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Case Report

Encapsulated Papillary Carcinoma in an Elderly Male Whose Diagnosis Was Difficult before Surgery: A Case Report

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Breast cancer · Male · Encapsulated papillary carcinoma · Papillary neoplasms

Abstract
Encapsulated papillary carcinoma (EPC) is a relatively rare histologic type of breast cancer. It is sometimes difficult to obtain a definitive diagnosis by needle biopsy, reflecting its characteristics as an intracystic lesion. Herein, we report a case of EPC in an elderly male that was difficult to diagnose before surgery. A 70-year-old man visited our hospital after a mass just under his right nipple that gradually increased in size. Ultrasonography revealed a well-defined intracystic 50 mm-sized tumor and a papillary-shaped solid component arising from the cyst wall. Cytology revealed small clusters of atypical cells suggestive of malignancy, but we did not reach a definitive diagnosis with subsequent vacuum-assisted needle biopsy due to the small amount of specimen. Given the imaging findings strongly suggested a malignant tumor, a mastectomy was performed. Histologically, there was a thick fibrous capsule and mildly atypical cells showed papillary growth, and we diagnosed the case as EPC (pTisNXM0). Sometimes, EPC is difficult to discriminate from intracystic papilloma before surgery, but clinicians should always keep in mind that this histological type exists with a certain frequency in male patients.

Introduction

Encapsulated papillary carcinoma (EPC) is a newly proposed histologic type in the WHO classification (4th Edition, 2012), replacing the so-called intracystic papillary carcinoma, and accounts for less than 0.5–1% of all breast cancers [1]. EPC is classified as papillary neoplasms in

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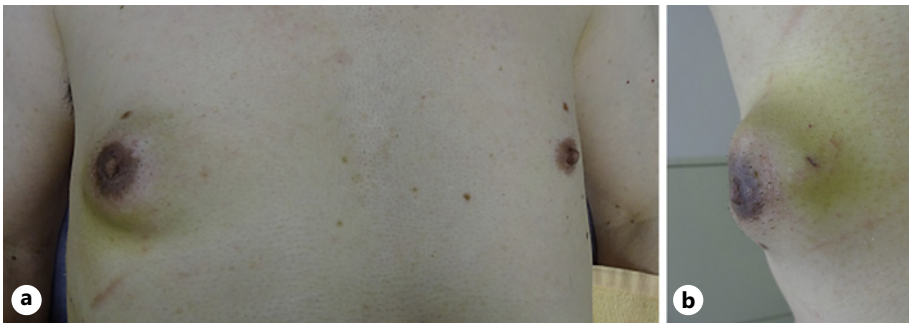

Fig. 1. Physical findings: front view (**a**) and lateral view (**b**). A mass with elasticity and hardness was observed just below the right nipple. Subcutaneous hemorrhage due to needle biopsy can be seen.

the WHO classification 5th Edition (2019) and is defined as follows: a carcinoma characterized by fine fibrovascular stalks covered by low/intermediate grade neoplastic epithelial cells, typically present within a cystic space and surrounded by a fibrous capsule, but usually no myoepithelial cells at the periphery of the lesion [2]. Because EPC has a very favorable prognosis, it is classified as ductal carcinoma in situ to avoid unnecessary systemic adjuvant therapy.

Typical findings on ultrasonography are a hypoechoic, anechoic, or mixed appearance reflecting solid or cystic components. Possible internal vascularity can be seen with Doppler imaging. Because the blood-rich solid component appears along the cystic wall, sometimes it is difficult to differentiate EPC from intracystic papilloma (ICP) [3].

Herein, we report a case of EPC in an elderly male that was difficult to diagnose before surgery. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see https://doi.org/10.1159/000533382).

Case Presentation

A 70-year-old man became aware of a mass just under his right nipple about 1 year ago, and as the mass had gradually increased in size, he visited our hospital. At the time, a golf ball-sized mass with elasticity and hardness was palpated. There were no abnormal findings on the skin or nipple, and no axillary lymph nodes were palpable (Fig. 1). No abnormal nipple discharge was observed.

