Metastatic lymph nodes in Intracystic papillary carcinoma of the breast: a case report and literature review

Glaoui. M, MD[1], Boulaamane. L, MD[1], Mahfoud. T, MD[1], Benzekri. A, MD[2], Jalil. A, MD[3], Benjaafer. N, MD[4], Errihani. H, MD[1]

(1) Medical oncology unit, National Oncology Institute, Rabat
(2) Anatomopathological unit, United-Nation Center, Rabat
(3) Surgery unit, National Oncology Institute, Rabat
(4) Radiotherapy unit, INO, Rabat

✉ *Corresponding Author: Dr Meryem Glaoui, MD*
National Institute of Oncology, Medical Oncology Department, Rabat
E-mail: m_glaoui@hotmail.com

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Abstract

Intracystic papillary carcinoma (IPC) of the breast is a rare malignant tumor, only a handful of cases with lymph node metastases were reported in the literature. The treatment guidelines for IPC are not well defined and the importance of lymph nodes dissection was not established.

An exhaustive integration of clinical, radiological and histopathological features is important to exclude misdiagnosis and inappropriate treatment.
We reported a case of a 57-year-old woman with an intracystic papillary carcinoma treated by segmental mastectomy and lymph node dissection. Adjuvant treatment consisting of chemotherapy followed by radiotherapy of the breast and endocrine therapy was indicated.

Introduction

Intracystic papillary carcinoma of the breast is a rare entity, known to occur in post menopausal women and is characterized by slow growth and has a better prognosis than ductal carcinoma (1). The presence of metastases in association with intracystic papillary carcinoma has rarely been reported. Lefkovvitz et al (2) described the presence of axillary lymph nodes or distant metastases in 3 out of 77 in situ papillary lesions.

In the current report, we discuss a case of a 57-year-old woman with an intracystic papillary carcinoma, no evidence of stromal invasion was shown and a complete absence of a myoepithelial cell layer around the periphery of the lesions was noted.

Surgical management consisted of a segmental mastectomy associated with an axillary lymph nodes dissection followed by systemic chemotherapy, local radiation and endorcrine therapy by an aromatase inhibitor.

Case presentation

A 57-year-old woman was admitted to our hospital with a 6 week history of swelling in her left breast. Physical examination found a 5.5cm non tender abnormality in the lower internal quadrant of the left breast with erythema, skin thickening and mobile left axillary lymph nodes.

Findings on the mammography showed a 5cm well-circumscribed bilobed mass in the left breast (Fig. 1). On the sonography, the mass had internal echoes and gently lobulated borders with some through-transmission associated with an axillary node that measured 3cm in greatest dimension and was homogenously hypoechoic.

Aspiration of the lesion revealed uniformly blood-stained fluid, and no evidence of malignancy was seen in the cytology analysis of the aspirate. A sonographically guided core needle biopsy suggested a fibrocystic mastopathy benign proliferative.

In view of the clinico-radiological and histolopathology discordance, the patient underwent left segmental mastectomy.

Histological analysis revealed a lesion 1.8cm × 1.5cm in size. The lesion comprised of papillary and solid proliferation of atypical cells within a large cystic space with a thick fibrous capsule (Fig 2). There was no stromal or fibrovascular invasion, and complete absence of myoepithelial cell layers around the periphery of the lesions was noted (fig 3).

The final histopathologic diagnosis was intracystic papillary carcinoma. Axillary dissection revealed one of 14 axillary lymph nodes with metastatic carcinoma. Immunohistochemistry showed high estrogen and progesterone receptor positivity with no overexpression of her-2/neu.

An adjuvant chemotherapy consisting of four cycles of AC60 regimen (doxorubicin: 60mg/m2, cyclophosphamide: 600 mg/m2) followed by external radiotherapy and letrozole was performed.

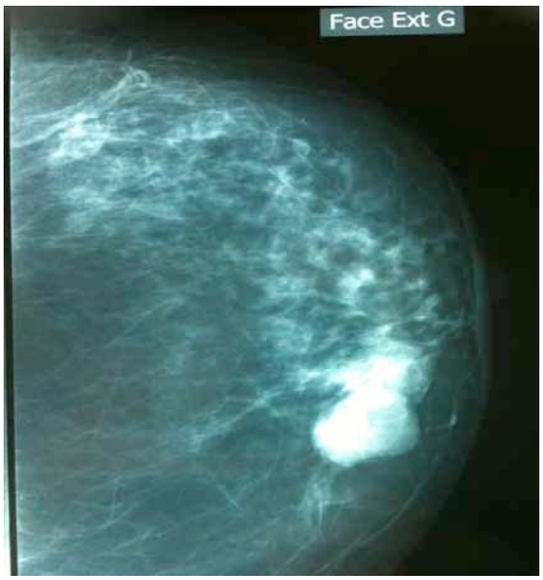

Fig 1: Mediolateral oblique mammogram of right breast shows bilobed mass

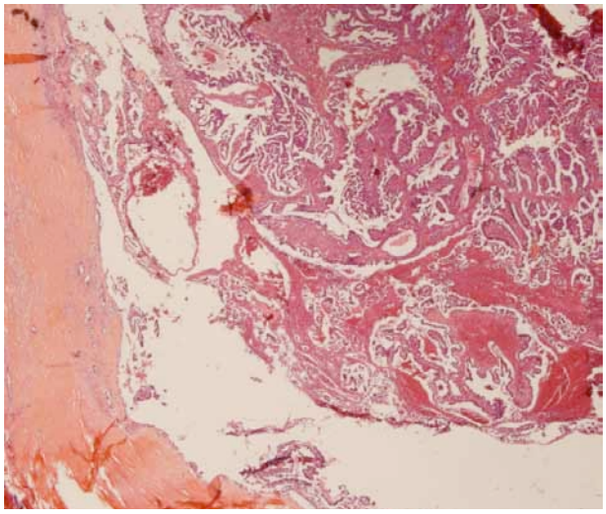

Fig 2: Low-power photomicrograph of a histopathologic specimen showing fibrotic cyst wall, hemmoragea and papillary proliferation

