



## Case Report

# Adenomyoepithelioma of the Breast and its Management: Case Presentation and Literature Review

V. Bassi<sup>1\*</sup>, M. Chevènement<sup>1</sup>, M. Gremaud<sup>2</sup>, A. Léger<sup>1</sup>

<sup>1</sup>Centre du sein, Réseau hospitalier Neuchâtelois, Switzerland

<sup>2</sup>ADMED Pathologie, Switzerland

\*Corresponding author: V. Bassi, Centre du sein, Réseau hospitalier Neuchâtelois, Rue de Maladière 45, Neuchâtel, Switzerland

Citation: Bassi V, Chevènement M, Gremaud M, Léger A (2023) Adenomyoepithelioma of the Breast and its Management: Case Presentation and Literature Review. J Community Med Public Health 7: 322. DOI: <https://doi.org/10.29011/2577-2228.100322>

Received Date: 25 May, 2023; Accepted Date: 01 June, 2023; Published Date: 05 June, 2023

### Abstract

Diagnosis of adenomyoepithelioma of the breast (AME) is challenging, with only four comprehensive studies and occasional cases (<200 cases) collected in the literature and with controversial clinical, radiological, and pathological features. Biopsy findings may be misleading because of tumor heterogeneity with proliferation of differentiated myoepithelial and epithelial cells. Normally, adenomyoepithelioma appears to have a benign course; however, aggressive proliferation with the possibility of metastases should be considered. We report a case of breast adenomyoepithelioma in a 50-year-old woman in good health and discuss its histological and anatomico-clinical features and its management.

There are no guidelines for the treatment of AME; therefore, surgical excision aimed at oncological radicality is the current therapeutic orientation owing to the high recurrence rate for benign histological type and aggressiveness for the malignant one.

**Keywords:** Adenomyoepithelioma; Pathology; Immunohistochemistry; Breast Surgery; Wide local excision

**Abbreviations:** AME: Adenomyoepithelioma; CK: Cytokeratin

### Introduction

Adenomyoepithelioma (AME) of the breast is a rare tumor that is characterized by dual differentiation into luminal and myoepithelial cells. These lesions can be diagnostically challenging, especially when core needle biopsy is performed, because of the heterogeneity of AMEs. It has been reported to occur between the 3<sup>rd</sup> and 9<sup>th</sup> decades but more commonly in the 5<sup>th</sup> and 6<sup>th</sup> decade. Malignancy has been reported in up to 40 cases, and imaging studies are not conclusive in providing a definite diagnosis; local excision is the best treatment and histopathology is the gold standard for diagnosis [1].

### Case Presentation

A 50-year-old woman (gravida 1, para 1) in good health was

admitted to our center after a screening mammogram. Her medical history revealed menarche at 13 years, regular 23-day cycles, one vaginal childbirth with a breastfeeding period of 1 year, no drugs or tobacco consumption. Family history was negative for breast cancer. The screening mammogram showed irregular opacities in the upper inner quadrant of the right breast. The patient then underwent mammography and breast ultrasonography. The mammogram showed parenchyma distributed slightly irregularly with small, well-delineated nodular areas of infra-centimeter size in the lower inner quadrant, upper outer quadrant, and deep central region, compatible with the area of the lesion detected during screening. No evidence of axillary lymph node metastasis was found.

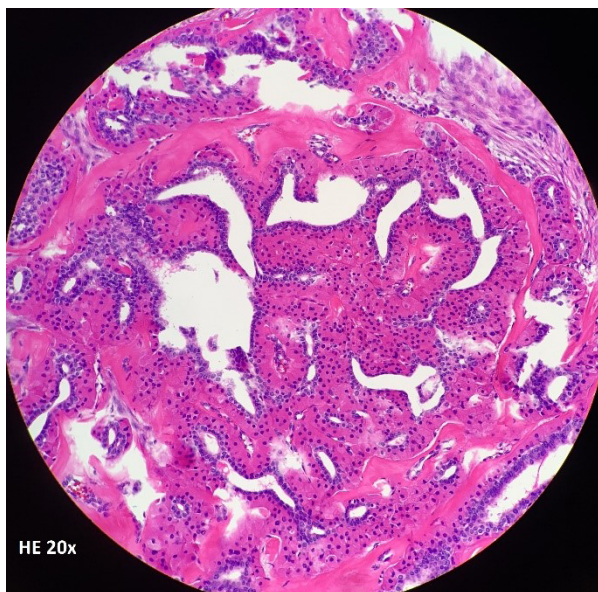
Ultrasound images showed multiple small, anechogenic, well-delineated oval lesions between the upper outer quadrant and lower inner quadrant of the right breast and circumscribed round, hypoechogenic, non-vascularized lesions measuring 5 mm with posterior acoustic shadowing of the upper inner quadrant. No evidence of axillary lymph node metastasis was found.

