Adenoid Cystic Carcinoma of The Breast: A Case Report

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(Received: 23 July, 2025; Revised: 31 October, 2025; Accepted: 4 December, 2025)

Abstract Adenoid cystic carcinoma (ACC) is an extremely rare tumor of the breast, accounting for approximately 0.1% of all cases of mammary malignancies. It is characterized by a slow-growing nature and an excellent prognosis, in contrast to most other forms of breast cancer. Diagnostic mammography and breast ultrasound play essential roles in the early detection and diagnosis of this uncommon tumor. Treatment options include lumpectomy with radiation therapy or mastectomy. Although late recurrence and distant metastasis have been reported in the literature, long-term surveillance remains crucial. This report describes the case of a 58-year-old woman with abnormal findings on screening mammography and ultrasonography, which were pathologically confirmed as adenoid cystic carcinoma of the breast, and includes a relevant literature review. *(Thai Cancer J 2025;45:190–197)*

Keywords: breast adenoid cystic carcinoma, mammogram, ultrasound

Introduction

Adenoid cystic carcinoma (ACC) of the breast is an exceedingly rare subtype of breast malignancy[1]**.** The incidence is less than 0.1% worldwide, including in Thailand. While ACC most commonly originates in the salivary glands, it can also occur in other parts of the body, including the breast[2]**.** Breast ACC typically exhibits an indolent clinical course and is associated with a relatively favorable prognosis when compared to ACCs originating in other locations. Radiological imaging modalities, including mammography and breast ultrasonography, play a pivotal role in the early detection of lesions, thereby facilitating prompt diagnosis and appropriate therapeutic planning. Definitive therapeutic strategies include either breast-conserving surgery (lumpectomy) followed by adjuvant radiotherapy or total mastectomy. Both modalities have demonstrated comparable local control and survival outcomes.[3] The choice of treatment depends on the individual characteristics of each patient. This report describes the imaging features of adenoid cystic carcinoma of the breast, with pathologic and clinical correlation.

Case report

A 58-year-old Thai female presented for a routine health check-up with no clinical symptoms. She had no known underlying medical conditions and denied any family history of cancer. Screening mammography revealed a 1.0 cm oval-shaped, isodense mass with microlobulated margin located in the mid-inner part of the left breast. No suspicious microcalcifications or architectural distortion were detected (Figure 1). Subsequent breast ultrasound demonstrated an irregular shape, parallel-oriented, heterogeneous hypoechoic mass with microlobulated and angular margins, accompanied by both posterior acoustic shadowing and enhancement, located at the 9 o'clock position of the left breast. No internal vascularity was observed on Doppler imaging (Figure 2). No abnormal axillary lymph nodes were identified. Radiologist assessment based on the Breast Imaging Reporting and Data System (BI-RADS) classified the lesion as BI-RADS category 4B, indicating a moderate suspicion of malignancy, with an estimated likelihood of breast cancer between 10–50%. A tissue biopsy was recommended.