Ultrasonography revealed a well-defined intracystic tumor, 50 mm in diameter, just below the right nipple. The solid component of the tumor arose from the cyst wall in a broadly basal fashion, with a papillary shape and abundant blood flow. The cyst contained a fluid level formation, suggesting retention of blood (Fig. 2). Magnetic resonance imaging showed a multifocal intracystic mass with a rapid-plateau pattern after contrast enhancement. Computed tomography scan showed no signs of invasion to the latissimus dorsi muscle, lymph node metastasis, or distant metastasis. There were no specific findings in his medical records or family history. Blood tests were normal, including tumor markers.

Fine needle aspiration revealed that the fluid component within the cyst was hematogenous and contained small clusters of atypical cells, and a malignancy was suspected. Subsequent vacuum-assisted biopsy showed ductal epithelium with mild nuclear atypia, but the amount of specimen was too small to form a definitive diagnosis. Considering the image findings strongly suggested a malignant tumor, a surgical procedure (mastectomy) was performed after consultation with the patient.

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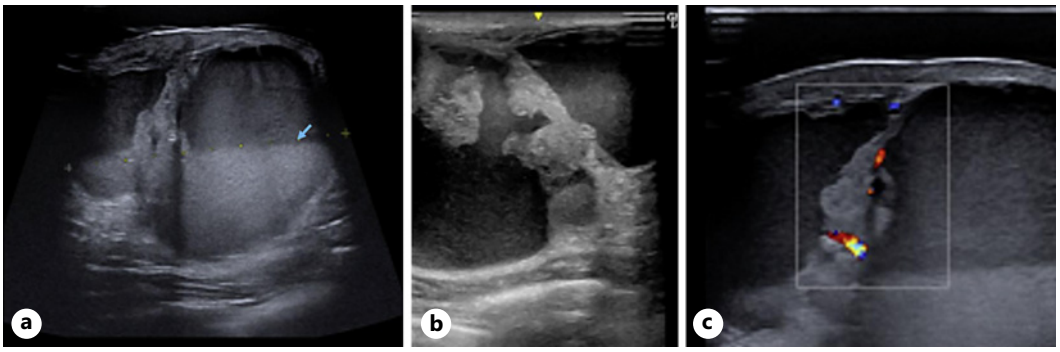

Fig. 2. Findings from ultrasonography: **a** a well-defined intracystic tumor formed a level of liquid (blue arrow), and blood collection was suspected. **b** The papillary lesions arose from the cyst wall. **c** By Doppler ultrasound, blood flow was observed in solid components.

