Original article

Malignant adenomyoepithelioma (mame) of breast: Review of literature

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Abstract:

Breast tumours are common but malignant adenomyoepithelioma (MAME) of breast is a rare entity. Here, we are presenting a case of 60 years postmenopausal lady complaining of swelling in the left breast since 2 years and blood stained nipple discharge since 4 days. From the results of histological and immunohistochemical examinations, this case was considered to be malignant adenomyoepithelioma. This case is being highlighted for its rarity and distinct morphological spectrum.

INTRODUCTION:

The ductal system of the breast is lined by epithelial cells surrounded by contractile myoepithelial cells. These myoepithelial cells help to propel milk from lobules towards the nipple. Breast tumours originating from myoepithelial cells are divided into 3 major categories: myoepitheliosis, adenomyoepithelioma, and myoepithelial carcinoma.[1] Adenomyoepithelioma of breast are uncommon tumours characterized by biphasic proliferation of epithelial and myoepithelial cells.[2] Malignant change can occur in one or both cellular components; this was first described by Hampel in 1970 and further classified by Tavassoli in 1991.[3] It usually occurs in older patients, unilateral, painless breast lump being the commonest presentation.[4] All reported cases were female except for two male patients.[2]Although these are benign tumours , local recurrence is reported, therefore wide local excision is the treatment of choice. Rarely malignant transformation can occur in one or both the components .[3]Differential diagnosis of this tumour includes sclerosing adenosis, fibroadenoma and tubular adenoma.[2]

CASE REPORT:

A 60 years postmenopausal lady presented to our hospital with complains of swelling in the left breast since 2 years and blood stained nipple discharge since 4 days.

Local examination of left breast revealed a firm to hard lump in lower quadrant of left breast measuring about 5x5cm which was non mobile with puckering of skin.

Mammography showed large ill-defined space occupying lesion with cystic changes and increased perfusion and no lymphadenopathy, BIRADS - IV

Trucut biopsy showed invasive breast carcinoma of the left breast.

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Modified radical mastectomy was done and the specimen was sent to our histopathology lab. On gross examination - Left MRM specimen measured 37x16x7 cm. Nipple-areolar complex appeared free from tumor. Cut surface showed a tumor occupying lower quadrant measuring 6x5.5x4cm with areas of hemorrhage, necrosis and cystic changes. Eleven lymph nodes were isolated. Surgical margins were not involved grossly.

Microscopically,breast tissue showed a well circumscribed neoplasm with tumour cells arranged in a lobulated pattern separated by fibrous septae.(Fig 1A) Tumour cells had dimorphic pattern with epithelial lined compressed tubules surrounded by layers of clear cells. (Fig 1B)Tubular epithelial cells showed pleomorphism with prominent nucleoli.Areas of hyalinization, necrosis and desmoplastic reaction were seen.Surgical margins and lymph nodes were free from tumour.Histopathological diagnosis was Malignant adenomyoepithelioma with malignant epithelial and myoepithelial component.

Immunohistochemistry was done showed low Ki67, CK positive in small tubules(Fig 1C), SMA (Fig 1D),S100 (Fig 1E)and p63 (Fig 1F)positive in clear areas which confirmed the diagnosis.

On follow-up patient is doing well now with no recurrence.

DISCUSSION:

Adenomyoepitheilomas have been divided into tubular, spindled or lobulated subtype depending upon their growth pattern .Tubular pattern is the commonest pattern showing epithelial cells in tubular or glandular arrangement surrounded by myoepithelial cells with clear cytoplasm. Malignant changes in one or both components have been described.[1]The exact etiology of breast adenomyoepithelioma is not known. All cases are sporadic and no positive family history is noted. Kiaer et al described a case with sequential change from adenomyoepithelial adenosis to adenomyoepithelioma which eventually became low grade malignant adenomyoepithelioma during a course of 18 years.[1]From these findings Choi et al proposed that adenomyoepitheliomas were derived from long standing myoepithelial breast lesions, such as adenosis and fibroadenoma.Though they are believed to be low grade malignancies, an aggressive clinical behaviour is documented in more than 50% of the cases with predominant haematogenous metatstases with a propensity for a local recurrence .[3]The myoeepithelial cells express tumour suppressors which include angiogenesis and proteinase inhibitors like maspin, p63 and WT-1.The presence of myoepithelial cells is considered a benign sign of a breast lesion, the numbers of which decrease with the advancing grades of neoplasia.[3] Myoepithelial cells are mitotically quiescent with a low proliferative index, but they can be transformed.The spectrum of these neoplasms include adenomyoepithelioma, malignant adenomyoepithelioma (with the degeneration of either or both the components) and a pure myoepithelial carcinoma which is extremely rare. Immunohistochemical confirmation of histologic diagnosis is always necessary. Immunohistochemistry shows that myoepithelial cells are positive for SMA, S100, p63 and desmin, vimentin, calponin, NGFR, CD 10, and EGFR, while epithelial cells show antibodies to cytokeratins based on the degree of differentiation.[5] Differential diagnosis includes tubular adenoma, sclerosing adenosis and fibroadenoma .[3]Tubular adenoma, sclerosing adenosis and fibroadenoma show less proliferative features. The prognosis of these patients is usually good. Wide local excision with adequate margins is the treatment of choice.High mitotic rate, cytologic atypia and infiltrative peripheral borders favour malignancy. Malignant myoepithelial tumours are either pure myoepithelial carcinoma or an adenomyoepithelioma with a component of myoepithelial carcinoma, epithelial carcinoma, sarcoma or carcinosarcoma.Distant metastasis is very rare in low grade tumours. [5]

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CONCLUSION:

Breast tumours are common, but adenomyoepithelioma are rare and this as in our case presenting as malignant adenomyoepithelioma is still more rare.It is useful to identify this tumour entity as it has low grade malignant potential and good prognosis with wide local excision.

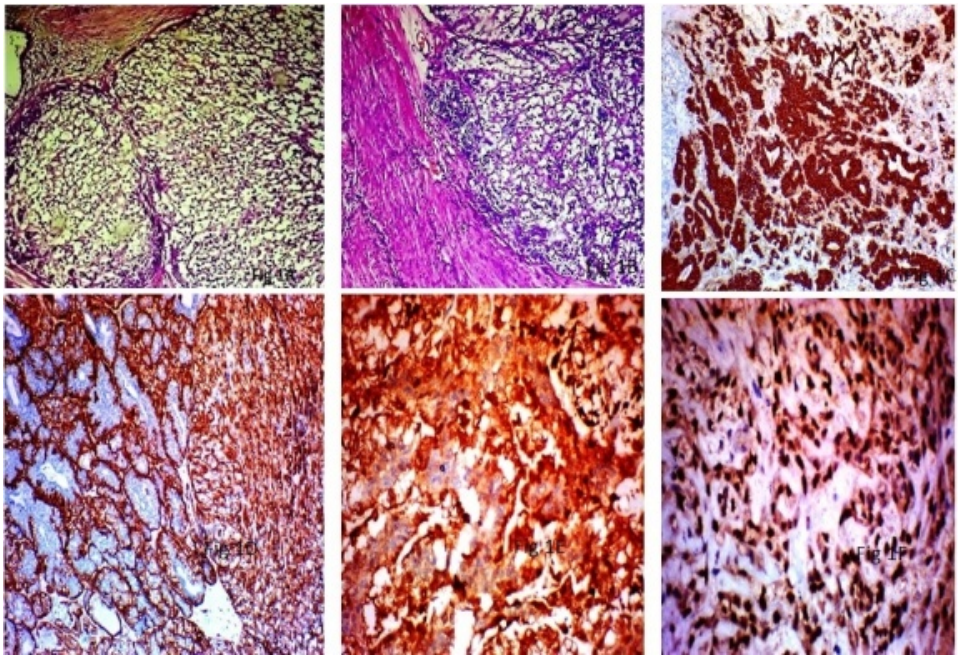

Fig 1A: Tumour cells arranged in a lobulated pattern separated by fibrous septae.

Fig 1B: Tumour cells with dimorphic pattern with epithelial lined compressed tubules surrounded by layers of clear cells.

Fig 1C: CK positive in small tubules.

Fig 1D: SMA positive.

Fig 1E: S100 positive.

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