A breast core biopsy was performed, which showed an AME in the upper inner quadrant that was partially sclerotic. Clinical breast examination showed no palpable lesions or lymph nodes.

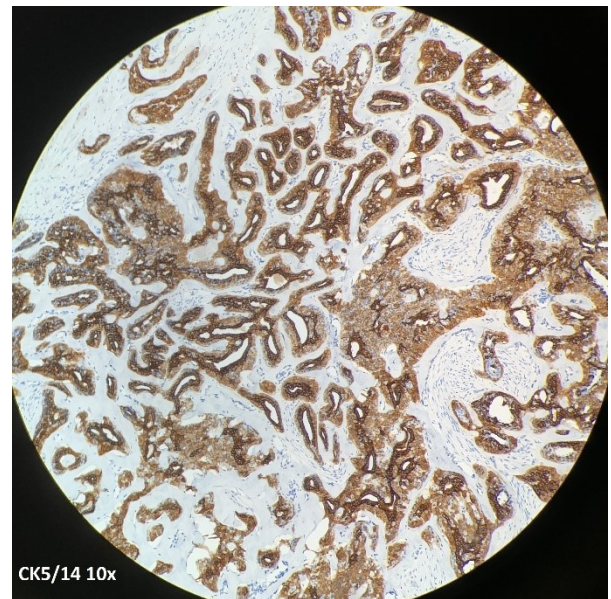
Tumorectomy with harpoon tracking was performed, and the patient recovered uneventfully. Negative surgical margins were achieved and the final pathology revealed a sclerotic AME and ductal hyperplasia without atypia or adenosis.

Microscopically, a well-defined biphasic tumor composed of irregular glandular structures with luminal epithelial cells surrounded by abundant myoepithelial cells and eosinophilic amorphous material was observed. The epithelial component was composed of a single layer of cubocylindrical cells without atypia. The predominant myoepithelial component was composed of cells with round nuclei and clarified or eosinophilic cytoplasm without atypia. In the periphery of the tumor, foci of usual hyperplasia without atypia or adenosis were observed.

Immunohistochemistry (estrogen receptor, cytokeratin CK5/14) confirmed the diagnosis (Figures 1 and 2) with heterogeneous positivity of estrogen and progesterone receptors, HER2-negative and focal accumulation of MIB-1-positive cells. During a consultation 2 weeks and 6 months after discharge, patient reported good health with no remaining deficits.



**Figure 1:** Tumorectomy.



**Figure 2:** CK5/6 and CK5/14 are immunohistochemical stains marking myoepithelial cells. In addition, in this case, light brown myoepithelial cells and dark brown luminal cells are marked by CK5/14.

## Discussion

AME of the breast is a relatively rare, benign tumor with a range of disordered epithelial-myoepithelial proliferations. Due to the morphological heterogeneity of this tumor, misinterpretation of the finding during needle biopsy may occur [2,3,4]. Most AME are benign, and malignant transformations have been reported in the literature [5-7]. Tavassoli reported the malignant potential of this tumor type in 1991 and divided it into benign and malignant lesions. In the latter, one or both components can have malignant features [8]. Due to the biphasic nature of the tumor, carcinomas may arise from ductal epithelial cells, myoepithelial cells, or both [9,10].

Fewer than 80 cases of malignant AME of the breast have been reported in approximately 40 papers thus far [11]. More than 50% of malignant AMEs invade or infiltrate the periphery, and mitotic figures are prevalent, occurring in up to 10 of 62 high power fields. Approximately twenty-three cases exhibited metastases in the lymph nodes, lung, liver, brain, bone, thyroid, kidney, and thoracic wall. There were no identifiable histopathologic features distinguishing the metastatic and primary tumors, and malignant manifestations such as pleomorphism and necrosis were observed in more than fifty of these cases [12].

It is important for breast surgeons to predict malignant potential and plan the operation. Tumor size (>16mm), mitotic figures, necrosis are characteristics that may be related to potential malignancy. However, the imaging techniques are not conclusive. Mammographic features and magnetic resonance imaging findings are non-specific and rarely show micro calcifications. Ultrasound features show a solid, oval, hypoechoic mass with irregular borders.

