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Adenomyoepithelioma of the Breast: Diagnosis, Treatment and Surveillance at Tertiary Cancer Center

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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**)**.

Adenomayoepithelioma is very rare case in 10 years, out of 19000 case evaluation & diagnosis we have encountered only 2 cases, 1 was referred case & 1 diagnosed at B.P. Koirala Memorial Cancer Hospital Breast Oncosurgery Department.

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:

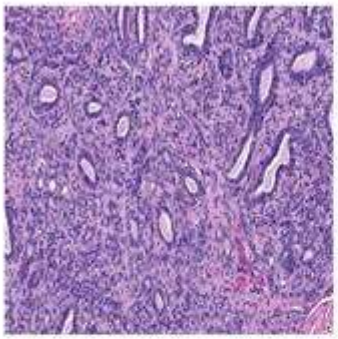
Radiology imaging is not specific and cannot distinguish between benign and malignant lesions **(**2**)**. Ultrasonography **(**USG**)** shows that adenomyoepitheliomas are typically solid or have a combined solid and cystic appearance. On mammography, they present as a lobulated dense mass with a partially

indistinct margin, with or without calcifications. Malignant adenomyoepitheliomas show irregular shapes with spiculated margins on mammography. **(**14**)**

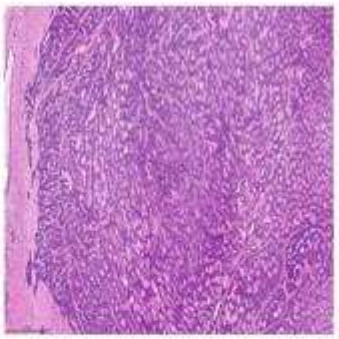
Pathology evaluation:

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**)**

Surveillance

Patient diagnosed with AMEs should be followed every 3 month for first 3 Year, then every 6 months for 2 years. After Year of surveillance and without any sigh of recurrence pt should have regular follow up early.

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.

References

1. Hamperl H. The myothelia **(**myoepithelial cells**)**. Normal state; regressive changes; hyperplasia; tumors. Curr Top Pathol. 1970;53:161-220. PMID: 4323195.

2. El-Helou E, Terro JJ, Kansoun A, Neaime GR, Mochairefa H, Ismail N, Naccour J, Zaarour M, Alam H. Breast adenomyoepithelioma, a case report. Int J Surg Case.

3. Intagliata E, Gangi S, Trovato C, Vecchio R, Strazzanti A. Benign adenomyoepitelioma of the breast: Presentation of two rare cases and review of literature. International Journal of Surgery Case

4. Reports. Pradhan S, Yadav RP, Adenomyoepithelioma of breast, case report, Journal of pathology of Nepal,2011

5. Krystel-Whittemore M, Wen HY. Adenomyoepithelioma. PathologyOutlines.com .

6. McLaren BK, Smith J, Schuyler PA, Dupont WD ,Page DL Adenomyoepithelioma: Clinical, Histologic, and Immunohistologic Evaluation of a Series of Related Lesions. Am J Surg Pathol 2005;29:1294-9.

7. Malcolm MH. Adenomyoepithelioma of the breast: a reviewstressing its propensity for malignant transformation J Clin Pathol2011;64:477-84

8. Tamura G, Monma N, Suzuki Y, et al. Adenomyoepithelioma**(**myoepithelioma**)** of the breast in a male. Hum Pathol 1993;24:678–81

9. Tavassoli F, Soares J. Myoepithelial lesions . In: Tavassoli FA,Stratton MR, editors. WHO Classification of Tumors: Pathology and Genetics, Tumors of the Breast and Female Genital Organs. Lyon:IARC, 2003.pp86-7

10. Rakha E, Tan PH, Ellis I, Quinn C. Adenomyoepithelioma of the breast: a proposal for classification. Histopathology. 2021 Oct;79**(**4**)**:465-479. doi: 10.1111/his.14380. Epub 2021 Jun 13. PMID: 33829532.

11. Parikh P, Jameel Z, Falcon S, Rosa M, Kiluk J, Hoover S, Soliman H, Ataya D. Adenomyoepithelioma of the breast: Case series and literature review. Clin Imaging. 2021 Jul;75:157-164. doi: 10.1016/j.clinimag.2021.02.002. Epub 2021 Feb 10. PMID: 33607537.

12. Kulka J, Madaras L, Floris G, Lax SF. Papillary lesions of the breast. Virchows Arch. 2022 Jan;480**(**1**)**:65-84. doi: 10.1007/s00428-021-03182-7. Epub 2021 Nov 3. PMID: 34734332; PMCID: PMC8983543.

13. Tay TKY, Tan PH. Papillary neoplasms of the breast-reviewing the spectrum. Mod Pathol. 2021 Jun;34**(**6**)**:1044-1061. doi: 10.1038/s41379-020-00732-3. Epub 2021 Jan 18. PMID: 33462367.

14. AlQurashi M, Abdel Hadi M, Binammar AA, Al Muhanna A, Kussaibi H, Al Shammary E. Adenomyoepithelioma of the Breast: A Report of 3 Cases. Am J Case Rep. 2022 Apr 11;23:e936070. doi: 10.12659/AJCR.936070. PMID: 35400730; PMCID: PMC9011969.

15. Xu J, Tang X, Iida Y, Fuchinoue F, Kusumi T, Yagihashi N, Kawachi K, Shimizu S, Masuda S. Adenomyoepithelioma with carcinoma of the breast: A report of two cases and a review of the literature. Pathol Res Pract. 2016 Feb;212**(**2**)**:130-4. doi: 10.1016/j.prp.2015.09.008. Epub 2015 Sep 8. PMID: 26596263.

16. Hayes, M. M. **(**2011**)**. Adenomyoepithelioma of the breast: a review stressing its propensity for malignant transformation. Journal of Clinical Pathology, 64**(**6**)**, 477–484. doi:10.1136/jcp.2010.087718.