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CASE REPORT

A Rare Case Of Adenoid Cystic Breast Carcinoma In A 19-Year-Old Girl

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ABSTRACT

A 19-year-old-girl presented to us with a lump in the right breast. The FNAC of the lump was reported as a benign disorder. Wide local excision was performed and histopathological examination of the specimen revealed an adenoid cystic carcinoma of the breast. Subsequently, she underwent axillary nodal dissection which was found to be free of metastases. As adenoid cystic carcinoma breast has a favourable prognosis compared to other histological types, the specific characteristics and diagnostic criteria are to be adhered to, for early and accurate detection and subsequent management.

Key Words: Adenoid cystic carcinoma, perineural invasion, breast

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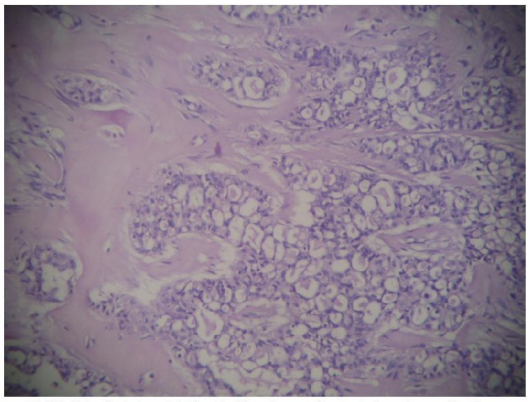
Introduction

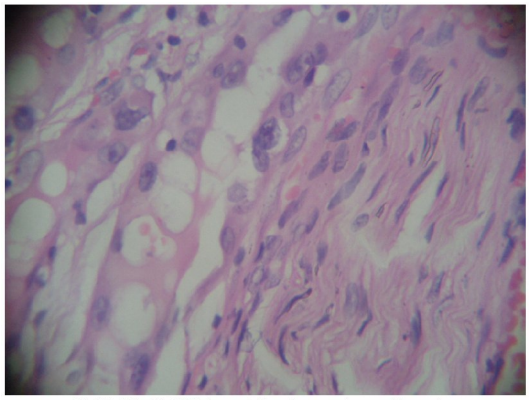
Adenoid cystic carcinoma of the breast is a rare neoplasm. It has a biological course of slow progression and a near absence of Iymph node metastasis. In contrast to extramammary adenoid cystic carcinoma, the prognosis of adenoid cystic carcinoma of the breast is excellent. Adenoid cystic carcinoma can be confused with other more common breast cancers such as intaductal carcinoma with a cribriform pattern. It is important to distinguish this cancer from other forms of breast cancer because of its excellent progrosis.

A 19-year old girl presented to the outpatient clinic in June 2008, with a six year old lump in the right breast. On examination, a firm nodule suggestive of fibroadenoma, was palpable in the central quadrant of the right breast. The FNAC of the lump (done elsewhere) showed benign epithelial hyperplasia, with a mild degree of nuclear atypia. Wide local excision was performed. A segment of breast tissue measuring 8 X 5 X 3 cm was removed and the cut surface showed a well circumscribed yellowish white nodule measuring 3 X 3 cm, with areas of haemorrhage. The specimen was fixed in formalin and processed routinely. Histological examination showed a circumscribed tumour with cribriform groups of epithelial cells [Table/Fig 1], separated by fibrocollagenous tissue. These groups were seen to surround the mammary ducts. Tumour nests showed a cribriform pattern with amorphous eosinophilic secretions that were focally PAS-positive. The cells showed mild atypia with vesicular nuclei and sparse mitosis. Perineural invasion [Table/Fig 2] was also noted. Immunostaining was negative for the oestrogen and progesterone receptors. Adenoid cystic carcinoma (ACC) of the breast of the

cribriform subtype and low grade was diagnosed. The surgical margins were free of tumour. Axillary dissection was performed, which was found to be free of nodal metastases. Staging investigations including chest X-ray and abdominal and pelvic ultrasonography were performed and showed no evidence of metastatic disease. The patient made good recovery after surgery. The patient received no adjuvant therapy and is well till date.



(Table/Fig 1) Adenoid Cystic Carcinoma breast showing cribriform pattern with eosinophilic secretions



(Table/Fig 2)ACC breast showing perineural invasion

Discussion

Adenoid cystic carcinoma of the breast is a rare neoplasm, accounting for less than 0.1% of all breast carcinomas. Nevertheless, its early recognition is mandatory as it has a favourable prognosis [1],[2].

ACCs of the breast occur predominantly in women and at a mean age of 50-60 years [3].

There has been only a single case reported previously of this type of tumour in a patient as young as 19-years of age [4].

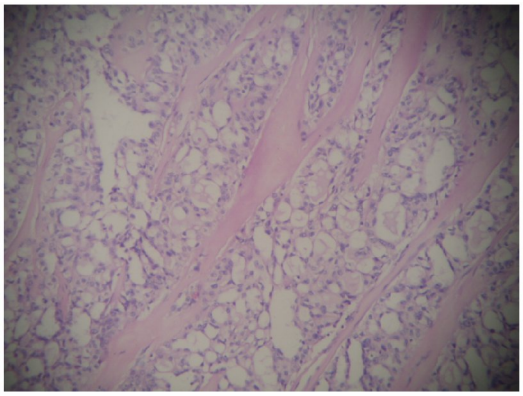
The clinical features were, a well-circumscribed palpable mass, which was occasionally tender on palpation [2]. The tumour is rarely fixed to the overlying skin, nipple, or pectoral muscle. Although any part of the breast may be involved, most ACCs are centrally located.

