ISSN Print: 2664-7591
ISSN Online: 2664-7605
Impact Factor: RJIF 5.2
IJAN 2024; 6(2): 06-09
www.pharmaceuticaljournal.in
Received: 10-05-2024
Accepted: 14-06-2024

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Encapsulated papillary carcinoma of the breast: A rare case report

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DOI: https://doi.org/10.33545/26647591.2024.v6.i2a.92

Abstract
Encapsulated Papillary Carcinoma (IPC) of the breast also known as 'encysted' or 'intracystic' papillary carcinoma, is a rare variant of papillary carcinoma affecting predominantly postmenopausal women. Patients present with a palpable mass and sometimes a bloody nipple discharge. As opposed to invasive papillary carcinoma of the breast EPC is a low grade carcinoma with or without stromal invasion and has an excellent prognosis regardless of whether these lesions are in situ or invasive. There are limited data about its epidemiology and only small studies focusing on the outcomes. Here is reported a rare case of encapsulated papillary carcinoma breast in a 70 year old female who presented with a four year history of a slow growing mass in the left breast.

Keywords: Encapsulated, papillary carcinoma, postmenopausal, bloody nipple discharge, stromal invasion

Introduction
Encapsulated papillary carcinoma (IPC), a variant of papillary carcinoma of the breast, comprises of less than 2% of breast cancers [1]. More recent nomenclature is 'encysted' papillary carcinoma [2].
It is more frequently seen in the elderly, median age being 69.5 yrs, more frequent in women (up to 96.5%) than in men (up to 3.5%) [3].
Recent information within the pathology literature, has demonstrated that, although IPC was once thought to be a variant of ductal carcinoma in situ (DCIS), some lesions may actually be low risk invasive tumour. Based on the indolent behaviour of the tumour the WHO task force recommended that intracystic papillary carcinoma be staged and treated as DCIS [5].
Histopathologically it is characterized by papillary carcinoma within a well-circumscribed cystic or distended duct with arborization of the fibro vascular stroma and unlike papillary DCIS these tumour lack myoepithelial cells at the periphery [1, 4, 5, 7].
Regardless of whether these lesions are in situ or invasive in nature, current studies have shown that they have non-aggressive biological behavior and excellent prognosis with only sufficient local therapy [3, 7-9].
Because of its rarity, there is paucity of literature about the tumour. Most studies are in the form of small case series and reviews. Only a limited number of large clinical studies safely assess its appropriate treatment and expected outcome [3, 4].

Case Report
A 70 year old female presented to the Surgery department with a large palpable mass in the left breast. Physical examination revealed an enlarged, tense and tender mass involving the whole of the left breast measuring about 15 x 12 x 8 cms with an area of ulceration oozing blood tinged fluid on the outer and lower quadrant of the breast measuring approximately 2 x 2 x 0.5 cms. Skin was thinned out and stretched but not fixed to the underlying swelling. Multiple left axillary lymph nodes were enlarged, largest measuring 1.5x1 cm. Right breast was normal.
The patient had initially presented with a small lump in the breast 4 years back in another hospital and was disclosed to have a diagnosis of cancer and advised excision of the lump but neglected medical advice for the same.

Because of the constant oozing of blood tinged fluid, the massive enlargement and associated discomfort the patient has now sought medical intervention.

FNAC was done and 15 ml of hemorrhagic aspirate was obtained which revealed a sparsely cellular aspirate suspicious for malignant cells. Ultrasonography of the Left breast was done which showed a thick walled irregular complex mass with septations and an echogenic component.

Modified radical mastectomy of the left breast was done. The gross examination of the specimen showed entire breast tissue with nipple and areola with a multi-loculated, thick walled cystic mass, totally measuring 17 x 9 x 9 cms. An area of ulceration measuring 2 x 2 x 0.5cms was seen in the lower outer quadrant. C/s showed cystic areas filled with dark brown blood clots, largest measuring 14 x 12 x 4 cms with an irregular, solid, whitish area measuring 1.8 x 1.5 cm protruding into the largest cystic space and also involving the posterior resection margin. The smallest cystic area measured 1 x 1 x 0.5 cms.

