

## Intracystic Papillary Carcinoma of The Breast a Rare Tumor in Male

Case Report

Safini F<sup>1\*</sup>, Benchakroun N<sup>1</sup>, Karkouri M<sup>2</sup>, Bouchbika Z<sup>1</sup>, Jouhadi H<sup>1</sup>, Tawfiq N<sup>1</sup>, Sahraoui S<sup>1</sup>, Benider A<sup>1</sup>

<sup>1</sup> MOHAMMED VI Cancer Treatment Center, Radiation Oncology Department, Ibn Rochd University Hospital, Morocco.

<sup>2</sup> Department of Cytology and Histopathology, Ibn Rochd University Hospital, Morocco.

### Abstract

Intracystic papillary carcinoma of the breast in men is an extremely rare disease with an excellent prognosis. Here, we report a new case of intracystic papillary carcinoma of the breast in a 79-year-old man. He was admitted in our department for a painless cystic lump in the left breast. He underwent a mastectomy without lymph node dissection. The pathologic examination objectified an intracystic papillary carcinoma without an invasion. There are no clear guidelines about the management of IPC. Most publications reinforce the importance of surgery is the mainstay of treatment, which can be either conservation or mastectomy.

**Keywords:** Breast; Intracystic Papillary Carcinoma; Man.

### Introduction

Intracystic papillary carcinoma (IPC) is a rare form of breast cancer, accounting for 0.5–1% of all breast cancers. Since its first description, IPC in male is a very rare tumor and few cases were published in the literature. It accounts for 5 to 7.5% of all male breast cancers. [1, 2, 3, 4]. A new case of IPC of the breast in a 79-year-old male is presented with review of the literature.

### Case Report

A 79-year-old male patient presented at the Mohammed VI Center for Cancer Treatment in Casablanca, Morocco, in July 2013 with a painless swelling in his left breast. He had no remarkable past medical or family history. On physical examination, ultrasonography coupled to mammography showed a retroareolar complex cyst lesion with a fleshy portion and this lesion was classified BI-Rads 4. Ultrasound-guided aspiration revealed a papillary carcinoma. He underwent breast-conserving surgery without axillary lymph node dissection. The lesion was 3 cm×2.8 cm×2 cm in size. Histopathology examination revealed a carcinoma-tous tumor with papillary architecture developed in a cystic cav-

ity lined by a fibrous wall, without necrosis or extension beyond the cyst wall (Figure1). The tumor was completely excised with clear margins. Immunohistochemistry showed negative HER2 with positive estrogen receptors (80%) and positive progesterone receptors (90%). The diagnosis of intracystic papillary carcinoma without invasion was made. An adjuvant hormone therapy was recommended by tamoxifen, without either chemotherapy or radiotherapy. The patient had no recurrence after 36 months of regular follow up.

### Discussion

Intracystic papillary carcinoma (IPC) of the breast in males is an extremely rare disease and few cases are reported in the literature [1]. The term "intracystic papillary carcinoma" refers to a solitary tumor in a cystic and dilated duct [1, 2]. IPC had been divided into three subgroups, which seems to correlate with the prognosis: IPC alone (pure form), IPC with ductal carcinoma in situ (DCIS), and IPC with invasion. It's often very hard to make a distinction between these three subtypes [1, 2]. The role of the pathologist is crucial, given the need for a very careful study of the entire specimen to not miss an invasive component or DCIS,

#### \*Corresponding Author:

Fatima Safini.

MOHAMMED VI Cancer treatment Center, Radiation Oncology Department, Ibn Rochd University Hospital, Morocco.

Tel: +212 22483030

E-mail: safinifatima@yahoo.fr

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Figure 1. Tumor Proliferation Without Underlying Stromal Invasion, Lined By a Fibrous Wall. Hematoxylin-Eosin, magnification  $\times 4$  (A).

