Discordant correlation of breast adenoid cystic carcinoma on imaging and pathology: A case report and literature review on surgical management

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ABSTRACT

INTRODUCTION: Adenoid cystic carcinomas (ACC) of the breast are extremely rare tumours, accounting for <0.1% of newly diagnosed breast cancer cases. Little data exist regarding the correlation of radiological findings with histology of this rare subtype. To our knowledge, gross size discrepancy between the 2 modalities has not been reported. We describe a case of ACC with appreciable size discordance between imaging and pathology report.

PRESENTATION OF CASE: A 71 years old lady presented with a painless right breast lump of a few months duration. Clinical examination revealed a 1.5 cm right breast upper quadrant mass. Axillary and systemic examinations were unremarkable.

Mammogram showed an asymmetric density in the right upper outer quadrant which corresponded to a suspicious nodule measuring about 2 cm on the ultrasound. Ultrasound of the axilla showed an indeterminate right lymph node. Core needle biopsy of the right breast nodule showed ACC while the lymph node biopsy was non-metastastic. Staging scans did not reveal any definite distant metastasis. Her naso-endoscopy and MRI of the neck were normal.

She underwent a right mastectomy and sentinel lymph node biopsy. Final histology returned as grade II 55 mm ACC. Lympho-vascular invasion was absent. The tumour was triple negative for Estrogen receptor (ER), Progesterone receptor (PR) and Human epidermal receptor 2 (HER 2). Sentinel lymph node biopsy was negative for metastasis.

She recovered well but declined adjuvant chemotherapy and radiation therapy. She is currently well 6 months post operation.

DISCUSSION: ACC is an extremely rare subtype, therefore there are limited reports in literature on its imaging and pathological characteristics. Of this sparse data, there was no mention that there might be a big size discrepancy between the 2 modalities.

This appreciable discrepancy has implications for pre-operative planning and the choice of breast surgery. It will be useful if the pathological extent of ACC could be determined more accurately radiologically.

However, there are no distinctive imaging characteristics for ACC. ACC can appear as a smooth round mass similar to that of a benign mass or as an irregular mass on mammogram. On ultrasound, ACC often manifested as a hypo-echoic heterogeneous mass with minimal vascularity on Doppler imaging and may have an indistinct margin. MRI has a higher sensitivity than mammogram and ultrasound in determining the true extent of the tumour, but there remains little data on its usefulness in ACC.

CONCLUSION: ACC can be extensively infiltrative and present much larger than its radiological size, as reported in our case. Use of better imaging modalities judiciously, in these cases, are needed to more accurately predict the true pathological size of ACC to prevent inadequate surgery.

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1. Introduction

Adenoid cystic carcinomas (ACC) of the breast are extremely rare tumours, accounting for <0.1% of newly diagnosed breast cancer cases [1,2]. There is therefore very little data regarding imaging characteristics of ACC and the correlation of the final pathological and imaging size of ACC. There has been no reported concerns of gross size discrepancy between imaging and histological size of ACC.

We describe a case of ACC with appreciable size discordance between imaging and pathology report. This case report was reported in accordance with the SCARE criteria [3].

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2. Presentation of case

A 71 years old lady presented to KK Women and Children Hos- pital Breast Clinic with a painless right breast lump of a few months duration. She has no known past medical history and was not on any regular medications. There was no reported family history of breast cancer. Clinical examination revealed a 1.5 cm right breast upper outer quadrant mass. Axillary and systemic examinations were unremarkable.

Mammogram showed an asymmetric density in the right upper outer quadrant (Fig. 1), which corresponded to a suspicious nodule measuring about 2 cm on the ultrasound (Fig. 2). Ultrasound of the axilla showed an indeterminate right lymph node. Core needle biopsy of the right breast nodule showed ACC while the lymph node biopsy was non-metastatic. Staging scans did not reveal any definite distant metastasis. Her naso-endoscopy and magnetic resonance imaging (MRI) of the neck were normal.

Surgical options of breast conservation or mastectomy with reconstruction were offered. She opted for a right mastectomy with sentinel lymph node biopsy. She was not keen for breast conservation as she was keen to avoid radiotherapy. The operation was done one month from presentation and clinical examination on the day of surgery did not reveal any appreciable change in tumour size. The operation was uneventful. Histology showed an extensively infiltrative tumour characterized by solid and cribriform nests, trabeculae of basoloid tumour cells intimately admixed with hyalinised and myxoid stroma (Fig. 3). Cribriform nests contained basophilic myxoid material with focal calcification and eosinophilic basement membrane material (Fig. 4). Immunohistochemistry revealed a dual population of intimately admixed luminal epithelial cells (CK7+, CD117+) and myoepithelial cells (p63+). The morphological features were those of an ACC. No ductal carcinoma in situ (DCIS) was seen. The tumour was grade II and measured 55 mm.