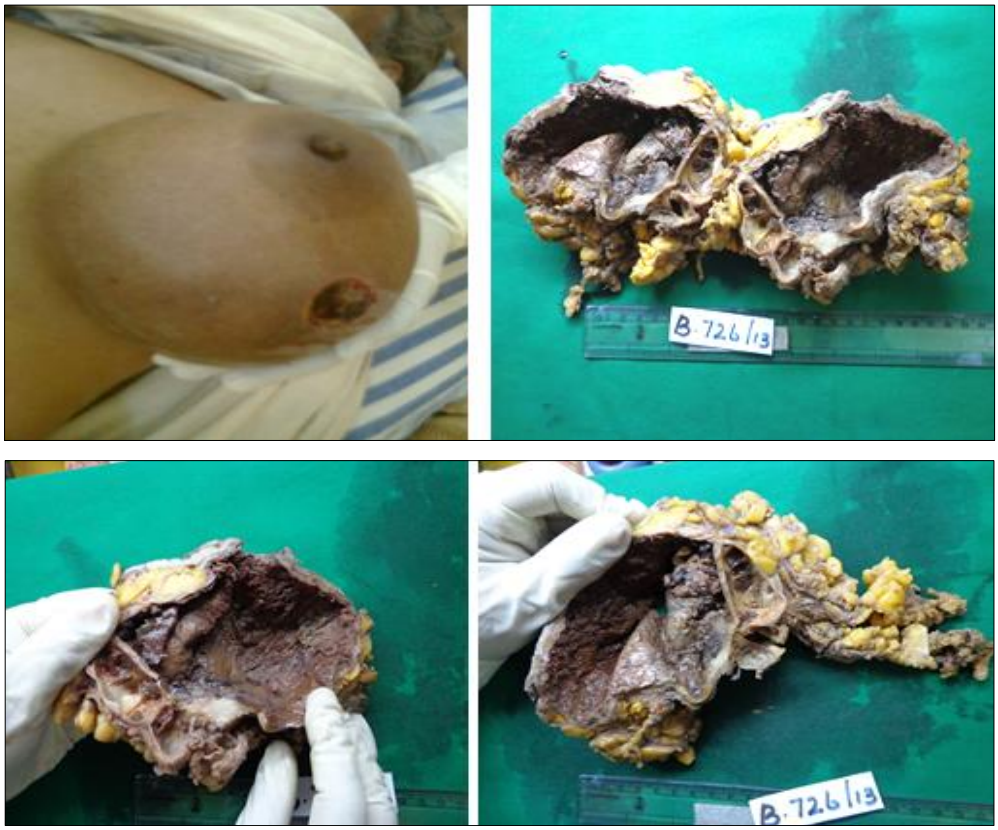

Fig 1: Gross examination of the modified radical mastectomy specimen showing the left breast with multi-loculated thick-walled cystic masses

Examination of the axillary tail revealed 14 lymph nodes largest measuring 1.5 x 1.5 x 1 cms, smallest measuring 0.5 x 0.5 x 0.5 cms. C/S of the nodes showed grayish white solid areas. Histopathological examination of the tumour showed a thick fibrous capsule and papillary structures with layering of round to oval cells with uniformity in the size and shape of the epithelial cells, loss of nuclear polarity, marked hyperchromasia, lack of a myoepithelial component with a stout fibro vascular core. The surrounding stroma was scanty. One area showed infiltration of the stroma by nests of ductal epithelial cells of intermediate grade malignancy with pleomorphism and mitosis.

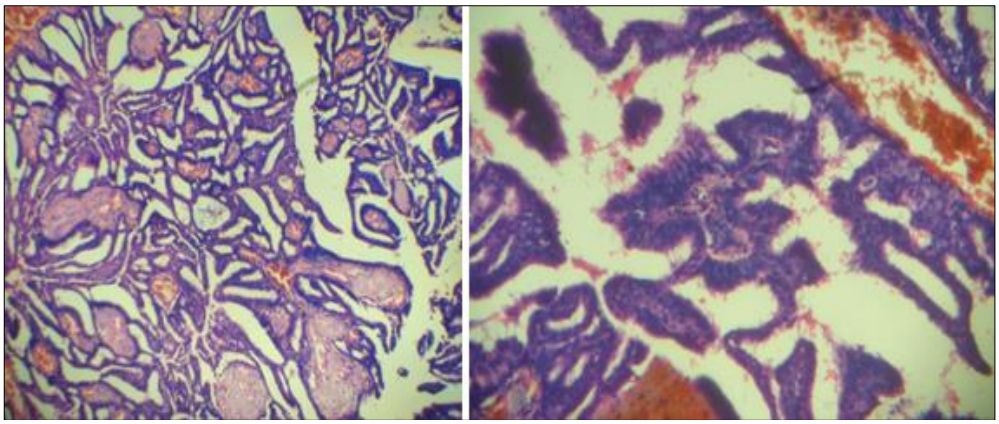

Fig 2: Histopathological image showing papillary structures with uniform epithelial cells and a stout fibro vascular core, characteristic of encapsulated papillary carcinoma.