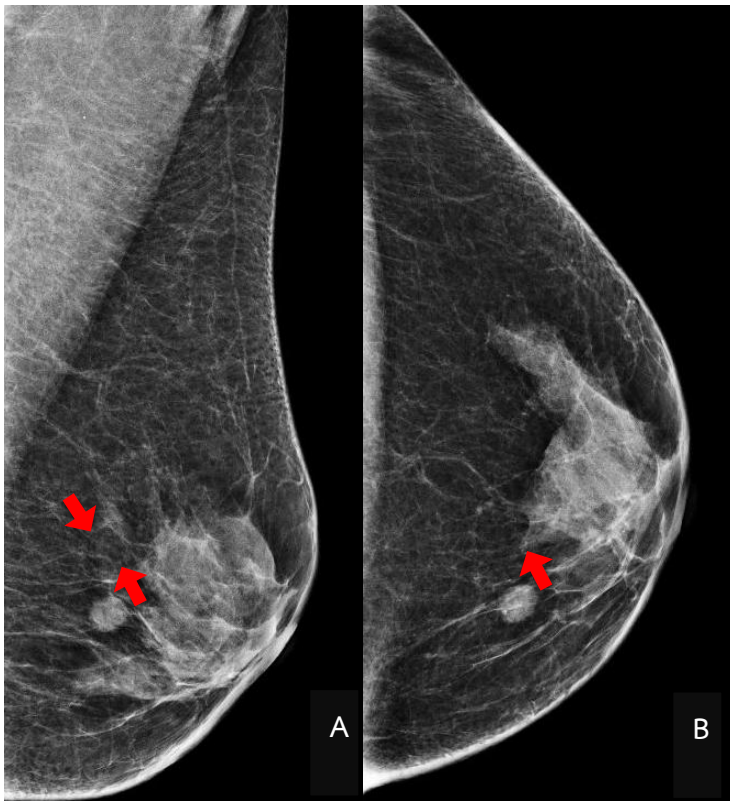

Figure 1 (A and B): MLO (A) and CC (B) mammographic views of the left breast demonstrate an oval-shaped, isodense mass with microlobulated margin (red arrow).

The patient underwent an ultrasound-guided core needle biopsy using a 14-gauge needle, with four core samples obtained from the lesion. Histopathological examination confirmed a diagnosis of breast adenoid cystic carcinoma.

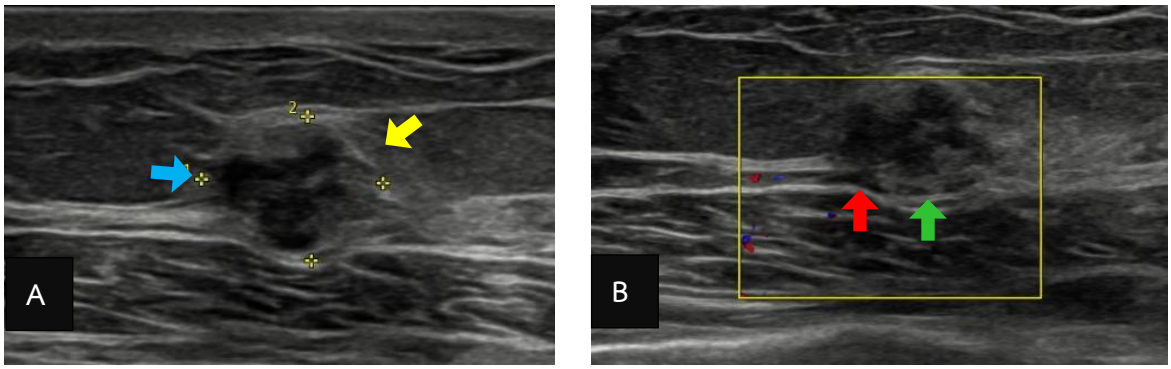

Figure 2 (A and B): Ultrasound images reveal an irregular shape, parallel, heterogeneous hypoechoic mass with microlobulated (blue arrow) and angular borders (yellow arrow). The lesion demonstrates a combined pattern of posterior acoustic shadowing (red arrow) and enhancement (green arrow), with no evidence of increased vascularity.

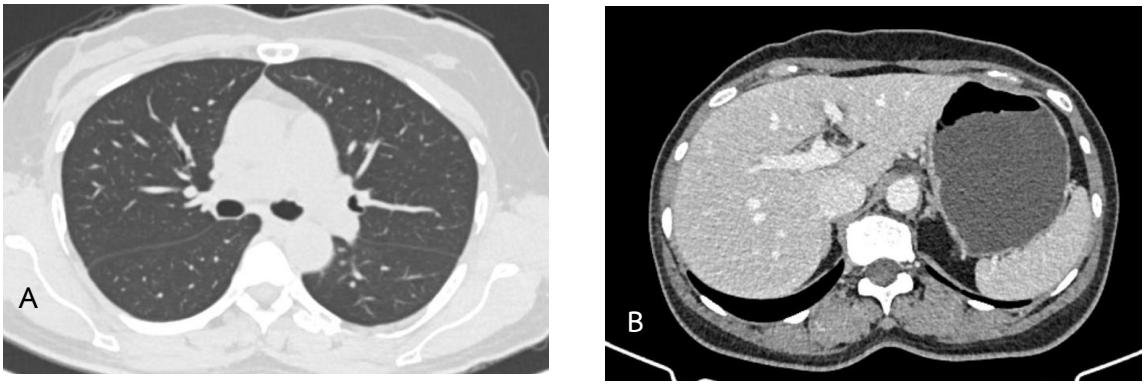

Figure 3 (A and B): CT imaging of the chest and abdomen demonstrates no evidence of regional or distant metastasis.

