



ADENOMYOEPITHELIOMA OF BREAST: A CASE REPORT

Pathology

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ABSTRACT

Adenomyoepithelioma is a rare benign breast neoplasm characterized by the proliferation of both epithelial and myoepithelial cells belonging to the breast lobules and ducts. Adenomyoepithelial tumors of the breast shows a spectrum of biological behavior. Radiologic findings are not specific and do not allow to distinguish the benign or malignant nature of the lesion Although benign, it has a potential for local recurrence; therefore, proper excision with margins free from disease is mandatory for diagnosis and treatment. Here we report a case of adenomyoepithelioma of breast in a young female patient, initially reported as lobular carcinoma in situ in a background of tubular adenoma.

KEYWORDS

Adenomyoepithelioma and biphasic breast tumor

INTRODUCTION

Adenomyoepithelioma is a rare benign breast neoplasm characterized by the proliferation of both epithelial and myoepithelial cells belonging to the breast lobules and ducts. Adenomyoepithelial tumors may be found in salivary glands and skin appendages, but very rarely presents in the mammary gland⁽¹⁾.

In the few cases reported previously, these tumors clinically show up as a single asymptomatic hard roundish shape irregular nodule and the differential diagnosis with breast cancer is challenging^(2,3,4,5,6,7).

Although benign, it has a potential for local recurrence; therefore, proper excision with margins free from disease is mandatory for diagnosis and treatment. Malignant transformation of adenomyoepithelioma is possible, and in this case, the tumor must be treated as a breast carcinoma^(8,9,10).

Here, we report a case of Adenomyoepithelioma of breast in a young female patient.

Case Report

A 20-year-old female noticed a lump in her right breast on self-examination, 3 weeks after which she reported to a local physician. The breast lump was not associated with any pain. She has a regular menstrual history. There was no significant past medical, surgical or family history. The physician referred her to the local hospital where she was examined and advised preliminary routine blood investigations and ultrasonography of the right breast lump.

The routine blood investigations were within normal limit. The ultrasonography showed a well-defined, oval shaped, heterogeneously hypoechoic, wider than taller SOL with smooth margin measuring 4.6 x 2.4 cm showing minimum vascularity on colour doppler study and having cystic and necrotic areas in subareolar region of right breast. The impression was given as: features suggestive BIRADS III lesion (? Fibroadenoma) in subareolar region of right breast with an advice of histopathological evaluation. The lump was excised thereafter and tissue was sent for HPE.

The histopathological report suggested to be lobular carcinoma-in-situ in a background of tubular adenoma. After receiving the report the patient was referred to State Cancer Institute, Gauhati Medical College and Hospital, Assam. She was advised a slide review in our department of Onco-Pathology before proceeding for further management of the patient.

On reviewing and re-sectioning of the received tissue blocks, we saw the tumor arranged in lobulated, tubular, trabecular and papillaroid

pattern. The tubules are lined by outer layer of polygonal to spindle shaped cells with clear cytoplasm and bland nuclei. The inner lumen is lined by cuboidal cells with moderate eosinophilic cytoplasm and bland central nuclei. The lobular areas are outlined by basement membrane material. Few of the cells show atypical features displaying high nucleocytoplasmic ratio with hyperchromatic nuclei. Therefore, the case was reported as myoepithelial neoplasm with a differential diagnosis of Adenomyoepithelioma right breast lump.

Immunohistochemistry for calponin, P63, CK-7, CK-5/6 was advised which stained positive complementing the histopathological diagnosis.

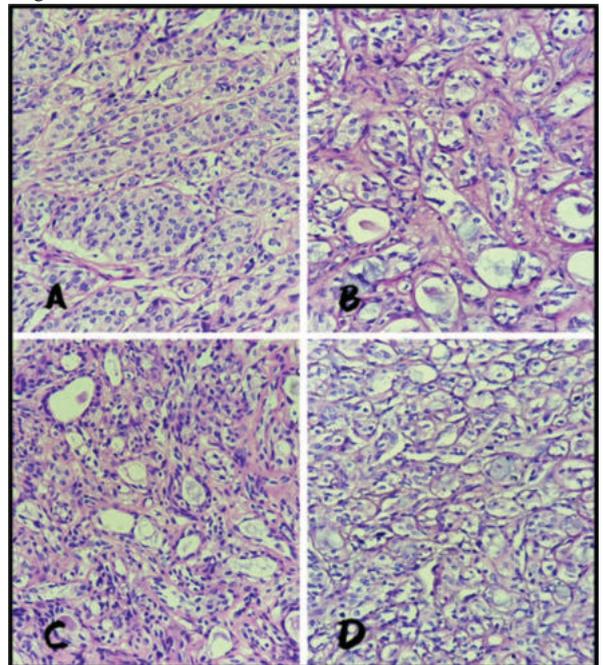


Figure 1- A- lobulated and trabecular pattern of cell arrangement (40x10 x, H & E stain), B- Prominent hyaline basement membrane material surrounding the tubules (40x10 x, H & E stain), C- Dual population of myoepithelial cells surrounding epithelial cells (40x10 x, H & E stain), D- Polygonal shaped myoepithelial cells surrounding the epithelial cells (40x10 x, H& E stain)