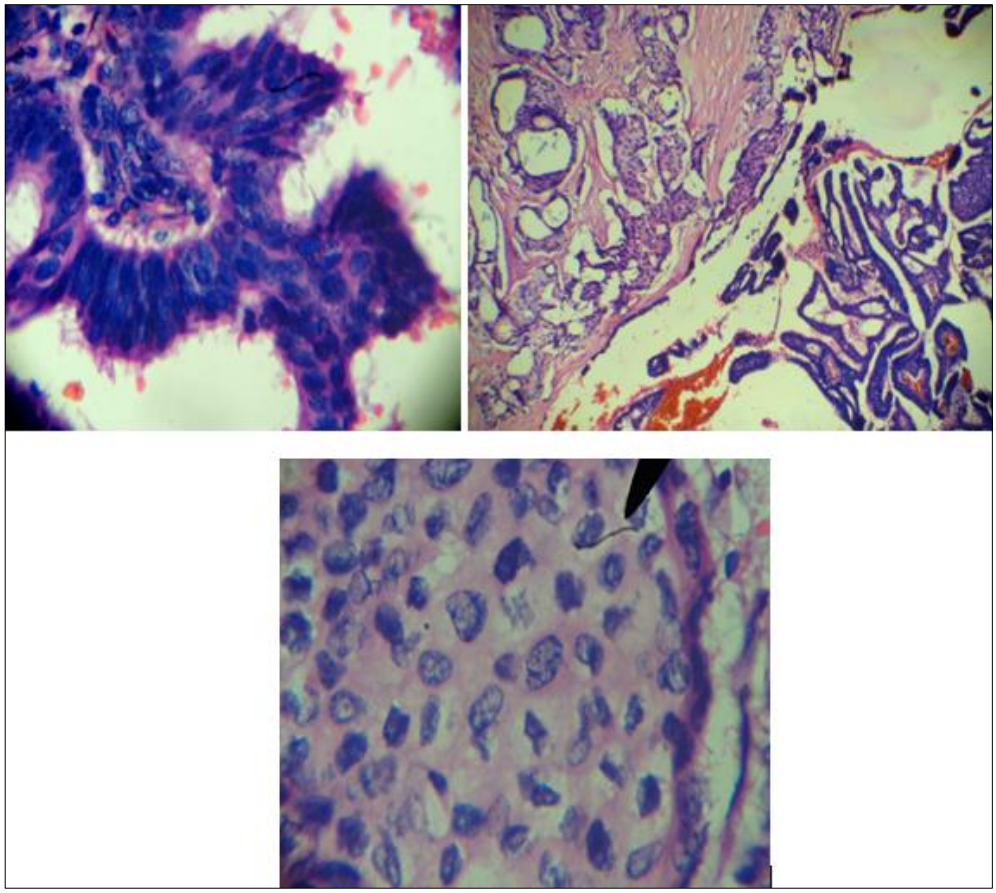

Fig 3: Histopathological section of the axillary lymph node showing metastatic carcinoma.

Examination of the 14 lymph nodes showed metastatic carcinoma in one of them. TNM stage T_4 N_1M_x. ER PR status not known.

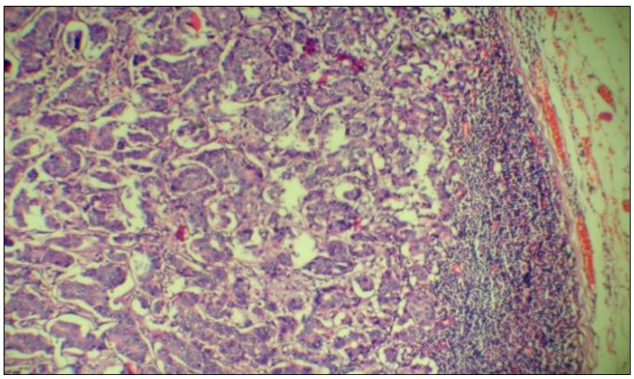

Fig 4: Ultrasonography of the left breast showing a thick-walled irregular complex mass with septations and an echogenic component

The patient refused chemotherapy and further investigations and was put on Tamoxifen citrate. After 16 months of follow up the patient remains disease free even though she did not receive the adjuvant chemotherapy.