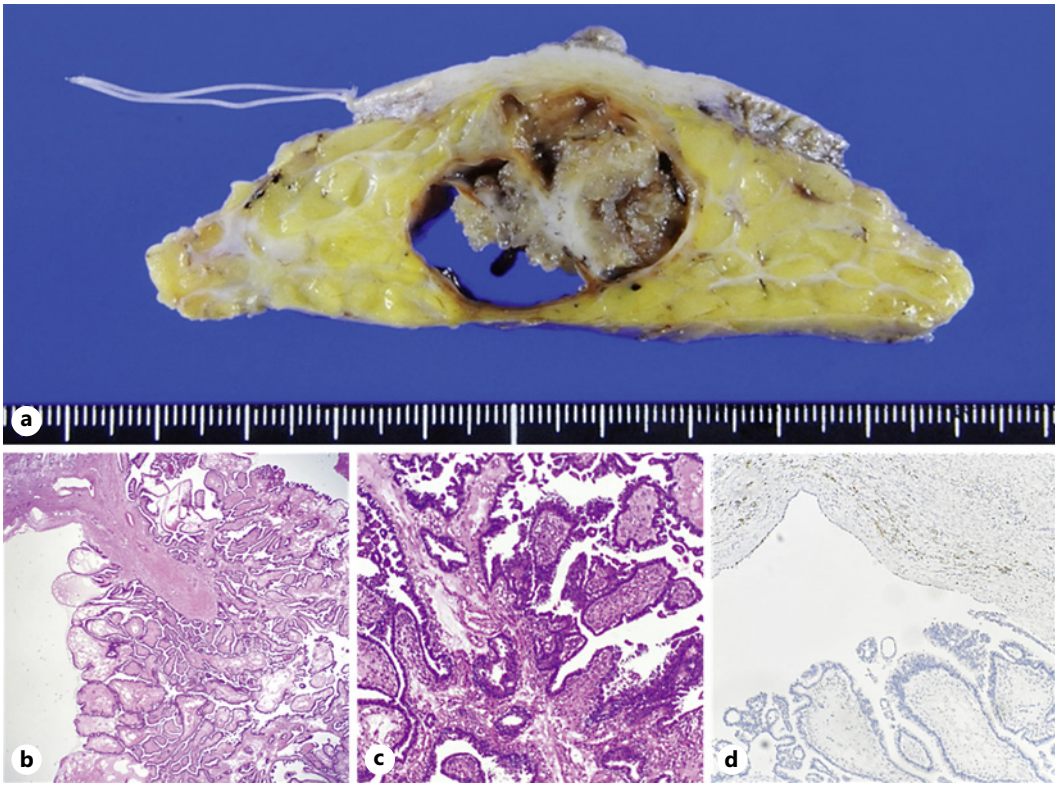

Fig. 3. Pathological findings: **a** the cut surface of the tumor. The papillary lesions arose from the cyst wall. **b**, **c** Hematoxylin and eosin staining. Mildly atypical cells showed papillary growth with fibrous stalk. **d** Immunohistochemistry for p63, showing a lack of staining in cancer cells.

Gross examination reveals a cystic structure 35 mm in diameter just below the nipple and an internal papillary lesion (Fig. 3). Histologically, there was a thick fibrous capsule, and mildly atypical cells showed papillary growth. Myoepithelial cells were sometimes absent at the peripheral borders of the compressively proliferating tumor (Fig. 3). Tumor cells were negative for p63 and CK5/6. Taken together, we arrived at a diagnosis of EPC, Ly0, V0, pTisNXM0 (ER: positive, PgR: positive, HER2: 1+).

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Postoperatively, the patient has been well. He was followed up without additional sentinel node biopsy or adjuvant systemic therapy. One year and a half has passed since then, and the patient has not shown any signs of recurrence.

Discussion

In the current case, it was difficult to reach a definitive diagnosis before surgery, although clinical findings of rapid growth and imaging findings suggested a malignancy. Considering the characteristics of EPC, there were some possible reasons why this case was difficult to diagnose: (1) it was difficult to obtain specimens by biopsy as with ICP and (2) the level of atypia of the cancer cells was low.

However, EPC represents around 3–4% of breast cancer cases in males, which is slightly more common than women [4, 5]. Moreover, ICP is, indeed, known to be very rare in males [6, 7]. Therefore, it is important to consider surgical intervention when there is a possibility of EPC in the absence of definitive diagnosis by needle biopsy. As to differential diagnosis from papilloma by immunohistochemistry, EPC is generally diffusely and strongly positive for ER and PR, and negative for high molecular weight cytokeratins, such as CK5/6 and CK14 [2, 8], consistent with what we observed in our case. The absence of myoepithelial cells in EPC is also useful in differentiating from papillary DCIS, as myoepithelial cells present around the ducts in the latter [9].

We report a case of EPC in an elderly male that was difficult to diagnose before surgery. Sometimes, EPC is difficult to discriminate from ICP before surgery, but clinicians should always keep in mind that this histological type exists with a certain frequency in male patients.

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Statement of Ethics

This report complies with the guidelines for human studies and includes evidence that the research was conducted ethically and in accordance with the World Medical Association Declaration of Helsinki. The authors have no ethical conflicts to disclose. Written informed consent was obtained from the patient for publication of this case report and any accompanying images. The study is exempt from Ethics Committee approval because the Ethics Committee of Saiseikai Kawaguchi General Hospital admits case reports without Ethics Committee approval.

Conflict of Interest Statement

The authors declare that they have no conflicts of interest to disclose.

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Author Contributions

M.S., F.M., Y.T., and K.O. treated this patient. H.S. conducted pathological diagnosis and provided histological information. M.S. and Y.H. wrote the manuscript. Y.I. reviewed and edited the manuscript. All authors contributed to discussions and agreed on the final version of the submitted manuscript.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

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