



## Case Report

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# Recurrent Encapsulated Papillary Carcinoma in the Ipsilateral Internal Mammary Lymph Node: a Case Report

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Encapsulated papillary carcinoma (EPC) is an uncommon breast malignancy that is known to be indolent and associated with an excellent prognosis. However, there is a rare possibility of locoregional relapse or metastasis. Here, we present a case of recurrent EPC in the ipsilateral internal mammary lymph node (IMLN) that was detected in the postoperative magnetic resonance imaging with abbreviated protocol (AB-MRI). AB-MRI could facilitate the early detection of recurrent disease in the IMLN and may provide prognostic gain for such patients.

**Keywords:** Breast neoplasms; Carcinoma; Papillary; Recurrence; Magnetic resonance imaging

## INTRODUCTION

Encapsulated papillary carcinoma (EPC) of the breast is a rare variant of papillary carcinoma, accounting for 0.5-1% of all breast cancers (1). This type of breast cancer is encysted within a dilated duct and surrounded by a well-defined, thick, fibrous capsule. EPCs typically lack myoepithelial cells within the papillae and around the periphery of the tumor, but have an indolent clinical course with excellent prognosis (2, 3). According to a study including the largest case series to date, the rates of lymph-node metastasis, locoregional recurrence, and distant metastasis of EPC were 2.6%, 6.2%, and < 1%, respectively (2). Another recent study reported five cases of recurrence and two cases of distant metastasis among 49 EPC cases (3). Most of the cases reported as locoregional recurrences were local recurrence in the breast or recurrence in the axillary lymph node or skeletal muscle of the chest wall, and there have been no reports on the recurrence of EPC in the internal mammary lymph node (IMLN). Breast cancer recurrence in the IMLN is uncommon and its prevalence is reported to be 1.5% on CT and 0.2-1.4% on PET-CT (4).

Given that it provides good diagnoses and acceptable positive predictive values, magnetic resonance imaging (MRI) of the breast is a valuable screening method for patients with a personal history of breast cancer. Although the widespread use of screening MRI has been limited because of its high cost and time requirements, the introduction of abbreviated breast MRI (AB-MRI) significantly reduced both the acquisition and the interpretation times, as well as the costs of the procedure (5). In addition, the diagnoses done by AB-MRI are comparable to those of conventional breast MRI for screening in women with previously treated breast cancer (5). Importantly,

AB-MRI enables thorough evaluation of breast-associated anatomical structures, such as the axilla, chest wall, and internal mammary lymphatic chain, in addition to intra-breast lesions.

Herein, we report a rare case of EPC that recurred in the ipsilateral IMLN and was detected using the postoperative screening AB-MRI.

