Case Report

Intracystic papillary carcinoma in the male breast: A diagnostic challenge

Veena Raja, Chinnaiya Subramaniam Babu Rajendra Prasad, Prashanth Murthy[1]

Department of Pathology, Sri Devaraj Urs Medical College, Sri Devaraj Urs Academy of Higher Education and Research, Tamaka, Kolar, [1]M.S. General Surgery, Murthy Nursing Home, Chikballapur, Karnataka, India

ABSTRACT

Breast carcinoma in men is rare, and "intracystic papillary carcinoma (IPC)" of a male breast is an extremely rare entity accounting for <1% of all breast malignancies. It represents a small distinctive subgroup of noninvasive breast cancer. We discuss a case of a 70-year-old male presented with left breast lump of 6 months duration which rapidly increasing in size over 10-day period showed cystic mass with a mural nodule on ultrasonography. A diagnosis of papillary neoplasm was awarded (suspicious for malignancy-C4) on cytology. The patient underwent simple mastectomy, and a final diagnosis of "IPC" was confirmed on histopathology. IPC of the male breast is an extremely rare entity with favorable prognosis. Triple assessment (clinical examination and radiological and histological assessment) is necessary to diagnose IPC. Pathologic diagnosis can be difficult at classical histological examination; thus, the absence of myoepithelial cells layer by immunohistochemical study can be useful.

Key words: Intracystic papillary carcinoma, male breast carcinoma, myoepithelial markers

INTRODUCTION

Breast carcinoma in men is rare, and intracystic papillary carcinoma (IPC) of the male breast is an extremely rare entity accounting for <1% of all breast malignancies.[1,2] It represents a small distinctive subgroup of noninvasive breast cancer.[3,4] The reported 10-year survival rate for IPC is 100%, and the recurrence-free survival rate is 96% and 77% at 2 and 10 years, respectively.[5] The microscopic examination of the lesion, together with the additional immunohistochemical studies, provides important information for the diagnosis, which cannot be made on clinical or macroscopical examination.[6,7]

CASE REPORT

A 70-year-old male presented with left breast lump of 6 months duration which rapidly increasing in size over 10 days period and is associated with pain. On examination, there was an enlarged left breast with soft to cystic in consistency, without nipple retraction or peau d'orange. The mass was free from the underlying structures. Nipple and areola are normal. There is also a keloid on the sternum. There is no history of nipple discharge [Figure 1]. The patient is not on any medications for some other diseases. He is nonalcoholic and nonsmoker. There is no significant ipsilateral axillary lymphadenopathy. There is no history of bone pain. Ultrasound of left breast showed cystic mass with a mural nodule and thickened wall at areas. Ultrasound of right breast was normal. Ultrasound of testis was not contributory.

Fine needle aspiration cytology

It yielded 5 ml of blood tinged fluid. Repeat aspiration done after evacuating the cyst, showed ductal epithelial cells in discohesion and a papillary structure exhibiting columnar cells with palisading, mild to moderate

Address for correspondence: Dr. Veena Raja,
Department of Pathology, Sri Devaraj Urs Medical College,
Sri Devaraj Urs Academy of Higher Education and Research,
Tamaka, Kolar, Karnataka, India.
E-mail: drveenaraja88@gmail.com

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anisokaryosis, nuclear overlapping, and moderate cytoplasm. Myoepithelial cells were absent.

A diagnosis of papillary neoplasm was awarded (suspicious for malignancy-C4) [Figure 2].

Surgeon went ahead with simple mastectomy without axillary clearance.

Histopathology report

Gross

Mastectomy specimen measured 8 cm × 5 cm × 3 cm. External surface is unremarkable. Cut surface of the mastectomy specimen shows cyst with mural nodule and papillary excrescences. Cyst is filled with blood clots and brownish fluid [Figure 3].

Microscopy

Section studied shows tumor cells arranged in a papillary pattern, cribriform pattern, and solid sheets. Epithelial cells were round to spindle, with moderate eosinophilic cytoplasm and pleomorphic nuclei. The nucleoli were evident. Mitosis was seen in 1-2/HPF. Necrosis was noted at a few areas. There was lack of myoepithelial cells. No invasion was seen [Figure 4].

Diagnosis-intracystic papillary carcinoma

Immunohistochemistry

Markers for myoepithelial cells were performed. Calponin, smooth muscle actin, p63, carcinoembryonic antigen (CEA), and S-100 protein were negative [Figure 5]. All these markers confirmed the absence of myoepithelial cell layer. Tumor cells were negative for estrogen receptor. Tumor cells are positive for progesterone receptor (0–5%, 1 + 20%, 2 + 35%, 3 + 40%).