Discussion

Encapsulated papillary carcinoma (IPC) of the breast is generally perceived as a malignancy of elderly women but can occur in males although exceedingly rare, accounting for 1% of all breast cancers [1, 2, 4, 6, 8, 12].

Approximately half of the EPC's arise in the retro areolar region of the breast and the usual clinical manifestation is a palpable mass or nipple discharge [4, 7].

FNAC may not be diagnostic in these cases due to cystic and haemorrhagic nature of the lesion. The aspirates are commonly hypo cellular and admixed with haemosiderophages due to the highly vascular nature of IPCs. If adequate material is obtained then it is highly cellular with complex papillae, nuclear hyperchromasia, stratification and foamy macrophages [3, 4, 7].

Radiological studies are helpful. IPC tends to be well defined on mammography, an irregular margin suggests the presence of invasion [6]. Ultrasonography typically reveals a hypoechoic area with soft tissue echoes projecting from the wall of the cyst [6]. Contrast entranced MRI may show marked enhancement of cyst walls, septations and mural nodules [6, 11].

In 1980, a classification system was described by Fisher *et al*. that divided papillary carcinomas into the invasive and non-invasive forms. The non-invasive form was then further subdivided into the diffuse form, the papillary variant of DCIS, involving multiple small and medium sized ducts, and the localized form, termed "intracystic" or "encysted" papillary carcinoma, confined to a dilated cystic space and surrounded by a fibrous capsule and characterized by thin fibro vascular stalks devoid of a myoepithelial cell layer [1, 4-7, 15]. Some pathologists may inadvertently report the tumour as invasive based on the lack of a myoepithelial layer or the presence of entrapped neoplastic cells within the fibrous capsule, which often have the appearance of invasive carcinoma [4, 10].

Encapsulated papillary carcinoma has been perceived as a slow growing tumour which has good prognosis. In a study Carter D *et al*., a series of 41 cases of EPC, 29 patients underwent mastectomy, 11 of them had axillary dissection. None of these patients had metastatic disease in the axillary

lymph nodes or recurrence in the follow up period which averaged 5 years. 11 pts. did not have mastectomy or radiotherapy. Eight of these patients (followed for an average of 10 yrs.) had no recurrence. The only patients who developed invasive carcinoma were those with DCIS as well as EPC in the excisional biopsy [4].

Grabowski *et al*., reported that about 50% of all EPC's are in situ lesions, whereas in the case of invasive EPC's, 10% are regionally extended and < 1% are metastatic [3].

Tumor progression continues as additional mutations occur over time within cells of the tumor population. Some of these mutations will consequently become dominant within the tumor population. The process is called clonal selection, since a new clone of tumor cells has evolved on the basis of its increased growth rate or other properties (such as survival, invasion, or metastasis) that confer a selective advantage [16].

Clonal selection continues throughout tumor development, so tumors continuously become more rapid-growing and increasingly malignant [16]. This forms the basis for this case of EPC with a four year history to evolve into a higher grade of malignancy with stromal invasion and subsequent lymph node metastasis.

In cases of EPC alone, EPC with DCIS, or EPC with invasion, complete local resection of the tumour with clear margins is the recommended surgical treatment. Since EPC rarely involves lymph nodes, partial mastectomy without axillary lymph node dissection is the standard treatment for patients with non-invasive EPC. But the association with invasive carcinoma and the difficulty in evaluating the focus of the invasive lesion may indicate sentinel axillary lymph node biopsy for EPC [3].

There is still no concensus on adjuvant radiation and endocrine therapy. A retrospective study of Fayanju *et al*. including 45 women with pure EPC indicates that only patients with pure EPC who are < 50 years should be considered for radiation therapy. But these tumour are strongly estrogen receptor positive and hormonal therapy could be considered. But in cases of EPC with DCIS adjuvant radiotherapy or chemotherapy could be useful given their low, but not insignificant possibility of loco regional relapse and/or metastases [3].

Conclusion

Encapsulated papillary carcinoma is a rare breast carcinoma with an excellent prognosis. Awareness, early and prompt diagnosis of this clinicopathological entity is important especially because of the good prognosis it carries.

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