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Case Report

Benign phyllodes tumor with osseous metaplasia: A rare case report

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ABSTRACT

Introduction: Phyllodes tumors are rare fibroepithelial neoplasms of the breast, classified as benign, borderline, or malignant based on histological features. Osseous metaplasia, characterized by the presence of bone formation within the tumor, is an uncommon finding. This case report highlights the rarity of osseous metaplasia in a benign phyllodes tumor.
Case Presentation: A 23-year-old female presented with a gradually enlarging lump in the left breast. Mammography and cytological diagnosis were suggestive of giant fibroadenoma. Surgical excision was done and a histopathological diagnosis of benign phyllodes tumor with osseous metaplasia was made.
Conclusion: The occurrence of osseous metaplasia in a benign phyllodes tumor is exceptionally rare. Differentiating this phenomenon is essential for accurate diagnosis and appropriate management. Further research is needed to understand the underlying mechanisms and clinical implications of osseous metaplasia in phyllodes tumors.

1. Introduction

Phyllodes tumor is a rare fibroepithelial neoplasm that accounts for approximately 0.3% to 1% of all primary breast tumors. These tumors are classified as benign, borderline, or malignant based on the features of the stromal component, according to the World Health Organization (WHO) classification.[1] It is worth noting that over 20% of phyllodes tumor cases are concurrently diagnosed with benign fibroadenomas, and a history of fibroadenomas is observed in around 12.5% of cases.[2]

Osseous metaplasia, characterized by the formation of heterotopic bone, can occur in both benign and malignant breast neoplasms. However, benign phyllodes tumor with osseous metaplasia is an extremely rare occurrence, and only a few cases have been reported in the existing literature.[3] The presence of osseous metaplasia adds an intriguing element to the diagnostic challenge posed by phyllodes tumors. In this report, we present a unique case of benign phyllodes tumor with osseous metaplasia.

Differentiating benign phyllodes tumors with osseous metaplasia from their malignant counterparts is of paramount importance in determining the appropriate management and prognosis for patients. Through this case report, we aim to enhance the understanding of this rare entity and emphasize the significance of accurate diagnosis and management in such cases.

2. Case Report

2.1. Clinical History

A 23-year-old female presented with a 4-month history of a progressively enlarging lump in her left breast. No history of pain, nipple discharge and lump in the opposite breast. On examination, the left breast appeared larger than the right breast, with dilated veins and stretched skin. Palpation

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revealed a firm, freely movable mass measuring 10 x 8 cm involving all the breast quadrants and the retroareolar region. No lymphadenopathy was observed in bilateral axilla.

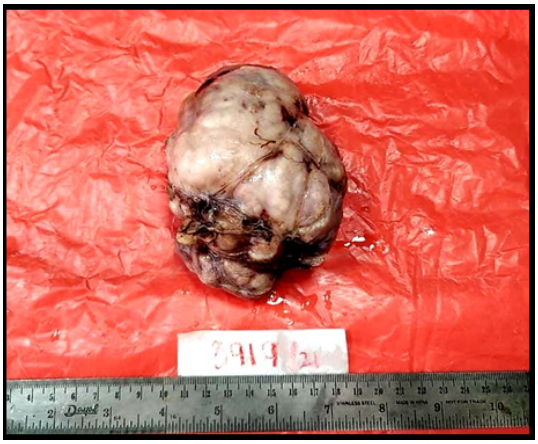

Fig. 1: Gross image showing firm and bosselated mass

Fig. 2: Cut surface of the mass

2.2. *Radiological findings*

A sonomammogram was performed, which revealed a well-defined lobulated hypoechoic lesion measuring 10.5 x 9.5 x 6.0 cm. The lesion showed posterior acoustic enhancement, internal vascularity, and a few linear cystic spaces. No calcifications were noted. Based on the imaging findings, a diagnosis of giant fibroadenoma was suggested.