## CASE REPORT

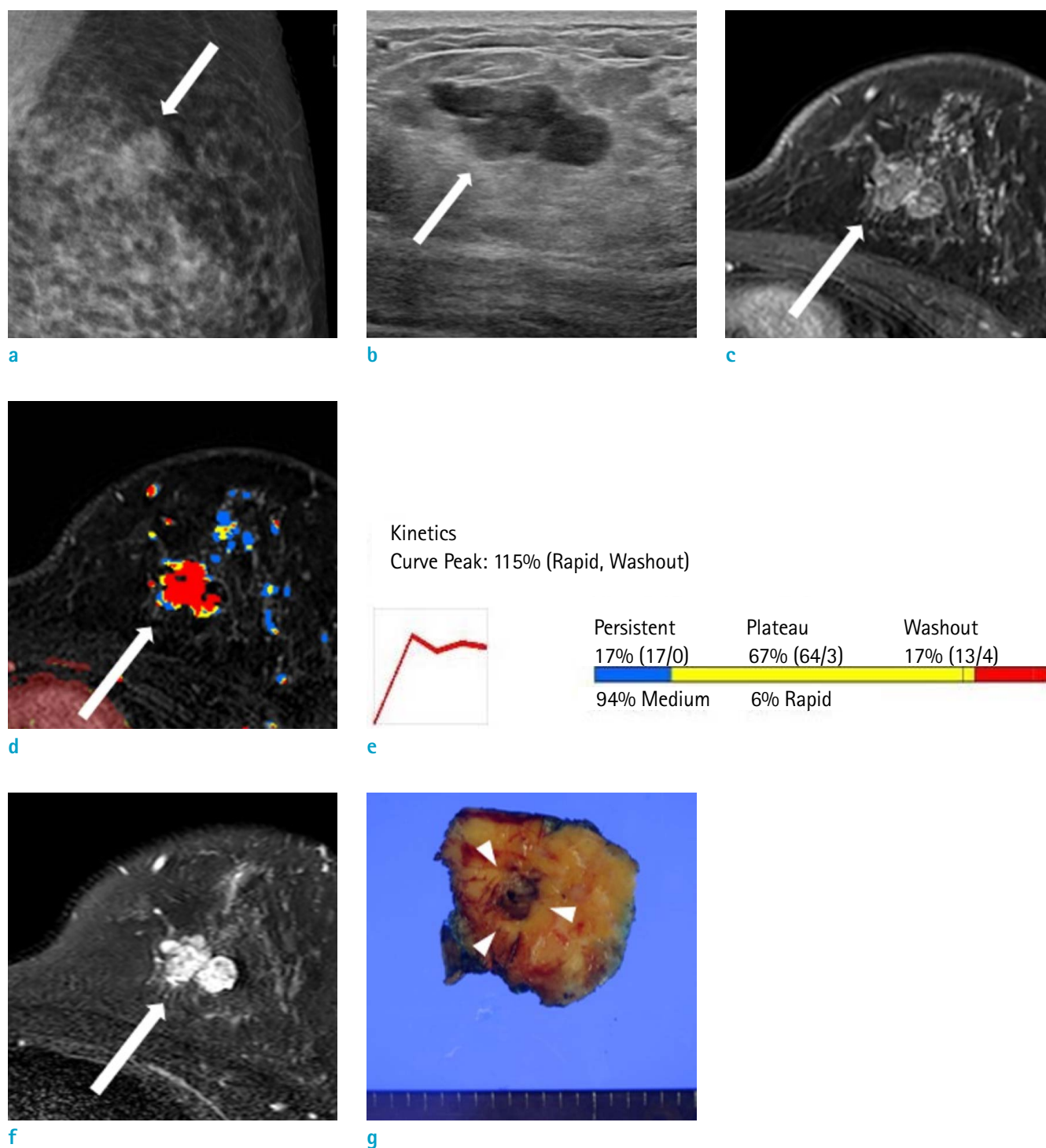
A 38-year-old woman presented with an incidentally detected mass in the left breast on screening breast ultrasound, by which a 31-mm irregular hypoechoic mass was detected with increased vascularity on color Doppler study, which we assessed as Breast Imaging Reporting and Data System (BI-RADS) category 5. We did a core-needle biopsy with a 14-gauge spring-loaded needle under ultrasound guidance. Histopathological examination of the biopsy specimen revealed an atypical papilloma. The pathologist recommended excision and histological reassessment of the lesion, considering the possibility of histologic underestimation of the biopsy specimen. Preoperative breast MRI revealed a 31-mm irregular heterogeneous enhancing mass with a washout component in the upper inner quadrant of the left breast, which was located 14 mm from the chest wall and 86 mm from the nipple (Fig. 1). There was no evidence of regional lymph- node metastasis in either the axillary or the internal mammary lymphatic chains. We did a breast-conserving operation, and diagnosed the tumor as an EPC with low nuclear grade, estrogen receptor (ER) positivity, progesterone receptor (PR) positivity, human epidermal growth factor receptor-2 (HER-2) negativity, and a Ki-67 proliferation index of 10-20%. The results of immunochemical staining for p63 highlighted the lack of myoepithelial cells. The tumor was also associated with an extensive low-grade ductal carcinoma *in situ* (DCIS) component, and there was no evidence of the tumor in the resection margins. A sentinel lymph- node biopsy revealed no metastases in one node. After the operation, the patient received oral tamoxifen and adjuvant radiation therapy of 5840 cGy in 32 daily fractions for 7 weeks.