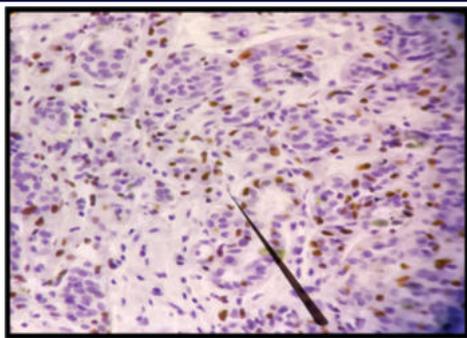


Figure 2- Myoepithelial cells showing calponin positivity on IHC

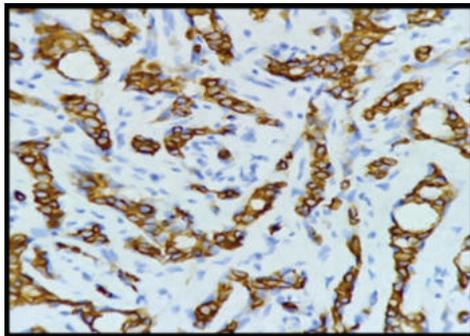


Figure 3- Epithelial cells showing CK-7 positivity on IHC

DISCUSSION

Breast adenomyoepithelioma was first reported by Hamperl in 1970⁽¹¹⁾. Adenomyoepithelial tumors of the breast are rare neoplasms; approximately 150 cases have been described in the Literature⁽¹²⁾.

Adenomyoepithelial tumors of the breast shows a spectrum of biological behavior⁽¹³⁾. Adenomyoepithelial tumors are characterized by proliferation of both epithelial and myoepithelial cells, which belong to the breast lobules and ducts⁽¹⁴⁾.

Benign adenomyoepithelioma has been classified as tubular, lobulated or spindle cell variants⁽¹²⁾.

Malignant transformation may occur in tumors with high mitotic rate, atypia, necrosis, cellular pleiomorphism and infiltrative borders. Malignant changes may involve most often the epithelial cells than the myoepithelial ones, and rarely both⁽¹⁵⁾. Malignant adenomyoepithelial tumors can be associated to mostly hematogenous rather than lymphatic metastasis⁽¹⁴⁾ to lung⁽¹⁶⁾, brain, jaws, liver and thyroid⁽¹⁷⁾.

Due to rarity of adenomyoepithelial tumor of the breast available imaging reports describe small patient cohorts and single patient studies only. Radiologic findings are not specific and do not allow to distinguish the benign or malignant nature of the lesion⁽¹⁸⁾. MRI might provide additional information about morphology and hemodynamic features⁽¹⁹⁾.

Fine needle aspiration are never diagnostic, as reported earlier^(3,5,14). It was not done in our case either. These lesions can be diagnostically challenging in core needle biopsy as well, because of its heterogeneity⁽⁹⁾.

On Histopathology, breast adenomyoepithelioma is a well circumscribed encapsulated or lobulated mass with proliferation of both epithelial and myoepithelial cells. Epithelial cells usually form glandular spaces. Myoepithelial cells are usually prevalent and may be polygonal shaped with clear cytoplasm or spindle shaped. Tubular structures can be occasionally noted. Malignant cases have infiltrative growth pattern, high mitotic rate or severe atypia which can be seen in epithelial or myoepithelial components. Malignant epithelial component can have features of invasive carcinoma no special type, invasive lobular carcinoma, metaplastic carcinoma. Malignant myoepithelial component shows features of myoepithelial carcinoma including overgrowth of myoepithelial cells, nuclear atypia and mitotic activity⁽²⁰⁾. Immunohistochemistry defines the epithelial component when markers cytokeratin AE1/AE3, CK7, CK8, EMA and CEA are found; the myoepithelial component is defined by markers calponin, S100, SMA, SMMCH, p63, CK5/14, smooth muscle myosin heavy chain^(4,5,21). There could be positivity for estrogen and/or

progesterone receptors⁽²²⁾.

There are neither guidelines for the treatment of benign or malignant adenomyoepithelioma nor mature clinical reference in the present scenario, since it is rare⁽²³⁾.

Benign adenomyoepithelioma has a good prognosis and radiotherapy has been used in few cases with local recurrence⁽²⁴⁾.

Prognosis is poor in malignant cases since, the disease is characterized by invasiveness, high recurrence rate and metastasis⁽⁷⁾, and chemotherapy has no significant success^(24,25).

CONCLUSION

The AME of the breast is a relatively rare, benign tumor that has a spectrum of disordered epithelial-myoepithelial proliferations. Due to the morphologic heterogeneity of this tumor, misinterpretation on a needle biopsy may occur^(26,27). Although most tumors are benign, malignant transformations have been reported. Cases with significant cytologic atypia, necrosis, and brisk mitotic rates raise the suspicion for malignancy^(28,29). Therefore, atypical features should be noted in the pathology report, and complete excision with adequate margins is recommended to decrease the potential for recurrence and metastasis.

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