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Case Report

Adenoid cystic carcinoma of the male breast: Case report and review of literature

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ABSTRACT

Background: Male synchronous bilateral breast cancer of Adenoid cystic carcinoma (ACC) is extremely rare.

Case Report: A 84-year-old male presented with a simultaneously, bilateral, rapidly growing lump in breast of three months duration. Physical examination right breast, revealed a single subareolar mass measuring 3.4 x 3 x2.8 cm. It was firm to hard, well-defined mass with retraction of nipple. The left breast mass was firm tender, measuring 3.1x2.5x2.1cm with irregular margins. Bilateral lymphadenopathy was noted. Sonommamography was suspicious of neoplastic lesion with secondary changes. The FNAC reported as positive for carcinoma cell of bilateral breasts. The bilateral modified radical mastectomy was done. On histopathology, we reported as bilateral breast cancer of Adenoid cystic carcinoma-solid, cribriform, tubular pattern, grade III in right breast and ACC with areas of invasive breast carcinoma and neuroendocrine features in left breast. All 24 right and left axillary lymph nodes were negative for metastesis. On immunohistochemistry staining tumor cells were negative for Estrogen Receptor, progesterone receptor, HER2/neu oncogene.

Conclusion: ACC of breast is a rare subtype of breast cancer. We present extremely rare case of male synchronous bilateral breast cancer of ACC for its clinical manifestations, radio imaging, and histopathological features.

1. Introduction

Male breast cancer is an uncommon. Here with was report an extremely rare case of synchronous bilateral breast cancer in male patient. On histopathological study diagnosed as bilateral Adenoid cystic carcinoma (ACC) breast. ACC of the breast, a rare type having less than 0.1% of all breast malignancies.[1] It is a low-grade malignancy. It shows a variety of morphological features which is having a prognostic impact. On immunohistochemistry these tumor are usually "triple negative". Although ACC in female patients is considered good prognotic value, in male it may be shows worse prognosis due to potentially aggressive behavior and the neglection of the tumor.[2]

2. Case Report

A case of a 84-year-old male with who presented with a simultaneously, bilateral, rapidly growing lump in breast of 3months duration. History of chest trauma was given. Patient was known case of hypertention of 15 years and on treatment of Novastast. He was known case of bilateral grade I chronic kidney disease. There was no any significant family history. Physical examination right breast, revealed a single subareolar mass measured 3.4 x 3 x2.8 cm. It

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was firm to hard, well-defined mass with retraction of nipple. The left breast mass was firm tender and measured 3.1x2.5x2.1cm with irregular margins. The bilateral axillary lymphadenopathy was noted.

On sonomammography right breast lesion was located in the subareolar region and measured 3.6 x2.5cm.It was hypo echoic with small cystic areas suggestive of? haematoma, ?fat necrosis ?neoplastic. Left breast lesion located in the subareolar region, measured 3.1 x2.8cm, was anechoic with infiltrative margin suggestive of neoplastic lesion. Bilateral lymphadenopathy was noted. On systemic examination there was no evidence of metastasis. The FNAC reported as positive for carcinoma cell of bilateral breast. The bilateral modified radical mastectomy and axillary lymph node dissection was done. On the histopathological evaluation, we received right and left modified radical mastectomy specimens. Right breast mass measured 3.4 x 3 x2.8 cm. It was single, well circumscribed, soft to firm tumor with focal areas of cystic changes (Figures 1 and 2). The right axillary eleven lymphnodes were noted. Left breast mass measured 3.5x2.5x2.5cm. It was single, well circumscribed, soft to firm tumor (Figures 3 and 4). The left axillary thirteen lymphnodes were noted. On histopathology reported as bilateral Adenoid cystic carcinoma-solid, cribriform, tubular pattern, grade III (Figures 5 and 6). In left breast along with ACC areas of invasive breast carcinoma were noted (Figure 7). All 24 right and left axillary lymph nodes were negative for metastesis. On immunohistochemistry staining tumor cells were negative for Estrogen Receptor, Progesterone Receptor, HER2/neu oncogene and positive for cytokeratin. The patient responded well to treatment and remained well on follow-up.