The patient underwent the first postoperative screening AB-MRI at six months after the operation in a 3T MR scanner (Ingenia, Philips Healthcare, Best, The Netherlands) using a dedicated 16-channel breast coil. The MRI sequences consisted of an axial T2-weighted sequence

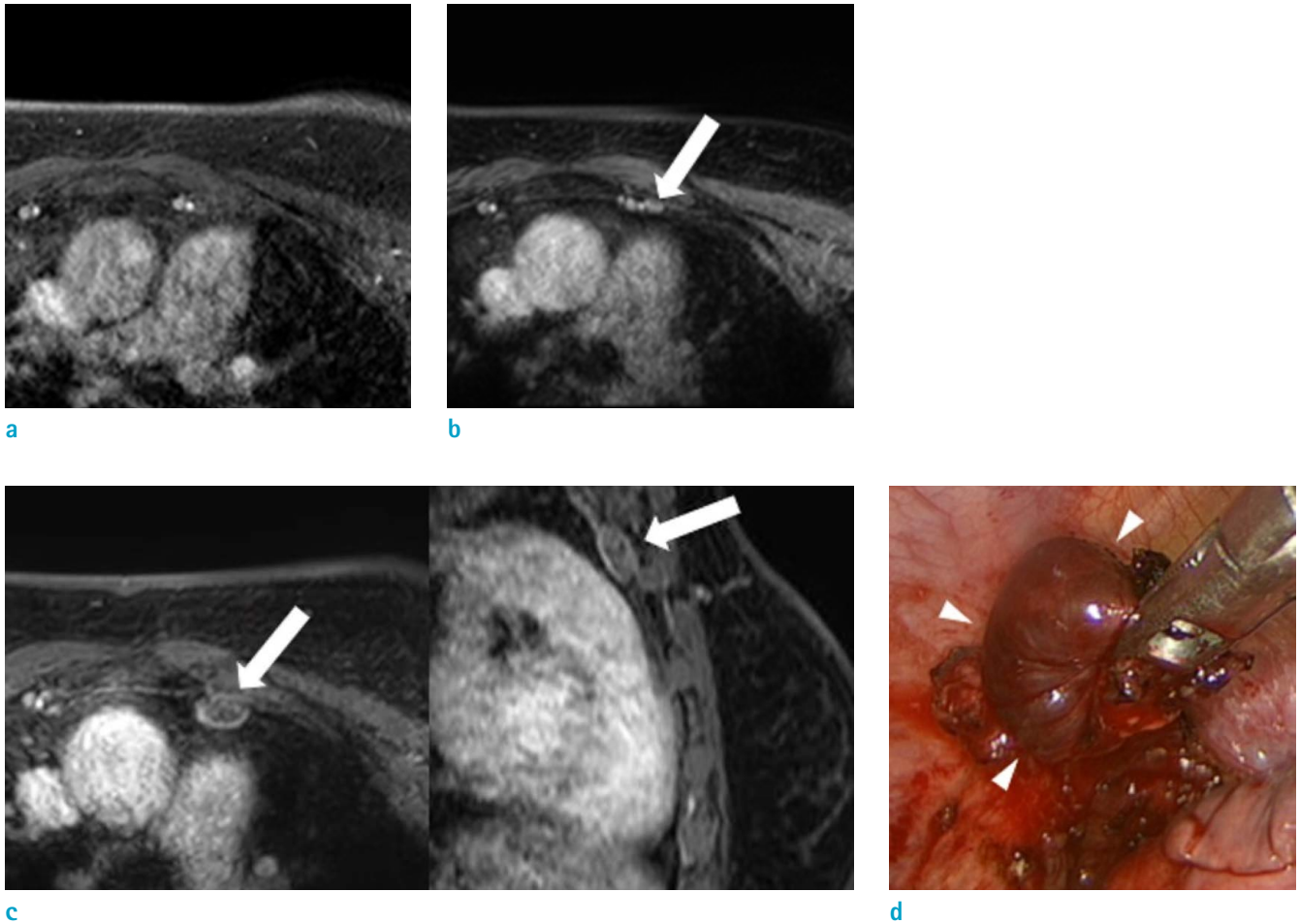
and an axial fat-suppressed dynamic 3D T1-weighted spoiled gradient-echo sequence of one unenhanced and four postcontrast-enhanced acquisitions. We administered an intravenous bolus injection of 0.1 mmol/kg gadoterate meglumine (Dotarem, Guerbet, Aulnay-Sous-Bois, Paris, France) at a flow rate of 2 mL/s using an MR-compatible power injector (Spectris, Medrad, Pittsburgh, PA, USA), followed by a 20 mL saline flush. On MRI, a post-operative change with architectural distortion was revealed in the upper inner quadrant of the left breast, and a post-radiation therapy change with diffuse parenchymal edema was revealed in the left breast. Notably, there was a 6-mm lymph node in the left first internal mammary lymphatic chain that had not been detected on the preoperative breast MRI. We considered this lymph node to be BI-RADS category 3 and recommended a follow-up examination with AB-MRI or breast ultrasound. The patient underwent a second AB-MRI using the same protocol a year after the first MRI; the left first IMLN had further increased to 12 mm (Fig. 2). We did an ultrasound-guided core-needle biopsy of the lymph node with a 14-gauge spring-loaded needle, and diagnosed it as metastatic EPC. We did surgical dissection with video-assisted thoracoscopic surgery, diagnosed it as metastatic EPC with ER positivity, PR positivity, HER-2 negativity, and a Ki-67 proliferation index of 5-10% from the surgical specimen. Immunohistochemical staining of p63, CK5/6, and CD10 again revealed a lack of myoepithelial cells in the papillae and around the periphery of the tumor.

