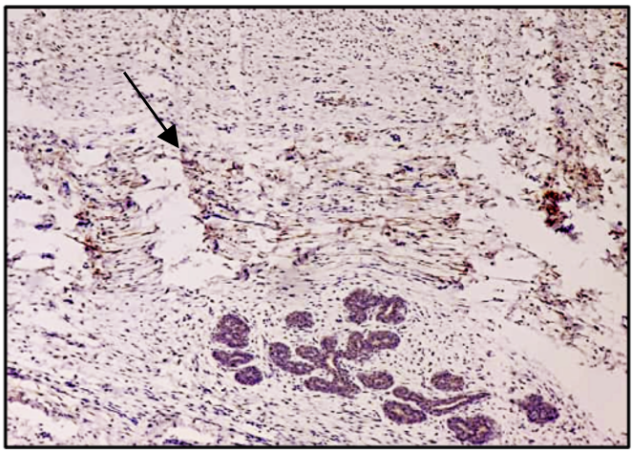

Fig. 6: IHC showing immunoreactivity for Desmin (arrow) [H &E -40X]

The pathogenesis involved in myoid component in myoid hamartoma remains controvertsial. The origin of the smooth muscle stroma has indeed been explained by a variety of different theories. The mammary tissues affected is considered that it possibly originates from the myoepithelial cells, stromal myofibroblasts, vascular smooth muscle cells, or stromal stem cells.[4–7]

MH masses are typically found in the upper outer quadrant of the breast, and are well-defined solid masses covered with partial or complete fibrous capsule. It's shape can be round, oval or lobulated, with gray-white and yellowish sections and textures ranging from soft to hard.[8] However, our case had an unusual presentation as the mass was massive with irregular margins mimicking a phyllodes tumor or giant fibroadenoma.

The mammographic findings of hamartoma are the presence a mass with well-defined borders with fat and soft tissue densities. A clearly defined solid mass with intralesional heterogeneous density that radiologically resembled fibroadenomas and phyllodes tumour was detected in our instance by radiography. Fibroadenomas are well-defined lesion with homogenous and hypoechoic areas. Whereas Phyllodes show a non-homogeneous echogenic structure with generally cystic areas.[9]

Histologically, MHs have different histological compositions based on the proportion of glandular, adipose, fibrous, and myomatous components. While in a minority of tumours, the myoid component mingle more diffusely with adipose and fibrous components as seen in our case, majority of the lesions have interlacing bundles of smooth muscle fibers forming focal leiomyoma.[10] The cytoplasms of the myoid cells are eosinophilic or lightly stained, and the nuclei are elongated and spindle-shaped with two blunt ends and fine chromatin. No mitosis or necrosis was noted. Core biopsy may aid in the diagnosis of myoid hamartoma.

Table 1: Histological differential diagnosis of myoid hamartoma

Leison	**Fibroadenoma**	**Spindle cell lesions of breast (leiomyomyoma, spindle cell tumor)**	**Myoid Hamartoma (Our case)**
Histological features	Intracanalicular or tubular glandular proliferation	Spindle cell proliferation and lack of breast ducts.	Normal ducts and lobules
Presence /absence of muscle component and adipose tissue.	Absence of both components.	Predominance of muscle components.	Presence of both components.
Immunohistochemistry			
Cytokeratin	Positive	Negative	Negative
SMA and Desmin	Negative	Positive	Strongly Positive
S100	Negative	Positive	Negative

However, several studies have recommended excisional biopsy to provide sufficient evidence for the pathological analysis in order to make a conclusive diagnosis of myoid hamartoma.[11]

Myoid hamartoma's clinical differential diagnosis includes phyllodes tumor, complex fibroadenoma, and various benign spindle cell tumors. The diagnosis of MH depends on the histomorphological and immunophenotypical examination of the masses.

Histologically, MH can be misdiagnosed as one of the following benign lesions as shown in Table 1.

From the clinical viewpoint, the management of an MH benign breast lesion is similar to that of fibroadenoma, and there are no particular concerns regarding recurrence. Thus, it does not require strict free margins or follow-up as in premalignant or cancerous lesions. However, as MHs are very rare breast lesions, it is of interest to carry out a detailed histopathological description and subsequent follow-up of these patients to achieve as much information as possible about the biological behavior of these breast tumors. In the case of patient concerns about tumor growth progression, pain, or breast asymmetry, a core biopsy is at least necessary to obtain the diagnosis and exclude possible aggressive lesions. Moreover, complete surgical removal is a viable solution.[5]

4. Conclusion

We conclude by describing a case of a large benign breast tumour termed as myoid hamartoma. Breast hamartomas are uncommon, benign tumours, despite their ability to grow to somewhat large sizes. Core needle biopsy, immunohistochemistry, and an adequate correlation of clinical and radiologic characteristics could all be used to establish a diagnosis. Since pathologists may categorize these lesions as fibroadenomas rather than hamartomas, breast hamartomas may go unreported. The actual incidence of MH may therefore be higher than what the literature suggests. More benign breast lumps may be correctly recognised as breast hamartomas with the use of immunohistochemistry & more widespread adoption of modern imaging modalities.[12]

In order to avoid this unusual phenomenon, this case report intends to emphasise its clinicopathological characteristics.

5. Source of Funding

None.

6. Conflict of Interest

None.

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