Further diagnostic work-up included a contrast-enhanced CT scan of the chest and abdomen, which revealed no evidence of regional or distant metastasis (Figure 3). A bone scan was also performed and showed no signs of bone metastasis (Figure 4).

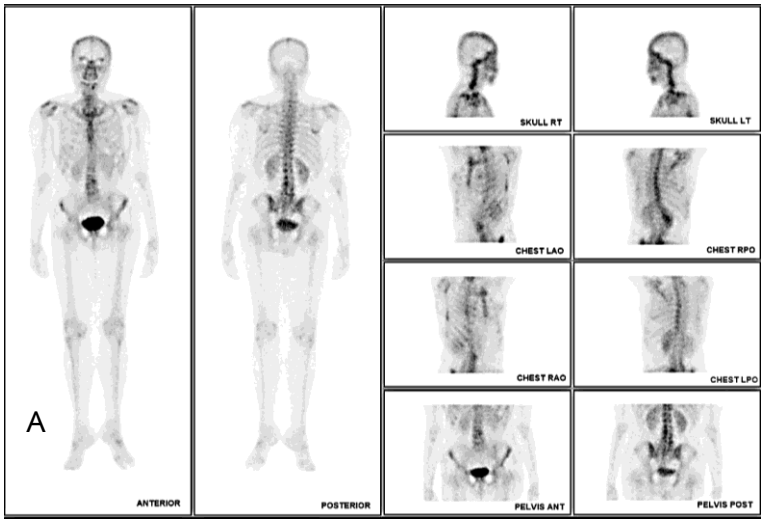

Figure 4 (A): Bone scan shows no abnormal radiotracer uptake indicative of bone metastasis.

Definitive surgical management was performed via breast-conserving surgery with sentinel lymph node biopsy. Surgical pathology of the lumpectomy specimen revealed a 1.2 cm tumor. Histopathologic analysis confirmed the diagnosis of the classic subtype of adenoid cystic carcinoma, with clear surgical margins and absence of lymphovascular invasion or perineural invasion. (Figure 5). All eight left axillary lymph nodes examined were negative for malignancy.

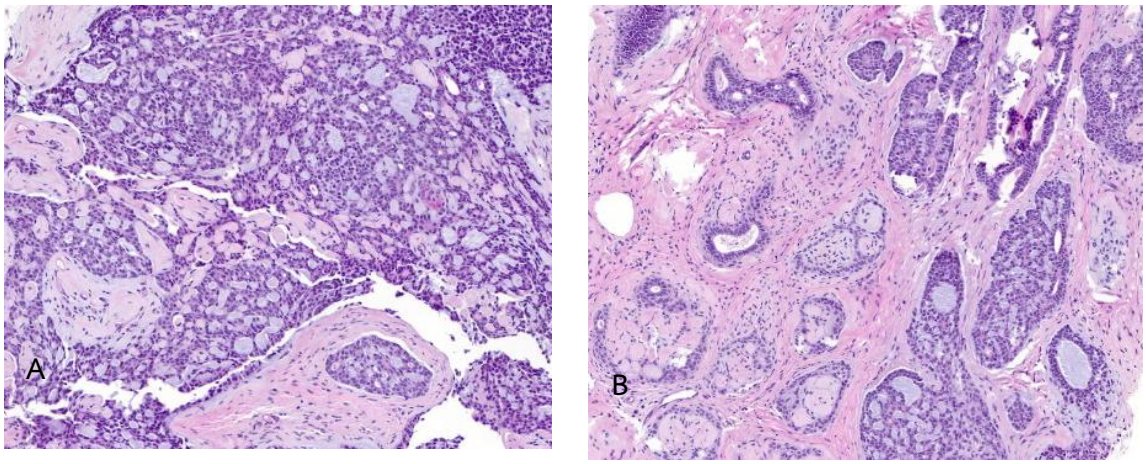

Figure 5 (A and B): Photomicrographs of the histopathologic specimen demonstrated cribriform, tubular and solid growth patterns (Grade 1).