## DISCUSSION

EPC is a rare breast tumor with non-aggressive biological features and excellent prognosis, with a 10-year survival rate of over 95% (6). It typically occurs in elderly women with a median age of 70 years; occurrence in patients younger than 40, as in our case, is uncommon (7). Symptoms of palpable masses or nipple discharges are present in up to 90% of patients (2, 7). EPC usually appears as a round or oval circumscribed mass on mammography and a complex solid and cystic mass on ultrasonography (7). The imaging features of EPC differ on MRI and often overlap with those of benign papillary lesions (7). EPCs usually express ER and PR positivity and HER-2 negativity in most cases, as in our case (7). Previous studies suggested that the diagnosis of EPC with either core-needle biopsy or vacuum-assisted biopsy may be challenging (7). Likewise, the diagnosis in this case was upgraded in surgical pathology, although



**Fig. 1.** A 38-year-old woman with an incidentally detected mass in the left breast. (a) A left mediolateral oblique mammogram shows a 31-mm irregular obscured isodense mass (arrow). (b) Ultrasonography shows a 31-mm irregular hypoechoic mass in the upper inner quadrant of the left breast (arrow). The axial fat-suppressed T1-weighted (c) early-phase dynamic contrast-enhanced magnetic resonance (MR) images of the left breast with (d, e) computer-aided diagnosis analysis show a 31-mm irregular heterogeneous enhancing mass in the upper inner quadrant, with a predominant curve type of washout in delayed phase (arrows). (f) The mass (arrow) shows a high signal intensity in the axial T2-weighted MR image. (g) A photograph of the gross surgical specimen reveals a 12-mm encapsulated papillary carcinoma that appears as an ovoid, pale-brown mass (arrowheads), surrounded by low-grade ductal carcinoma *in situ* on microscopic examination.