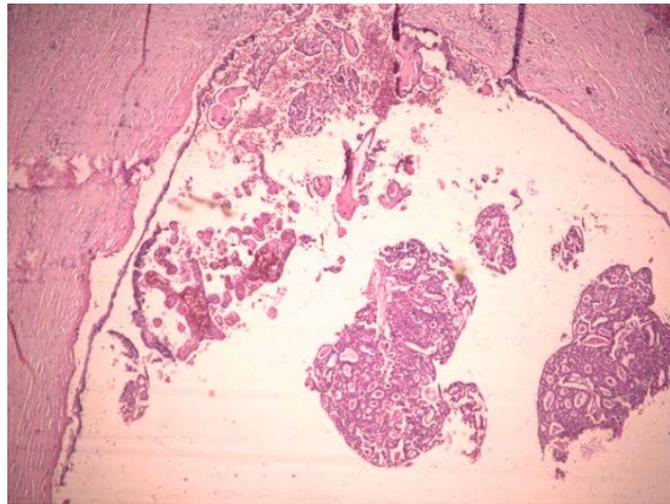
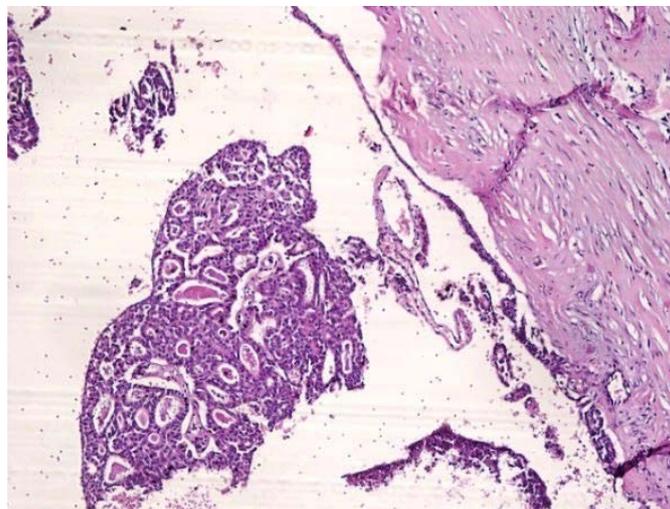


Figure 2. Atypical Tumor Cells. Hematoxylin-Eosin, Magnification X 20 (B).



because the detection of associated lesions is primordial for treatment decision [3, 4]. Using myoepithelial cell staining, Hill et al., suggest considering the cases of IPC as a form of progression between in situ and invasive disease. The lack of a basal MEC layer suggests that these tumors might behave in an invasive manner and might explain the distant metastases from IPC reported in the literature [5].

The use of the Calponin, smooth-muscle myosin heavy chain (SMM-HC) cytoplasmic stains can identify the presence of an intact basal MEC layer orienting to the absence of invasive component [6].

This rare entity is commonly reported in old men with a mean age in males of 68, 2 years (67 to 84 years) [3, 7]. Patients often diagnosed with a palpable breast mass, as was the case in our patient, other clinical abnormalities can revealed IPC like mild pain, bloody nipple discharge and/or pruritus [8]. The imaging is based on mammography and ultrasound. The radiologic presentation of IPC is not specific; and on ultrasonography, it can appear like a pure cyst, a solid mass or a mixed lesion [4, 9].

Because the invasive component and DCIS occur at the periphery, cytological diagnosis can give a false negative result and exci-

sion biopsy of the papillary lesions is recommended to exclude invasive cancer or associated adjacent DCIS [1, 2, 10]. Some authors suggested that core biopsy can also distinguishing papillary neoplasms from other diseases and benign papillomas from papillary carcinoma [11, 12].

Immunohistochemical studies found that IPC breast cancer in males has higher rates of oestrogen and progesterone receptor positivity than are found in women, this explain the good prognosis of this tumor, but the HER2 was almost negative [9, 13].

The optimal management of IPC remains controversial and there is currently no consensus about the treatment of IPC, but all authors indicate that it should be based on the associated pathology and the decision must be taking in the context of a multidisciplinary discussion.

Surgery remains the mainstay of treatment, which consists of either a mastectomy or lumpectomy with complete local excision of the tumor with clear margins [1, 2].