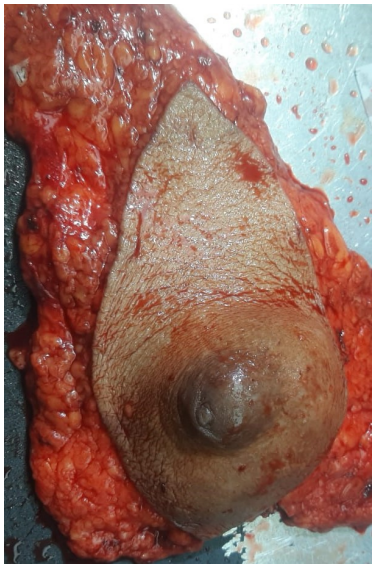

Figure 1: Right breast showing single subareolar mass measuring 3.4 x 3 x2.8 cm

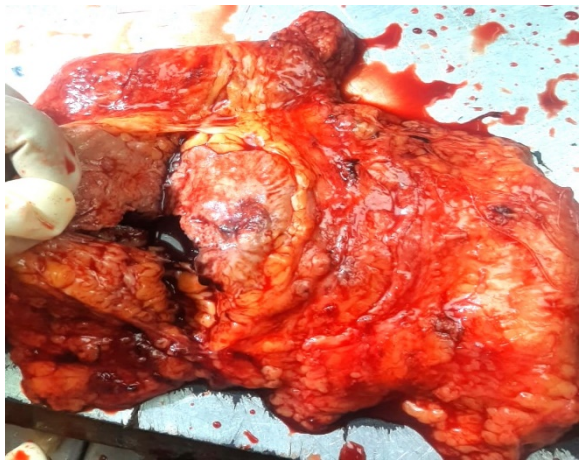

Figure 2: Right breast cut section showing firm to hard, well-defined mass measuring 3.4 x 3 x2.8 cm with tiny cystic areas

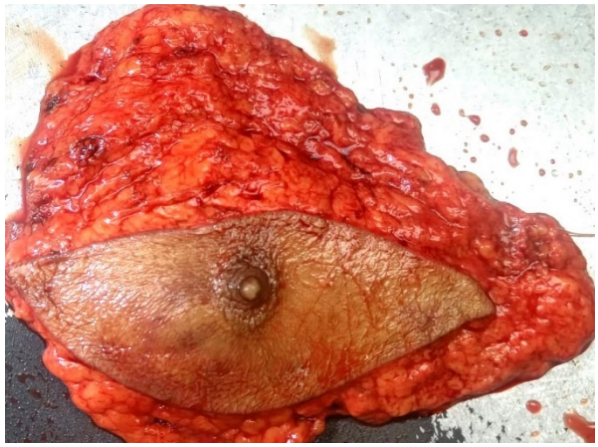

Figure 3: Left breast showing single subareolar mass measuring 3.5x2.5x2.5cm

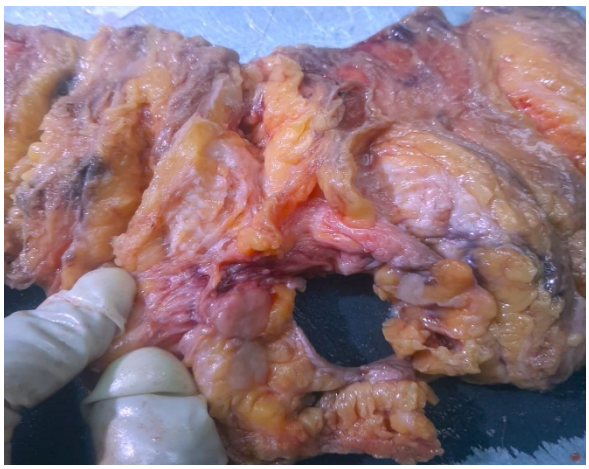

Figure 4: Left breast mass cut section showing firm well-defined measuring 3.1x2.5x2.1cm

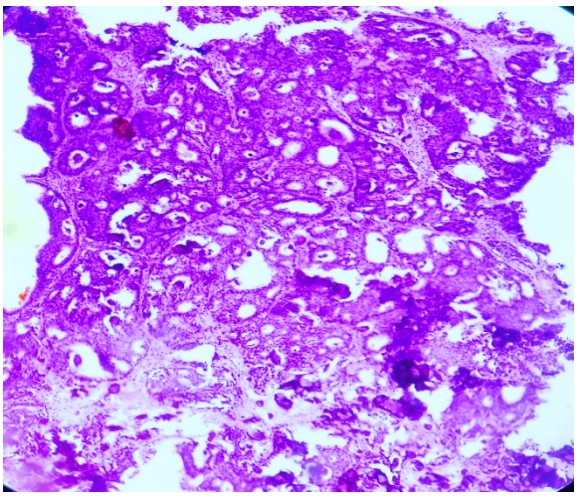

Figure 5: Microscopic of right breast adenoid cystic carcinoma showing a cribriform, tubular and solid growth patterns.(H&E staining, 40x)