Lympho-vascular invasion was absent. The tumour was triple negative for estrogen receptor (ER), progesterone receptor (PR) and human epidermal receptor 2 (HER 2). Sentinel lymph node biopsy was negative for metastasis. She recovered well but declined adjuvant chemotherapy and radiation therapy. She is currently 12 months post operation and is well. She will be on lifelong follow up with the breast surgeon.

3. Discussion

ACC is an extremely rare subtype, therefore there are limited reports in literature on its imaging and pathological characteristics. From this sparse data, there is no mention that there might be a big size discrepancy between the 2 modalities. However, microscopic extent of ACC had been described to extend beyond a macroscopic well defined border in 50–60% of cases [4]. We report a case of ACC with gross size discrepancy between the radiological and pathological measurement, emphasizing that the radiologic findings could underestimate the true extent of this rare subtype, especially if it was of an extensively infiltrative tumour as seen in our case.

There are no distinctive imaging characteristics for ACC. ACC may appear as a smooth round mass similar to that of a benign mass or as an irregular mass on mammogram [5], like in our patient. Micro-calculcations are rarely seen on mammogram [6]. On ultrasound, ACC often manifests as a hypo-echoic heterogeneous mass with minimal vascularity on Doppler imaging and may have an indistinct margin [7,8], similar to our case. In our case, the tumour measured 20 mm on imaging, much smaller than its final histological size of 55 mm. This appreciable discrepancy has implications for pre-operative planning and the choice of breast surgery. It would be useful if the pathological extent of ACC could be determined more accurately radiologically. She was fortunate to have chosen a mastectomy as her margins may have been involved if a wide excision was done based on an expected 20 mm tumour size.

MRI has higher sensitivity [8] than mammogram and ultrasound in determining the true extent of the tumour, but there is little data on its usefulness in ACC. On MRI, ACC can present as a mixed solid-cystic mass with indistinct margin associated with internal septations on T2 weighted image, that exhibits delayed enhancement on MRI [7]. However, there is currently inadequate data to support the routine use of MRI for pre-operative planning. The application of MRI in this rare subtype needs to be further defined.
Pathologically, ACC had been associated with distinctive features such as having prominent perineural invasion [9], absence of lympho-vascular invasion [4,9], triple negative in nature, rare nodal and distant metastasis [10,11], as seen in our patient. Despite its triple negative status, ACC tends to have a favourable outcome [12] with an excellent prognosis even when it recurs. The recurrences tend to be late, in contrast to early recurrences in the other triple negative tumours [13]. Involved surgical margins were associated with a higher risk of recurrence [14].

Surgically, both options of mastectomy or lumpectomy with radiotherapy had been associated with equivalent survival in ACC [12]. In view of the large size discrepancy between imaging and pathology noted in our case, a review of the re-excision rates of
ACC following breast conservation was done. However, this was not well documented as there were limited reports on re-excision rates. Hodgson et al., [14] reported that in patients with ACC undergoing breast conservation surgery, positive margins can occur in at least 42% of patients, requiring further operations. They however, did not specifically correlate between the imaging and pathological size to account for the high positive margin rates.

While radiological and pathological size discrepancy could be common in tumours, with some histological subtypes such as lobular invasive carcinoma (ILC) more prone to it, this is the first reported case of ACC, which shows an appreciable size discrepancy between the 2 modalities. This highlights the need for better imaging modalities to determine the true extent of ACC.

Radiotherapy decreases local recurrence for patients who had undergone breast conservation surgery [13,16] and a recent study also demonstrated the beneficial effects of radiotherapy on overall survival and disease free survival. Though it was recommended at our breast tumour board for our patient to undergo radiation and chemotherapy she was not keen. There is very few published data on the benefits of chemotherapy but systemic adjuvant chemotherapy is proposed for patients with tumours bigger than 3 cm and with axillary lymph node metastasis [17].

4. Conclusion

In conclusion, ACC can be extensively infiltrative and be much larger than its measured radiological size, as reported in our case. Use of better imaging modalities judiciously, in these cases, is needed to more accurately predict the true pathological size of ACC to prevent inadequate surgery.

Conflicts of interest

There are no conflicts of interest.

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Ethical approval

Ethical approval is exempted as this is a case report and not a research study.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

All authors contributed to the writing of the paper.

Registration of research studies

Not applicable.

Guarantor

Guarantor is Dr Yan Zhiyan.

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