On mammography, these tumours often appear as moderately circumscribed, lobulated nodules [3]. The histological appearance of ACCs in the breast is similar to that of ACCs of the skin adnexa and of the salivary glands, but most authors agree that ACCs of the breast have a much better prognosis than ACCs of the salivary glands [1],[2].

The most striking histological feature of this neoplasm is the presence, at least focally, of cribriform nests of cells. These nests are typically composed of two cell types: small basaloid myoepithelial cells and ductal epithelial cells. The cribriform gland-like spaces are filled with homogenous amorphous eosinophilic PAS-positive material or granular basophilic material [5]. A variety of microscopic growth patterns can be found in ACCs. These configurations have been described as cribriform, solid, glandular (tubular), reticular (trabecular) and basaloid. ACCs of the breast are usually receptor negative, the prognostic significance of this feature remaining uncertain. Immunohistochemical stains are positive for S-100 protein and actin in the basaloid cells and positive for cytokeratin in the ductal epithelial cells. A previous study [2] suggested that histological grading of the breast, as

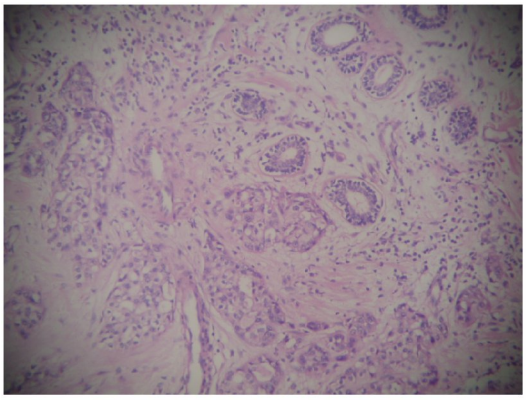
described for salivary gland ACCs, has some prognostic value, but this has not been confirmed by two other studies [6],[7]. The origin of the tumour is unknown, with evidence of both ductal epithelial and myoepithelial cell derivation.

Differential diagnoses include cribriform intraductal carcinoma and invasive ductal carcinoma with a cribriform pattern, and benign collagenous spherulosis. If a fine needle aspiration is performed, the differential diagnosis between ACCs and collagenous spherulosis is rather difficult. ACCs show small aggregates with occasional larger sheets, in which a cribriform appearance can sometimes be recognized. Sparse epithelial tubules are seen, and myoepithelial cells are present in most of the tumour cells. The diagnostic feature is the finding of eosinophilic hyaline globules [Table/Fig 3] of basement membranes surrounded by tumour cells. These cytological findings can also be seen in collagenous spherulosis, although the cells of ACCs usually show moderate nuclear enlargement and hyperchromatasia, while in collagenous spherulosis the cells are more regular. The spherules in collagenous spherulosis contain elastin, periodic acid-Schiff(PAS)-positive material, and type IV collagen. Collagenous spherulosis occurs most often in ducts involved by hyperplasia, but it can also develop in papillomas and sclerosing adenosis. It has been reported that calponin and CD117 could also be helpful in the differential diagnosis of collagenous spherulosis from adenoid cystic carcinoma. Calponin was present in the myoepithelial cells of collagenous spherulosis, but not in adenoid cystic carcinoma. Moreover, expression of CD117 was seen in the epithelial cells of adenoid cystic carcinoma, but not in simple collagenous spherulosis [8].



(Table/Fig 3) Hyalin Bands in ACC breast

Despite well-defined gross borders, more than half of these tumours show microscopic infiltration of the adjacent adipose tissue and breast parenchyma, [Table/Fig 4] making complete tumour resection difficult [5].



(Table/Fig 4) ACC cells infiltrating into a lobule

Guidelines for treatment of this neoplasm are not well-established in the literature. It's rarity makes the comparison of treatment options difficult. Recurrence in the breast has been described after treatment by local excision alone. Therefore, these types of tumours are best managed by wide excision or quadrantectomy. A low axillary dissection or sentinel lymph node mapping should be performed if the clinical examination suggests lymph node involvement, if the breast contains another type of invasive carcinoma, for high-grade adenoid cystic lesions, and for tumours larger than 3 cm. Systemic adjuvant chemotherapy is recommended for patients with axillary

lymph metastases and may be considered in patients with high-grade lesions, or if the tumour is larger than 3 cm.
In conclusion, other more common forms of breast cancer such as intraductal carcinoma may mimic the appearance of adenoid cystic carcinoma. Adenoid cystic carcinoma of the breast has a favourable prognosis.

Therefore, the accurate diagnosis and treatment is clinically important. Although it is rare, local recurrence and distant metastases may occur long after initial treatment and close follow-up is mandatory .

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