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Case Report

A recurrent case report of encapsulated papillary carcinoma in the male breast

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Abstract: Breast papillary lesions are a group of heterogeneous lesions and present a major difficulty both in clinical and pathological diagnosis. This disease often shows nipple discharge or a lump. Immunohistochemical staining can be used to distinguish between benign and malignant breast papillary lesions. Here we report a recurrent case of encapsulated papillary carcinoma (EPC), a rare neoplasm of the breast, in a 61-year-old man at our institution. EPC is a rare clinical type of breast cancer, accounting for 0.5% to 1% of cases and predominantly occurring in post-menopausal women. The concept of EPC has not been unified in various international documents. Only a few cases have been reported in males.

Keywords: Breast cancer, papillary lesions, encapsulated papillary carcinoma, male

Introduction

Breast papillary lesions are a group of heterogeneous lesions which present a major difficulty for pathological diagnosis. Histologically, they are characterized by different scales of the finger-like protrusions or leaf-like structure and the presence of arborescent fibrovascular cores covered with myoepithelial cell. Normal breast gland tissue does not show papillary morphology, and related studies on the mechanism of papillary appearance are rare [1]. The absence of myoepithelial cells is an important feature to distinguish benign and malignant papillary lesions. EPC is a rare clinical type of breast cancer, accounting for 0.5% to 1% of breast cancer and predominantly occurring in post-menopausal women. Only a few cases in males have been reported [2]. The tumor often presents as a cystic lesion on ultrasound [3]. This report is of a recurrent case of EPC in a 61-year old man at our institution. The terminology used in reports of EPC varies confusingly in the international literature.

Case presentation

A 61 year-old man presented with complaints of a 2-year history of swelling and nipple discharge in the left mammary region in July 2013. He had a left-sided tumor biopsy in a private hospital that suggested hematoma. He did not have any significant medical history or family history of breast cancer. Six months after the biopsy, the patient developed a mass that progressed in size on his anterior chest wall. Physical examination revealed a 3.5 × 2.0 cm well-defined and freely mobile with respect to the underlying muscular plane mass, situated 2 cm away from the left areola at the 2 o'clock position. Additionally, there was no regional lymph node involvement. Routine laboratory investigations were within normal limits. A core needle biopsy of the breast performed during the present admission showed evidence for intra-ductal papillary carcinoma. Therefore, the tumor was completely excised with the overlaying skin and preserving the nipple. The tumor was well-circumscribed and measured approximately 3.0 × 2.0 cm. The cut surface of the mass was reddish-yellow with blood clots. A microscopic examination indicated that: a small number of free ductal epithelium showed papillary hyperplasia with mild dysplasia. Immunohistochemical examination revealed a lack of myoepithelial cells in the papillary fronds and cyst wall. Furthermore, the mass showed a

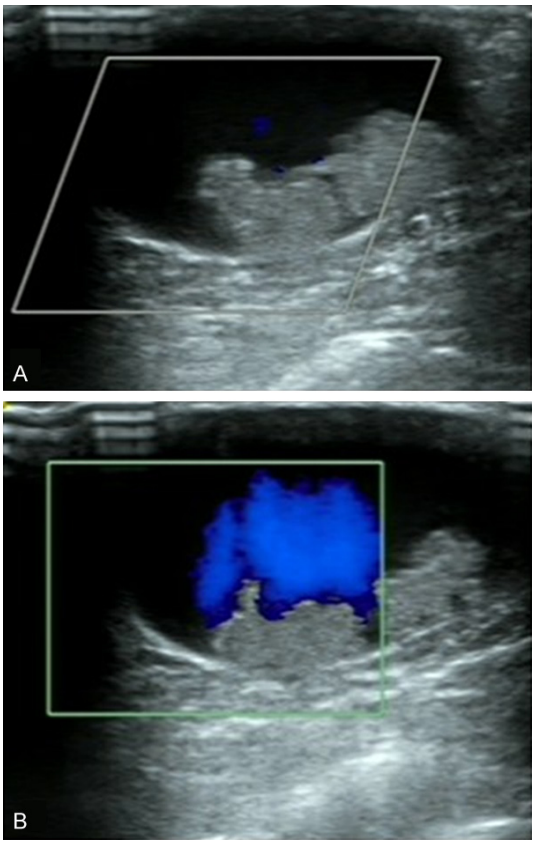

Figure 1. A and B. Ultrasound scan shows an obvious papillary lesion.

