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Adenomyoepithelioma of the breast: A Rare Case Report from B.P. Koirala Memorial Cancer Hospital

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Introduction

Adenomyoepithelioma is a rare, benign breast neoplasm characterized by the proliferation of both epithelial and myoepithelial cells within the breast lobules and ducts. Although it is most commonly seen in salivary glands and skin, Hamper reported the first case in the breast in the 1970 **(**1**)**. While this benign tumor is predominantly found in older females, rare cases have been identified in young women as well. Adenomyoepithelioma is notorious for local recurrence, and malignant metastasis has been reported **(**2**)**.

Case Presentation

A 22-year-old female presented to the outpatient department with the chief complaint of a painless lump in the upper outer quadrant of the right breast that had been present for the past 3 months. The patient had no history of pain, nipple discharge, skin changes, or an increase in the size of the mass. There was no family history of breast tumors, and the patient had no personal history of smoking, alcohol intake, or oral contraceptive pill **(**OCP**)** intake.

Upon examination, a 2 x 2 cm, firm, well defined, round, partially mobile lump with a smooth margin and free from underlying structures was palpated over the upper outer quadrant of the breast, with no skin changes noted.

Ultrasound revealed an ill-defined, heterogeneous lesion measuring 21 x 16 x 13mm in the fibroglandular tissue of the right breast at the 8 to 9 o'clock position, with minimal vasculature noted within the lesion. A fine-needle aspiration cytology **(**FNAC**)** was performed and showed features consistent with a benign proliferative disease of the breast, specifically a fibroadenoma.

The patient underwent wide local excision under intravenous anesthesia and was discharged on the same day with no postoperative complications. The histopathology report revealed adenomyoepithelioma, with immunohistochemistry markers showing P63 diffusely immunoreactive in the myoepithelial component, S-100 diffusely immunoreactive in the myoepithelial component, and HMWCK immunoreactive in the epithelial component. The proliferation index Ki-67 was 1-5%. ER was patchy immunoreactive in 10-15% of neoplastic cells.

The patient has been advised to undergo strict six-monthly follow-up with ultrasound of the breast and axilla to monitor for local recurrence and metastatic spread.

Discussion

Adenomyoepithelioma can be classified into three types: tubular, lobulated, and spindle cell. Tubular adenomyoepithelioma has an ill-defined margin, similar to tubular adenoma. Lobulated adenomyoepithelioma presents as nests of myoepithelial cells surrounding compressed epithelial-lined spaces. Spindle cell adenomyoepithelioma shows sparse epithelial-lined spaces that resemble leiomyoma-like cells. **(**3**)**

Diagnosis:

Adenomyoepitheliomas typically presents as a single, palpable, well-circumscribed, firm mass, with dimensions of up to 8 cm. Microscopic satellite lesions may be present in the periphery of the tumor at times, but it is mostly located in the central portion. Rarely, adenomyoepitheliomas may also present in phyllodes tumor or fibroadenoma. Pain and nipple discharge are rare symptoms associated with adenomyoepitheliomas. **(**15**)**

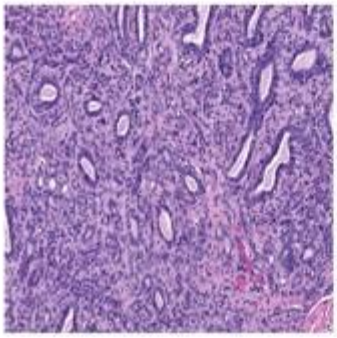
Radiology evaluation:

Radiology imaging is not specific and cannot distinguish between benign and malignant lesions **(**2**)**. Ultrasonography **(**USG**)** shows that adenomyoepitheliomas are typically solid or have a combined solid and cystic appearance. On mammography, they present as a lobulated dense mass with a partially indistinct margin, with or without calcifications. Malignant adenomyoepitheliomas show irregular shapes with spiculated margins on mammography. **(**14**)**

