

## Case Report

# Adenoid cystic carcinoma of the right breast combined with bilateral mammary ductal ectasia: a case report

Xingxing Wang, Yong Hu

Department of Pathology, The NO. 105th Hospital of PLA, Hefei, Anhui, PR of China

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**Abstract:** Adenoid cystic carcinoma (ACC) of the breast is a very rare and indolent tumor with a favorable prognosis. Breast that harbors ACC can also contain other types of lesion, though rare. In this report, we present a case of breast ACC associated with bilateral mammary duct ectasia, which is the first known case.

**Keywords:** Adenoid cystic carcinoma, breast, mammary ductal ectasia, case report

### Introduction

Adenoid cystic carcinoma of the breast is a rare breast cancer variant that usually formed into classic tubular or cribriform architecture by epithelial and myoepithelial cell, however, it characteristically present with a very low axillary lymphnode metastases and a better prognosis than most forms of breast cancer [1]. According to all published medical literature, there are only several reports that reference breast ACC coexistence with other benign breast lesions such as microglandular adenosis, tubular adenosis, adenomyoepithelioma and fibroadenoma [2-5]. Here we present a case of right breast ACC combined with bilateral mammary duct ectasia.

### Case report

A 33-year-old female was admitted to NO. 105th Hospital of PLA, China, complaining of bilateral nipple sero-sanguineous discharge with breast lumps and right breast pain in the past 5 years. The family history was positive for breast carcinoma as her sister suffered from invasive breast carcinoma of no special type 6 months ago. The breast examination revealed a mass in the upper-outer quadrant 2 cm away from the right nipple, with an approximate size of 5 cm×4 cm. The mass was tough and irregular, unclear in boundary and slightly adhesive to surrounding tissues. There was no erythema,

ecchymosis, ulceration or dimpling identified. Both of the nipples were on the same horizontal line without retraction. The superficial lymph nodes were not palpable in the bilateral axillary and clavicular fossa.

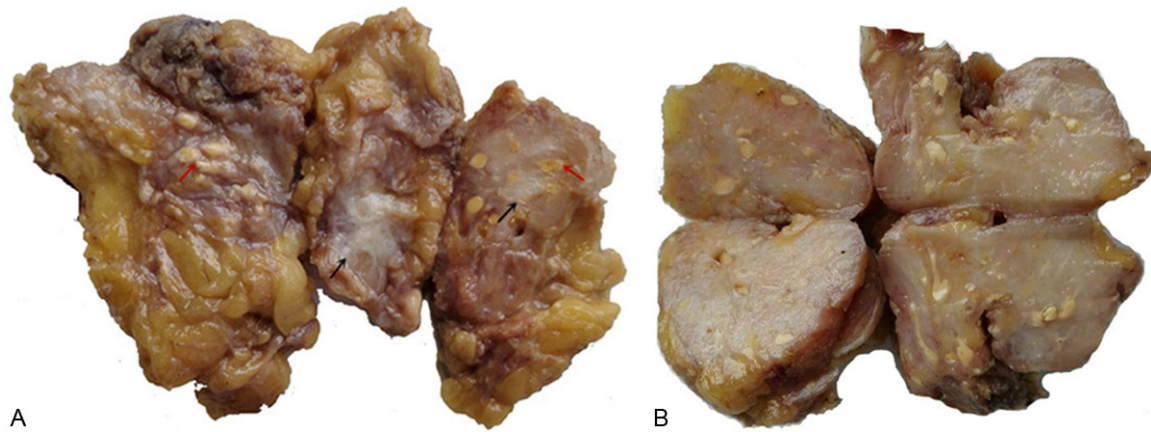
Ultrasonography revealed ill-defined masses in both breast. The right breast mass is solid with a non-homogenous, disturbance internal echo and surrounded by dilated mammary duct while the left breast mass with a clear boundary and surrounded by dilated mammary duct.

Lumpectomies were performed, and the histopathological diagnose of right breast adenoid cystic carcinoma combined with bilateral mammary duct ectasia were reported.