DISCUSSION

Intracystic papillary breast carcinoma is rare in females and exceedingly rare in males. The spectrum of lesions

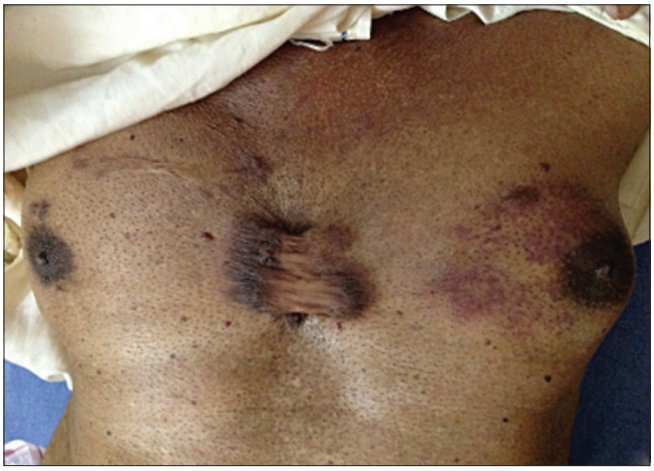

Figure 1: Clinical picture showing enlarged left breast without nipple retraction of peau d'orange. There is also a keloid on the sternum

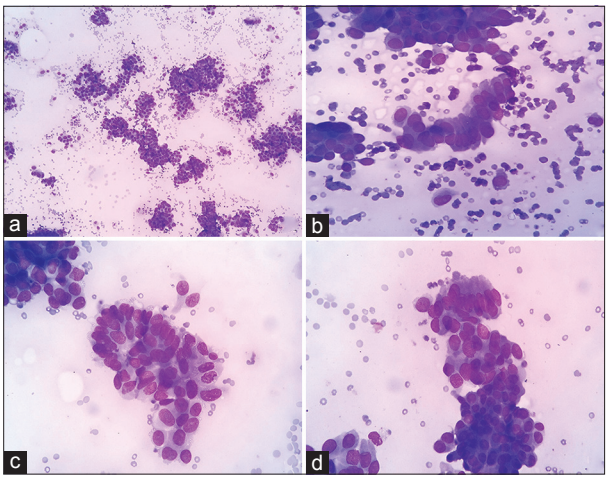

Figure 2: (a) Highly cellular smear showing papillary structures and also discohesive cells, (b) columnar cells with nuclear palisading and mild anisokaryosis, (c and d) columnar cells with palisading, overlapping of nuclei and mild anisokaryosis

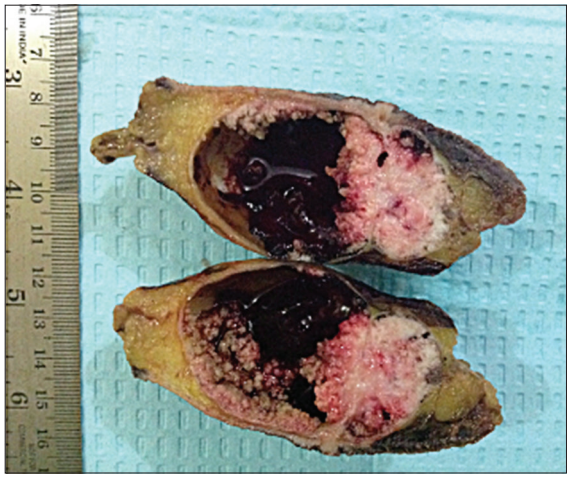

Figure 3: Cut surface of the mastectomy specimen showing cyst with mural nodule and papillary excrescences. Cyst filled with blood clots and brownish fluid

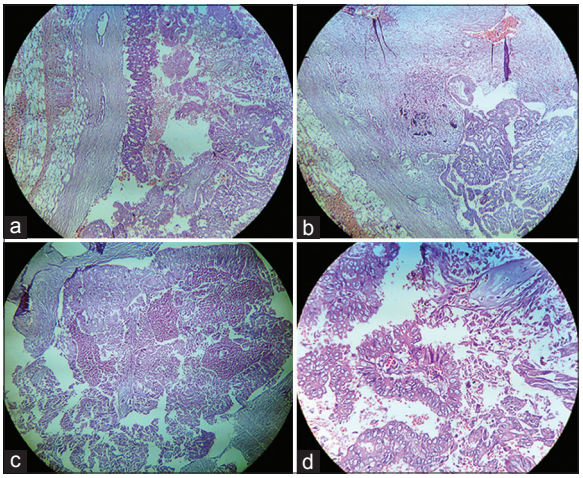

Figure 4: (a) Well-circumscribed lesion showing solid pattern (b) focal areas of calcification, (c) cribriform pattern (d) papillary pattern