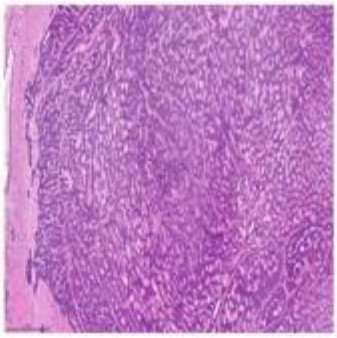
Pathology evaluation:

FNAC and core needle biopsy are also rarely diagnostic due to the heterogeneity of the tumor, with many cases falsely diagnosed as fibroadenoma in the literature. However, the presence of tightly aggregated glands arranged in compact nodules and prominent clear cell or spindle cell myoepithelium can be clues to the diagnosis. **(**4**) (**8**) (**13**)**

Micrscopic description of adenomyoepithelioma: **(**10**)**



Myoepithelial cells surrounding glands



Compressed tubules

Immuno Histo chemistry (IHC):

IHC is an important tool in the diagnosis of adenomyoepithelioma. The panel of markers, including p63, S-100, and HMWCK.P63 is a transcription factor that is expressed in the nuclei of myoepithelial cells and is a very sensitive and specific marker for identifying these cells. In adenomyoepithelioma, P63 staining is typically positive and shows a consistent rim of staining around the epithelial cells, which can help to distinguish it from other breast tumors. Ki-67 proliferation index is also useful in predicting the recurrence rate of the tumor. ER, PR, and HER-2 are usually negative in malignant adenomyoepithelioma. **(**6**) (**7**) (**9**) (**12**)**

Differential diagnosis:

- **Sclerosing adenosis:** Sclerosing adenosis is a condition where there is an increase in the number of epithelial glands along with hardening of the surrounding stroma, which can lead to an abnormal shape of the glands. It is usually not associated with the formation of a well-defined mass, and there is no significant presence of myoepithelial cells.

- **Intraductal papilloma:** Refers to a papillary growth consisting of an increase in epithelial cells along with fibrovascular cores, without a significant presence of myoepithelial cells.
- **Invasive carcinoma:** Malignant growth of epithelial cells with clear evidence of invasion. Absence of myoepithelial markers such as p63/p40, SMMHC, calponin, and CK5.
- **Nipple adenoma:** Growth of epithelial cells within the collecting ducts of the nipple. There is no significant presence of myoepithelial cells.
- **Tubular adenoma:** well-defined border and the proliferation of tubules with a single layer of myoepithelial cells surrounding the epithelial component without expansion of the myoepithelial component characterize the tubular adenoma. **(**16**)**

Treatment

No specific guidelines have been established for the treatment of adenomyoepithelioma. However, wide local excision with negative margins is recommended due to the local recurrence nature of the tumor. Some cases of malignant adenomyoepithelioma have also been reported in the literature, where mastectomy and sentinel lymph node biopsy are recommended. **(**10**) (**11**)**

Prognosis

Benign adenomyoepitheliomas generally have a good prognosis but require close monitoring and follow-up. In cases of local recurrence, radiotherapy has been used with positive results. However, malignant adenomyoepithelioma has a poor prognosis due to its low-grade invasiveness, high recurrence rate, and resistance to chemotherapy. Malignant tumors are more likely to metastasize via hematogenous spread to distant organs such as the brain, liver, and lungs rather than through the lymphatic system. **(**10**) (**11**)**

Conclusion

Adenomyoepitheliomas **(**AMEs**)** are uncommon benign breast tumors that should be considered in the differential diagnosis of solid breast lumps. While imaging features are not specific, suspicion of benign or malignant nature can be raised through radiologic examination. FNAB is often not diagnostic. Currently, there are no established guidelines for treating either benign or malignant AME. Surgical excision with negative margins is recommended for both types of AME due to the high recurrence rate

for benign tumors and the aggressiveness of malignant ones. Close monitoring and follow-up are necessary for benign cases, while malignant cases have a poor prognosis and are characterized by low grade, invasiveness, and high recurrence rate. Chemotherapy is not effective for treating malignant AME, and metastasis occurs through hematogenous spread to organs such as the brain, liver, and lungs.

Keywords: Adenomyoepithelioma; Asymptomatic; Benign mass; Case report; Surgical excision; Treatment; surveillance.

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