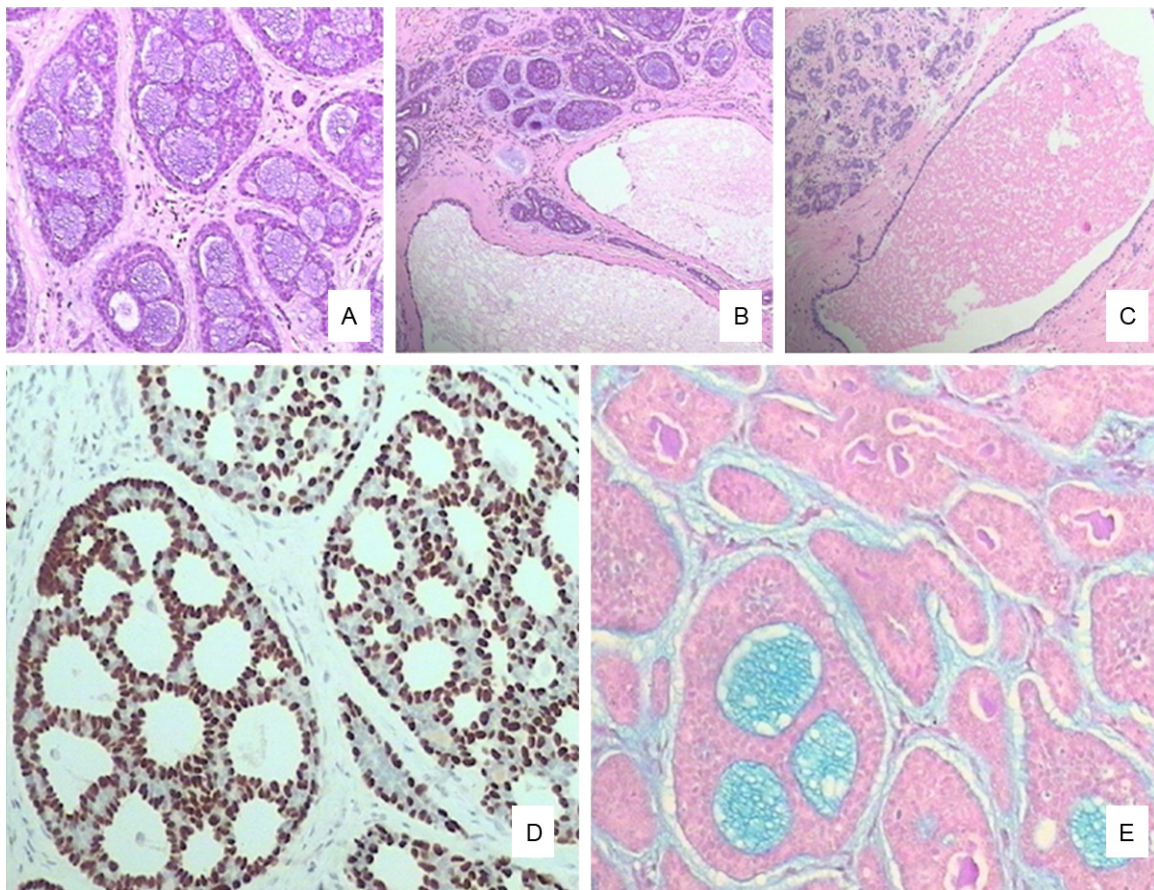
Macroscopically, the right breast mass displayed an ill-defined lesion with an irregular and satellite growth pattern in the central area, which was surrounded by dilated duct flushed with grey-white and grey-yellow pastes (**Figure 1A**). The left breast mass was characterized by dilated duct with ooze, as was the right (**Figure 1B**).

Histologically, ACC of the breast is similar to that of their salivary gland, lung and skin counterparts and is formed by epithelial and myoepithelial cell types arranged into classic tubular or cribriform architecture (**Figure 2A**). In the right breast, the dilated mammary duct coexist with

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**Figure 1.** A. Scar shaped neoplasam (black arrow) surrounded by dilated mammary duct filled with grey-white and grey-yellow pastes (red arrow) in the right breast. B. Dilated duct with ooze creamy in the left breast.



**Figure 2.** A. Epithelial and myoepithelial cells arranged into classic tubular or cribriform architecture. H&E staining, Original magnification,  $\times 200$ . B. Breast ACC combined with mammary ductal ectasia. H&E staining, Original magnification,  $\times 40$ . C. Dilated mammary duct adjacent to lobules of mammary gland. H&E staining, Original magnification,  $\times 40$ . D. Neoplastic myoepithelial cells express P63. Immunostaining staining, magnification,  $\times 200$ . E. The true glandular spaces and pseudolumina which were formed by neoplastic cells stained PAS (eosinophil) and AB (blue) separately. AB-PAS staining, Original magnification,  $\times 200$ .

the ACC cells; in the left breast, the extremely dilated mammary duct, full of eosinophilic

material, is adjacent to lobules of mammary gland (**Figure 2B, 2C**). The neoplastic cells were

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polarized around two types of structures: true glandular spaces and pseudolumina. The true glandular spaces were small, difficult to see, which contained neutral mucin that stained for Periodic Acid-Schiff (PAS) while the pseudolumina seemed large and round shaped, were positive staining for Alcian blue (AB) (**Figure 2D**).