2.3. *Cytological findings*

Ultrasound-guided fine-needle aspiration cytology (FNAC) of the left breast mass was performed. Microscopic examination of the cytology slides showed benign ductal epithelial cells arranged in staghorn tridimensional clusters, monolayered sheets, and a cribriform pattern. The myoepithelial cell layer appeared intact. The background

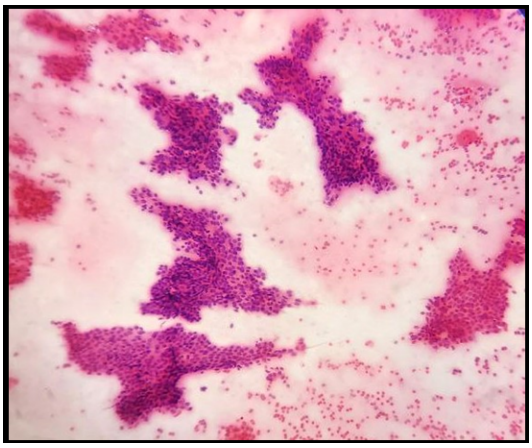

Fig. 3: Low power view of FNAC of left breast mass displaying benign ductal epithelial cells arranged in staghorn clusters and monolayered sheets (H and E 10 X magnification)

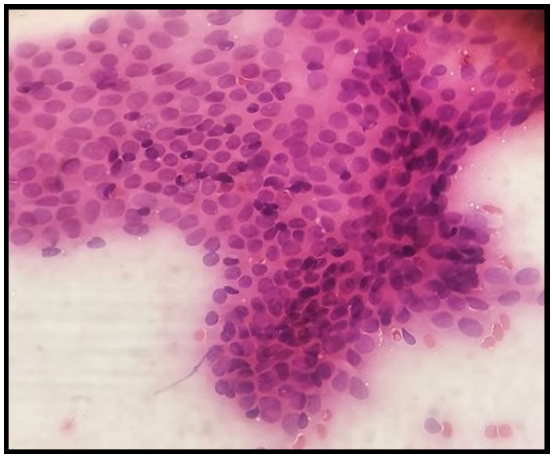

Fig. 4: High power view of FNAC showing intact myoepithelial cells (H and E 40 X magnification)

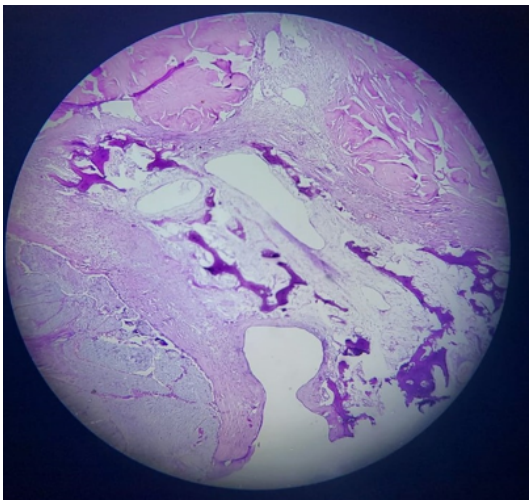

Fig. 5: Histopathological image of left breast mass showing osseous metaplasia component (H and E 10 x magnification)

revealed fibromyxoid stroma with plenty of bare benign nuclei admixed with hemorrhage. Based on these features, a cytological diagnosis of fibroadenoma was made. (Figures 3 and 4).

2.4. Histopathological Findings

Excisional biopsy of the left breast mass was performed, and the specimen was sent for histopathological examination. Gross examination revealed a pale white to pale brown globular mass measuring 13 x 9 x 5.3 cm. The outer surface was firm in consistency and bosselated (Figure 1). The cut surface revealed a well-circumscribed, pale white to tan white predominantly solid tumor with lobulations bulging above the surface. Areas of calcification were also noted (Figure 2).

