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Case report

# Adenoid cystic carcinoma: A case of rare breast cancer ☆

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ABSTRACT

Adenoid cystic carcinoma is a rare form of breast cancer accounting for 0.1%-1.0% of all mammary malignancies. It is characterized by an indolent clinical course and favorable prognosis, contrary to other breast cancers. Diagnostic mammogram and breast ultrasound play a pivotal role in the early detection and diagnosis of breast adenoid cystic carcinoma. Treatment may consist of lumpectomy and radiation therapy vs mastectomy alone. Even though rare, late disease recurrence and metastasis has been reported in the literature thus long-term surveillance is of utmost importance for these patients. We will review the literature and discuss the case of a 52-year-old female who presented with a palpable lump of the right breast, which was pathologically proven to be adenoid cystic carcinoma of the breast.



## Introduction

Adenoid cystic carcinoma is an extremely rare tumor of the breast. It is a rare variant of adenocarcinoma accounting for less than 1.0% of all cases of mammary malignancies [1]. The most common site of adenoid cystic carcinoma occurrence is in the salivary glands. However, multiple other sites of occurrence outside of the salivary glands have also been reported in the literature [1]. Adenoid cystic carcinoma of the breast has an indolent clinical course and excellent prognosis when compared to ACC of the salivary glands [2]. Given the rarity of this tumor, diagnosis remains a challenge. Imaging plays a critical role by early identification of the lesion. Treatment may consist of either lumpectomy with radiotherapy, or mastectomy; with both surgical options associated with equivalent survival [3]. There is no clear evidence on the literature on the efficacy of adjuvant systemic therapy for the treatment of ACC of the breast [3] and it is offered to patients on a case-to-case basis.

## Case report

The patient is a 52-year-old African American female with no personal or family history of breast cancer, who presented to our facility with a palpable non-painful lump in the right

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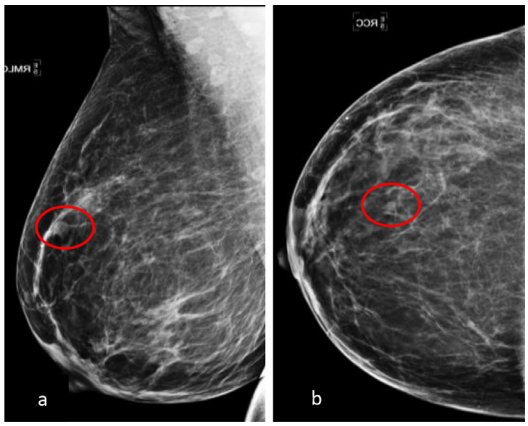


**Fig. 1 – A and B: MLO (A) and CC (B) mammographic views of the right breast demonstrate an obscured mass in the right upper outer breast (circled in red) at the site of palpable abnormality.**

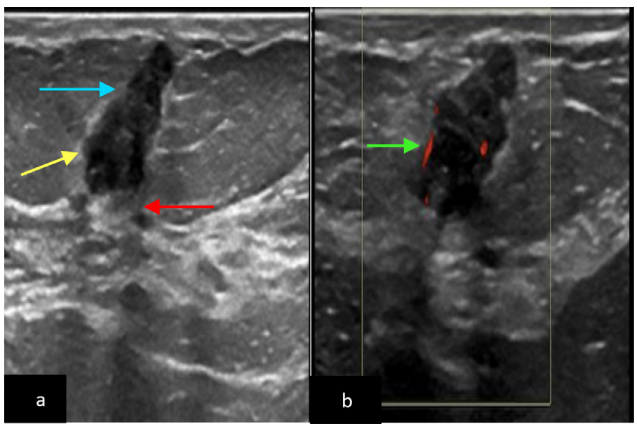


**Fig. 2 – A and B: Ultrasound images demonstrate an irregular, not parallel, solid, hypoechoic mass (blue arrow) with microlobulated borders (yellow arrow), posterior acoustic shadowing (red arrow) and peripheral vascularity (green arrow).**

breast. Diagnostic mammogram images demonstrated an approximate 1.0 cm obscured mass in the upper outer right breast, 10 o'clock position, corresponding to the site of the palpable abnormality (Fig. 1A and B). Ultrasound demonstrated an irregular, not parallel, solid, hypoechoic mass with microlobulated margins, echogenic rim, posterior acoustic shadowing, and peripheral vascularity (Fig. 2A and B). There were no abnormal axillary lymph nodes. Given the suspicious features, the final assessment of the diagnostic mammogram and ultrasound was a BIRADS-4 and recommendations were given for tissue diagnosis with ultrasound-guided core biopsy. Biopsy was performed with five passes with a 12-gauge Celero vaccum-assisted device under ultrasound guidance. Pathology report yielded adenoid cystic carcinoma of the breast. MRI of the breast was not used in surgical planning as the extent of disease was clearly defined by mammogram and ultrasound imaging. The patient underwent needle localization