Immunohistochemically, the neoplastic luminal cells express Keratin 7, Keratin 5/6 and CD117 while the neoplastic myoepithelial cells express smooth muscle actin, P63 (**Figure 2E**) and calponin. Approximately 5% of the cells express estrogen receptor (ER) and nearly 1% of the cells express progesterone receptor (PR). None of the cell expresses human epidermal growth factor receptor 2 (HER-2). Ki-67 is positive in about 20% of the cells.

The patient experienced modified radical mastectomy with right side axillary lymph node dissection, which was later, proved to be free of metastasis. Two weeks after surgery, she received four courses of chemotherapy (CEF) and remained in a good condition throughout the 14-month follow-up period.

### Discussion

ACC of the breast is a very rare breast cancer variant, accounting for less than 0.1% of all breast carcinomas. Predominantly, it affects women at an average age of 64 years although some authors report it may occur at a very young age, even juvenile young male [6-8]. According to literature, it mainly occurs unilaterally without predilection for laterality and about 50% of cases are found in the subareolar region [9]. The present case depicting ACC occurred in the upper-outer quadrant of the right breast accompanied by bilateral mammary duct ectasia.

So far, there are several reports suggesting an association between breast ACC and various breast benign lesions including microglandular adenosis, tubular adenosis, adenomyoepithelioma, and fibroadenoma. Here, we present a breast ACC combined with bilateral mammary duct ectasia that makes it the first case to report referring breast ACC and mammary duct ectasia. Whether breast ACC is an independent lesion that coexists with mammary duct ectasia

or a secondary lesion to mammary duct ectasia is currently not clear.

Spontaneous pain is one of the typical clinical feature in some patients due to perineural invasion that is similar to ACC of the salivary [10]. Most of the patient did not have such a symptom, but some reported their patients have experienced slight to severe pain in palpable mass within breast [11-14]. In this case, the patient complained of pain.

Nipple discharge is a not-infrequent complaint among women and is often the first indication of underlying breast pathology [15]. A variety of diseases such as intraductal papillomas, mammary duct ectasia, breast cancer, pituitary adenomas, breast abscesses can manifest as nipple discharge. About 40% of the cases are associated with benign breast lesion, among them, 25% are secondary to mammary duct ectasia. Only about 1-5% of all breast cancers are presented as nipple discharge [16]. Bloody secretion is usually related to breast carcinoma. Nevertheless, what makes this case rarity is that the female patient has had sero-sanguineous discharge for almost 5 years.

Most breast ACC are grouped as "triple negative breast carcinoma" or "basal-like breast carcinoma" by expressing CK5/6 and HER-2, but not expressing ER and PR [17-19]. ACC is of special interest because of its distinctive histological appearance and favorable prognosis, as lymph node involvement and distant metastases are very rare [20, 21]. In this case, what makes it uncommon is that about 5% of neoplastic cells stained for ER and 1% stained for PR that is consistent with a few reports [22, 23].

Surgery is the main stay of treatment of breast ACC, besides it, radiotherapy, adjuvant therapy and hormonal therapy are also beneficial. It was recommended that radiotherapy should be performed following lumpectomy irrespective of surgical margins [24]. However, some authors argued that the role of adjuvant therapy and hormonal therapy is controversial since they do not increase the survival rates of patients. In the present case, Tamoxifen and 4 courses of chemotherapy were administered to the patient following modified radical mastectomy given the patient's positive endocrine receptor and young age.



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In conclusion, breast ACC is a rarely observed neoplasm, when accompanied by bilateral mammary duct ectasia is even more unusual. Whether it arises from mammary duct ectasia or only coexists with mammary duct ectasia is unknown. More data should be collected in order to observe the association between breast ACC and mammary duct ectasia.

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### Disclosure of conflict of interest

None.

**Address correspondence to:** Dr. Yong Hu, Department of Pathology, The NO. 105th Hospital of PLA, Hefei, Anhui, PR of China. Tel: 86 0551 6596 6253; E-mail: huyong105@126.com

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