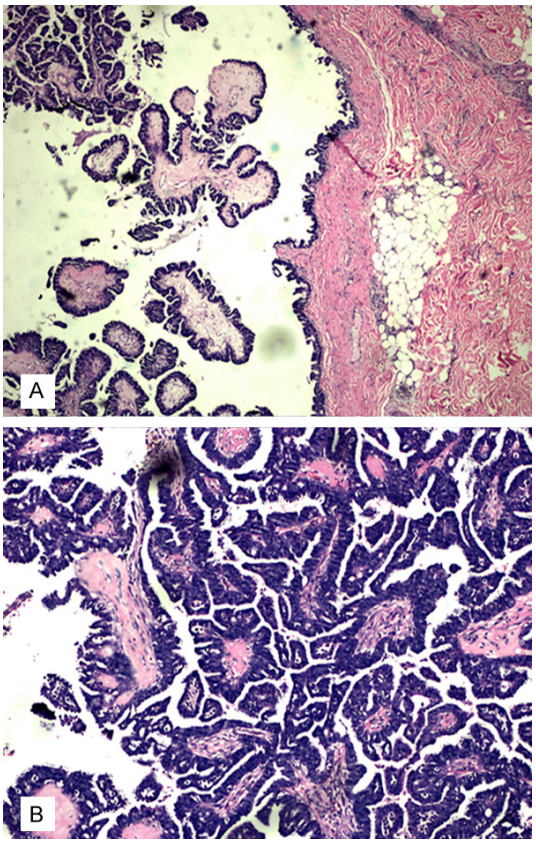

Figure 2. Pathological diagnosis of the resected specimen: cells in the papillary growth with a thin stem of fibro-vasculature as the axis. Hematoxylin & Eosin stain, 200 ×.

negative reaction with p63 and ck5/6, but was positive with 34βE12. Based on the morphology and immunohistochemical results, a diagnosis of intraductal papillary carcinoma was made.

The patient was followed for 3 years, but in July 2016 the patient developed a left-sided tumor again with swelling and pain. Clinical examination revealed a 3.5 × 3.0 cm palpable lump on the chest wall of the left breast. Ultrasonography displayed a mixed tumor (**Figure 1**). Due the history of malignancy and the possibility of a tumor, a excision was performed. The patient underwent a modified radical mastectomy and sentinel lymph node biopsy. Frozen pathology revealed: a tendency of intraductal papillary carcinoma. Lymph node metastasis was not seen in SLNB (0/3). Postoperative pathology indicated: EPC without vascular tumor thrombus and nerve invasion (**Figure 2**). Immunohistochemical staining yielded the following results: negativity for AR, p63, CD1, Calponin, CK5/6 and positivity for P53 (+), EGFR (++), HER-2 (1+), ER (+) 80%, PR (+) 90%, and Ki-67 index 20%. During the 14-month follow-up, no evidence of recurrence or metastasis was observed.

Discussion

The male patient was diagnosed as intraductal papillary carcinoma (IPC) in 2013. The pathological diagnosis of recurrence in 2016 was that of encapsulated papillary carcinoma (EPC), both were malignant papillary lesions. IPC is a malignant, non-invasive, neoplastic, fibrotic papillary hyperplasia with complete or near-complete (>90%) absence of myoepithelial cell [4]. The tumor is also known as papillary ductal carcinoma *in situ*, a non-invasive papillary carcinoma. There was a layer of myoepithelial cells around the catheter, including varying degrees of compression. The terminology used in reports of EPC varies confusingly in the international literature. In 2012, the International Agency for Research on Cancer (IARC) [5] regarded EPC as a variant of papillary carcino-

ma, as synonymous with intraductal papillary carcinoma, non-specific intraductal papillary carcinoma and cystic papillary carcinoma. However, no consensus has been reached on the definition and diagnostic criteria of EPC. Currently, EPC is considered to be a low grade infiltrative growth of invasive carcinoma. Wynveen [6] defined it as fibrous envelope surrounded by the tumor with absence of myoepithelial cell both within the papillae as well as around the periphery of the tumor. There are no myoepithelial cells in the nipple of EPC, however, the myoepithelial cells surrounding the lumen of the IPC. This suggests that EPC may not be carcinoma *in situ*, but an invasive breast carcinoma. Immunohistochemical stains, such as p63, actin, calponin, can promote the identification of myoepithelial cells, therefore, they are useful in distinguishing EPC from other breast papillary lesions [7]. Some scholars believe that EPC is a ductal carcinoma *in situ* or a subtype of IPC because of the clear boundaries with surrounding gland tissue and the good prognosis [8, 9]. However, Collins argued that EPC is evolved from IPC [10] and it is considered to be a transition from *in situ* to invasive carcinoma. Due to the low incidence of EPC, and no clear diagnostic criteria, there is currently no uniform treatment. In general, surgery is still the main treatment. EPC has a good prognosis. The treatment is correlated with pathological results. Relevant studies have shown that a surgical approach has nothing to do with the prognosis and recurrence of EPC. Therefore, local excision is the right choice, both to remove the lesion also avoid over-diagnosis and treatments. The axillary lymph node metastases of EPC is almost never reported. To avoid axillary lymph node dissection, Akladios [11] thought sentinel lymph node biopsy is a better approach. Therefore, according to the size of the lesion to select a safe surgical area with the sentinel lymph node biopsy is the most common surgical choice.

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Disclosure of conflict of interest

None.

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