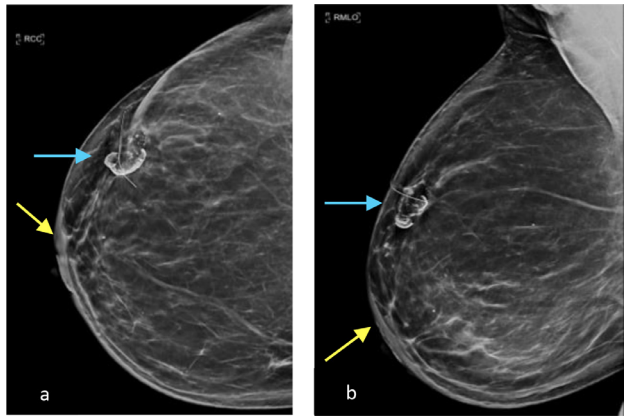


**Fig. 3 – A and B: The follow up screening mammogram, seven years after lumpectomy. CC (A) and MLO (B) mammographic views of the right breast demonstrate post-operative scarring at the lumpectomy site (blue arrow) and post-radiation skin thickening (yellow arrow). There is no mammographic evidence of disease recurrence.**

of the tumor, lumpectomy, and sentinel lymph node sampling of the right axilla. Surgical pathology of the lumpectomy specimen measured 1.5 cm and yielded adenoid cystic carcinoma; solid, basaloid type, grade II, with margins free of tumor. Three right axillary lymph nodes were submitted for examination, all of which were proven to be free of malignancy. The tumor was triple negative for estrogen receptor (ER), progesterone receptor (PR), and human epidermal receptor 2 (HER2). The patient underwent balloon catheter brachytherapy with subsequent whole breast radiation therapy at an outside facility. Yearly follow-up diagnostic mammograms and breast ultrasounds over a period of 7 years has demonstrated stable post-operative and post-radiation changes of the right breast and axilla without disease recurrence (Fig. 3A and B).

## Discussion

Adenoid cystic carcinoma has been recognized as a rare cause of breast cancer accounting for less than 0.1 % of all mammary malignancies [1]. The tumor is commonly found in the salivary glands but may also arise in other tissues such as the breast, nasopharynx, lung, and uterus [1]. Breast adenoid cystic carcinoma occurs predominantly in women during the 5th and 6th decade of life [3]. It usually presents as a palpable lump of the breast, slowly growing, with predilection for the subareolar region and upper outer quadrant [3]. Breast pain associated with the palpable mass has been reported to occur in approximately 14% of cases [9]. It is rarely bilateral and nipple discharge is uncommon [4]. There are two distinct cell types of breast ACC based on immunohistochemical staining, luminal and basaloid types [2]. Growth patterns have been described as cribriform, glandular, trabecular, and solid [2] with solid carrying a higher malignant potential [9]. Grading is based on percentage of solid component; grade I tumors have minimal solid component, Grade II with less than 30% and grade III

is composed of greater than 30% solid component [9]. Higher grade tumors are associated with recurrence and increased risk of metastasis [2].

The literature on the radiographic appearance of adenoid cystic carcinoma of the breast is often nonspecific, with no distinctive radiologic imaging characteristics of ACC. On mammogram, ACC can present as an irregular lesion, or as a circumscribed round mass with margins similar to a benign lesion. On ultrasound, it often presents as an irregular hypoechoic mass with angular, indistinct, or microlobulated margins and with mild peripheral vascularity [5]. Contrast enhanced MRI aids in pre-operative planning and has shown to have a high sensitivity in determining the true extent of the tumor [10]. The MRI appearance of ACC has been described as a round, well-circumscribed lesion with rapid enhancement and no washout [5].

Adenoid cystic carcinoma of the breast has an indolent clinical course. Regional lymph node invasion, distant metastasis and disease recurrence is uncommon [7]. On the contrary, ACC of the salivary glands is an aggressive tumor that often metastasizes with a poor prognosis and an approximate 30% 5-year survival rate [8]. Adenoid cystic carcinoma of the breast has been reported to be ER/PR and Her2 protein negative [6]. Clinically, a triple negative breast tumor is more aggressive and does not respond well to hormonal therapy. However, contrary to other triple negative breast malignancies, the prognosis of ACC is favorable with a 10-year survival rate of 90%-100% [6].

In most cases of adenoid cystic carcinoma of the breast, good local control may be achieved with lumpectomy plus whole breast radiation or simple mastectomy [7]. Local recurrences have been reported in cases where radiotherapy was not administered in conjunction to lumpectomy. Axillary lymph node dissection is usually not performed due to the low incidence of nodal metastasis and may be considered only if there are clinical signs of nodal invasion [3]. Given the favorable prognosis of ACC of the breast, adjuvant systemic chemotherapy has not been proven to provide a clinically significant benefit [7]. However, it may be offered to patients on a case-to-case basis and has been proposed for higher grade lesions, greater than 3 cm and with axillary lymph node metastases [6]. There have been reported cases in the literature with local and distant recurrence as late as 10 years after onset, with lungs as the most common metastatic site [7]. Therefore, long-term surveillance with physical exam, diagnostic mammogram and breast ultrasound is recommended for all patients with ACC of the breast.

## Conclusion

Adenoid cystic carcinoma of the breast is a rare type of breast cancer with an excellent prognosis because of the low incidence of distant metastasis, lymph node invasion, and recurrence. Treatment often consists of local resection in conjunction with radiation. Due to the late onset of local relapse, long-term follow-up after surgery with diagnostic mammogram and ultrasound is of utmost importance for these patients. The rarity of this tumor poses a true diagnostic challenge due to varying imaging manifestations. Although extremely rare, recognition of adenoid cystic carcinoma of the breast is important as early diagnosis ensures excellent prognosis.

## Patient consent

The patient has provided verbal and written consent for case report publication.

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