Axillary node metastasis can occur in up to 14% of the cases; therefore, an axillary dissection is recommended by some authors in patients with IPC associated with invasive component but not

**Table. Published Case Reports of Intracystic papillary carcinoma of the Breast in males**

No.	Reference	Age (years)	Symptom	laterality	Tumor size	Histology	Surgical treatment	Adjuvant treatment	Follow up (month)	outcome
1	Imoto S et al., (1998) [10]	62	mass	right breast		non-invasive IPC	lumpectomy	No	12	NED
2	Pacelli A et al., (2002) [12]	67	painless mass	right breast	15 mm	non-invasive IPC	Simple mastectomy+axillary sentinel node biopsy	NA	NA	NA
3	Kinoshita T et al., (2005) [11]	71	Mass+ bloody nipple disch	right breast	6 cm × 5 cm	non-invasive IPC	simple mastectomy	No	NA	NA
4	Sinha et al., (2006)	75	painless mass	right breast		non-invasive IPC	lumpectomy	No	12	NED
5	Romics L Jr et al., (2009) [16]	44	swelling	left breast	2.5 cm × 1.8 cm × 1.2 cm	IPC -DCIS	mastectomy with sentinel lymph-node	Adjuvant radiotherapy (40 Gy in 25 fractions)	NA	NA
6	Pandey P et al., (2009) [8]	50	painless lump	left breast	5 cm × 4 cm × 4 cm	pure IPC	lumpectomy	No	15	NED
7	Hussain A et al., (2012) [7]	58	painless	right breast	3.5 cm	IPC small focus of invasive ductal carcinoma	total mastectomy and sentinel node biopsy	Adjuvant hormone therapy (tamoxife)	12	NED
8	Muallaoglu S et al., (2012) [3]	48	painful swelling	right breast	2cm × 3 cm	non-invasive IPC	lumpectomy	No	24	NED
9	Taha et al., (2013) [4]	68	mass	right breast	20 × 11 mm	IPC	Lumpectomy+axillary lymph node dissection	NA	NA	NA
10	Hariprasad S et al., (2013) [9]	50	mass	left breast		non-invasive IPC	Simple mastectomy	No	24	NED
11	Vagholkar K et al., (2013) [13]	55	blood nipple discharge+mass	right breast	7cm	IPC with invasive component	radical mastectomy+ axillary lymph node dissection	NA	6	NED
12	Al Salloom (2015) [14]	53	mass	left breast	7x6x5 cm	non-invasive IPC	radical mastectomy+ axillary lymph node dissection	No	24	NED

*IPC: Intracystic Papillary Carcinoma; DOD: Died of Disease; NED: No Evidence of Disease.*

indicated in pure IPC or IPC with DCIS [1, 4]. It is also important to be pointed out that the role of sentinel lymph node biopsy has not been evaluated in IPC but it may be an excellent alternative to axillary dissection in these patients [14].

Also, there is currently no clear indication for adjuvant hormonal treatment in patients with oestrogen-receptor positive. For some authors, it does not seem to have impacted the prognosis [1]. Fay-anju et al., found that patients with IPC in association with DCIS or invasive disease were more likely to receive radiotherapy and tamoxifen [15].

The role of radiotherapy remains undefined [14, 1]. The place of radiotherapy in IPC remains undefined and many authors recommend adjuvant radiotherapy for IPC associated with invasion and/or DCIS in those undergoing breast conserving surgery but not after mastectomy [15, 16].

The prognosis of pure IPC is excellent with a relative survival rate of 100% at 10 years and a disease-free survival rate of 91%,

whatever the chosen treatment, long-term monitoring is necessary [2, 10, 16].

## Conclusion

This is the first report from Morocco of a male having intracystic papillary breast carcinoma. This tumor is a very rare in man and this makes it difficult to derive standard treatment from prospective trials. The role of pathologist is primordial for eliminate an IPC with invasion. The surgery is the mainstay of treatment, and adjuvant therapy (chemotherapy, radiotherapy) is indicated if IPC is associated to DCIS or invasive carcinoma. Despite its good prognosis and whatever the chosen treatment, long-term monitoring is necessary.

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