Further immunohistochemical (IHC) analysis characterized the tumor as triple-negative breast cancer, with negative expression for estrogen receptor (ER), progesterone receptor (PR), and human epidermal growth factor receptor 2 (HER2). The Ki-67 proliferation index was approximately 1–10%, consistent with the typically low proliferative activity observed in ACC. Postoperatively, the patient received adjuvant radiotherapy.

Discussions

Adenoid cystic carcinoma of the breast is an exceptionally rare form of breast cancer, accounting for less than 0.1% of all breast cancer cases.[4] Typically, this type of cancer is more commonly found in the salivary glands, but it can also occur in the breast, larynx, lung, or uterus. Breast ACC predominantly affects women, with most cases diagnosed in the fifth to sixth decade of life.[5] Clinically, it may present as a solitary, slowly growing, palpable mass and typically not associated with breast pain or nipple discharge.[6] Histologically, ACC is characterized by a biphasic cellular composition consisting of epithelial and myoepithelial cells, typically arranged in cribriform, tubular, and solid growth patterns.[7] The tumor is histologically graded into three levels according to the proportion of solid components present, with higher-grade lesions correlating with an increased likelihood of local recurrence and distant metastasis.[8]

At present, there is no specific radiologic pattern that distinctly characterizes ACC of the breast. On mammography, lesions may appear with either circumscribed or not circumscribed borders, and typically without suspicious calcifications. On ultrasound, the tumor may have irregular shape with indistinct, angular, or microlobulated margins. Internal vascularity may or may not be present on Doppler study.[9] Contrast-enhanced magnetic resonance imaging (MRI) aids in pre-operative planning and delineating the true extent of the tumor. The MRI appearance of ACC has been described as a round or oval shape, well-circumscribed lesion characterized by rapid initial enhancement without subsequent washout.[10] Various imaging appearances have been described; however, none are pathognomonic for breast ACC. In this case, imaging studies revealed that the margin was not well circumscribed, which made the lesion easy to categorize as BI-RADS category 4 for further biopsy.

To our knowledge, the clinical course of breast ACC is generally indolent. Lymph node metastasis and local invasion are uncommon, and the recurrence rate is low. This contrasts with ACC in the salivary glands, which tends to behave more aggressively, frequently invading adjacent tissues and having a 5-year survival rate of approximately 30%.[11] Breast ACC typically

lacks expression of estrogen receptor (ER), progesterone receptor (PR), and HER2 protein, classifying it as triple-negative breast cancer.[5] While most triple-negative breast cancer has poor prognosis due to resistance to hormone therapy, breast ACC generally has a favorable clinical outcome, with reported 5-year survival rates of 95% and 10-year survival rates of 90%.[12]

In most cases, local disease control can be effectively achieved through breast-conserving surgery followed by radiotherapy or total mastectomy.[13] There are case reports of local recurrence in patients who underwent lumpectomy alone without adjuvant radiotherapy.[13] Given the generally excellent prognosis of breast ACC, there is currently no strong evidence supporting a significant benefit from adjuvant chemotherapy.[13] However, chemotherapy may be considered for high-risk patients, such as those with tumors larger than 3 cm or with axillary lymph node involvement.[5]

Case reports in the radiologic literature have documented local recurrence and late distant metastasis, occurring more than 10 years after initial treatment. The lungs are the most common site of distant spread.[14] Therefore, long-term follow-up involving physical examinations, mammography, and ultrasound imaging at regular intervals is essential.

Although the long-term prognosis is generally favorable, continued surveillance remains necessary. The limitation of this report is that the patient is still undergoing treatment. Consequently, the long-term follow-up data, including local recurrence, metastatic potential, and treatment response, are not yet available. Continued monitoring is required to assess the eventual prognosis.

Conclusion

Adenoid cystic carcinoma of the breast is a relatively rare subtype of breast cancer but is generally associated with a favorable prognosis, characterized by a relatively high survival rate and a low incidence of metastatic potential. The primary treatment modality is local control through surgical excision combined with radiotherapy. Despite its indolent nature, there have been reported cases of local relapse and distant metastasis in the medical literature, emphasizing the importance of regular post-treatment surveillance. Regular follow-up with mammography and breast ultrasound is essential for the early detection of disease recurrence. Although no pathognomonic radiologic features have been definitively established, early and accurate diagnosis remains critical, as it substantially contributes to optimal treatment planning and improved long-term outcomes.

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