**Fig. 2.** (a) The axial fat-suppressed contrast-enhanced T1-weighted image of the preoperative MR image shows no significant lymphadenopathy in the left first internal mammary lymphatic chain. (b) The first post-operative MR image done six months after the operation with the same sequence reveals a newly developed 6-mm lymph node in the left first internal mammary lymphatic chain (arrow). (c) The second post-operative MR image done a year after the first post-operative MR shows that the node enlarged to 12 mm (arrows). (d) Intraoperative view during the video-assisted thoracoscopic surgery demonstrates a 12-mm round and soft lymph node in the left first internal mammary lymphatic chain, which correlates with the MR imaging findings (arrowheads).

ultrasound-guided core-needle biopsy showed atypical papilloma.

In the latest edition of the WHO Classification of Tumors of the Breast, EPCs are subdivided according to the presence of an invasive component (8). Whereas EPC with invasion is staged and managed according to the characteristics of the invasive component, the prognosis of pure EPC is excellent, in that local recurrence or distant metastasis is very rare, and local excision with negative surgical margins appears to be a sufficient treatment that may minimize the unnecessary expenses of cancer therapy (7). However, our case revealed recurrent disease even after surgery with negative resection margins. There have been only a few

case reports of EPC that had no invasive component in the pathological examination of the initial surgical specimen, but later developed recurrence (1).

Although not consolidated, because of the paucity of cases, efforts have been made to reveal predictors related to the recurrence of EPC. A few studies have highlighted that the presence of associated adjacent DCIS, as in our case, or associated invasive carcinoma increases the risk of recurrence and metastasis (7). Another study observed no association between the presence of associated DCIS and a higher recurrence rate (6). Most EPCs, including this one, are low-grade tumors, but high-grade tumors are reported to be more frequently associated with invasion, tumor recurrence,



and distant metastasis (7). Another study reported that larger tumors may be associated with recurrent disease (1). The size of the tumor in our case was close to the median size of EPCs described in previous reports (2). One study also suggested that the older age of patients and higher Ki-67 levels are associated with the invasiveness of EPC, but another study reported contradictory results showing that a younger age of patients is related to EPCs with invasive components (7). Currently, no independent association between these factors and tumor recurrence or metastasis has been confirmed.

IMLN is a secondary lymphatic drainage area in patients with breast cancer. In particular, large tumors with a medial location and axillary lymph-node metastasis have a higher rate of IMLN metastasis (4). The prognostic significance of IMLN metastasis is well documented in the literature (4). Mammography and ultrasonography are the two most commonly used imaging modalities for surveillance of patients treated for breast cancer, but they are limited in the evaluation of extra-axillary nodes, such as the internal mammary lymphatic chain, infra- and supraclavicular region, and interpectoral space. Because isolated regional lymph-node recurrence may be potentially curable, early detection of extra-axillary lymph-node metastases may improve patient prognoses. There have been a few studies of the value of PET-CT in postoperative surveillance of breast cancer, including extra-axillary node evaluation, but it has low sensitivity and specificity for intra-breast lesions and may risk excessive radiation exposure (9). MRI not only is the most sensitive imaging modality for the detection of breast malignancy, but also covers the adjacent anatomical structures in its scan area, facilitating thorough investigation of breast-associated extra-axillary nodes. For the detection of IMLN metastasis with MRI, 90.7% accuracy, 93.3% sensitivity, and 89.3% specificity have been reported based on a size of  $\geq 5$  mm (10).

However, the routine use of conventional MRI for postoperative surveillance of breast cancer is limited because of its long scan time, interpretation time, and high cost. As such, AB-MRI may be cost-effective for early detection of both local and regional recurrence in postoperative surveillance. The AB-MRI protocol used in our case consisted of one unenhanced and four contrast-enhanced acquisitions of fat-suppressed T1-weighted sequences, but a much simpler protocol, such as an unenhanced plus single postcontrast-enhanced image, revealed promising diagnostic performance in the literature

(5).

In conclusion, we report the case of a 38-year-old woman who presented with recurrent EPC in the ipsilateral IMLN on postoperative AB-MRI. IMLN metastasis of EPC is extremely rare, but it adversely affects patient prognosis. Careful postoperative image surveillance with AB-MRI may facilitate early detection and proper management of such cases.

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