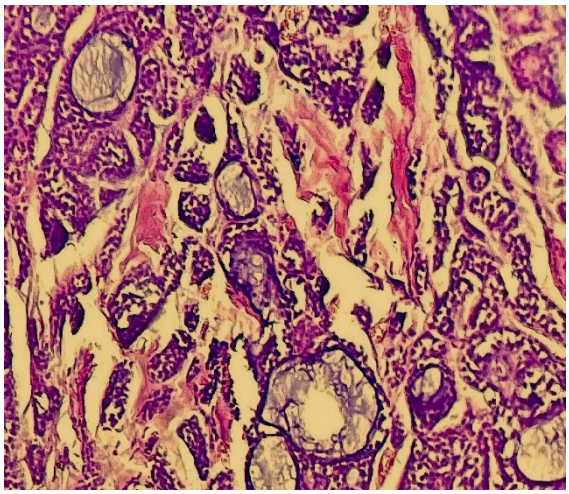

Figure 6: Microscopic of breast adenoid cystic carcinoma showing a, tubular and solid growth patterns.(H&E staining, 100x)

3. Discussion

Male breast cancer is a rare neoplasm, accounting for about 1% of breast cancers overall.[3]

The term "synchronous" refers to the presence of primary tumors in both breasts, which are diagnosed simultaneously. The bilateral synchronous breast cancer presentation is extremely rare having incidence of 1.5%-2% of all male breast cancers.[4] The cases of metachronous bilateral breast cancer presentation of breast cancer were observed.[5]

Most cases of male breast cancer are detected between the ages of 60 and 70years.[6] ACC of breast is a rare subtype of breast cancer. On histomorphology it is identical to salivary gland counterpart. ACC is commonly seen in the major and minor salivary glands. The rarely cases are reported in other sites like breast, skin, cervix, lung, lacrimal gland, etc.

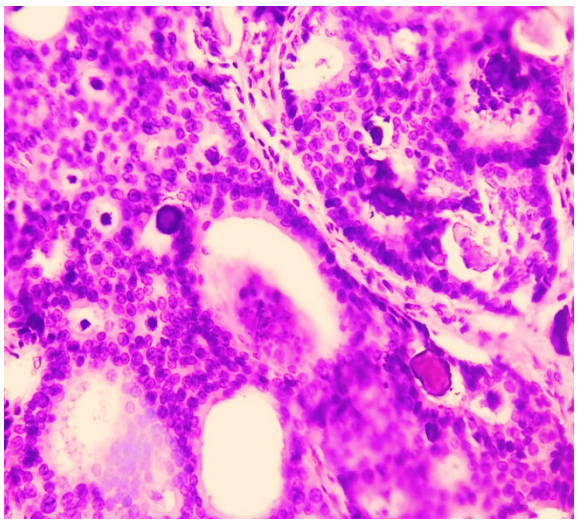

Figure 7: Microscopic of left breast adenoid cystic carcinoma showinga solid and basaloid growth patterns, lumens containing eosinophilic cylinders comprised of basement membrane material (H&E staining, 400x)

The most frequent histological type in male is invasive breast carcinoma (85-90%), other rare types are invasive papillary (4.5%), mucinous (2.8%), and papillary carcinoma.[7] The large tumor size, axillary lymph node metastasis and on histopathology grade II/ III invasive breast carcinomas shows poor prognostic criteria.[8] In our case, it was bilateral solid - basaloid variant ACC.

On histopathological examination ACC of breast characterised by a dual cell population of luminal and basaloid cells and it is composed of both epithelial and myoepithelial components.