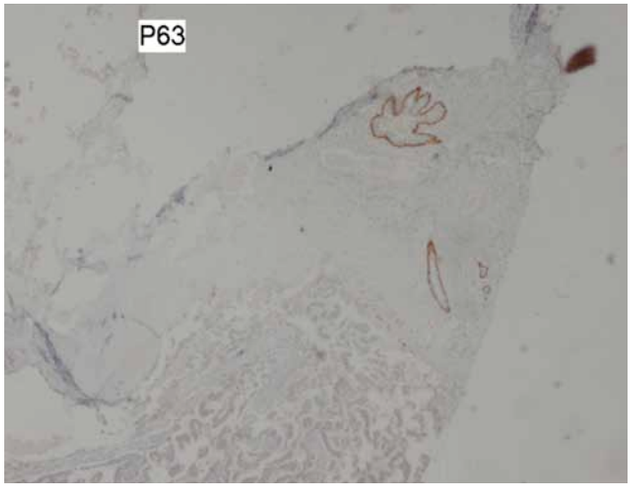

Fig 3: Intracystic papillary carcinoma. Immunohistochemistry for myoepithelial cells (p63) is negative at the periphery of the lesion

Discussion

IPC is a rare malignancy of the breast. It typically occurs in older women and has an excellent prognosis. The reported 10-year survival rate for IPC is 100%, the recurrence-free survival rate is 96% and 77% at 2 and 10 years respectively (3). The terminology applied to describe papillary breast lesions in the literature is relatively confusing. The traditional term "intracystic papillary carcinoma" generally refers to a localized lesion, *in situ* to a cystically dilated duct. Given the often marked stromal response surrounding these lesions, the distinction between *in situ* and invasive papillary carcinoma can be very difficult to make. Therefore, IPC has been divided into three subgroups, which seems to correlate with the prognosis: IPC alone, IPC plus DCIS, and IPC with invasion. In this manner, the term "papillary DCIS" would refer to a more diffuse process that involves multiple ducts as opposed to a localized lesion (4).

Recently, Hill et al., using myoepithelial cell staining, suggested a spectrum of progression from *in situ* disease to invasive disease, signifying that what appears to be DCIS on histology may potentially cause distant metastases (5).

The lack of an intact basal myoepithel cell layer can be identified by calponin, smooth-muscle myosin heavy chain (SMM-HC) cytoplasmic stains, and by p63 nuclear stains. This "gold standard" method has a relatively high sensitivity and denotes the invasiveness of the tumor cells in malignant papillary breast lesions (5).

Clinical appearance is commonly benign; women with intracystic papillary carcinoma may have no symptoms, a palpable mass, or may present with bloody nipple discharge. On the mammography, intracystic papillary carcinoma is often seen as a round or oval circumscribed mass, most frequently in the retroareolar region. Sonography usually reveals a cystic mass, with or without septations, with solid papillary masses projecting into the cyst lumen (6, 7). Although most papillary carcinomas are cystic masses, some may be mixed in composition with predominantly solid components .Sonography may be useful for showing wall thickening and adjacent anechoic and hyperechoic areas that may represent hemorrhage resulting from ruptured capillaries within the cyst wall or hemorrhagic infarction of the tumor cells (1). Fine-needle aspiration cytology and core biopsy are usually performed. However, the false negative results with cytology are relatively frequent (8). Therefore, excisional biopsy is suggested when papillary lesions are suspected.

There is still no clear consensus regarding optimal treatment of IPC.
Several studies have shown that prognosis is excellent and recurrence rates are low, regardless of the intervention. The surgical management of IPC appears to parallel that of invasive ductal carcinoma, with a lower rate of breast conservation in older data. However, more recent studies show a tendency towards breast conservation, with no significant increase in tumor recurrence or cancer related deaths (3, 9, 10).

Axillary lymph node dissection or sentinal node biopsy is often performed in patients in whom invasion is likely (11, 12, 13, 14). However, the current case, as well as several other studies, does demonstrate axillary lymph node involvement even in cases without reported adjacent invasive ductal carcinoma (15, 16).
In addition to surgical excision, several studies have reported the use of adjuvant radiation and/or endocrine therapy in the management of IPC (3, 9, 10). Recently, Fayanju et al. reviewed the usual adjuvant treatment applied for IPC and found that patients with DCIS or microinvasive disease in association with IPC were more likely to receive radiotherapy and tamoxifen (9).

Given the controversial and evolving understanding of the invasiveness of the tumor and the presence of Axillary lymph node metastases associated with IPC in our case, a multimodal management including surgery followed by adjuvant chemotherapy, local radiotherapy and endocrine therapy were performed.

Conclusion

Axillary lymph nodes metastases are uncommon in pure intracystic papillary carcinoma with no guidelines regarding their management.

Surgical excision is currently the default standard. Given the evidence provided by the current case and others previously reported, it may be prudent to perform Sentinel lymph node biopsies to evaluate axillary involvement even in pure Intracystic papillary carcinoma. However, the roles of local radiation and systemic endocrine therapy in the treatment of IPC with lymph nodes metastasis are still controversial.

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