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# BREAST ADENOMYEPITHELIOMA: A CASE REPORT AND LITERATURE REVIEW

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#### ABSTRACT

We report a case of adenomyepithelioma (AME), a rare tumor of the breast due to a proliferation of differentiated myoepithelial and epithelial cells in a 68-year-old patient. This lesion, controversial on the radio-clinical and especially on the anatomical-pathological level, evolves in most cases in a benign mode with an aggressive potential characterized by recurrences sometimes multiple recurrences. A malignant evolution with metastases is exceptional; explaining why some authors describe adenomyoepithelioma as a member of the group of myoepithelial tumors while others consider it a rare breast carcinoma.

**KEYWORDS:** Adenomyoepithelioma, breast, epithelial tumor, mammography, surgery.

### INTRODUCTION

Adenomyepithelioma (AME) is a relatively rare tumor of the breast characterized by a proliferation combining both myoepithelial and differentiated epithelial cells. It is included in the group of myoepithelial tumors consisting of myoepitheliosis and myoepithelial carcinoma.<sup>[1]</sup> The combination of myoepithelial and glandular (epithelial) cells is found in exocrine glands such as lacrimal, salivary, skin, mammary and Bartholin glands. This tumor is more frequent in the salivary gland than in the mammary gland where Hamperl described it only in 1970.<sup>[1-4]</sup>

Based on a clinical case, we have performed a review of the literature.

The nosology of this lesion is controversial on the radioclinical and especially on the anatomopathological level. In most cases, it evolves in a benign mode with an aggressive potential characterized by multiple recurrences; exceptionally, it evolves in a malignant mode with metastases, which explains its classification in the group of myoepithelial tumors for some authors, in the group of rare breast carcinomas for others.<sup>[5]</sup>

#### **OBSERVATION**

This is a 68-year-old patient, G6P6, postmenopausal with no hormone therapy or personal or family history of breast disease. She had consulted our department for a painless node of the left breast, discovered by chance by self-palpation since one week. The clinical examination revealed a painless node, movable in relation to the superficial plane and adherent to the deep plane with skin retraction, measuring 3 cm in diameter and located in the left superior-external quadrant. The lymph nodes were free.

Mammography showed the presence of a mass with spiculated contours, of high tone, located at the level of the SEQ of the left breast, containing suspicious microcalcifications. This mass is located 60 mm behind the nipple. The ultrasound performed in addition, confirmed the presence of a nodular formation, with irregular contours, attenuating, located in the SEQ measuring 20 mm in major axis. The lesion was classified as BIRADs 5.

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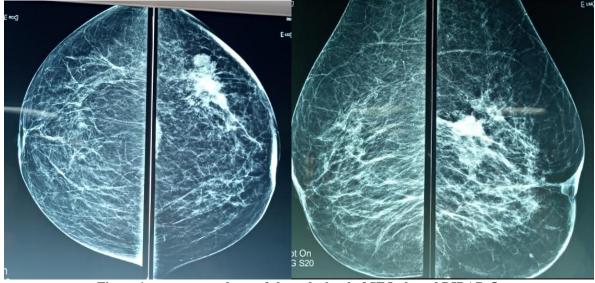


Figure 1: mammography: nodule at the level of SEQ classed BIRADs5.

A biopsy was performed and anotomopathology suggested the possibility of an adenomyoepithelioma.

After the tumorectomy of the palpable node of the left SEQ, the anatomopathological study of the surgical piece confirmed a spindle cell adenomyoepithelioma without cyto-nuclear atypia.

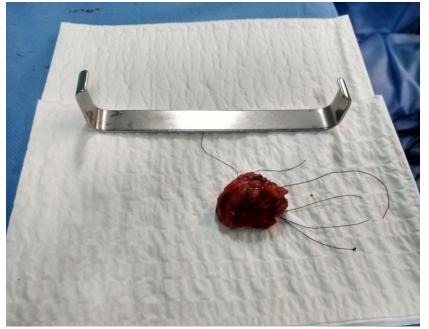


Figure 2: macroscopic aspect of the tumor.

The postoperative course was simple.

## DISCUSSION

In terms of nosology, AME was described in 1970 by Hamperl-Bässer. After the publication of series of 13, 18 and 27 cases<sup>[1-4]</sup>, which allowed the clinical and histopathological characteristics of this lesion to be defined; most of the current publications concern small samples. Less than 80 cases were described in 1995 in the literature.<sup>[4]</sup> It is a complex tumor with several histological variants. Its cellular biological profile is also

variable with a large proportion of benign tumors, which may recur in the long term and give rise to metastases.<sup>[4,5]</sup>

The average age of onset described in the literature<sup>[7]</sup> is 56 years, close to that of our patient.

No risk factors or personal history has been reported for this tumor, except a family history of breast cancer found in the series of Van Haverbeke et al.<sup>[2, 4, 7]</sup> The classic clinical presentation described by Van Haverbeke et al<sup>[2,4,7]</sup> is the same as that described in our patient (incidental discovery of a mass with benign characteristics). The mammogram shows a wellcircumscribed round, oval or macrolobulated opacity of central or peripheral location, eggshell calcifications or microcalcifications regular punctate or regular punctiform microcalcifications. On ultrasound examination, the lesion is hypoechoic tissue, nonabsorbing, with well circumscribed or polycyclic contours, with a large transverse axis of mixed type, consisting of tissue and microcystic or macrocystic structures with sometimes small vegetations.

The diagnosis of certainty is only provided by histology. On the anatomopathological level macroscopically, it is a mostly solid tumor, well limited, of firm or indurated consistency, of variable size, sometimes composed of small confluent nodes (lobulated), grayish-beige or pinkish in color. Some foci of hemorrhage, necrosis, small cysts or macrocysts cysts or macrocysts with vegetations are sometimes visible.

Therapeutically, adenomyepithelioma of the breast is mostly a benign tumor, it is recommended to remove it in the normal zone.

Due to the risk of local recurrence after 2 years. This recurrence would be attributed either to an excision incomplete, either in a multifocal form or probably to intense mitotic activity associated with nuclear atypias and proliferation large number of epithelial cells that discuss malignant adenomyepithelioma or myoepithelial carcinoma when the component myoepithelial is important. Mastectomy followed lymph node dissection formerly performed in some cases are currently of very limited indication.

Regarding the course and prognosis, several authors have published numerous cases of recurrences. These recurrences often only occur in the tubular type: Young et al. report a case that had recurred 3 times at 36, 54 and 84 months.

## CONCLUSION

Adenomyepitheliomas are rare tumors. They occur during the perimenopausal period. The clinic is that of a benign mass. Mammography and ultrasound allow a classification in ACR 3 or 4 and allow biopsy. Surgical excision in the normal zone is the recommended treatment. The diagnosis of certainty is histological showing a myoepithelial and epithelial component. Evolution of these tumors is sometimes marked by recurrence, possibly multiple requiring regular clinical and radiological monitoring.

Very rare metastases have been described involving the nosological problems for this tumor.

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