Adenoid cystic carcinoma (ACC) was first discovered by Robin and Laboulbene in 1853 and described as "tumeur heteradenique". Then ACC was first used in breast oncology by Geschikter in 1945.[9] Male breast ACC is very rare, first reported in 1969, since then a total of 17 cases have been reported.[10]

ACC in males breast clinical present as firm to hard palpable subareolar mass. It may be cystic. The mass is accompanied by tenderness. In late presentation skin ulcers, nipple dischages, nipple retraction are noted.[11]

ACC occurs commonly in age of 38-81 years (median age of 60 years. The average size of tumor in ACC is 3.0 cm.[12] ACC noted equally on the left and right side and usually located in the subareolar region. occurring equally on the left and right sides.[13]

The breast ACC on mammography manifests as an irregular, lobulated, heterogeneous mass with indistinct or microlobulated edges.[14]

3.1. Histological characteristics and differential diagnosis

The histopathological features are diagnostic for breast ACC. Breast ACC is a kind of biphasic tumor composed of basaloid and luminal epithelial cells. The tumor cells are distributed around small cysts, forming both true glands and pseudoglandular spaces containing eosinophilic basement membrane material and basophilic mucin. The sub-type are mostly of three different structural patterns: tubular, cribriform, and solid, either alone or in combination.[15] May be associated with microglandular adenosis. The breast ACC rarely shows nerve infiltration

The various pattern on histopathological findings were given as cribriform, solid, tubular or trabecular architectural patterns. In our case it was predominantly soid and tubular patten. Also depending on glandular lumnina two types of lumens are a) True glandular lumina lined by ductal epithelial cells and b) Pseudolumens containing eosinophilic cylinders comprised of basement membrane material. In our case both were noted.

The various histologic variants are a) Classic variant having low to intermediate nuclear grade and composed of mixture of 3 different architectural patterns -tubular, cribriform and solid.[16]This was observed in our case in right side breast. It is most common most variant. b) solid - basaloid variant composed of mixture of solid nests and basaloid features. These tumor type is more aggressive having increased mitotic rate, areas necrosis, with increased proliferatve index of Ki67 > 30%.[17] c) Other is ACC with high grade transformation. It is associated with invasive breast cancer of NST type. This was observed in our case on left side breast mass.

Ro et al. first classified breast ACC into three grades based on the ratio of solid components within the lesion. Grade I - numerous glands and cystic components, without solid growth. In grade II tumors-solid growth is <30%, while in grade III tumors solid growth is >30%. Ro et al also proposed that grade II and III tumors are larger in size and are more prone to relapse.[18]

The histological differential for ACC of breast which presents as cribriform or tubular pattern is invasive cribriform/tubular carcinoma, and cribriform ductal carcinoma in situ. The cribriform carcinoma lacks dual cell population. Cribriform ACC should be differentiated from invasive cribriform carcinoma and adenomyoepithelioma. The benign collagenous spherulosisis another differential.

3.2. Immunohistochemical characteristics

Breast ACC is a type of basal-like breast cancer that is generally triple negative for Estrogen Receptor, Progesterone Receptor and Human Epidermal growth factor Receptor 2 (HER2/neu). Unlike other triple-negative breast cancers, however, breast ACC has a favourable prognosis.

The glandular epithelial cells express cytokeratin CK7, CK8/18, epithelial membrane antigen and CD117 (C-kit). Myoepithelial cells are immunoreactive for CK5/6, CK14, CK17, P63, smooth muscle actin, calponin, and vimenti.[15] Basement membrane markers, e.g. collagen IV or laminin, can be used.

ACCs of breast show a low proliferation activity by standard Ki-67 labeling index. Axillary nodal metastases are rare, more frequently seen in the solid variant.

3.3. Treatment and prognosis

The prognosis of male breast cancer remains uncertain because of the late diagnosis, unpredictable course, and high potential for metastasis.[19] According to Ro JY grading system, local excision is recommended for grade 1, simple mastectomy is recommended for grade 2, and mastectomy and axillary dissection is recommended for grade.[3,15]

The management for male breast cancer is mastectomy (simple, radical or total mastectomy), while chemotherapy, radiation and hormonal treatment are infrequently used. An adjuvant radiotherapy may be beneficial for positive surgical margins. For patients with high-grade lesions, axillary lymph node or distant metastasis recommend systemic adjuvant chemotherapy.[20,21] There is limited information about the prognosis of breast ACC in males related to the extremely low numbers of reported cases. ACC has a relatively favorable prognosis as compared with other invasive breast carcinoma.Veeratterapillay R et al, observed the breast ACC is a rare malignant neoplasm with an excellent prognosis having 5-year survival rate in 98% cases.[22] Few cases of recurrence and metastasis observed during long-term follow-up related to positive surgical margin, variant like ACC with high grade transformation.

4. Conclusion

ACC of breast is a rare subtype of breast cancer. We present extremely rare case of male synchronous bilateral breast cancer of ACC for its clinical manifestations, radio imaging, and histopathological features.

5. Source of Funding

None.

6. Conflict of Interest

None.

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