In our case, the screening mammography revealed irregular opacities that were identified on ultrasound, presenting as a round, well demarcated, hypoechoic lesion, not vascularized, with a rear shadow cone.

Benign AME can be treated with wide local excision, as it rarely recurs locally. In contrast, malignant tumors are more likely to recur locally and have a 30-40% chance of metastases, commonly through a hematogenous route to the lungs, brain, thyroid, and chest wall. However, metastases to axillary lymph nodes are rare [12].

## Conclusion

AME of the breast was first described by Hamperl in 1970 and the second case was reported several years later. It should be considered in the differential diagnosis of other solid breast lesions, such as sclerosing adenosis, adenoma, adenoid cystic carcinoma, fibroadenoma, intraductal papilloma with myoepithelial hyperplasia, low-grade adenosquamous carcinoma, metaplastic carcinoma, tubular carcinoma malignant myoepithelioma, and papillary carcinoma.

Treatment of AME remains controversial owing to the absence of prospective studies and guidelines; therefore, surgical excision aimed at oncological radicality is the current therapeutic orientation owing to the high recurrence rate for benign histological type and aggressiveness for the malignant one.

## Conflicts of Interest

The authors declare no conflicts of interest.

## Acknowledgements

We would like to thank the doctors and staff at our institute for their continuous support and guidance.

## Authors' Contributions

AL and VB are the surgeons who operated on the patient. VB and MC drafted the manuscript. AL supervised the preparation of this case report. MG performed the histopathological analysis.

All authors have read and approved the final manuscript.

## Funding

This research did not receive any specific grants from funding agencies in the public, commercial, or not-for-profit sectors.

## References

1. Adejolu M, Wu Y, Santiago L, Yang WT (2011) Adenomyoepithelial tumors of the breast: imaging findings with histopathologic correlation. *AJR Am J Roentgenol* 197: W184-W190.
2. Hoda SA, Rosen PP (2004) Observations on the pathologic diagnosis of selected unusual lesions in needle core biopsies of breast. *Breast J* 10: 522-527.
3. McLaren BK, Smith J, Schuyler PA, Dupont WD, Page DL (2005) Adenomyoepithelioma: clinical, histologic, and immunohistologic evaluation of a series of related lesions. *Am J Surg Pathol* 29: 1294-1299.
4. Ahmed AA, Heller DS (2000) Malignant adenomyoepithelioma of the breast with malignant proliferation of epithelial and myoepithelial elements: a case report and review of the literature. *Arch Pathol Lab Med* 124: 632-636.
5. Dwarakanath S, Lee AK, Delellis RA, Silverman ML, Frasca L, et al. (1987) S-100 protein positivity in breast carcinomas: a potential pitfall in diagnostic immunohistochemistry. *Hum Pathol* 18: 1144-1148.
6. Saad RS, Richmond L, Nofech-Mozes S, Ghorab Z (2012) Fine-needle aspiration biopsy of breast adenomyoepithelioma: a potential false positive pitfall and presence of intranuclear cytoplasmic inclusions. *Diagn Cytopathol* 40: 1005-1009.
7. Tavassoli FA (1991) Myoepithelial lesions of the breast. Myoepitheliosis, adenomyoepithelioma, and myoepithelial carcinoma. *Am J Surg Pathol* 15: 554-568.
8. Han JS, Peng Y (2010) Multicentric adenomyoepithelioma of the breast with atypia and associated ductal carcinoma *in situ*. *Breast J* 16: 547-549.
9. Han B, Mori I, Nakamura M, Wang X, Ozaki T, et al. (2006) Myoepithelial carcinoma arising in an adenomyoepithelioma of the breast: case report with immunohistochemical and mutational analysis. *Pathol Int* 56: 211-216.
10. Hayes MM (2011) Adenomyoepithelioma of the breast: a review stressing its propensity for malignant transformation. *J Clin Pathol* 64: 477-484.
11. Zhai DY, Zhen TT, Zhang XL, Luo J, Shi HJ, et al. (2021) Malignant adenomyoepithelioma of the breast: Two case reports and review of the literature. *World J Clin Cases* 9: 9549-9556.
12. Lari EA, Lari AA, Alsaeed T (2020) Malignant adenomyoepithelioma of the breast: A case report. *Int J Surg Case Rep* 72: 56-58.