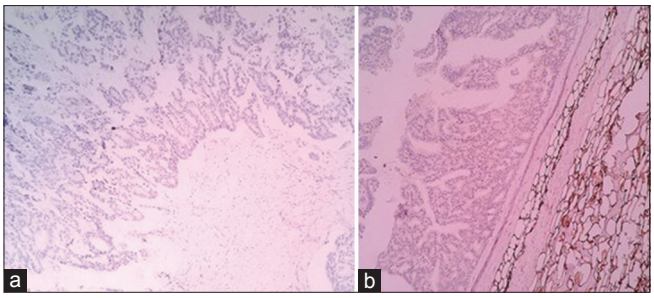

Figure 5: (a) Carcinoembryonic antigen (b) S-100. Both were negative for myoepithelial cells

ranges from benign intraductal papilloma to intraductal papillary carcinoma and invasive papillary carcinoma. The diagnosis of IPC of the male breast should be made carefully. Triple assessment is essential-clinical examination, radiological and histological assessment. The most common symptom is growing fullness in the breast or gynecomastia in males. Nipple discharge can also be a presenting symptom.[1-3] Fine needle aspiration cytology in cystic lesions of the breast may not offer the same sensitivity and specificity as it does in solid lesions of the breast. This could lead to misdiagnosis of cancer.[1,4] Radiological studies are helpful. IPC tends to be well defined on mammography; an irregular margin suggests the presence of invasion. Ultrasonography of these lesions typically reveals a hypo-echoic area (representing the cyst) with soft tissue echos projecting from the wall of the cyst (intracystic tumor).[1,4,5] Histologically, IPC is divided into three subgroups: Pure form, IPC associated with ductal carcinoma *in situ*, and IPC associated with invasive carcinoma.[3,4] IPC may show four cellular patterns: Cribriform, compact columnar epithelial, stratified spindle cell or a transitional cell form resembling urothelium, or a combination of two or more of these patterns may be seen.[4] Intraductal papillomas are characterized by an arborescent structure composed of fibrovascular stalks covered by a layer of myoepithelial cells with overlying epithelial cells. Invasive papillary carcinoma is characterized by thin fibrovascular stalks with neoplastic epithelial cell population and devoid of the myoepithelial cell layer.[1,2] Myoepithelial cell staining suggests a spectrum of progression from *in situ* disease to invasive disease. The lack of an intact basal myoepithelial cell layer can be identified by calponin, smooth muscle myosin heavy chain cytoplasmic stains, and p63 nuclear stains. This "gold standard" method has a relatively high sensitivity and denotes the invasiveness of the tumor cells in the malignant papillary breast lesions.[4,6,7] Other markers which can be used for identification of myoepithelial cell layer are cytokeratin (CK14 and CK17), CEA, S-100 protein, and factor VIII.[6,7] Tsuda *et al*. reported that loss of heterozygosity (LOH) on chromosome 16q was a useful marker for IPC since intraductal papilloma showed no LOH. Using the polymerase chain reaction, the malignant potential of intracystic papillary lesions may be more clearly determined.[8] There is currently no consensus regarding the management of IPC. A study done by Harris *et al*. reviewed patients who underwent varying combinations of local excision, radiotherapy, and tamoxifen. The absence of axillary involvement and low recurrence rate after local excision suggests that wide local excision without axillary dissection is currently the treatment of choice for pure IPC. IPC associated with DCIS or invasive cancer or both should be treated on the basis of this associated pathology. Prognosis of pure IPC is usually excellent because the malignant potential and the proliferative activity of the cancer are low.[9]

CONCLUSION

IPC is an extremely rare lesion of the male breast. The diagnosis of IPC of the male breast should be made carefully. Triple assessment is essential (clinical examination and radiological and histological assessment). The diagnosis of an intracystic malignancy should be suspected when ultrasonography shows intracystic mural nodule, the mass persists or recurs after fine needle aspiration or cellular atypia is identified in the aspirate specimen. The most important feature for the diagnosis of IPC is the absence of a myoepithelial cell layer in the papillary processes. The mainstay of treatment is surgical excision, with adjuvant therapy if associated to DCIS or invasive carcinoma.

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Conflicts of interest

There are no conflicts of interest.

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