Microscopic examination revealed a biphasic tumor with exaggerated intracanalicular morphology, producing a leaf-like epithelial pattern. The stroma displayed uniform spindle cells with elongated nuclei. Within the stroma, areas of osseous metaplasia were identified, with bony trabeculae rimmed by osteoblasts. Osteoclastic giant cells were also seen. Additional features included areas of myxoid change, calcification, and hyalinization (Figure 5). Resected margins (painted) were negative. Based on these findings, a diagnosis of benign phyllodes tumor with osseous metaplasia was made.

3. Discussion

Phyllodes tumors of the breast are a fascinating group of fibroepithelial neoplasms, accounting for 2-3% of all fibroepithelial neoplasms.[4]These tumors exhibit a wide spectrum of histological features and are classified into benign, borderline, and malignant categories based on a combination of various parameters, including stromal cellularity, stromal overgrowth, nuclear atypia, mitotic count, and tumor margins.[5]

Osseous metaplasia, the presence of bone tissue in neoplastic stroma, is an uncommon finding but has been reported in various organs, including the breast, thyroid, parathyroid, lung, gastrointestinal tract, and pancreas.[6] While osseous metaplasia is rare in benign tumors, it has been observed in cases of fibroadenoma, pleomorphic adenoma, phyllodes tumors and amyloid tumor of the breast.[3]

Norris and Taylor reported five cases of osseous metaplasia occurring in phyllodes tumors, highlighting its occurrence in this specific type of neoplasm.[7] Additionally, approximately 200 cases of osseous metaplasia have been reported, with a significant number arising in cases of phyllodes tumors, fibroadenomas, and sarcomas.[8]

Osseous metaplasia in phyllodes tumors presents as a rare phenomenon and can pose diagnostic challenges. Mammographic and ultrasound findings vary depending on the underlying cause. In benign neoplasms like fibroadenoma and benign phyllodes tumor, osseous metaplasia can manifest as a well-defined oval lesion with calcification.[9]On the other hand, in malignant neoplasms, it may appear as a circumscribed mass with calcification or a spiculated malignant appearance.[3,10]

Numerous theories have been proposed to explain the genesis of heterotopic bone and cartilage.The mechanism for the occurrence of heterotopic ossification in non-neoplastic processes would seem to be the metaplasia of stromal tissues in response to chronic inflammatory or degenerative alterations.[11]

Accurate diagnosis of osseous metaplasia in phyllodes tumors requires careful evaluation and histological examination. The differentiation between benign and malignant lesions is crucial for appropriate patient management. Surgical excision biopsy plays a significant role in reducing sampling errors and improving diagnostic accuracy. Further research is needed to understand the underlying mechanisms and clinical implications of osseous metaplasia in breast tumors, as well as its potential associations with other conditions.

4. Conclusion

Benign Phyllodes tumor with osseous metaplasia is an exceedingly rare neoplasm, particularly within the realm of benign breast tumors. The majority of cases exhibiting cartilaginous and osseous metaplastic changes are observed in malignant tumors. The presence of osseous metaplasia in phyllodes tumors can pose diagnostic challenges, as it may mimic malignancy in both imaging and histological evaluation. Accurate differentiation between benign phyllodes tumors with osseous metaplasia and their malignant counterparts is crucial to guide appropriate patient management and prevent overtreatment.

Recognition and characterization of benign phyllodes tumors with osseous metaplasia contribute to our understanding of the diverse spectrum of breast neoplasms. Further investigation and reporting of such cases are essential to expand our knowledge regarding the clinical behavior, optimal treatment strategies, and prognosis of this rare variant. A multidisciplinary approach involving radiologists, pathologists, and clinicians is vital in achieving accurate diagnoses and providing personalized patient care.

In conclusion, our case report highlights the rarity and diagnostic significance of benign phyllodes tumors with osseous metaplasia. It underscores the importance of distinguishing them from their malignant counterparts to avoid unnecessary interventions and ensure appropriate management. Continued research and the accumulation of evidence in this field will aid in refining diagnostic criteria, improving treatment outcomes, and enhancing patient care for individuals with this unique variant of phyllodes tumors.

5. Conflicts of interest

There are no conflicts of interest.

6. Source of Funding

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